Mothers of children with intellectual disability or autism spectrum disorder: pre-existing differences, health and quality of life

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The health of our mothers underpins the well-being of our children, our society and ultimately the world.
ABSTRACT

Introduction

People with intellectual disability (ID) have an IQ of less than 70 and deficits in adaptive functioning. Autism spectrum disorder (ASD) is a debilitating neuro-developmental condition with severe deficits in sociability and behaviour. Researchers consistently report poorer health and quality of life (QoL) in the mothers of children with ID or ASD. However these mothers may have different socio-demographic and health profiles from other mothers prior to their child’s birth. While mothers of minority race-ethnicities have been reported to be less likely to have a child with ASD than Caucasian mothers, there may be a complex inter-relationship between maternal race-ethnicity, immigration, region of birth and the risk of having a child with ASD compared to mothers with no ID or ASD. Mothers with a child with ASD have been reported to have more mental health problems both before and after the birth. I wanted to investigate how much of the psychiatric ill health in mothers of children with ID or ASD was pre-existing, possibly due to genetic factors, and how much was associated with the burden of caring.

Linkable data available in Western Australia (WA) would enable me to examine maternal mental health before and after the birth of a child according to the categories of ID or ASD. I could access de-identified records of births including socio-demographic characteristics, hospitalisations, deaths, and information on disability status. By interviewing mothers of children with ID or ASD, I could relate my research to the context of living with a child with ID or ASD. Therefore, my aims were to:

1. Explore pre-existing differences between mothers of children with ID or ASD compared to other mothers
2. Explore the health of mothers of children with ID or ASD compared to other mothers
3. Explore the health of mothers of children with a sub-type of ID or ASD compared to that of mothers of children with a different sub-type of ID or ASD
4. Interview mothers of a child with ID or ASD regarding their QoL.
Methods

I linked data from state-wide registries of women with a live-born child in WA between 1983 and 2005 inclusive. A mother was assigned to a case or comparator group according to the status of her index child. In case mothers, the index child was the eldest child with ID or ASD born during the study period. Case children were grouped according to the following categories: *Mild or moderate ID of unknown cause, Severe or profound ID of unknown cause, Down syndrome, Other ID of known cause, ASD with ID and ASD without ID.* In the comparator group, mothers who had no child with ID or ASD the index child was the eldest child born during the study period. I analysed data using multinomial logistic, Cox or negative binomial regression. The qualitative study was conducted by personal interview with 16 mothers.

Results

Mothers of children with ASD were less likely to be immigrant or of minority race (particularly Indigenous). Black women from East Africa had a higher prevalence of ASD with ID in their children. Women with psychiatric disorders were twice as likely to have a subsequent child with ID or ASD. After the birth, mothers of children with ID or ASD had a greater risk of death than other mothers. Apart from mothers of children with Down syndrome, these mothers had an increased risk of psychiatric disorders after the birth. Qualitative data suggested that caring for a child with ASD and comorbid ID reduced maternal QoL and exacerbated any mild autistic traits.

Conclusion

Mothers of children with ID or ASD had poorer mental health before the birth of their child and poorer mental health, earlier mortality and a lower QoL after the birth than mothers of children without these disabilities. Their increased risk of a prior psychiatric disorder suggests that their poorer mental health could be related to genetic factors. Their increased incidence of psychiatric disorders after the birth and their earlier mortality suggest that their poorer health is also likely to be related to the burden of caring for their children. Further research is needed to replicate these findings in other countries with linked data capabilities.
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DECLARATION FOR THESES CONTAINING PUBLISHED WORK AND/OR WORK PREPARED FOR PUBLICATION

This thesis contains published work and work prepared for publication, all of which has been co-authored. The bibliographical details of the work and where it appears in the thesis are outlined below.

Publication 1 (Chapter 2)

Ms Fairthorne planned and conducted the review and wrote the initial draft. Dr Leonard and Dr de Klerk provided advice and reviewed and revised the initial manuscript.

Publication 2 (Chapter 4)

Ms Fairthorne planned and conducted the review and wrote the initial draft. Dr Langridge, Ms Bourke and Dr Leonard provided advice and reviewed and revised the initial and subsequent manuscripts.

Publication 3 (Chapter 5)
Fairthorne J, de Klerk N, Schieve L, Leonard H, Yeargin-Allsopp M. Race-ethnicity, immigrant status, birth region and the odds of a child with ASD. Submitted to co-authors prior to submission to the Office of the Director of the Centers for Disease Control and Prevention for clearance to submit to a journal.
Ms Fairthorne conceived the study, performed the analyses and wrote the initial draft. Dr Yeargin-Allsopp provided initial advice and reviewed and revised the initial manuscript. Dr de Klerk provided statistical advice and all co-authors assisted with subsequent manuscripts.

**Publication 4 (Chapter 5)**

Ms Fairthorne and Dr Hammond conceived the study and Dr de Klerk provided modifications. Ms Fairthorne performed all analyses under the guidance of Dr Hammond and Dr de Klerk who also advised on the statistical methods. Ms Fairthorne wrote the initial draft and all co-authors provided input into subsequent drafts and approved the final version.

**Publication 5 (Chapter 5)**

This paper is tangential to the study resulting in Publication 9. It was conceived by Ms Fairthorne and she was guided by Dr Whitehouse and Dr Fisher in the writing of the manuscript. All authors approved the final manuscript.

**Publication 6 (Chapter 6)**

**Publication 7 (Chapter 7)**
Publication 8 (Chapter 7)

Publications 7 and 8
Ms Fairthorne conceived each of the studies and Dr Jacoby provided modifications. Ms Fairthorne performed all analyses and Dr de Klerk and Dr Jacoby advised on the statistical methods. Ms Fairthorne wrote the initial draft and all co-authors provided input into subsequent drafts and approved the final version.

Publication 9 (Chapter 8)

Ms Fairthorne conceived the study and recruited and interviewed the study participants under the guidance of Dr Fisher. Ms Fairthorne transcribed all interviews and wrote the original draft. All co-authors assisted with subsequent drafts and approved the final submission.

Overall statement of candidate’s contribution
Ms Fairthorne planned the research reported in this thesis, undertook the research and wrote the thesis. We confirm that permission has been obtained from all co-authors to include these manuscripts in this PhD Thesis.

Signature of student

Signature of co-ordinating supervisor
OTHER PUBLICATIONS ASSOCIATED WITH THIS THESIS

Publication 10 (Chapter 5, 6 and 7)

Ms Fairthorne wrote a section of this chapter based on her experiences using population-based linked data to explore characteristics of the mothers of children with ID and ASD.

Publication 11 (Chapter 8)

Ms Fairthorne wrote this short article which was then approved for submission by Dr Fisher.
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<tr>
<td>ABA</td>
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<td>ADHD</td>
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<td>BAP</td>
<td>Broad Autism Phenotype</td>
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Chapter 1: Introduction

First, I detail the mode of this thesis, iterate the aims and describe how the research within was developed in relation to these aims. Second, I provide the reader with background information to underpin the area of my research. Within this context, I describe the effect of intellectual disability (ID) and autism spectrum disorder (ASD) at the individual level, followed by descriptions of the diagnosis, sub-groups and prevalence of each disorder.

1.1 This thesis

This thesis presents my research both as a story of discovery and as a series of papers. In order to achieve this, I have written the majority of chapters as a modified version of one or more of the published or submitted papers. The first page of published papers and the title page and abstracts of manuscripts have been included as appendices.

1.2 Intellectual disability, autism spectrum disorder

Persons with ID have an IQ of less than 70 along with deficits in adaptive functioning which have been present prior to the age of 18 years. The deficits associated with the debilitating neuro-developmental condition, autism spectrum disorder (ASD) are present from early childhood and severely impact functioning in the areas of sociability and behaviour. The effect of these disorders on a person’s ability to function productively and independently within their family and society varies according to the severity of the individual’s condition.

1.3 Family and societal impact of ID and ASD

The burden associated with the care of a child with ID or ASD may be considerable and, in turn, this may affect maternal health and family quality of life. Poorer health in the mothers of children with ID or ASD has been correlated with child and maternal characteristics such as challenging behaviours, reduced family support and lower socioeconomic status (SES). There is a high societal cost...
associated with ID and ASD and this can be measured across educational, medical, residential and other sectors of our society. (8, 9)

1.4 Aims

My three overarching aims and fourth primary aim were to:

1. Explore any pre-existing differences between mothers of children with ID or ASD and mothers of typically developing children
2. Explore the health of mothers of children with ID or ASD compared to the health of mothers of typically developing children
3. Explore the health of mothers of children with a sub-type of ID or ASD compared to the health of mothers of children with a different sub-type of ID or ASD
4. Interview mothers of a child with ID or ASD regarding factors they perceived as affecting their QoL.

1.5 Existing research

In order to design the research to best achieve my overarching aims, I conducted a critical review of the literature that explored the health of mothers of children with ID or ASD compared to the health of mothers of typically developing children. This review comprises Chapter 2. Here, I review the literature that compared both the health of mothers of children with ID or ASD to that of mothers with typically developing children and that which compared the health of mothers of children with ID or ASD according to the diagnostic sub-group of their child. Finally, I examine research that explored the correlates of poorer health in these mothers. In this way, I came to some understanding of the reasons for poorer health in the mothers of children with ID or ASD. The results of this review enabled me to make an informed choice regarding the research that would best explore the health of mothers of children with ID or ASD. This process is described in Chapter 3 and I present my research in Chapters 4–8.

1.6 Sub-groups

The classification of the level of severity of ID has traditionally been based on the Intelligence Quotient (IQ) and generally categorised as mild or moderate (mild–
moderate) in persons with IQ between 35–40 and 69 and severe or profound (severe) in persons with IQ less than 35–40. In more recent years, for the provision of services, adaptive behaviour scores have been used for assessing levels of support or funding needed. In terms of aetiology, ID can be broadly divided into cases of known biomedical causes and cases of unknown cause. Approximately 80% of all ID has no known biomedical cause and cases of mild–moderate ID are more likely to be of unknown cause than severe ID (90%). Known or biomedical causes of ID are either genetic or non-genetic. Within the genetic group, Down syndrome is the most common, comprising around 40%, whereas other genetic causes include Fragile X and Rett syndromes. Non-genetic causes include fetal alcohol syndrome (FAS), infections and adverse events such as head injury. Figure 1 shows the genetic and non-genetic causes of ID and their inter-relationships.

In most cases, the aetiology of ASD is unknown. However, research has implicated a strong genetic basis, possibly involving a gene–environment interaction. On occasions, researchers have grouped those with ASD according to the presence of ID. Those with ASD and comorbid ID are within the group, ASD with ID and those with ASD without comorbid ID are within the group, ASD without ID. From 30 to 60% of persons with ASD have comorbid ID. In my research, I have included people with ASD and comorbid ID in the ASD and not the ID group.

1.7 Diagnosis

Currently, clinicians in WA diagnose children with ID using the three criteria provided by the American Association on Intellectual and Developmental Disability. In some cases a decision may be made on the basis of the presenting clinical diagnosis being consistent with ID, such as Down syndrome. On the other hand, for an ASD, clinicians use the criteria provided by DSM-4 (Diagnostic and Statistical Manual of Mental Disorders, fourth edition) and impairments are required within the three strands of Reciprocal social interaction, Communication, and Repetitive behaviours or interests.

The age of diagnosis for ID varies considerably. For example, Down syndrome and some other conditions associated with ID can be diagnosed before birth, whereas persons with mild–moderate ID may not be diagnosed until they are of school age.
Further, ID caused by infection or trauma will be diagnosed after the event. The age of diagnosis for ASD is reported to be decreasing. (29) Studies published in 2011(30, 31) reported that the median age of diagnosis was around four and a half years. As expected, children with less severe symptoms tend to be diagnosed later. (29, 32)

Figure 1: Aetiology of intellectual disability

ID of all causes

- Known causes
  - Genetic causes
    - Down syndrome
    - Fragile X syndrome
    - Rett syndrome
  - Other genetic causes
- Unknown causes
  - Non-genetic causes
  - Fetal alcohol syndrome
  - Other teratogenic causes
  - Infections
    - Meningitis
    - Other
  - Other adverse events such as head injury

ID, Intellectual disability

1.8 Prevalence

The range of prevalence estimates of ID and ASD is wide and likely to be influenced by the diagnostic criteria, ascertainment methods, exclusion criteria, the age of the study population and the geographical region. (33) For example, with ID, studies that use a child or adolescent population usually have a higher prevalence than those with an adult population. (33) If persons are identified through service providers, the estimated prevalence will be less than in those derived from a population study because some people with ID may never have received services. Moreover, the prevalence of ID in lower-income countries is usually higher than in wealthy countries, possibly because of poorer maternal health, (33) more frequent birth injury (33) and unidentified environmental determinants. (34) Further, with ASD, the diagnostic prevalence is increasing (35-36), which means that earlier studies are likely to have lower estimates.

Prevalence estimates of ID ranging from 37 to 142 per 10,000 births (Table 1). The meta-analysis of Maulik et al. (33) with a median rate of 104 per 10,000 births appears...
to be the best international estimate as it included high-, median- and low-income countries and, unlike other studies, did not exclude particular groups such as those with cerebral palsy.(34)

<table>
<thead>
<tr>
<th>Study, year</th>
<th>Criteria</th>
<th>Study size</th>
<th>Ages Birth cohort</th>
<th>Region</th>
<th>Prevalence estimate</th>
</tr>
</thead>
<tbody>
<tr>
<td>Kelly et al., 2011(40)</td>
<td>Mild–moderate and severe ID included</td>
<td>Not given (26,484 cases)</td>
<td>All ages</td>
<td>Ireland</td>
<td>36.9</td>
</tr>
<tr>
<td>Westerinen et al., 2014(41)</td>
<td>ICD-9, ICD-10 codes for ID &amp; codes associated with ID</td>
<td>5.18 million (36,053 cases)</td>
<td>All ages</td>
<td>Finland</td>
<td>69.6</td>
</tr>
<tr>
<td>Maulik et al., 2011(33)</td>
<td>ICD 8-10, DSM-3 &amp; 4, AAMR classification &amp; others</td>
<td>Meta-analysis, 1980–2009</td>
<td>All ages</td>
<td>International</td>
<td>104</td>
</tr>
<tr>
<td>Leonard et al., 2002(34)</td>
<td>IQ &lt; 70, information from registries</td>
<td>240,358 (3,426 cases)</td>
<td>6–16 years</td>
<td>Western Australia</td>
<td>142</td>
</tr>
<tr>
<td>Croen et al., 2001(35)</td>
<td>Enrolled with state disability service provider &amp; no CP/ASD</td>
<td>4,590,333 (27,547 cases)</td>
<td>5–22 years</td>
<td>California</td>
<td>52</td>
</tr>
</tbody>
</table>

ICD, International Classification of Diseases; AAMR, American Association on Mental Retardation; CP, cerebral palsy

Prevalence studies of ASD provide estimates of between 51 and 147 per 10,000 (Table 2). Due to the later age of diagnosis of children with ASD without ID,(29, 42) studies that include only younger children may miss diagnoses within this group. While the estimate of 147 per 10,000 by the US (United States) Centers for Disease Control and Prevention(25) is likely to be a best estimate for the US, it is an outlier compared to the others. For this reason, I considered that the best estimate of the diagnostic prevalence of ASD in other Western countries would be around 60 per 10,000, which is near the median of the other estimates.

Intellectual disability and ASD are each diagnosed according to behavioural criteria. Commonly, sub-groups of ID are formed according to the level of disability, whether the cause is known and whether it is a genetic condition. The division of ASD is most often according to the presence or absence of ID. The prevalence of ID is stable at around 104 per 10,000 births and the prevalence of ASD in 2008 was around 60 per 10,000.
Table 2: Prevalence estimates of ASD (per 10,000 births)

<table>
<thead>
<tr>
<th>Study, year</th>
<th>Criteria</th>
<th>Study size</th>
<th>Ages</th>
<th>Region</th>
<th>Prevalence</th>
</tr>
</thead>
<tbody>
<tr>
<td>Frieden et al., CDC, 2012(25)</td>
<td>DSM-4-TR</td>
<td>337,093 (3,820 cases)</td>
<td>• 8 years</td>
<td>US</td>
<td>147</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>• 2008</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Isaksen et al., 2012(43)</td>
<td>ADOS-G</td>
<td>31,015 (158 cases)</td>
<td>• 6–12 years</td>
<td>Norway</td>
<td>51</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>• 1996–2002</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Parner et al, 2011(44)</td>
<td>ICD-10</td>
<td>404,816 (20,022 cases)</td>
<td>• 5–10 years</td>
<td>Denmark</td>
<td>68.5</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>• 1994–1999</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Parner et al., 2011(44)</td>
<td>DSM-4</td>
<td>151,048 (678 cases)</td>
<td>• 5–10 years</td>
<td>Western Australia</td>
<td>51.0</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>• 1994–1999</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Fernell et al., 2010(45)</td>
<td>DSM-IV/DSM-4-TR &amp; ICD-10</td>
<td>23,566 (147 cases)</td>
<td>• 6–7 years</td>
<td>Stockholm, Sweden</td>
<td>52–72</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>• 2002</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Chakrabarti et al., 2005(46)</td>
<td>DSM-4</td>
<td>10,903 (64 cases)</td>
<td>• 4–6 years</td>
<td>Midlands, UK</td>
<td>58.7</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>• 1996–1998</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

CDC, Centers for Disease Control and Prevention; ICD-10, International Classification of Diseases-tenth revision; DSM-IV, Diagnostic and Statistical Manual of Mental Disorders, fourth edition; TR, Trial; ADOS-G, Generic Autism Diagnostic Observation Schedule; ASD, autism spectrum disorder

1.9 References


In this chapter, my aim is to organise and review the literature relating to health and its correlates in the mothers of children with intellectual disability (ID) or autism spectrum disorder (ASD). In 2.1, I review the current literature pertaining to the health and quality of life (QoL) of mothers of children with these disabilities and state the aims of my review. In 2.2, I describe the methods of the review and in 2.3, the results. 2.4 is a discussion of the research included in the review and in 2.5, I close with a discussion of the implications for further research. The abstract and authors of the paper arising from this chapter comprise Appendix 1.

2.1 Introduction

The deficits associated with ID and ASD may negatively impact maternal health (1.3). Further, correlates of poorer maternal health have been documented in these mothers (1.3). Within this review, I aimed to compare the health of mothers of children with sub-types of ID or ASD compared to the health of mothers of typically developing children. Further, I aimed to identify the stronger correlates of poorer health in these mothers. Hence, I have been able to identify sub-groups of vulnerable mothers and the risk factors for their poorer health. In combination, these results would provide valuable information to those planning services and supports for mothers with children from a particular sub-group(s) of ID or ASD.
2.2 Methods

I searched the Medline, Web of Knowledge, Scopus and Google scholar databases using combinations of the following search terms:

1. Terms associated with ID, ASD or diagnostic sub-groups of ID or ASD: 
   *intellectual disability, mental retardation, autis*, *pervasive development disorder*, *disab*, *Down, Asperger*
2. Terms associated with health: *health, depression, physical, mental, psych* *phenotype* 
3. Terms associated with carers: *mothers, parents, care*.

A paper was included in the review if it met all of the following criteria:

1. It was published in a peer-reviewed journal between 1 January 1990 and 30 June 2014 inclusive
2. It described original research (not a review) in English and was a full-length paper
3. The underlying research utilised a cohort, case–control, correlation or cross-sectional study
4. I considered that the methods of ascertainment and measurement of the characteristic(s) of interest were unlikely to lead to bias
5. The study population was >20
6. It compared the health of mothers, parents or carers of children with ID or ASD with that of mothers, parents or carers of children without disability or with a population norm
7. It compared the health of mothers, parents or carers of children with a sub-type of ID (such as Down syndrome) or ASD with that of mothers, parents or carers of children with another sub-type of ID or ASD.

2.3 Results

I retained 58 papers for the core review (Table 3) and acknowledge that searching with other search terms or combinations of terms may have provided a different basis for my review. Papers were sorted into the two groups of *Comparisons with the general*
population and Comparisons by the child’s disability. For a supplementary review, I used an additional 20 articles to provide maternal and child characteristics that were associated with poorer health or QoL in mothers (or other primary carers) of children with ID or ASD (Table 4). These articles enabled me to explain some of the intergroup health disparities that had emerged from the core review. In Tables 3 and 4, I have provided a summary of each paper, which includes a five-level measure of the quality of the evidence provided,(1) along with the methods of recruitment and data collection, the number of cases and controls in the study population and the disability groups of interest. An explanation of the assessment of the quality of the evidence is given in Table 5.
<table>
<thead>
<tr>
<th>Paper (in order of citing)</th>
<th>Strength of evidence</th>
<th>Data collection</th>
<th>Recruitment</th>
<th>Country</th>
<th>Study population</th>
<th>Case groups</th>
<th>Comparison group</th>
</tr>
</thead>
<tbody>
<tr>
<td>Eisenhowser et al., 2013, (2)</td>
<td>Medium</td>
<td>Single-item measure</td>
<td>Service providers</td>
<td>US</td>
<td>116 cases, 129 controls</td>
<td>DD</td>
<td>TD</td>
</tr>
<tr>
<td>Cantwell et al., 2014, (3)</td>
<td>High</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>45 cases, 45 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Gallagher et al., 2009, (4)</td>
<td>Medium</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>218 cases, 127 controls</td>
<td>DD</td>
<td>TD</td>
</tr>
<tr>
<td>Sullivan et al., 2012</td>
<td>Medium</td>
<td>Questionnaires</td>
<td>Service providers</td>
<td>US</td>
<td>45 cases, 45 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Cantwell et al., 2009, (5)</td>
<td>Medium</td>
<td>Single-item measure</td>
<td>Service providers</td>
<td>US</td>
<td>106 cases, 106 controls</td>
<td>DD</td>
<td>TD</td>
</tr>
<tr>
<td>Smith et al., 2012, (6)</td>
<td>Medium</td>
<td>Questionnaires</td>
<td>Service providers</td>
<td>US</td>
<td>106 cases, 106 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Magaña et al., 2006, (7)</td>
<td>Medium</td>
<td>Six-item survey</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Yamaki et al., 2009, (8)</td>
<td>Medium</td>
<td>Six-item survey</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Gallagher et al., 2013, (9)</td>
<td>Medium</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Emerson et al., 2003, (10)</td>
<td>High</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Morgan et al., 2012, (11)</td>
<td>High</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Daniels et al., 2008, (12)</td>
<td>Medium</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Sullivan et al., 2012, (13)</td>
<td>High</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Jokiranta et al., 2013, (14)</td>
<td>High</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Mouridsen et al., 2007, (15)</td>
<td>High</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Larsson et al., 2005, (16)</td>
<td>High</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Bolton et al., 1998, (17)</td>
<td>Medium</td>
<td>Questionnaires</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Caldwell et al., 2008, (18)</td>
<td>Medium</td>
<td>Questionnaires</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Gupta et al., 2007, (19)</td>
<td>Medium</td>
<td>Questionnaires</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Harvey et al., 1997, (20)</td>
<td>Medium</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Veisson et al., 1999, (21)</td>
<td>Medium</td>
<td>Questionnaires</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Blacher et al., 1997, (22)</td>
<td>Medium</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
<tr>
<td>Browne et al., 1998, (23)</td>
<td>Medium</td>
<td>Linked data</td>
<td>Service providers</td>
<td>US</td>
<td>100 cases, 100 controls</td>
<td>ASD, ADHD</td>
<td>TD</td>
</tr>
</tbody>
</table>

Table 3: P apers in the core review
<table>
<thead>
<tr>
<th>Paper (in order of citing)</th>
<th>Strength of evidence</th>
<th>Data collection</th>
<th>Recruitment</th>
<th>Country</th>
<th>Study population</th>
<th>Case groups</th>
<th>Comparison group</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bourke et al., 2008, (33)</td>
<td>Medium</td>
<td>SF-12</td>
<td>Service providers</td>
<td>Australia</td>
<td>363 cases</td>
<td>DS</td>
<td>Population norm</td>
</tr>
<tr>
<td>Hedov et al., 2000, (34)</td>
<td>Medium</td>
<td>SF-36</td>
<td>Service providers</td>
<td>Sweden</td>
<td>207 cases</td>
<td>DS</td>
<td>Population norm</td>
</tr>
<tr>
<td>Scott et al., 1997, (35)</td>
<td>Medium</td>
<td>Depression &amp; other scales</td>
<td>Service providers</td>
<td>Canada</td>
<td>108 cases, 188 controls</td>
<td>DS</td>
<td>TD</td>
</tr>
<tr>
<td>Jeans et al., 2013, (36)</td>
<td>Medium</td>
<td>Questionnaires</td>
<td>Previous study</td>
<td>US</td>
<td>“100 cases,” “11,000 controls”</td>
<td>ASD</td>
<td>TD</td>
</tr>
<tr>
<td>Baker-Ericzén et al., 2005, (37)</td>
<td>Medium</td>
<td>120-item stress index</td>
<td>Schools</td>
<td>US</td>
<td>37 cases, 23 controls</td>
<td>ASD</td>
<td>TD</td>
</tr>
<tr>
<td>Zablotsky et al., 2013, (38)</td>
<td>Medium</td>
<td>One-item health, three-item stress indices</td>
<td>Previous study</td>
<td>US</td>
<td>1,114 cases, “6,000 controls”</td>
<td>ASD</td>
<td></td>
</tr>
<tr>
<td>Montes et al., 2007, (39)</td>
<td>Medium</td>
<td>Two-item scale</td>
<td>Previous study</td>
<td>US</td>
<td>364 cases, 61,408 controls</td>
<td>ASD</td>
<td>TD</td>
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<tr>
<td>Schieve et al., 2011, (40)</td>
<td>Medium</td>
<td>Aggravation scale</td>
<td>Previous study</td>
<td>US</td>
<td>872 cases, 11,100 controls</td>
<td>ASD</td>
<td>no DD</td>
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<tr>
<td>Watt et al., 2012, (41)</td>
<td>Medium</td>
<td>36-item stress/90-item symptom indices</td>
<td>Service providers</td>
<td>Canada</td>
<td>50 cases, 50 controls</td>
<td>ASD</td>
<td>TD</td>
</tr>
<tr>
<td>Mugno et al., 2007, (42)</td>
<td>Medium</td>
<td>Validated questionnaire</td>
<td>Service providers</td>
<td>Italy</td>
<td>115 cases, 42 controls</td>
<td>PDD,</td>
<td>TD</td>
</tr>
<tr>
<td>Rusk et al., 2011, (43)</td>
<td>Medium</td>
<td>SF-12</td>
<td>Service providers</td>
<td>US</td>
<td>33 cases</td>
<td>ASD</td>
<td>Population norm</td>
</tr>
<tr>
<td>Norlin et al., 2013, (44)</td>
<td>Medium</td>
<td>Well-being scale, stress questionnaire</td>
<td>Previous study</td>
<td>Sweden</td>
<td>58 cases, 178 controls</td>
<td>ID</td>
<td>TD</td>
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<tr>
<td>Olsson et al., 2008, (45)</td>
<td>Medium</td>
<td>Postal surveys</td>
<td>Service providers</td>
<td>Sweden</td>
<td>62 cases, 183 controls</td>
<td>ID</td>
<td>TD</td>
</tr>
<tr>
<td>Khanna et al., 2011, (46)</td>
<td>Medium</td>
<td>SF-12 &amp; other scales</td>
<td>Service providers</td>
<td>US</td>
<td>304 cases</td>
<td>ASD</td>
<td>Population norm</td>
</tr>
<tr>
<td>Lee et al., 2005, (47)</td>
<td>Medium</td>
<td>Multiple surveys</td>
<td>Service providers</td>
<td>US</td>
<td>89 cases, 46 controls</td>
<td>HFASD</td>
<td>TD</td>
</tr>
<tr>
<td>Stoneman et al., 2007, (48)</td>
<td>Medium</td>
<td>Multiple indices</td>
<td>Service providers</td>
<td>US</td>
<td>29 cases (DS), 21 comparisons</td>
<td>ID not DS</td>
<td>DS</td>
</tr>
<tr>
<td>Griffith et al., 2010, (49)</td>
<td>Medium</td>
<td>Stress index, depression/affect scales</td>
<td>Previous study</td>
<td>UK</td>
<td>19 DS, 19 ASD, 19 ID</td>
<td>ASD, ID</td>
<td>DS</td>
</tr>
<tr>
<td>Dumas et al., 1991, (50)</td>
<td>Medium</td>
<td>Stress &amp; depression indices</td>
<td>Service providers</td>
<td>Canada</td>
<td>(30, 30, 30) cases, 60 controls</td>
<td>ASD, DS, Behav problems</td>
<td>TD</td>
</tr>
<tr>
<td>Eisenhower et al., 2005, (51)</td>
<td>Medium</td>
<td>Depression scale, Family impact scale</td>
<td>Service providers</td>
<td>US</td>
<td>(14, 12, 43) cases, 136 controls</td>
<td>ASD, DS, DD</td>
<td>TD</td>
</tr>
<tr>
<td>Sanders et al., 1997, (52)</td>
<td>Medium</td>
<td>Stress &amp; Family indices</td>
<td>Service providers</td>
<td>US</td>
<td>(18, 18) cases, 18 controls</td>
<td>ASD, DS</td>
<td>TD</td>
</tr>
<tr>
<td>Lenhard et al., 2005, (53)</td>
<td>Medium</td>
<td>Questionnaires</td>
<td>Service providers</td>
<td>Germany</td>
<td>(411, 66) cases, 69 controls</td>
<td>DS, ID of unknown cause</td>
<td>TD</td>
</tr>
<tr>
<td>Stores et al., 1998, (54)</td>
<td>Medium</td>
<td>Stress index</td>
<td>Service providers</td>
<td>UK</td>
<td>91 cases, 78 controls</td>
<td>DS, ID not DS</td>
<td>TD</td>
</tr>
<tr>
<td>Hamlyn-Wright et al., 2007, (55)</td>
<td>Medium</td>
<td>Stress index</td>
<td>Service providers</td>
<td>UK</td>
<td>(265, 223) cases, 131 controls</td>
<td>ASD, DS</td>
<td>TD</td>
</tr>
<tr>
<td>Piven et al., 1991, (56)</td>
<td>Medium</td>
<td>Questionnaire</td>
<td>Service providers</td>
<td>US</td>
<td>42 ASD, 42 DS</td>
<td>ASD</td>
<td>DS</td>
</tr>
<tr>
<td>Estes et al., 2009, (57)</td>
<td>Medium</td>
<td>Stress &amp; distress indices</td>
<td>Service providers</td>
<td>US</td>
<td>51 ASD, 22 DD</td>
<td>ASD</td>
<td>DD</td>
</tr>
<tr>
<td>Olsson et al., 2001, (58)</td>
<td>Medium</td>
<td>Depression inventory</td>
<td>Service providers</td>
<td>Sweden</td>
<td>(145, 52) cases, 204 controls</td>
<td>ASD, ID without ASD</td>
<td>TD</td>
</tr>
<tr>
<td>Totika et al., 2011, (59)</td>
<td>Medium</td>
<td>General health questionnaire</td>
<td>Previous study</td>
<td>UK</td>
<td>(47, 51,590) cases, 17,727 controls</td>
<td>ASD, ASD with ID, ID only</td>
<td>No ASD or ID</td>
</tr>
</tbody>
</table>

PH, physical health; SF-36, Short Form 36-item survey; DD, developmental disability; ID, intellectual disability; TD, typically developing; IDD, intellectual and developmental disability; Neuro, neurodevelopmental disorders; PDD, pervasive developmental disorder; ASD, autism spectrum disorder; ADHD, attention deficit hyperactivity disorder; SF-12, Short Form 12-item survey; yo, years old; SF-8, Short Form 8-item survey; AD, autistic disorder; AS, Asperger syndrome; NOS, not otherwise specified; psych, psychiatric; DS, Down syndrome; HFASD, high-functioning ASD; Behav, behaviour; Psych, psychiatric disorder; diag., diagnosis
Table 4: Papers in the supplementary review

<table>
<thead>
<tr>
<th>Paper (in order of citing)</th>
<th>Level of evidence</th>
<th>Data collection</th>
<th>Recruitment</th>
<th>Country of origin</th>
<th>Study population</th>
<th>Disability group(s)</th>
<th>Method of comparison</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bromley et al., 2004, (60)</td>
<td>Low</td>
<td>Validated questionnaires</td>
<td>Service providers</td>
<td>UK</td>
<td>68 mothers</td>
<td>ASD</td>
<td>Health compared by family/child traits</td>
</tr>
<tr>
<td>Corrice et al., 2009, (61)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Service providers/community</td>
<td>US</td>
<td>120 mothers</td>
<td>DD not DS</td>
<td>DS</td>
</tr>
<tr>
<td>Paynter et al., 2013, (62)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Community</td>
<td>Australia</td>
<td>43 parents</td>
<td>ASD</td>
<td>Health compared by family/child traits</td>
</tr>
<tr>
<td>Firth et al., 2013, (63)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Service providers/community</td>
<td>Australia</td>
<td>109 parents</td>
<td>ASD</td>
<td>Health compared by family/child traits</td>
</tr>
<tr>
<td>Baghdadi et al., 2014, (64)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Previous study</td>
<td>France</td>
<td>165 parents</td>
<td>ASD</td>
<td>Health compared by family/child traits</td>
</tr>
<tr>
<td>McStay et al., 2014, (65)</td>
<td>Low</td>
<td>Questionnaires</td>
<td>Autism Research Centre</td>
<td>Australia</td>
<td>196 parents</td>
<td>ASD</td>
<td>Health compared by family/child traits</td>
</tr>
<tr>
<td>Cotton et al., 2004, (67)</td>
<td>Medium</td>
<td>Questionnaires</td>
<td>Previous study</td>
<td>Australia</td>
<td>136 parents</td>
<td>ASD, DS, FID</td>
<td>TD</td>
</tr>
<tr>
<td>Adams et al., 2014, (68)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Service providers</td>
<td>US</td>
<td>311 children</td>
<td>ASD</td>
<td>QoL correlated with child traits</td>
</tr>
<tr>
<td>Ji, et al., 2014, (69)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Service providers</td>
<td>China</td>
<td>273 carers</td>
<td>ASD</td>
<td>Health compared by family/child traits</td>
</tr>
<tr>
<td>Emerson et al., 2010, (70)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Previous study</td>
<td>UK</td>
<td>&gt;15,000 parents</td>
<td>Cognitive delay</td>
<td>Health compared by family demographics</td>
</tr>
<tr>
<td>Dardas et al., 2014, (71)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Community</td>
<td>Jordan</td>
<td>184 parents</td>
<td>Autistic disorder</td>
<td>Correlated variables relating to family variables</td>
</tr>
<tr>
<td>Hoppes et al., 1990, (72)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Service providers</td>
<td>US</td>
<td>38 mothers</td>
<td>ASD</td>
<td>DS were comparator group</td>
</tr>
<tr>
<td>Werner et al., 2014, (73)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Convenience sampling</td>
<td>Israel</td>
<td>171 carers</td>
<td>ID, ASD,</td>
<td>Children with PD comparator</td>
</tr>
<tr>
<td>Woodgate et al., 2008, (74)</td>
<td>Low</td>
<td>Interviews</td>
<td>Service providers</td>
<td>Canada</td>
<td>16 families</td>
<td>ASD</td>
<td>Qualitative study</td>
</tr>
<tr>
<td>Kelso et al., 2005, (75)</td>
<td>Low</td>
<td>Focus groups/interviews</td>
<td>Service providers</td>
<td>Australia</td>
<td>26 carers</td>
<td>Disabilities</td>
<td>Qualitative study</td>
</tr>
<tr>
<td>De Grace et al., 2014, (76)</td>
<td>Low</td>
<td>Interviews</td>
<td>Community</td>
<td>US</td>
<td>7 families</td>
<td>ASD</td>
<td>Qualitative study</td>
</tr>
<tr>
<td>Losh et al., 2009, (78)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Community</td>
<td>US</td>
<td>83 case, 32 controls</td>
<td>ASD</td>
<td>No ASD</td>
</tr>
<tr>
<td>Kunihira et al., 2006, (81)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>University students</td>
<td>Japan</td>
<td>1,364 students</td>
<td>NA, students &amp; Populations norm</td>
<td></td>
</tr>
<tr>
<td>Ingersoll et al., 2011, (84)</td>
<td>Medium</td>
<td>Validated questionnaires</td>
<td>Service providers</td>
<td>US</td>
<td>149 parents</td>
<td>NA, investigating autistic traits &amp; depression etc.,</td>
<td>Population norm</td>
</tr>
</tbody>
</table>

ASD, autism spectrum disorder; DS, Down syndrome; DD, developmental disability; PD, physical disabilities; FID, familial intellectual disability; TD, typically developing; ID, intellectual disability; NA, not applicable; SF-12, Short Form 12-item survey; Psych, psychiatric disorder; AD, autistic disorder; AS, Asperger syndrome; PDD(NOS), pervasive developmental disorder (not otherwise specified); Neuro, neurodevelopmental disorders; HFASD, high-functioning autism; ADHD, attention deficit hyperactivity disorder; NA, Not applicable.
Table 5: Strength of evidence for studies in Tables 3 and 4

<table>
<thead>
<tr>
<th>Strength of evidence</th>
<th>Study-type</th>
<th>Strength of evidence</th>
<th>Population studies</th>
</tr>
</thead>
<tbody>
<tr>
<td>High</td>
<td>Randomised control trial</td>
<td>High</td>
<td>Longitudinal design with control</td>
</tr>
<tr>
<td>Medium–high</td>
<td>Control trial without randomisation</td>
<td>Medium–high</td>
<td>Longitudinal design without control</td>
</tr>
<tr>
<td>Medium</td>
<td>Cohort or case control analytic study</td>
<td>Medium</td>
<td>Cross-section design with control</td>
</tr>
<tr>
<td>Lower medium</td>
<td>Multiple time series</td>
<td>Lower medium</td>
<td>Cross-section design without control</td>
</tr>
<tr>
<td>Low</td>
<td>Descriptive studies</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Note: Table adapted from Description of levels of evidence grades and recommendations(4)

2.4 Discussion

First, I report on physical and mental health, followed by the overall health and QoL of the mothers of children with ID or ASD compared to other mothers. Second, I summarise research that compared the health of mothers of children from different diagnostic sub-groups of ID and ASD. Next, I examine the associations with poorer health in these mothers and provided an insight as to how these associations might affect the health of mothers of children from particular sub-groups. Finally, I summarise my findings and discuss the implications of this review.

2.4.1 Comparisons with the general population

In this section, I compare the health of mothers of children with ID or ASD using the three categories of Physical health, Mental health, and Overall health and quality of life (Figure 2).

Physical health

In two studies(2, 3) that assessed physical health using item-based scales, mothers(2) and parents(3) of children with developmental disabilities reported poorer physical health than controls. In a third study, a comparison of blood samples(4) taken before and after vaccination revealed that parents of children with ID or ASD had a poorer antibody response than mainstream parents. The results of another case–control study, extracted from population data(5) demonstrated that parents that lived with their adult
children with ID or ASD had higher rates of cardiovascular problems than matched mainstream parents. Carers of children with neurological disorders reported a higher prevalence of asthma, back problems and migraines than carers of children without these problems. By way of surveys, older caregivers of persons with ID or ASD also reported an increased prevalence of arthritis compared to older caregivers in the general population. Moreover, the second study also identified excesses of high blood pressure, obesity and limited mobility in persons over 40 years and a higher prevalence of diabetes and high blood cholesterol in 40–59 year olds compared to age-matched controls.

Carers who lived with their child with ID reported more headaches, sleep disturbances, gastro-intestinal problems and respiratory infections than controls. Further, parents co-residing with their child with ID or developmental disability had a higher average Body Mass Index (BMI) than similar parents who were not co-residing. Mothers of three year olds with developmental delay without ASD assessed their physical health as poorer than did mothers of other three year olds. Likewise, during interview, parents of children with ID reported poorer physical health and more frequent visits to their family doctor than the control group.
Other researchers found lower levels of self-reported physical health(11) and more episodes of physical illness(12) in the parents of children with ASD compared to the parents of children without disabilities. Mothers of adolescents with ASD reported more headaches, backaches, muscle soreness, tiredness and hot flushes than mothers of adolescents with no ASD.(13) Using the Short Form 12-item Health Survey (SF-12), mothers of children with ASD without ID reported poorer physical health than the population norm.(14)

Three research groups(15, 16, 17) failed to differentiate between the physical health of carers of children with ID or ASD and the carers of typically developing children. The first(15) used the Short Form 36-item Health Survey (SF-36) and surveyed the health of middle-aged and older mothers of adults with ID. The second(16) compared older parents or carers of adults with ID to that of their counterparts in the general population. Reasons for their null findings might be that these two study populations included only carers of children with ID whereas all but one of the first mentioned studies included children with ASD or developmental disabilities. In a third study,(17) mothers provided information that suggested the physical health limitations of the 526 case mothers of children with ID or ASD were not greater than that of the 14,444 mothers of typically developing children.(17) This might be because this comparison was made after controlling for mental health and also because the children were only five years old. Hence, mothers would not have had longer-term exposure to the challenges presented by their children.

Mental health

Psychiatric disorders

Using case–control methodology, researchers(18) interviewed 50 parents who had a child with ID and 50 parents with a healthy child. Case parents were assessed as having significantly higher levels of neuroticism than control mothers.(18) A secondary analysis of data collected from a British National Survey of nearly 10,000 mother–child dyads used a validated assessment to measure maternal well-being. The 245 mothers of children with ID had a slightly lower self-reported rate of psychiatric disorders than other mothers.(19) Using data linkage,(20) Australian researchers identified that the mothers with chronic schizophrenia, bipolar disorder or unipolar major depression had a
higher rate of ID in their children compared to the children of unaffected mothers. In the British study,(19) data were self-reported psychiatric disorders in mothers who were included in the secondary analysis because they had a child with ID. Mothers with more severe psychiatric disorders may have been less likely to be included since they might not have been well enough to respond to invitations to participate or well enough to self-report on their condition. In contrast, the Australian study(20) relied on linked data and mothers were included in the study because they had a severe and lifelong psychiatric disorder. Hence, the psychiatric disorders were not self-reported and the study population is more well-defined than the previous two studies. This may explain the difference.

A population-based case–control study(21) used record linkage to examine the diagnoses associated with hospitalisations in mothers of a child with ASD. These mothers were more likely to have been hospitalised with an associated diagnosis of depression, personality disorders or schizophrenia than mothers of children without ASD.(21) Three other studies, (22, 23, 24) also using record linkage, reported that parents of a child with ASD were more likely to have been diagnosed with schizophrenia spectrum disorders,(22, 23) affective disorders,(23) bipolar disorders(22) or personality disorders(24) than parents of children without ASD. An earlier Danish study(25) found that pre-existing parental psychiatric disorders were associated with increased rates of ASD in the offspring.(25) An examination of psychiatric disorders in the family of probands with ASD compared to those with Down syndrome revealed that the families of the probands with ASD had significantly more obsessive–compulsive and affective disorders than the families of probands with Down syndrome.(26)

**Other aspects of mental health**

As with specific psychiatric disorders, the majority of other research on mental health in the carers of children with ID or ASD found that carer health was compromised. Using the SF-36, both middle-aged and older women caring for adult relatives with developmental disabilities reported poorer mental health compared to population norms.(27) Parents of children with developmental disabilities had higher scores on a stress index than the parents of typically developing children.(28) Depression was also commonly reported. Using the Beck Depression Inventory (BDI), mothers of children
with developmental delay exhibited more depression than mothers of children without these disabilities. (29) Another study (30) also used the BDI and mothers of disabled children had more depressive symptoms than comparison mothers. With the same inventory, Latina mothers of children with ID scored higher levels for depression than Latina mothers of typically developing children. (31) Using standardised stress scales, two independent research groups (6, 32) found more stress in the parents of children with ID than in controls. In studies using the SF-12 (33) or SF-36, (34) mothers of children with Down syndrome exhibited poorer mental health than recognised population norms. In a survey study, (35) researchers matched parents of an infant with Down syndrome to the parents of infants with no disability by SES and found that the Down syndrome group experienced more distress.

A study using survey methodology (36) indicated that 200 mothers of children diagnosed with ASD and aged four years and nine months exhibited higher levels of depression and stress than mothers of age-matched children without ASD. In other case–control studies, also with data derived from surveys, mothers (or parents) of children with ASD reported more stress, (37–39) more aggravation (40) and poorer mental health (38, 39) than other mothers (or parents). Other researchers identified greater stress in parents of children with ASD than in parents of typically developing children. (41) Again using self-report, the psychological health of parents of a child with ASD was compromised compared to mainstream parents. (42) Similarly, others found that the self-reported mental health-related QoL of carers of children with ASD was lower than the population norm. (43) Others reported that the parents of children with ASD had elevated concentrations of a pro-inflammatory biomarker for psychological distress. (19)

Four research groups (7, 15, 16, 5) found no difference between the mental health of parents of children with ID or ASD. First, African American mothers of children with developmental disabilities were no more depressed than the African American mothers of typically developing children. (7) Another group (15) used the SF-36 to survey mental health in middle-aged and older mothers of adults with ID and found no difference to population norms. (15) The remaining two studies stratified by carer age. One of these (16) used the SF-12 to investigate the mental health of older parents caring for their children with ID. Compared to population norms, parents aged 55–64 years (but
not older parents) reported poorer mental health.\(^{16}\) The remaining study, using similar methodology, reported no difference between middle-aged parents of children with ID and developmental disabilities and other parents in either self-reported depression levels or well-being.\(^{5}\) However, by their mid-sixties, those who remained caring for their child now reported higher levels of depressive symptoms.\(^{5}\) In a similar way to the studies that reported on physical health, the studies with null results for mental health related to mothers of children with ID. The first of the two studies\(^{16}\) stratified by carer age and identified poorer mental health in parents older than 64 years. The second\(^{6}\) reported more depression in parents in their mid-sixties who remained caring for their child with a disability.

**Overall health and quality of life**

In most instances, studies reported lower levels of overall health and QoL in parents of children with ID or ASD. Mothers of children with ID reported poorer well-being than other mothers.\(^{44, 45}\) Similarly, families with a member with ID were found to have a poorer QoL\(^{32}\) and poorer perceived overall health\(^{18}\) than the control group. Others reported that the overall health of carers of children with Down syndrome was poorer than the comparator group.\(^{34}\)

Carers of children with ASD were found to have a worse perception of their QoL\(^{11}\) and health-related QoL\(^{46}\) than carers of children with no disability. In a similar way, a comparison of mothers of children with ASD to those with no disability found that case mothers had a larger proportion of days with negative health symptoms than the controls.\(^{13}\) A secondary analysis from a British cohort study\(^{17}\) included nearly 15,000 mothers, and researchers accessed data from questionnaires to assess maternal well-being. Mothers of children with ASD were deemed to have a similar level of well-being to the mothers of children with behaviour problems and no ASD.\(^{17}\) This finding would be consistent with behaviour problems being the main correlate of poorer health in mothers of children with ASD. Finally, comparisons of the parents of children with ASD without ID to parents of children without disabilities indicated a lower QoL in the case parents.\(^{47}\)
Summary

In the vast majority of studies, poorer physical health was demonstrated in the mothers of children with ID or ASD compared to other mothers. Findings ranged from poorer overall physical health to a higher prevalence of specific conditions such as asthma, arthritis and diabetes. There was a similar picture with aspects of mental health and QoL. Mothers of children with ID or ASD most often had an increased prevalence of psychiatric disorders, more stress, depression and poorer overall mental health. Furthermore, the poorer physical health demonstrated in those with mental health issues (48) is likely to exacerbate the association between poorer physical and mental health in these mothers. Last, in all but one instance,(16) researchers reported that mothers of children with ID or ASD had a lower perception of their QoL than other mothers.

2.4.2 Maternal comparisons by child disability

In many instances, comparisons involved the health of mothers of children with ASD and the mothers of children with Down syndrome. Other studies compared the mothers of children with ID but not Down syndrome to mothers of children with Down syndrome. All reports described that the mothers of children with ASD had the poorest outcomes and mothers of children with Down syndrome the next best after mothers of typically developing children. When looking at intergroup comparisons, it is important to consider the possibility of confounding by socio-demographic variables, as many are not randomly distributed among disability groups. For example, one study(49) compared the parents of children with Down syndrome to the parents of children with ID of other aetiologies. Before adjusting for income level, the well-being of the Down syndrome group was higher. After adjustment, this advantage disappeared. An analysis of survey data(45) indicated that mothers of children with ID had poorer well-being than mothers of children without ID and differences in economic hardship were a major risk factor.(45) Compared to parents of children with other disabilities,(50–53) parents of children with Down syndrome had less self-reported stress. Results from another case-control study,(54) indicated that mothers of children with ID of unknown cause exhibited more anxiety, guilt and emotional burden than the mothers of children with Down syndrome and that the Down syndrome group was indistinguishable from the mothers of typically developing children.(54) Notably, none of these studies adjusted
for SES and this may have biased the results. Another study(55) concluded that the mothers of the children with Down syndrome were less stressed than other mothers, even after adjusting for SES.

Further comparisons involved other aspects of mental health. For example, using questionnaires, authors(56, 57) reported lower levels of anxiety and depression in the parents of children with Down syndrome than in the parents of children with ASD. In the first study,(56) the authors commented that the parents of children with ASD were of higher SES than the Down syndrome group but nevertheless did not adjust for SES. In a third study(58) exploring validated stress and psychological distress measures, mothers of children with ASD exhibited higher levels of both stress and distress than mothers of children with developmental delay without ASD. Another study used the BDI to compare the levels of depression in mothers of children with ASD to those of children with ID without ASD.(59) In an attempt to adjust for SES, groups were matched according to geographical area of residence. The mothers of children with ASD displayed more severe depression than the mothers of children with ID but not ASD.(59)

Three studies(11, 17, 60) compared aspects of the mental health of parents of children with ASD with and without ID using a validated questionnaire. Parents of children with ASD without ID demonstrated a lower QoL and more stress than the parents of children with ASD with ID.(11) Mothers of five-year-old children with ASD without ID also reported higher levels of maternal emotional disorder than the mothers of children with ASD with ID.(17) Although the higher level of emotional disorder in the mothers of children with ASD without ID may reflect a higher burden of care, it could also be indicative of a greater predisposition to mental health problems. The same research group(60) examined emotional disorder in the mothers of children from 5 to 16 years old with ASD with and without ID and found no difference between the two ASD groups but higher levels in the ASD group than in the mothers of children with ID only.

In summary, research suggested that the burden of caregiving had least effect (if the reported associations are causal) on the health of the parents of children with Down syndrome.(50–57) The second least affected group was the parents of children with others forms of ID.(58, 59) The parents of children with ASD were most often assessed
as having the poorest health,(59, 60) particularly those parents of a child with ASD without ID(11, 17) (Figure 3).

**Figure 3:** Ranking of case groups as suggested by studies in the review

<table>
<thead>
<tr>
<th>Down Syndrome</th>
<th>ID not Down Syndrome</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
</tr>
</thead>
</table>

ID, intellectual disability; ASD, autism spectrum disorder

### 2.4.3 Correlates of poorer health and quality of life

**Child behaviour and sleep patterns**

The most common correlate with poorer parental health-related QoL outcomes was challenging child behaviour. Researchers identified that better social skills(37) and fewer behavioural symptoms(6, 29, 33, 61–64) in children with ID or ASD were correlated with better maternal mental health. Others(65) administered a validated QoL scale to the parents of children with ASD and identified that challenging behaviour in a child was an independent risk factor for poorer QoL. Along similar lines, three research groups(58, 66, 67) reported that the behavioural or emotional problems in a child had a greater effect on parental health than the diagnosed disability of the child. Another research group(2) reported that challenging behaviour in young children with developmental disabilities contributed to declining maternal health. Moreover, behaviour problems in these children had a greater effect on maternal health than similar levels of behaviour problems in typically developing children.(2)

One study(68) used parent report to compare sleep problems in children from different disability groups and a typically developing group. More sleep problems were found in the children with ASD than in those with Down syndrome or other forms of ID who, in turn, had more sleep problems than the typically developing children.(68) Another research group(69) reported that sleep problems exacerbated challenging behaviours in
children with ASD. Only one study(3) concluded that problem behaviours in children with a developmental disability were not predictive of poorer maternal physical health.

**Support**

Scores on a questionnaire to assess health-related QoL indicated that family support reduced the carer burden in families with a child with ASD.(70) Two other research groups(38, 61) reported that higher levels of family and neighbourhood support reduced the risk of poorer mental health in mothers of children with ASD. Using the SF-12 and other questionnaires, researchers(16) found that improved mental health in older parents of children with ID was associated with having a partner and having a large and supportive network of family, friends and neighbours. Conversely, single mothers of a child with ID were more likely to be depressed(59) and to have a low sense of coherence in their families.(66) Further, poorer mental health in the mothers of adults with developmental disabilities was associated with a lack of services.(27) Using a validated questionnaire, parents of children with ASD demonstrated more impairments in social relationships than other parents.(11) This would implicate lower levels of informal supports in these mothers. Using an online questionnaire, a research group(3) concluded that associations between stress and physical health issues were moderated by social support in parents of children with developmental disabilities. In a national survey,(40) less emotional support was associated with increased scores on an aggravation scale in the parents of children with ASD.

**Socioeconomic status**

Reports suggest that lower socioeconomic status (SES) impacts negatively on maternal health and that this is mediated by lower levels of support. For example, researchers(71) identified that mothers of children with early cognitive delay and low SES had an increased risk of a psychiatric disorder and a higher burden of care compared to other mothers of a child with a similar disability but high SES. In a similar vein, others(72) concluded that parents of children with autistic disorder and higher incomes had lower distress and an improved QoL than similar parents with lower incomes. Another study(19) in which parents of children with ID were identified from a national survey of more than 10,000 children, reported that poverty increased the burden of a child’s ID.
Finally, a research group(61) found that mothers with poor housing reported lower levels of support. This suggests that some of the effects of low SES on maternal health might be mediated by lower levels of support.

**Rewards and stigma**

In a comparison of mothers of children with Down syndrome and mothers of children with ASD, researchers(73) identified greater attachment and gratification in the mothers of children with Down syndrome. Another study(74) found that affiliate stigma or self-stigmatisation was higher among carers of persons with ASD compared to carers of persons with ID or physical disabilities. In the 1950s and 60s, clinicians sometimes believed that the mother’s lack of early bonding might contribute to the development of autistic features in the child.(75) Perhaps some of this attitude persists with increased self-blame among mothers of children with ASD and more societal stigma towards them than parents of children with other developmental disabilities.(76) Also, there may be additional societal stigma towards the parents of children who look normal but are clearly manifesting abnormal behaviours.(76)

**Diagnostic issues**

Diagnostic uncertainty and an unclear prognosis is often the scenario with ASD. It is this uncertainty that is likely to produce the greatest emotional trauma for parents.(54, 75, 76, 77) Parents of children with no diagnosis are disadvantaged(77) as they are denied anticipatory and reproductive guidance and support from networking with parents of children with a similar diagnosis. Further, there may be no abatement of the pressure to search for a biomedical cause for their child’s condition. In one investigation,(54) German researchers compared three groups—parents of children with Down syndrome, parents of children with ID of unknown cause, and parents of typically developing children—with respect to anxiety, guilt and emotional burden. They concluded that uncertainty was a major cause of stress in parents of children with ID of unknown cause. Similarly, after interviewing parents of children with ASD, researchers concluded that the uncertainty and lack of information around the time of diagnosis was overwhelming and all-consuming for parents.(79, 80)


**Broad Autism Phenotype**

In 1943, Leo Kanner(81) was the first to report mild autistic traits in the mothers of children with ASD. The concept of mild autistic-like traits in the relatives of a person with ASD has developed considerably this century and has been labelled the Broad Autism Phenotype (BAP).(82) Validated questionnaires such as the Autism Spectrum Quotient (AQ) and Broad Autism Phenotype Quotient (BAPQ) are used to quantify the extent of this trait on a linear scale.(83) Two research groups found that persons exhibiting features of the BAP were at increased risk of depression.(84, 85) If the BAP is immutable and not exacerbated by the burden of caring, this association may partly explain the increased prevalence of depression and associated disorders demonstrated in parents of children with ASD.(84, 85) Alternatively, scores of the Social Skills sub-scale of the AQ could be increased by the burden of caring: since researchers have verified that parents of children with ASD become socially isolated due to the burden of caring for their child.(75)

**Explaining intergroup disparities**

Mothers caring for children with ASD were most often described as having poorer health and mothers of children with Down syndrome better health in comparison to other mothers of children with ID or ASD. The health of mothers associated with other diagnostic groups appeared to lie somewhere in between (Figure 3). Some research might explain these discrepancies. For example, research has demonstrated that children with Down syndrome have more social competence and less problem behaviours than those with ASD.(50) Others(60) reported more severe behavioural and emotional problems in children with ASD than in those with ID. Another research group(86) suggested that the mothers of children with Down syndrome feel as rewarded as those of children without disabilities, whereas others(62) found that these mothers were more rewarded than the parents of children with other developmental disabilities. In other words, the associations of maternal health with child behaviour and rewards might explain some of our preliminary rankings of poorer maternal health. Compared to controls, parents of a child with ID reported lower levels of social support.(9) In a similar way, the existence of diagnostic issues for their child and their own personality traits might also contribute to the poorer health of mothers of children with ASD.
2.5 Implications for further research

My core review identified 58 original research papers investigating the health of mothers of children with ID or ASD, compared either to mothers of typically developing children, population norms or mothers according to the sub-group of their child’s disability. Further research into the health of mothers of children with ID or ASD is implicated in mothers of children with ID with groups sub-divided according to the level and cause of the ID. Further research is also implicated in mothers of children with ASD according to the presence of comorbid ID in their child. The timing of health differences is important and for this reason, research should differentiate between health issues with onset before and after the birth of the child and/or in relation to the diagnosis of the child. Further research into the correlates of poorer health would inform those developing supports and interventions to assist these mothers to improve their health. For example, as challenging behaviours are a strong correlate of poorer health, workshops to provide mothers with behaviour management programs to assist their child’s development are implicated. Similarly, providing more support—such as home services—for mothers with health issues might make the way for their improved health.

2.6 References


Chapter 3: The proposed research in the context of existing knowledge

In Chapter 2, I reviewed the research into the health of mothers of children with intellectual disability (ID) or autism spectrum disorder (ASD) compared to the mothers of typically developing children, and the research that compared the health of mothers according to the diagnostic sub-type of their child. As a result, I have identified gaps and areas where a modified or alternative approach would be likely to increase our understanding.

In this chapter, I first summarise the current state of research into the health of mothers of children with ID or ASD. Second, I expose the limitations and gaps in the existing research and third, I describe my research program, which follows as a logical consequence. Finally, I describe the structure of the ensuing chapters of this thesis. In Part 1, I summarise the characteristics of the research papers cited in my review (Chapter 2). This enables me to ascertain the limitations and gaps in this area of research and my opportunities to address some of the unanswered research questions. Part 2 describes the components of my research, its aims and the maternal groups used for comparisons. In Part 3, I describe the organisation of the remaining chapters of this thesis.
3.1 Opportunities to add to current knowledge

3.1.1 Existing research

In Chapter 2, I identified 58 papers that explored the health of mothers of children with ID or ASD and fulfilled my inclusion criteria. A list of these papers and their properties were provided in Table 3 and a summary of the distributions of these properties is in Table 6.

Table 6: Properties of studies exploring the health of mothers of children with ID or ASD

<table>
<thead>
<tr>
<th>Strength of evidence*</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Medium (Case control analytic study)</td>
<td>52 (90%)</td>
</tr>
<tr>
<td>High (Longitudinal population study with control)</td>
<td>6 (10%)</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>58</strong></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Data collection</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Interviews/questionnaires/diaries</td>
<td>Linked data</td>
<td>Other</td>
</tr>
<tr>
<td>50 (86%)</td>
<td>6 (10%)</td>
<td>2 (3%)</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>58</strong></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Recruitment</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Service providers/general community</td>
<td>Linked data</td>
<td>Previous study</td>
</tr>
<tr>
<td>38 (66%)</td>
<td>5 (8%)</td>
<td>15 (26%)</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>58</strong></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Country of origin</th>
<th>UK</th>
<th>Other Europe</th>
<th>North America</th>
<th>Australia</th>
<th>Asia</th>
</tr>
</thead>
<tbody>
<tr>
<td>8 (14%)</td>
<td>15 (26%)</td>
<td>28 (45%)</td>
<td>6 (10%)</td>
<td>1 (2%)</td>
<td></td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>58</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Number of cases</th>
<th>&lt; 50</th>
<th>50-99</th>
<th>100-499</th>
<th>500-999</th>
<th>&gt;1,000</th>
</tr>
</thead>
<tbody>
<tr>
<td>10 (17%)</td>
<td>35 (26%)</td>
<td>22 (38%)</td>
<td>5 (10%)</td>
<td>6 (10%)</td>
<td></td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>58</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Grouping of carers by child disability(s) and/or onset of disorders for comparisons</th>
<th>Combinations of ID/ASD/DD/IDD /Neuro</th>
<th>ASD+ – ID</th>
<th>Down</th>
<th>ASD+ –ID &amp; psych before /after birth</th>
<th>ID/ASD/Down</th>
<th>ASD+ –ID &amp; before/after diagnosis</th>
</tr>
</thead>
<tbody>
<tr>
<td>37 (64%)</td>
<td></td>
<td>3 (6%)</td>
<td>4 (6%)</td>
<td>2 (3%)</td>
<td>11 (19 %)</td>
<td>1 (2%)</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td></td>
<td><strong>58</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

ID, intellectual disability; ASD, autism spectrum disorder; DD, developmental disability; IDD, intellectual & developmental disability; Neuro, neurological disorder with and without behaviour problems; ASD+ — ID, ASD with or without ID; psych, psychiatric disorder; Down, Down syndrome

* Strength of evidence criteria are explained in Table 5

3.1.2 Limitations and gaps in knowledge

Ethnicity and immigration variables

Ethnicity and immigration status are quite independent. Ethnic status relates to membership of a group within a society that is formed according to culture and race,(1)
whereas immigration status relates only to being born abroad. I found multiple studies (2–5) that reported on the risk of ID or ASD in the children of immigrant women and/or women of a minority ethnicity but few that looked at these traits independently. Further, I located multiple studies (5–8) that did not adjust for the year of the child’s birth. This is important since birth year might be a potential confounder due to the increasing rate of ASD and the changing immigration rates in many countries.

**Timing of the child’s birth**

There are three reasons why it is important to differentiate between the health of mothers of children with ID or ASD prior to and after the onset of caring. First, prior maternal health would be likely to affect health after the onset of caring. Second, pre-existing health issues might provide insight into genetic and modifiable risk factors for ID or ASD. Third, the onset of health issues after the birth would provide a measure of the burden of care. Failure to take into account the timing of the onset of health issues limits our ability to use research findings to understand possible risk factors and causes of ID or ASD and limits our ability to measure the effect on maternal health of caring for their children. In turn, measures of the effect on maternal health of caring are able to inform services and interventions aimed at improving maternal health. Apart from one study that examined the risk of ID in the children of mothers with severe and chronic psychiatric disorders, (9) I found no research investigating the pre-existing health of mothers of children with ID without relying on maternal or family recall. With ASD, only three studies (10–12) ascertained whether the psychiatric disorder existed prior to the birth (or diagnosis of the disability) of the child.

**Psychiatric disorders**

All four studies investigating psychiatric disorders in the mothers of children with ASD demonstrated an increased incidence compared to other mothers. (10–14) The prevalence of pre-existing maternal psychiatric disorder in the mothers of children with ID or ASD might be used to investigate genetic predisposition, environmental exposure or an interaction between the two in these mothers. On the other hand, the prevalence of psychiatric disorders with onset after the child’s birth might be largely attributable to the caregiving burden in mothers of children with ID or ASD. This being said, it is
important to remember that some conditions (such as Schizophrenia) have expected ages of onset in the late teens and throughout the twenties. This onset could confound associations in some women who have a diagnosis of schizophrenia after the birth of a child, but nevertheless independently of this birth.

**Sub-type of the disability**

No research took into account the cause or severity of the child’s ID. This might be important as the effect on mothers of caring for children with varying degrees of ID may be different. For example, a child with mild or moderate ID of unknown cause (mild–moderate ID) and functional speech may, to a degree, socialise and function independently, rendering this child more open to bullying and accidental misfortunes. While the lesser independence of a child with severe or profound ID of unknown cause (severe ID) might provide a shield from these negatives, the higher incidence of physical disability and medical problems would be likely to be different sources of stress for mothers of children with severe ID. Hence, the challenges faced by mothers are likely to differ according to the level of the disability. Not having a cause for their child’s disability might be an additional source of stress. For children with ASD, only three studies stratified case groups according to the presence of comorbid ID in the child. This stratification is important since these sub-types of ASD may have different aetiologies and mothers of children with ASD with ID and mothers of children with ASD without ID may experience different stressors in relation to their child.

**Bias**

Of the 58 reviewed studies, only eight (13%) used objective measures of data collection such as registry records or cortisol levels (Table 6). Others relied on interviews or questionnaires and hence there may have been recall or other biases. For example, depressed mothers may view their children as more symptomatic and thus any positive correlation between maternal depression and child behaviour might be an artefact. On the other hand, at interview, cultural pressures might result in participants describing their children in more positive terms. Further, researchers have
expressed concerns regarding self-report in terms of the limits placed on responses by social desirability.(20)

Fifty-three (92%) studies recruited through service providers, the general community or previous studies (Table 6) and hence may have been subject to selection bias. For example, when recruiting through newspaper advertisements, a bias may result as the healthier carers would be more likely to respond and participate.(19) Alternatively, those with more issues might want an opportunity to air their grievances.(20) In five (8%) studies (Table 6), linked registry data provided the study populations and for these, large numbers and an inherent objectivity of inclusion and measurement provided advantages in relation to representativeness, power, and in reducing bias and confounding. As confounders are risk factors associated with both the outcome and exposure of interest,(21) confounding might be associated with inherent differences between the mothers of the disability sub-groups. For example, the physical health of the mothers of children with severe ID and those of children with ASD may differ from each other and the general population prior to the birth.(22) Up to 16 (28%) of the studies included research conducted in a language other than English (Table 6). In some of these, particularly those using interviews and questionnaires, translation of related documents may have limited the meaningful transfer of results.(23)

In summary, only a few studies have differentiated between maternal health before and after the birth of their child. With maternal psychiatric disorders, examining those with onset before the birth might provide an insight into genetic and environmental causes of ID or ASD. Examining those with onset after the birth might provide an insight into the burden of caring for a child with ID or ASD. Finally, most of the existing research papers (Table 6) report measurement and recruiting methods that may have been subject to bias.

### 3.1.3 Measures of health

In my research, I wanted to choose an efficient and valid way of measuring maternal health. I found six studies that used linked registry data to measure aspects of health (Table 6). For example, parental hospital admissions with an associated psychiatric diagnosis were linked to the presence or absence of a diagnosis of ASD in the children.
This linking of parents and children enabled researchers to assess the odds of psychiatric disorders in parents of a child with ASD compared to control parents. (12) Others (22) accessed maternal antenatal morbidity data from midwife records and linked the maternal records to those of their children in a disability database. This enabled an investigation of the relationship between common obstetric conditions and subsequent ID in the offspring. Using linked data in this way has distinct advantages over the use of questionnaires in terms of accuracy, economy and the size of study population. For example, in WA more than 300,000 mothers gave birth from 1983 to 2005. This greatly increases the chance of being able to identify small effects within a population. Further, there is no reliance on participant recall.

Most often, using linked data substantially increases the sample size of a study. Three of the studies (10, 12, 13) using linked registry data included more than 1,000 cases. Apart from those relying on recruits from previous studies, only one of the studies (24) that used questionnaires was able to include more than 1,000 cases (Table 3). Another advantage of linked registry data is a reduction of potential bias, particularly selection bias since people are not recruited. Further, there is no reliance on participant recall or perspective since data have been recorded by others at the time of the event. Another advantage is the ability to explore a patient’s medical history in the decades preceding a particular event (such as the birth of a child). Importantly, there is an inherent respect of patient privacy since there are no face-to-face meetings of researcher and patient, and all data are de-identified.

Some researchers found that a failure to adjust for socio-demographic factors confounded results when comparing parental case groups based on child disability status. (25, 26) Linked data provides an obvious means to avoid this dilemma since socio-demographic information, age, maternal parity and prior medical history can often be extracted from registries. Apart from those studies exploring maternal age as a risk factor, most population studies adjusted for this potential confounder. (10, 11, 12) Exceptions were an international report comprising discrete studies from three nations where only one described adjusting for maternal age. (13) Another reported mean and range of maternal age in cases and controls but seemed not adjust for it. (14) Additionally, with linked data, where statutory databases recording death are linked to
other registries, mortality rates become a possible way to measure the health of a population sub-group.

### 3.1.4 Linked health data in Western Australia

Since the late 1970s, record linkage has enabled data relating to the same individual, family or place to be combined from multiple sources. This has been enabled by the personnel of the Health Information Linkage Branch (formerly the Data Linkage Unit) who use personal alpha–numeric identifiers to preserve the privacy of the population. In WA, databases available for potential linkage include the Midwives Notification System, the Births and Deaths Registries, the Hospital Morbidity Data System, the Mental Health Information System and the Intellectual Disability Exploring Answers IDEA Database (Table 7). Researchers in WA are in the privileged position of having access to data from the IDEA database, which contains information on the severity of the ID, whether or not the cause is known and the presence or absence of ID in persons with ASD.

<table>
<thead>
<tr>
<th>Table 7: Description of databases</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Database</strong></td>
</tr>
<tr>
<td>Midwives Notification System(29)</td>
</tr>
<tr>
<td>Births Registry(29)</td>
</tr>
<tr>
<td>Deaths Registry(29)</td>
</tr>
<tr>
<td>Hospital Morbidity Data System (HMDS)(29)</td>
</tr>
<tr>
<td>Mental Health Information System (29)</td>
</tr>
<tr>
<td>IDEA Database(28)</td>
</tr>
</tbody>
</table>

DOH, Western Australian Department of Health; WA, Western Australia; DSC, Disability Services Commission; IDEA, Intellectual Disability Exploring Answers; ID, intellectual disability; ASD, autism spectrum disorder; ICD-9, International Classification of Diseases, ninth revision; International Classification of Diseases, tenth revision

### 3.1.5 Quality of WA’s linked data

Western Australia’s data linkage system was established in the 70s and consequently there have been many years to work towards optimising the quality of the records. The
Health Information Linkage Branch, amongst other things, work to remove duplicate records and correct data artifacts. Very importantly, high quality personal identifiers are used to link data across data-bases. There are now about 20 staff members and each has specific responsibilities for quality control and liaison with data custodians and researchers. (27)

The quality of the MNS database, is optimised by using multiple procedures. (30) Firstly, data is checked for completeness and formatting by the relevant health providers or midwives. This happens prior to submission to the Maternal and Child Health Unit where the data are stored. Secondly, during the uploading or data, records with errors are rejected and suspect data is flagged. Subsequently, both erroneous and suspect data are returned to the providers for checking and/or correction. Thirdly, where possible, records from a database are cross-checked with matching records from another database. Any unusual changes or occurrences are checked and investigated as required. These include changes in frequency or unusual combinations of events. Lastly, this database is subject to a five-year audit and, as feasible, recommendations made to improve the quality of the data are implemented. (30) For the 13 year period from 1980 to 2012, 100% of notifications to the MNS were certified as complete and valid. (31)

Data quality in the HMDS database is maintained by the Inpatient Data Collections Unit which provides the support and education for the personnel in this area. (32) The MHIS database houses the details of all outpatient mental health contacts. Data are regularly serviced and amongst other things, duplicate records removed and records with links to death records are closed as required. (33) The IDEA database is located at the Telethon Kids Institute and has an international reputation for promoting high quality research. (34) It is overseen by the IDEA Advisory Council which is a group of stakeholders who feed into or benefit from the use of the database. (34) The database was initiated in 2002 and contains population-based information on individuals born since 1983 who have been identified through the state-based Disability Services Commission (DSC) and the WA Education Department. Updates have occurred biannually and data are checked and uploaded by personnel at the Telethon Kids Institute under the supervision of the Data Custodians. (28)
3.1.6 Unanswered questions

I found no population study measuring the physical and mental health or QoL in mothers of children with ID or ASD, with a comparison to each other and the general population. I found no research that focused on health issues before and after the birth of a child with ID. I found only one study(9) that explored the relationship between maternal psychiatric disorder and having a subsequent child with ID. I found no study exploring the relationship of a child with ID and maternal psychiatric disorder. Moreover, only four studies(13, 32–37) took account of whether the ASD was associated with ID. In addition, I found no research that examined the mortality or the most common causes of death in the mothers of children with ID or ASD compared to other mothers. Mortality rates are an ultimate measure of health. Causes of death in mothers of children with ID or ASD might provide an insight into genetic, environmental or gene–environment issues associated with the disabilities of their children. My proposed research has been tailored to overcome the limitations of the existing research and to provide more reliable answers to previous research questions and reliable answers to new research questions.

3.2 My research within this thesis

The story of my research continues in the remainder of this thesis. Subsequent chapters detail the stages of my exploration of the health of mothers of children with ID or ASD.

3.2.1 Review

My review of the health of mothers of children with ID or ASD in Chapter 2 impressed upon me the importance of identifying whether these mothers might be different from other mothers before the birth of their child in terms of health or other characteristics. Hence, my first study was a review of the literature pertaining to pre-existing differences in the mothers of children with ID or ASD. This review comprises Chapter 4. As a result, I identified that maternal immigration and ethnicity influence the risk of having a child with ASD. I found no study that explored maternal immigration, ethnicity and region of birth as independent risk factors for having a subsequent child
with ASD. Therefore, I decided to conduct a study designed to explore these variables as risk factors for ASD in WA.

### 3.2.2 Population studies using linked data

Five of my studies exploited linked registry data to explore pre-existing differences, psychiatric disorders and mortality. Due to the associations of socio-economic status, maternal age and parity to the risk of a child with ID or ASD (4.3), I adjusted for each of these characteristics in each study using linked data. A fourth covariate, which allowed for the year of the index birth was also included. This allowed for the increasing prevalence of autism (1.6) over the collection period of each study. The first study examined the relationships between maternal ethnicity, immigration status, birth region and the risk of having a child with ASD with and without ID. This study comprises the first part of Chapter 5. The second study aimed to investigate the relationship between maternal psychiatric disorder and the risk of a subsequent child with ID or ASD and forms the second part of Chapter 5. An ultimate expression of ill health is death. Therefore, I embarked on a study of mortality and cause of death in the mothers of children with ID or ASD. This third study compared mortality and the cause of death in the mothers of children with ID or ASD to the mothers of children with no child with ID or ASD and comprises Chapter 6.

Once I had established that mothers of children with ID or ASD had increased odds of a pre-existing psychiatric disorder, I reasoned that it was important to examine maternal psychiatric disorders with onset after the birth of a child with ID or ASD. In the last two of the population studies, I aimed to investigate the onset of maternal psychiatric disorders in mothers of children with ID or ASD after the birth of their child. This has been written as two papers, which are presented in Chapter 7.
Maternal case groups

I formed the six core case groups of mild–moderate ID, severe ID, Down syndrome, ID of known cause (not Down syndrome), ASD with ID and ASD without ID (Figure 4). I chose these groups carefully as inappropriate groupings could mask differential morbidity or mortality within a sub-group. For example, it was important to separate the mothers of children with Down syndrome from the mothers of children with other ID of known cause. First, this was because Down syndrome is the most common known cause of ID. Second, Down syndrome is not an inherited disorder and so mothers would not have any related, genetically determined effects on their health, as might occur with other conditions such as Fragile X or neurofibromatosis. Third, in some other mothers of children with ID of known and unknown cause, a particular exposure (such as heavy alcohol consumption or severe anaemia) might be associated with their child’s disability and also influence their own morbidity. The comparator group consisted of all mothers with no child with ID or ASD. Where numbers were small, I combined core case groups to form composite case groups. For instance, for some analyses, I combined mothers of children with mild–moderate and severe ID to form the group of mothers of children with ID of unknown cause. On others, I formed a general case group consisting of all mothers of children with either ID or ASD, by combining all of the core case groups (Figure 4). These provided larger numbers for analyses as required. In some studies, only ID or ASD were considered and here only the relevant case groups were examined.²

# All children who had ASD with ID were considered only as an ASD case group and not as an ID case group.

¹ In Publications 5 and 8 only ASD with ID and ASD without ID were examined. In Publication 7 only the four ID case groups were examined (pp. iii and iv).
Index child

Each mother was assigned an index child and in mothers of children with ID or ASD, the index child was their eldest child with a disability born between 1983 and 2005*. In the comparator group, the index child was the eldest child born between 1983 and 2005. Mothers were assigned to a case group according to the disability of their index child. In Figure 4, comparison, composite and core case groups and their inter-relationships are shown. All core groups are mutually exclusive. For instance, persons with both ASD and ID are excluded from ID case groups and placed only within the ASD with ID case group. Similarly, persons with Down syndrome and ASD are only within the Down syndrome group.

* In Publication 4, the study population consisted of the mothers of children born in WA from 1983 to 1999 (p. iii).
Figure 4: Inter-relationships of the comparator, core and composite case groups

Mothers of all children born from 1983 to 2005

Any ID or ASD ☼

No ID or ASD*

Any ID▼

ID of unknown cause☼

Mild or moderate ID☼

Severe or profound ID☼

ID known cause (~ Down)☼

Down syndrome☼

ID known cause:

Any ASD▼

ASD w/out ID☼

ASD with ID☼

ID, intellectual disability; ASD, autism spectrum disorder; w/out, without; Down, Down syndrome; ~, not
* comparator group; ☼ composite case group; ☼ core case group
3.2.3 Qualitative study

Although population studies have considerable advantages in terms of objectivity and the ability to look at patient history prior to a health-related event, I thought it important to hear mothers’ own perspectives of their QoL. In Chapter 8, I describe the qualitative study in which I collected data through interviewing 16 mothers of children with ASD with ID. I chose this sub-group as I had found no qualitative research that had explored the QoL of these mothers. Here, I provide an account of their insights into factors influencing their QoL.

After transcribing and analysing the interviews, I was impressed by the fact that nearly half of the mothers described personality changes that had occurred following the onset of their child’s ASD. Mothers’ descriptions in this area led to a Hypothesis Paper entitled

*Is the Broad Autism Phenotype in mothers of children with autism spectrum disorder exacerbated by the challenges of caring for their children?*

This paper provides a challenge to the concept that the *Broad Autism Phenotype* is pre-existing and immutable. Therefore, I have included this study as the third part of Chapter 5, which explores pre-existing differences.

3.2.4 Ethics

For the population studies, permission for the research was granted by the WA Department of Health Human Research Ethics Committee (#2011/64). I obtained permission to conduct the qualitative study from The University of Western Australia Human Research Ethics Committee (RA/4/1/6107)

3.3 Discussion

Finally, I present Chapter 9. Here I review my findings in terms of the achievement of my aims, make conclusions and discuss the future implications of my research.
3.4 References

34. Leonard H. Inaugural Report of the IDEA Database: Intellectual Disability in Western Australia: Telethon Institute for Child Health Research; 2004
Chapter 4: Pre-existing differences of mothers of children with ID or ASD—a review

The primary aim of this chapter is to explore and classify the literature pertaining to the pre-existing characteristics of mothers of children with intellectual disability (ID) or autism spectrum disorder (ASD) which may or may not differ from those of mothers of typically developing children. This chapter is divided into six major parts. In the introduction, I discuss pre-existing differences in relation to the aetiology of ID or ASD and the socio-demographic associations. In Part 2, I describe the literature search and selection. Each of the next three parts is devoted to a specific sub-group of characteristics, entitled Socio-demographic factors, Immigrant status and ethnicity and Health and associations. In the sixth part, I summarise the pre-existing maternal differences identified, and discuss the research implications. Finally, I report the emergent associations with ID and ASD and their relevance to aetiology, future research, reducing prevalence and ultimately improving the lives of families with a child with ID or ASD. The first page of the published book chapter has been included as Appendix 2.

4.1 Introduction

In Chapter 1, I described the genetic and non-genetic causes of ID. The aetiology of ASD is complex and much is yet to be understood,(1) though research has implicated a strong genetic basis(2–5) involving multiple genes(3, 5, 6) and gene–environment interactions.(7–9) Advances in chromosomal microarray analysis and gene sequencing technologies are continuing to improve our understanding of all developmental disorders, and aetiologies of ASD are continuing to be uncovered.(8) In such a way, a child presenting with autistic symptoms may be found to have a certain genetic mutation (or combination of mutations) that accounts for the true underlying biological diagnosis. For example, a diagnosis of Rett syndrome would be confirmed when a girl with ASD with ID was found to have a mutation of the MECP2 gene on the X-chromosome.(10)
Researchers have identified relationships between ID, ASD and socio-demographic factors such as maternal education,(11, 12) immigration(13, 14) and ethnicity.(15) Other reported associations involve aspects of a mother’s health including physical characteristics,(16) physical health,(17, 18) mental health(19, 20) and health behaviours.(21, 22)

Intellectual disability and ASD commonly coexist with 30–80% of persons with ASD also having ID.(14, 23) Currently, the relationship between ASD and comorbid ID is poorly understood.(24) However, it is known that phenotypically, persons with these disorders can be grouped into three categories: ID only, ASD with ID and ASD without ID.(24)

My literature review in Chapter 2 left me concerned that many researchers had not separated the pre-existing characteristics and characteristics due to the effects of caregiving in mothers of children with ID or ASD. I considered this most likely in regard to the prevalence of psychiatric disorders and the presence of milder autistic traits (Broad Autism Phenotype). These factors, along with the socio-demographic associations and associations with autoimmune disorders provided me with a guide for choosing terms for my literature search. Inherent characteristics of mothers of children with ID or ASD could be associated with the genetic, environmental or genetic–environmental aetiology of their child’s condition. It was therefore important to separate pre-existing factors, particularly in relation to mental health, since morbidities such as depression(25) might also develop due to the more intense demands of caring for a child with ID or ASD.

The aim of this study is to review research on the pre-existing characteristics that differentiate mothers of children with ID or ASD of unknown cause from each other and from mothers of children without these disabilities. Such an investigation may help to further clarify the determinants of ID or ASD, including the role of genetic and modifiable risk factors. Improving our understanding of such risk factors may reduce the future burden of these disabilities by hastening the development of effective prevention and treatment strategies.
4.2 Literature search and selection

I searched the Medline, Web of Knowledge, Scopus and Google scholar databases and used combinations of the following search terms:

1. Terms associated with ID or ASD: *autis*, pervasive development disorder*, intellectual disability, mental retardation, disab*
2. Terms associated with ID or ASD: aetiology: immigra*, migra*, ethnic*, age, socio-demographic, prenatal, perinatal, auto*, immune*, anti*, psych* and phenotype*
3. Terms associated with mothers of children with ID or ASD: traits, characteristics, parents, mothers, children, persons.

A paper was included in the review if it:

- Was accepted for publication between 1 January 1990 and 31 October 2012 inclusive
- Was a full text article in English
- Described new research published in a peer-reviewed journal
- Described the results of a cohort, case–control, correlation or cross-sectional study of at least 15 subjects
- Compared a characteristic of parents or mothers of children with ID or ASD with parents or mothers of children without disability, or with a population norm
- Assessed characteristics that were pre-existing and not likely to be a result of caring for a child with ID or ASD
- Used methods of ascertainment and measurement of the characteristic(s) of interest that were assessed as unlikely to lead to bias.

Eighty papers were retained for the review. These papers are not the entire literature pool in the area. Had I chosen different search terms or used different combinations of terms in my searches, the basis for my review may have been different. The three broad categories for my analyses were Socio-demographic factors, Immigrant status and ethnicity, and Health and associations; and I sorted papers into one or more of these categories. An additional 61 articles were used to provide background information and possible explanations of some of the reported associations.
4.3 Socio-demographic factors

There are a number of considerations that impinge on the effect of socio-demographic factors on the prevalence of ID or ASD. First, in persons with ASD with ID and those with ASD without ID, some features overlap. A child with ASD with ID, particularly in the past, may often have been diagnosed with only ID as there have been secular changes in the identification of children with ASD. Second, persons who could be diagnosed with ASD without ID are most often able to function independently and may remain undiagnosed in a range of scenarios. Third, the process through which children are assigned a diagnosis of ASD is much more complex than for ID. Although elsewhere the gold standard might be the considered judgement of an expert clinician who had seen many patients with ASD,(26) in Western Australia (WA) a diagnosis of ASD in a child requires an assessment by a team comprising a paediatrician, psychologist and speech pathologist.(27) Waiting times for this assessment can be prolonged in the public system(28) and in the US.(29) As a result, in some countries a child whose parents are socioeconomically disadvantaged may often be diagnosed with ID when a diagnosis of ASD with ID would be more appropriate.

4.3.1 Socioeconomic status

Thirteen studies investigated the parental SES of children with ID or ASD(11, 12, 14, 30–39) and all but two(33, 34) reported a different association of SES with ID than ASD. Children with ID were more likely to be from lower SES families whereas children with ASD were more likely to be from higher SES families.

Low SES was often identified as a risk factor for ID(12, 35–37, 39), especially mild or moderate ID.(11, 14, 39) One of these studies(39) was a cross-sectional study of over five million children that included nearly 250,000 with an intellectual or developmental disability. These researchers concluded that children with mild–moderate ID had an increased risk of exposure to social conditions that were detrimental to their development.(39) Another study examined SES and ID prevalence in the 1966 and 1985–86 Finnish birth cohorts.(37) The researchers concluded that the association of low SES with ID persisted over time. Plausible hypotheses for this ongoing association
are that there had been no improvements in antenatal and obstetric care in those of lower SES over the 20 years in question. Alternatively, one could postulate that there is a prominent genetic involvement in the aetiology of ID. Another is that the higher risk of exposure to a developmentally unfavourable environment has persisted over the 20-year interval in the children of mothers of lower SES.(39)

In contrast to ID, a range of measures of high SES were consistently associated with ASD. In a large telephone interview study in the US,(31) family wealth was used as a measure of SES. The researchers found that children from higher income families were more likely to have a diagnosis of ASD. Similarly, others(38) using family income as a marker for SES found a significant association between high family SES and ASD in the offspring. Further analyses, using the dual markers of high family income and more years of maternal education, found a particular association between high SES and ASD without ID.(38) Using population data and deriving SES from mother’s place of residence at the time of the child’s birth, Australian researchers also found that ASD, ASD with ID and particularly ASD without ID were associated with higher SES.(14)

The overall association between high SES and ASD without ID could result from the increased empowerment of parents of high SES to pursue a diagnosis when their children have a milder variant of ASD.(40) In families of low SES, higher-functioning children with autistic traits might be informally labelled by family and contemporaries as unusual, difficult or emotionally damaged. In a comparable way, lower-functioning children with autistic traits might formally or informally be given a diagnosis of ID. Further, children of parents of lower SES might be more likely to be diagnosed at a later age than those of higher SES and hence not be included in studies of ASD and SES with lower ages of cut-off.(40)

Further evidence of the possible social contributions to the likelihood of an ASD diagnosis was found in a large multi-based national study in the US.(30) Undiagnosed children that met the criteria for ASD had a lower SES than previously diagnosed children. Moreover, SES and the prevalence of ASD were positively associated in a dose–response fashion.(30)
Factors influencing the likelihood of an ASD diagnosis were examined using data on around five million births in Californian cohorts from 1992 to 2000. The researchers found that an interaction between high- and low-level SES measures influenced the likelihood of an ASD diagnosis. Medi-Cal is a program providing medical assistance to the needy in California and these researchers employed family use of Medi-Cal and property values in the area of a mother’s residence as a measure of SES. They reported that children whose families were enrolled with Medi-Cal births and living in wealthier neighbourhoods were two and a half times more likely to receive a diagnosis of ASD than their counterparts living in poorer areas. This could indicate that for parents with limited resources, living in a higher SES neighbourhood had benefits in terms of the likelihood of their child being diagnosed with ASD. This may result from the parents’ increased access to support persons such as paediatricians and child health nurses, and to educational programs such as parent classes and interventions for children, compared to that of similar parents in less affluent areas. In contrast, a Danish study accessing linked population data used maternal education and parental wealth as a measure of SES and found no association between SES and ASD diagnosis. In neighbouring Sweden, a population-based study used low income, manual occupation and less education as measures of low SES. The researchers concluded that low, not high SES, was a risk factor for ASD. There may be a number of reasons for the differing findings of these studies. The universal healthcare and routine screenings offered in Denmark and Sweden may eliminate the ascertainment bias associated with high SES that may exist in other Western countries.

In total, ten studies used education alone as a measure of SES. With ID, all research identified a negative association between high maternal education and the risk of ID in the offspring. For instance, with children with ID and developmental delay without ASD (which may include those with known genetic syndromes), seven studies concluded that mothers were of a lower educational status. One of these, a population study, established that mothers of children with ID were less likely to have more than 13 years of education.

The association of maternal education with varying levels of ID has been investigated. On the basis that risk factors for Down syndrome differed from those of other forms of
ID, children with Down syndrome were excluded. Mothers of children with severe ID were found to be more likely to be of lower educational status than other mothers. Comparable results were found for mothers of children with mild–moderate ID with these mothers having increased odds of a lower educational status than mothers in the general population. One of these studies used Californian service agency records and a sample of more than 27,000 mothers of children with mild–moderate ID or severe ID. They concluded that lower maternal education was also associated with an increased risk of severe ID in the offspring. The relationship was reversed with maternal education and ASD with all four of the studies investigating ASD reporting positive associations between high maternal education and the risk of ASD in the offspring. Three of these studies were from California and each reported that parents of children with ASD were more educated than the general population. The fourth reported that mothers with more than 16 years education were more than twice as likely to have a child with ASD without ID than mothers of a child with only 12 years education.

4.3.2 Marital status

Four groups of researchers examined marital status in relation to the odds of ID or ASD. All but one concluded that mothers of children with ID were more likely to be without partners at the time of their child’s birth. However, in Finland, the negative association between living with a partner and the odds of ID in the offspring, present in a 1966 birth cohort, was absent in the 1985–86 cohort. The reduction of the association in the second cohort may have been a reflection of the improved SES of single mothers over the 20-year period.

In terms of ID, women without a partner had increased odds of having a child with ID and particularly mild–moderate ID. Similarly, a cohort study using UK data, concluded that compared to typically developing children, those with early cognitive delay were less likely to have their biological parents living together during the first five years of their lives compared to families with a typically developing child.

A retrospective cohort study assessed marital status in terms of living with a partner. At the time of the child’s birth, women living with a partner were 35% more
likely to have a child with ASD and particularly ASD with ID. On the other hand, a similar Canadian study found that mothers not living with a partner at the time of their child’s birth were 19% more likely to have a child with ASD than those mothers who were living with a partner.(45)

4.3.3 Parental age

In most studies, increasing maternal age, sometimes along with increasing paternal age, was associated with ASD. A minority of studies found relationships only with paternal age or found no association with either maternal or paternal age. Contrasting results were reported with ID where teenage mothers were more likely to have children with mild–moderate ID and older mothers were particularly likely to have children with severe ID. Socio-demographic and biological explanations are offered.

All ten studies investigating the association of maternal age with the odds of a child with ASD found that advanced maternal age was associated with ASD(14, 21, 38, 47–52) and ASD without ID.(14, 38) Four of these studies(14, 47, 49, 52) reported an additional association with paternal age. For instance, a population-based study using data from multiple sites throughout the US found associations with both maternal and paternal age after adjustment for the other parent’s age, birth order and maternal education.(49)

Four of the cited studies(53–56) reported an association between paternal but not maternal age and ASD in the offspring. In one of these,(56) a case–control study (N = 84), advanced parental age was associated with nearly twice the risk of ASD without ID. Another, population-based cohort study(54) reported more than five times the risk of ASD in the children of men aged more than 40 years compared to men younger than 30 years. The remaining studies used population-based data from Sweden(55) and Denmark.(53) After an adjustment for maternal age, the Swedish researchers identified a linear association of increasing paternal age and the risk of ASD. They reasoned that paternal age could be a risk factor; as generally the male was considered to be the origin of new mutations and these increased with age.(53)
By comparison, two population studies (33, 34) from Scandinavia and a British cohort study \((N = 5,246)(57)\) did not identify advancing maternal or paternal age as risk factors for ASD. As with SES, the results from Scandinavia might reflect the different model of health service provision in Scandinavia. Moreover, there is evidence that children with ASD are diagnosed later in younger mothers.\((58)\) Thus there may be a bias of ascertainment in some studies where younger children are included. In the British study,\((57)\) younger mothers may been included more often since they were recruited when pregnant. Further, a diagnosis of ASD was not required for their child but instead, a parent completed the *Social and Communication Disorders Checklist*. In other studies from the US,\((32, 38, 49, 50)\) Canada\((21)\) and Australia,\((14, 48)\) ASD may be under-ascertained in the children of younger parents, possibly as a result of their lesser confidence to be proactive in the diagnostic process.

Maternal age had a dual association with ID of unknown cause. First, teenage mothers were more likely to have children with mild–moderate ID.\((11, 12, 14)\) Second, older women were more likely to have a child with severe ID.\((12, 59)\) The results of a Finnish cohort study that investigated ID of both known and unknown cause\((37)\) were discounted because of the inclusion of ID of known cause. With Down syndrome, the most common cause of ID, it is known that the risk increases very abruptly with advancing maternal age.\((60)\) This might explain the researchers’ finding of an association between increased maternal age and ID in the offspring seen in the 1966 birth cohort.\((37)\) The finding that the association no longer existed in the 1985–86 cohort may have been due to the introduction or increased uptake of prenatal screening for Down syndrome.

The association of parental and particularly maternal age with ID or ASD suggests that both social and biological forces are operating. Younger parents may find a diagnosis of ASD more difficult to obtain for their children because of inexperience and the demanding navigational requirements of many local systems. Thus, some of the ID diagnoses of their children may be undiagnosed cases of ASD. Further, the excess of older mothers of children with ASD, and to a lesser extent ID, may result from increased *de novo* mutations in older women and their partners\((61)\) or the increase of epigenetic mechanisms that are associated with ageing.\((62)\)
4.3.4 Parity

Parity describes the number of live-born children and stillbirths at more than 20 weeks gestation for a woman. (63) Strong relationships of high parity with ID and low parity with ASD were demonstrated in the majority of studies. Mothers of higher parity had increased odds of having a child with mild–moderate ID (11, 12, 14) whereas one of the research groups (11) concluded that fourth or subsequent children had an increased risk of mild–moderate ID. The Finnish study of two birth cohorts, 20 years apart, found that high parity persisted as a risk factor for ID over time. (37) A large cohort study (12) compared the parity of the mothers of Californian children with ID to the parity of mothers of typically developing children born between 1987 and 1994 and reported that mothers of parity of three or more were 30–50% more likely to have a child with mild–moderate ID or unspecified ID. (12) Both this study and another Californian study reported that mothers of children with severe ID had an elevated but not significantly increased parity compared to mothers of typically developing children. (12, 43)

In women of lower parity, the risks of ASD, (21, 47, 64) ASD with ID (14) and ASD without ID (14, 64) were found to be increased in a number of studies. One of these was a Canadian cohort study using linked databases and with nearly 1,000 case mothers. (21) The authors identified that women having their first child were at the greatest risk of having a child with ASD. Moreover, a national, population-based study in the US reported that older women having their first child with older partners were around three times more likely to have a child with ASD. (49)

Two studies (17, 33) found different associations between parity and the risk of ASD. The first, a prospective cohort study (17), of more than 110,000 mothers in the US asserted that mothers of parity greater than two were more likely to have a child with ASD than other mothers. The second, a Danish case–control study nested in population data, found no association. (33) The first study (17) involved nearly 120,000 nurses that were followed via mailed questionnaires over 16 years. Hence, all mothers were educated, and due to their involvement with nursing could be expected to have more knowledge of ASD than other mothers, on average. Further, parity was assessed as a binary variable with the two values of greater than two and less than or equal to two. Commonly, other studies have defined parity as either a continuous variable or one with
more than two levels, which might account for variations in the findings. Possibly, socio-demographic factors were again operating in the second study. In relation to SES and the odds of ASD, it is possible once again that the disparate findings of this same Danish study may have been due to less ascertainment bias, setting them apart from other studies in the area.

4.3.5 Summary

Socio-demographic factors often operate quite differently for ID and ASD. For example, high parental SES was negatively associated with the risk of ID and positively associated with the risk of ASD in the offspring. Marital status had different associations. At the time of their child’s birth, mothers of a child with ID were less likely to be living with a partner than were mothers of typically developing children. On the other hand, there was no consistent association of marital status with mothers of a child with ASD. Parity had reverse associations for ID and ASD. Compared to mothers of typically developing children, mothers of high parity were more likely to have a child with ID and mothers of low parity were more likely to have a child with ASD. Similar patterns existed for maternal age: mothers of younger age were more likely to have a child with ID than older mothers. However, an additional association existed with older mothers being more likely to have a child with severe ID. In contrast, mothers of an advanced age were more likely to have a child with ASD than were younger mothers.

To some extent, an over-ascertainment of ID and to a greater extent, an under-ascertainment of ASD due to social factors could be contributing to the socioeconomic effects seen with ID and ASD. For instance, in terms of the severity of ASD, researchers in California, with birth cohorts from 1992 to 2000, divided the children with ASD into two groups of equal size where the less severe group comprised children in the top 50% of cases according to level of functioning and the most severe group was the lower 50%. They found that the children from the less severe group were more often found in neighbourhoods that housed wealthier and more educated individuals. Conversely, the same researchers reported that where low SES was measured by a Medi-Cal payment for the birth, the ratio of more severe to less severe cases was always greater than one. The researchers’ interpretation was that the most difficult to diagnose
cases of ASD—that is the less severely affected—were under-ascertained in lower SES populations. (32)

The association of high SES with ASD also might be compounded by some of the characteristics known to be related to mothers of children with ASD. Older women with the support of a partner and with fewer children would seem more likely to achieve a more complex diagnosis requiring more assessments for their child than younger single mothers. Socio-demographic associations with ASD in most Western countries do not appear to operate as strongly and might even be absent in some Northern European countries. This might be due to a different social welfare structure in this region and specifically related to the universal screening for developmental disability. In addition to these and other social factors that could bias ascertainment, biological factors may be operating with older parents.

4.4 Immigrant status and race-ethnicity

The term immigrant describes mothers who give birth while residing in a country that is not their own country of birth. Ethnic describes mothers who belong to a minority race and culture-based group (65) whose country of residence may or may not be their country of birth. To some extent, the groups of immigrant mothers and mothers of minority race-ethnicities overlap. When examining social forces in relation to ID and ASD, it is important to take into account the often-complex process associated with making a diagnosis of ASD.

4.4.1 Immigrant status

Immigrant mothers and intellectual disability

Overall, immigrant mothers were 20–50% less likely to have a child with mild–moderate ID than were non-immigrant mothers. (11, 12, 14) In Australia, immigrant mothers from Asia were less likely to have a child with mild–moderate ID than were non-immigrant mothers. (11, 14) The reversal with mild–moderate ID might be due to the higher SES of immigrants from Asia compared to other immigrant groups. (66–68)
Differing results were found with the association of ID in the children of Mexican immigrants. A study of children with severe ID, and born in California, found that immigrant mothers from Mexico, who would have been likely to be Hispanic, were nearly twice as likely to have a child with severe ID compared to parents born in the US. (43) The idiosyncrasies associated with this immigrant group, compared to those immigrants from more distant locations, might explain this finding. They are likely to be less empowered than their non-immigrant counterparts and their immigration was less likely to be regulated. Hence, from a socio-demographic viewpoint, they could be more likely to present with low SES, which is a risk factor for ID.

Immigrant mothers and autism spectrum disorder

All eight studies of immigrant mothers of children with ASD (13, 14, 22, 48, 53, 69–71) concluded that immigrant mothers were more likely to have a child with ASD and particularly ASD with ID. (13, 14) In relation to immigrant mothers from Asia, an Australian study used birth records and active surveillance to ascertain children with ASD. The authors found that immigrant mothers born in South-East or North-East Asia were more likely to have a child with ASD than were other immigrant mothers. (48) A Western Australian study, using linked population data, also found that immigrant mothers from South-East or North-East Asia were at increased risk of having a child with ASD with ID. (14) A similar situation was described in Sweden where immigrant mothers from East Asia were more than three times as likely to have a child with ASD. (70) As mentioned in relation to ID, this might be due to the higher SES of immigrants from Asia compared to other immigrant groups.

Black immigrant mothers and immigrant mothers from developing countries were more likely to have a child with ASD compared to other immigrant mothers. A study from the UK (69) and another from Sweden (70) reported that Black immigrant mothers (69) and mothers from sub-Saharan Africa (70) were much more likely to have a child with ASD compared to non-immigrant mothers. Further, a small Swedish case–control study compared the prevalence of autistic disorder and pervasive development disorder not otherwise specified (PDD–NOS) in Black children with at least one parent born in Somalia to the prevalence in children without a Somali background. (71) The
researchers reported that these 17 Black women were three to four times more likely to have a child with ASD compared to mothers without a Somali background.(71)

There is some evidence that the intensity of the mother’s skin colour is related to her risk of having a child with ASD. A Swedish study compared the risk of ASD in the children of immigrants from each of North, East and other parts of Africa.(13) The mothers from North Africa were predominantly Moroccan and hence were probably fairer than the other two groups of mothers. For example, the East African group was predominantly from Somalia and Ethiopia while the race-ethnicity of the group from other parts of Africa was not described. The risk of ASD in the North African group was elevated (1.5) but not significantly higher than that of non-immigrant parents. On the other hand, the risk in the East African mothers and mothers from other parts of Africa of having a child with ASD was 2.5 and 3.5.(13)

Immigrant mothers from distant countries and those who emigrated during pregnancy were more likely to have a child with ASD than were other immigrant mothers. For instance, researchers from the UK(69) and Denmark(53) found that immigrant mothers born outside of Europe were more likely to have a child with ASD. Similarly, a Swedish study found that immigrant mothers who were not from either the US or Europe were nearly three times more likely to have a child with ASD compared to mothers from Scandinavian countries.(22) Another Swedish study(13) ascertained that immigrant mothers who emigrated during pregnancy were even more likely to have a child with ASD than mothers who emigrated at other times.

There is evidence that immigrant mothers are at different risks of ASD with ID and ASD without ID. Two Swedish studies found that immigrant mothers (with the exception of those from neighbouring Northern Europe), were less likely to have a child with ASD without ID(13) and Asperger syndrome(70) compared to non-immigrant mothers. One of these studies,(13) along with an Australian study,(14) reported that immigrant mothers were more likely to have a child with ASD with ID. In addition, the Swedish study(13) found that the African immigrant mothers were more likely to have a child with ASD with ID compared to non-immigrant mothers. Similar results were found in a small Swedish case–control study,(71) where all 17 of the Somali children with ASD presented with ASD with ID.
Using data from the 2007 National Survey of Children’s Health it was found that non-immigrant Hispanic children had about twice the prevalence of ASD of immigrant Hispanic children. These results contrasted with those in previous studies of immigrant mothers. The lower likelihood of ASD in immigrant Hispanics compared to non-immigrant Hispanics could be explained by the relative ease of access of Mexican Hispanics to the US. In many countries, immigrants must meet stringent criteria prior to entry and some of these relate to the health of their offspring, their age, wealth, education and occupation. However, Mexican Hispanics would be less likely to experience the same stress, climatic change and exposure to new infections as most other immigrant groups. Also this group was more likely to be of a lower SES than other immigrant groups. Moreover, immigrant parents in other studies have usually relocated from more distant locations. For example, one reported findings that related to immigrants from Somalia to Sweden, another to non-European immigrants to Britain and another to immigrants to the isolated continent of Australia.

Overall, immigrant mothers, and particularly Asian and Black immigrant mothers from distant or developing countries and those who travelled while pregnant, were at a higher risk of having a child with ASD. The mothers at highest risk of a child with ASD were from groups who would be expected to experience the most stress. For example, those relocating from a developing country and those pregnant at the time might be expected to experience higher stress than mothers relocating from a developed country or who are not pregnant. This stress, along with the environmental changes associated with immigration, may have specific and negative effects on the developing fetal central nervous system.

The risk of immigrant mothers having a child with ASD might be further exacerbated by an increased exposure to novel viruses and intrauterine infections. Other hypotheses to explain this association relate to low vitamin D levels and these have been further fuelled by animal studies. A study of rat pups with gross vitamin D deficiencies reported structural brain abnormalities similar to those in children with ASD. Further, ASD was particularly common in the children of Black immigrant women or women of Asian race-ethnicity and darker women more often have a vitamin D deficiency. Generally, immigrant mothers are more likely to have a child with
ASD with ID and less likely to have a child with ASD without ID. This may indicate different aetiologies for these sub-groups.

Along with these biologically based hypotheses, social factors may affect the likelihood of an immigrant mother having a child diagnosed with one of ASD, ASD with ID or ASD without ID. For instance, a diagnosis of ASD without ID would be particularly difficult where the child’s parents were in an unfamiliar country, with a different language and where unusual behaviours might be explained by cultural differences.(13) In Australia, excluding the relatively small group of refugees, and in the US, immigrants from Asia and their children are more often of a higher SES than other immigrant mothers.(66, 67) This might explain why the association with ASD was greater in this group than in other immigrant groups.

4.4.2 Race-ethnicity

In many studies, immigrant status and race-ethnicity were not differentiated. Therefore, within the ethnic group of mothers, there would have been mothers who, due to their country of birth, were both ethnic and immigrant. A recurrent trend in the research is that mothers from minority ethnic groups were more likely to have a child with ID and less likely to have a child with ASD than mothers who were not from minority ethnic groups. Overall mothers from ethnic minority groups were more likely to have a child with ID. Asian mothers giving birth in California were 40% more likely to have a child with severe ID, although this result was not significant.(12) Hispanic mothers were more likely to have a child with either mild–moderate or severe ID than were Caucasian mothers. Again, in each of the ID groups, the results narrowly failed to achieve significance.(12) By comparison, Australian Aboriginal mothers were more than three times as likely to have a child with mild–moderate ID and were 60% more likely to have a child with severe ID.(11, 14)

Compared to ID, there was a reverse situation with ASD with epidemiologists finding that mothers from ethnic minorities and particularly Aboriginal mothers were less likely to have a child with ASD. For example, in the US, Hispanic mothers were less likely to have a child with ASD compared to non-Hispanic mothers.(15, 50, 75) In New York, the prevalence in Latinos was around half that in non-Latinos.(31) Some of these
mothers from minority ethnic groups would have also been immigrant and, as reported, immigrant mothers usually have higher rates of ASD. This means that mothers from ethnic minority groups and who are native to their country might be expected to have the lowest likelihood of a child with ASD. This is the case in both Australia and Canada, where Aboriginal mothers had about half the odds of having a child with ASD compared to non-Aboriginal mothers.(14, 21)

The higher rates of ID and the lower rates of ASD found in most ethnic minority and particularly Aboriginal communities may relate to the differing gene frequencies of these groups from the general population. However, differences could be exacerbated by environmental factors such as maternal alcohol consumption,(76) without this being specifically identified as an aetiological factor.(77) Another consideration could be that marginalised groups are less empowered than others to pursue a diagnosis of ASD in contrast to a diagnosis of ID, and that the infrastructures established for diagnostic assessment do not meet their needs. This second factor is likely to account for the higher prevalence of ID and lower prevalence of ASD in the children of Australian Aboriginal mothers.(78)

In two Californian studies, contrasting findings were found for the previously described associations of race-ethnicity with ASD. First, a cohort study found that Hispanic mothers were no less likely to have a child with ASD with ID than were white mothers.(43) The same study also reported that Black Californian mothers were more than five times as likely to have a child with ASD with ID than were white mothers.(43) Further, Californian Asian mothers were almost four times as likely to have a child with ASD with ID than were white mothers.(43) Again, this may be a reflection of the higher proportion of immigrants in these groups. A second explanation in relation to the Asian mothers could be the fact that Asian mothers in the US tend to have a higher SES than most other mothers of minority race-ethnicities. The second of the two studies reported that Asian mothers giving birth in California were 30% less likely to have a child with mild–moderate ID.(12) This may also be a reflection of their higher SES.
4.4.3 Summary

Generally, non-immigrant mothers and particularly Aboriginal mothers were more likely to have a child with ID (especially mild–moderate) than were mothers who were not ethnic. Immigrant mothers, and especially Black immigrant mothers and immigrant mothers from Asia, were more likely to have a child with ASD compared to non-immigrant mothers. Further, immigrant mothers were more likely to have a child with ASD with ID and less likely to have a child with ASD without ID compared to non-immigrant mothers. Immigrant mothers from distant or developing countries and mothers who emigrated when they were pregnant were even more likely to have a child with ASD. By contrast, in the US, Hispanic immigrant mothers were less likely to have a child with ASD than were non-immigrant Hispanic mothers.

4.5 Health and associations

4.5.1 Mental health

The World Health Organization describes mental health as a state of mental well-being.\(^{(79)}\) This state of well-being can be enhanced by the prevention of mental disorders and the treatment and rehabilitation of those with mental disorders. Compromised mental health has been reported in the mothers of children with ID, and to a greater extent in the mothers of children with ASD than other mothers. However, this difference may be due to the paucity of research on psychiatric disorders in mothers of children with ID, which was identified in Chapter 3.

Studies have most commonly investigated the mental health of mothers of children with disabilities rather than the developmental outcomes in children born to mothers with mental health diagnoses. In one case–control study, the latter approach was employed and linked data from population-based registries were used to compare the likelihood of ID or ASD with ID in the children of more than 3,000 mothers with schizophrenia, bipolar disorder or unipolar major depression to the likelihood of these disorders in the children of control mothers.\(^{(80)}\) Children of mothers with either schizophrenia, unipolar major depression, bipolar disorder or a combination of these disorders were about three times as likely to have a child with ID as mothers without these disorders.\(^{(80)}\) Further,
mothers with ID themselves were more likely to have a child with ID compared to mothers with no history of psychiatric disorder or ID. The same study found that around 1,300 mothers had bipolar disorder and these were assessed as nearly ten times more likely to have a child with ASD with ID than mothers without these disorders. However, there were only four children with a mother with pre-existing bipolar disorder so these large odds are associated with particularly wide confidence intervals, and only just reached significance.

Research has also found that mothers of a child with ASD were more likely to have a pre-existing psychiatric or personality disorder than mothers of typically developing children. Further, parents of a child with ASD were more likely to have affective disorder, obsessive–compulsive disorder, anxiety, paranoia and somatization than the parents of typically developing children. One of these studies was conducted by a Californian team and recruited 269 parents of children with ASD via an existing university research program and control parents (students, or their contacts, at the university) of typically developing children. Self-reported mental health measures were obtained via questionnaire. Other reported associations with parents of a child with ASD were increased rates of schizophrenia, psychosis and depression compared to the parents of typically developing children. Mothers of a child with ASD were more likely to have had pregnancies complicated by depression than mothers of typically developing children. When the rates of mental disorders in parents of people with ASD were compared to those in parents of people with Down syndrome, it was found that the rates of anxiety disorder were higher in the former group.

### 4.5.2 Broad Autism Phenotype

The Broad Autism Phenotype (BAP) was introduced in Chapter 2. It is measured in the five domains of social skills, communication, attention to detail, attention switching and imagination. Researchers from the UK conducted a case–control study comprising parents of children with and without ASD from more than 1,500 families. Parents of children with ASD were more likely to exhibit autistic-like traits in all domains, except that of attention to detail, than other parents. Further, researchers found that a BAP occurred more commonly in parents of children with simplex ASD (where only
one family member has ASD) and multiplex ASD\(\text{(90, 91)}\) (where more than one family member has ASD) than in parents of typically developing children. A dose–response effect was also described with parents in multiplex ASD families expressing a BAP significantly more often than parents in simplex ASD families.\(\text{(92)}\)

Some factors associated with maternal mental health may have a deleterious effect on the fetus and increase the likelihood of a child developing ID or ASD. For example, mothers with schizophrenia may remain on antipsychotic drugs during their pregnancies and these drugs, perhaps along with lower levels of self-care (such as diet and medical care) and genetic factors related to the disease may adversely affect development in the fetus. The milder autistic features in the parents of children with ASD might also be attributable to genetic factors associated with ASD.\(\text{(93)}\) In their affected children, these factors, along with additional genetic factors from the other parent, may sometimes produce the clinical phenotype of ASD.

### 4.5.3 Height and weight

Compared to the mothers of typically developing children, differing associations between maternal height and the mothers of children with ID or ASD were identified. Using population data, researchers identified that shorter women and those of medium height were more likely to have a child with mild–moderate ID than other mothers.\(\text{(11)}\) Further, the shortest group of women were more likely to have a child with severe ID than other women.\(\text{(11)}\) The same study also identified that mothers of children with mild–moderate ID were significantly shorter than the mothers of typically developing children. However, the mothers of children with severe ID were not significantly shorter than the mothers of typically developing children.\(\text{(14)}\)

Using linked data, researchers identified that mothers of children with ASD were significantly taller, particularly those of children with ASD without ID compared to the mothers of typically developing children.\(\text{(14)}\) Another study\(\text{(85)}\) found that mothers of children with ASD were both taller and heavier than mothers of typically developing children. Similarly, a Canadian population study found that among non-smoking women, taller and heavier women were more likely to have a child with ASD compared
to the mothers of typically developing children. However, associations have also been described between SES and height, and so the observed height differences may just be a reflection of the different mean SES of mothers of children with ID, ASD or no disability.

4.5.4 Health behaviours

Smoking

Researchers reported limited associations of smoking during pregnancy and ID or ASD in the offspring. A population study in the US ascertained that mothers who smoked 20 or more cigarettes a day were more likely to have a male child with ID than mothers who did not smoke during pregnancy. A large Finnish cohort study found that mothers who smoked after two months of pregnancy were no more likely to have a child with ID than mothers who did not smoke after this time. The definition of smoking was broader than in the first study so the likelihood of identifying an association between maternal smoking and ID would have been reduced. Further, neither smoking during the first two months of pregnancy nor gender of the fetus was considered for inclusion in the model. An alternative study, with a more stringent definition of maternal smoking and that addressed these omissions would have been more likely to discern an association.

The findings in relation to ASD are also limited. Researchers reported that mothers who smoked during pregnancy were more likely to have a child with ASD than non-smoking mothers. A Swedish nested case–control study using medical registry data, found that mothers who smoked during early pregnancy were 70% less likely to have a child with ASD (but not Asperger syndrome) and twice as likely to have a child with Asperger syndrome. This raises the possibility that Asperger syndrome has a distinct aetiology from other forms of ASD. On the other hand, differing results were found in a US population-based, case–cohort study. Data from more than six million mothers and their children were adjusted for potential confounders such as maternal age, education and marital status. The definition of smoking during pregnancy was not described, so presumably this encompasses all mothers who had admitted to smoking one or more cigarettes. The researchers reported that mothers who smoked during pregnancy were no more likely to have a child with ASD than mothers who did...
Chapter 4

not. Using two large cohort studies, researchers found no association between mothers who smoked during pregnancy and ASD. (21, 97) However, the first study examined only associations between maternal smoking and ASD generally. (21) Consequently, any association between the relatively small group of mothers of children with Asperger syndrome may have been lost in the broader analysis. In addition, smoking was defined as any smoking during pregnancy, which may have lessened the likelihood of an association with ASD. There were different outcomes in the second study, (97) where mothers of children with ASD with ID and ASD without ID were considered separately and mothers who smoked 10 or more cigarettes a day were a distinct group from the less intense smokers. (97) Researchers asserted that, in Sweden at least, smoking during pregnancy was not associated with increased risk of ASD, either with or without ID. They considered associations reported in previous studies as being attributable to confounding by socio-demographic variables. (21)

**Alcohol consumption**

Mothers who consumed excessive alcohol during pregnancy were assessed as more likely to have a child with ID. This cause of ID is termed fetal alcohol syndrome (FAS) or more broadly, fetal alcohol spectrum disorder (FASD). Studies from Sweden (98) and the US (98) attributed between 2–10% of mild–moderate ID to FAS or FASD, and researchers who conducted a US study considered that a further 3% of severe ID was associated with these diagnoses. (99) In WA, heavy prenatal alcohol exposure was found to be an important cause of ID, accounting for 2.5% of non-genetic ID. (76) Further, under-ascertainment is likely due to the stigma women associate with the disclosure of heavy alcohol consumption during pregnancy. (100) In addition, perhaps due to inadequate training, (101) clinicians may lack awareness and confidence in making this diagnosis (100) or believe that it would not be beneficial to the mother or child.

**4.5.5 Summary and conclusion**

Large cohort studies and linked data have provided researchers with the opportunity to study populations of mothers and their children with and without ID or ASD. Data can also be adjusted for a range of possible confounders such as SES and age. This enables
the identification of new risk factors for ID or ASD and the elimination of others. For example, the association of smoking during pregnancy with ID or ASD in the offspring has weakened in the most recent studies using linked population data. Persisting associations are an increased risk of Asperger syndrome and PDD–NOS in the children of mothers who smoked during pregnancy and an increased risk of ID in the male children of mothers who smoked heavily during pregnancy. Maternal alcohol consumption during pregnancy remains a risk factor for ID. The remaining associations of maternal smoking with ID or ASD in the offspring could result from the effect of this exposure on overall fetal development and particularly growth restriction,(102, 103) preterm birth(102) and low birth weight(104) Moreover, sub-optimal fetal growth has been associated with mild–moderate ID in Caucasian children.(105) The association of maternal alcohol consumption with ID might be due the multiple effects of alcohol on the fetus and placenta,(106) For example, alcohol can induce oxidative stress in placental villous tissue. Other demonstrated effects are an increase in neural tube defects and increased heart rate and cortisol levels in the exposed infant.

4.6 Physical health

The research literature has provided evidence that maternal physical health, both prior to and during pregnancy is related to the likelihood of a mother having a child with ID or ASD. Various pre-existing conditions in the mother and related or unrelated complications of her pregnancy increase the likelihood of a mother having a child with ID or ASD compared to mothers who do not have the condition.

4.6.1 Obesity

Pre-pregnancy obesity is an example of a condition that increases the likelihood of a woman having a child with ID or ASD. Obese women were more likely to have a child with ID(16, 37) or ASD(16, 45, 107) than women who were not obese. Of these, a Finnish study used linked data from the birth cohorts of 1966 and 1985–86 and around 250 mothers of children with ID were in each of the cohorts.(37) In both cohorts, mothers with prior obesity were more likely to have a child with ID than those without. Interestingly, the association of ID with pre-pregnancy obesity was an increasing risk
reflected in the greater odds of the later cohort (2.4 versus 1.8). Another studies(107) reported that women with an early age of menarche were more likely to have a child with ASD than other women. The increased odds associated with early menarche and pre-pregnancy obesity could indicate the possibility of maternal hormonal involvement in the risk of ID and ASD.(107) Then again, the relationship with ID may be resulting from confounding by the association between socioeconomic disadvantage and obesity in highly developed countries.(108) In the light of the increasing prevalence of obesity in these countries, these associations with ID and ASD are an important direction for future research.(109)

4.6.2 Autoimmune and immune function

With respect to maternal autoimmune disorders, the association with subsequent children with ID (described as a developmental disability other than ASD) was weak compared to that with ASD. For example, in a case–control study using linked data with more than 1,200 cases, mothers with an autoimmune disorder were 60% more likely to have a child with ASD than mothers without an autoimmune disorder.(18) Researchers conducted a case–control study (N = 61) using questionnaires to evaluate the frequency of autoimmune disorders in families of children with ASD compared to other families.(110) They reported a dose–response association. In families with one autoimmune disorder the risk of a child with ASD was about double and in families with three autoimmune disorders this climbed to more than five. Other studies(111, 112) have found that women with a particular autoimmune disease were more likely to have a child with ASD than women who did not have the disease. Results of a larger case–control study with 407 cases(111) indicated that women with psoriasis were more likely to have a child with ASD than mothers without this disorder.(111) Another research group(112) used linked data with more than 3,000 mothers of a child with ASD and nearly 700,000 control mothers. They reported that women with rheumatoid arthritis or coeliac disease were more likely to have a child with ASD than mothers who did not have the one of these disorders.(112)

Women with pre-existing or gestational diabetes were more likely to have a child with ID or ASD. For instance, two studies(16, 113) identified an association between gestational diabetes and ID or a condition similar to ID. One was a large retrospective
cohort study(113) using linked registry data. Here the researchers found that mothers with diabetes during pregnancy were nearly 70% more likely to have a child with mild–moderate ID compared to mothers without this disorder. The other research group(16) found that mothers with diabetes during pregnancy were nearly two and a half times more likely to have a child with developmental disability other than ASD (IQ<70) than mothers without diabetes during pregnancy. Other researchers compared more than 160,000 mother–child dyads and identified that mothers with pre-existing diabetes were more than 10% more likely to have a child with ID.(114)

The association with ASD was also found by multiple independent research groups. For example, researchers(21) reported that women with pre-existing diabetes were more likely to have a child with ASD than women without pre-existing diabetes. Two studies(17, 113) found that mothers with diabetes during pregnancy were more likely to have a child with ASD(17) and ASD with ID(113) than mothers without the disorder. The first of these was a US population study(17) that included nearly 800 cases of ASD and more than 66,000 births. Mothers who developed gestational diabetes were associated with a 76% increased risk of ASD compared to women who did not develop the condition.(17) The second was an Australian population study(113) that found that mothers who had diabetes during pregnancy were nearly three times as likely to have a child with ASD with ID than mothers without diabetes.(113) More-attenuated results were reported by Californian researchers(16) that conducted a case–control study (500 controls) and compared the mothers of children with ASD and mothers of children with developmental disabilities other than ASD, to typically developing children.(16) They found that mothers with gestational diabetes were more likely (but not significantly) to have a child with ASD than mothers without the disorder. Similarly, a case–control study (408 controls) used linked data from national birth and inpatient registries and reported that women with pre-existing diabetes had nearly twice the odds of having a child with ASD than other mothers but again the results were not significant.(22) The lack of significance may be due to the reduced power of these smaller studies.

Maternal immune dysfunction has been associated with the neurological development of the fetus.(115) A case–control study(116) identified that mothers of children with ID or ASD were more likely to have aberrant cytokine (immune response regulator) profiles compared to the mothers of typically developing children. In this study, the
concentration of serum cytokines at mid-pregnancy in the mothers of children with developmental disabilities other than ASD, ASD with ID, and typically developing children were compared. Mothers of a child with a developmental disability other than ASD were more likely to have higher concentrations of three particular cytokines than mothers of a typically developing child. Further, mothers of a child with a ASD with ID were more likely to have higher concentrations of three different cytokines (to the non-ASD group) than mothers of typically developing children.(116) Differences have been identified relating to the immunological status of mothers of children with ASD prior to their pregnancies. The results of an independent case–control study(117) showed fetal brain antibodies in mothers of children with ASD but not in control mothers. This study found that mothers of children with ASD were significantly more likely to have an auto-antibody reactivity pattern for human fetal brain proteins than mothers of typically developing children.

Autoimmune diseases and idiosyncrasies of the immune system might impinge on the immature nervous system of the developing fetus. This could have a deleterious effect on future cognitive function(118) and increase the likelihood of ID and ASD.(116)

4.6.3 Pre-eclampsia and symptoms

Pre-eclampsia is a condition occurring in about 8% of first pregnancies(119); common symptoms are hypertension and oedema. Women that experienced pre-eclampsia were more likely to have a child with ID or ASD. For instance, researchers conducting a population-based, retrospective cohort study in the US(120) concluded that women that suffered pre-eclampsia were nearly 60% more likely to have a child with ID.

Three groups of researchers found that women with pre-eclampsia(17, 21, 121) and those suffering oedema(84) during their pregnancies were more likely to have a child with ASD than other pregnant women. A smaller case–control study (206 controls), found that women with pre-eclampsia had reduced (though not significantly so) likelihood of a child with ASD.(51)

Hypertension during pregnancy was associated with an increased risk of a child with ASD but not ID. Three studies(16, 22, 45) found that women with hypertension during
pregnancy were more likely to have a child with ASD than women without hypertension. In one of the studies,(22) Swedish researchers conducted a nested, matched case–control study with data from over 400 children with ASD and over 2,000 controls. Records of children’s hospitalisation over 10 years were linked to birth records. The researchers concluded that mothers who suffered a hypertensive disease during pregnancy were 60% more likely to have a child with ASD than other mothers. In contrast, a large cohort study of more than 65,000 nurses found that mothers with hypertension during pregnancy were no more likely to have a child with ASD than mothers without hypertension during pregnancy.(17) This study population consisted entirely of nurses. Perhaps, with their increased medical knowledge, these nurses more often sought treatment before their blood pressure reached a level that would have been damaging to the unborn child.

4.6.4 Epilepsy

A retrospective cohort study was conducted of nearly 3,000 mothers of children with ID of unknown cause or ASD and around 237,000 mothers of typically developing children using linked population data from medical registries.(113) This established that mothers with epilepsy during pregnancy were more than three and a half times as likely to have a child with mild–moderate ID and more than four and a half times as likely to have a child with ASD with ID compared to mothers without epilepsy during their pregnancies. A case–control study in the US(110) had only 61 control mothers of a child with ASD. Here mothers who had experienced seizure prior to their pregnancies were nearly six times as likely to have a child with ASD. However, possibly due to the small size of the study, results did not reach significance.

4.6.5 Medication use

There is always a risk that the use of certain medications during pregnancy may have adverse effects on a developing fetus. Sometimes, medications initially considered safe have been later implicated to adversely affect the unborn child. For instance, five studies(55, 85, 122–124) found that the children of mothers who used anti-depressants,(55, 122, 123) anti-convulsants,(124) psycho-active drugs,(55) prescribed
medications (45, 85) and medications generally (124) had a higher risk of a child with ASD. One of these studies (55) was a population-based case–control study in Sweden. Using registry data, the researchers assessed that mothers who took psycho-active drugs or anti-depressants during their pregnancies were more than four times as likely to have a child with ASD. (55)

It is also possible that the increased use of prescribed medications in mothers of children with ASD may have resulted from a bias in data collection. In one of the studies finding an increased use of prescribed medications, mothers were recruited via their association with a support agency. (85) This method might have resulted in a bias in the direction of a high SES that, in turn, may have produced an increased use of prescribed medications in the case mothers. Nonetheless, the study that found an increased use of medications generally was a population study using medical registries. (124) The reported associations are likely to be mediated by a complex interaction of factors. For instance, in addition to possible SES bias, there could be a genetic association such as the familial link of depressive disorders or epilepsy with ASD. Another possibility is an environmental effect that results from the physiological effect of medication use on the uterine environment.

### 4.6.6 Other correlates

Mothers who experienced a range of other conditions pertaining to their physical health during pregnancy were found to be more likely to have a child with ID or ASD than other mothers. For example, asthma during pregnancy was associated with ID, with pregnant women with asthma being more likely to have a child with mild–moderate ID than mothers without this condition during pregnancy. (113) An Australian population study found that women who had renal or urinary conditions during pregnancy were more than twice as likely to have a child with mild–moderate ID as women without these conditions during pregnancy. (113) Further, women who suffered anaemia during their pregnancies were more than five times as likely to have a child with severe ID than women without anaemia during pregnancy. (113)

Infections during pregnancy were associated with ID. For example, one study (125) reported that mothers who suffered trichomoniasis during pregnancy were more likely
to have a child with ID than mothers without this condition during pregnancy. A cohort study used Medicaid claims and linked infant records to investigate the association of treated and untreated urinary tract infections during pregnancy with later ID in the child. The researchers reported that pregnant women with untreated urinary tract infections were 30% more likely to have a child with ID than other pregnant women. Moreover, mothers with untreated urinary tract infections were 22% more likely to have a child with ID than mothers with antibiotic-treated urinary tract infections.

Other health issues during pregnancy were also associated with a higher risk of ASD. For instance, researchers reported that women who had allergies, asthma, bleeding or high body temperature during pregnancy were more likely to have a child with ASD than other women. Infections during pregnancy were also associated with ASD with researchers reporting that women with pregnancies complicated by urinary tract and other infections were more likely to have a child with ASD than mothers without infections.

### 4.7 Summary

Before the birth of their affected children, certain socio-demographic, health and physical attributes differentiate mothers of children with ID or ASD from those of mothers in the general population. Further, these attributes often vary by the disability group of their child. In Tables 8–10, these differences are grouped into categories according to their associations with groups of mothers. An examination of Table 8 shows that with socio-demographic factors, the relationships with ID and ASD are most often reversed. First, low SES was most often associated with the ID; and high SES, with ASD.

Different associations of marital status were found for ID and ASD. With all but one study, single mothers were at increased risk of mild–moderate ID and unspecified ID, compared to women who were living with a partner. With ASD, the only two studies found in the area reported contrasting results.
With maternal age and ID, two associations emerged. Younger mothers had an increased risk of bearing a child with mild–moderate ID, but severe ID was associated with increased maternal age. In the majority of the studies, increased maternal age, along with increased paternal and parental age, were associated with ASD and ASD without ID.

Higher parity had a consistent positive association with mild–moderate ID and unspecified ID in most studies. However, with severe ID, there was no association. With, ASD, the relationship was reversed and the association was with lower parity.

In Table 9, the associations with immigrant status and race-ethnicity are summarised. Most often, immigrant mothers are more likely to have a child with ASD and ASD with ID than non-immigrant mothers. On the other hand, ASD without ID was associated with non-immigrants, excepting those immigrants from nearby countries. The Mexican/Hispanic immigrant mothers in the US were a separate group since these mothers were less likely to have a child with ASD than Mexican/Hispanic non-immigrant mothers. With race-ethnicity, the associations differed from those with immigrant status, in spite of the overlap between the groups. Except for Asian mothers, mothers from ethnic minority groups (and particularly Aboriginal mothers) were at an increased risk of a child with ID. With the exception of Black mothers or those of Asian race-ethnicity, the situation was reversed for ASD since mothers from ethnic minority groups were at a lower risk of children with ASD compared to Caucasian mothers.

Table 10 shows the associations of health and other characteristics that are common to mothers of children with ID and mothers of children with ASD. The quite small proportion of characteristics common to mothers of children with ID and mothers of children with ASD are highlighted in the same colour. With mental health, only one study found an association with the mothers of children with ID. In contrast, ten research groups reported associations with ASD. Autistic-like traits were associated only with the parents of children with ASD.

As with other socio-demographic factors, ID and ASD had an overall reverse association with height. Shorter women were more likely to have offspring with ID whereas taller and heavier women were more likely to have offspring with ASD. The
associations with maternal smoking during pregnancy were minimal. Excessive alcohol consumption during pregnancy was only associated with ID whereas obesity was associated with both ID and ASD.

Both ID and ASD had associations with autoimmune and immune function, though associations with ASD were more often described. Both pre-existing diabetes and diabetes during pregnancy were associated with ID and ASD. Further, abnormal levels of cytokines during pregnancy were also associated with each of ID and ASD. Other associations with only ASD were autoimmune dysfunction generally, psoriasis, rheumatoid arthritis, celiac disease and maternal fetal brain antibodies.

Only one study associated pre-eclampsia with ID whereas seven studies associated hypertension, oedema and pre-eclampsia with ASD. Epilepsy and asthma had associations with both ID and ASD but no other associations during pregnancy were common to both disorders. Medication use during pregnancy was associated with ASD.

4.8 Conclusion

This chapter provides a review of the research pertaining to the pre-existing characteristics of mothers of a child with ID or ASD. Some consistent and enduring associations have emerged. With socio-demographic factors, these are the contrasting associations of maternal education, age, immigrant status and race-ethnicity with ID and ASD. With maternal health, aspects of mental health and autoimmune function have weaker associations with the mothers of children with ID than ASD. Further, the BAP and medication use during pregnancy only have associations with ASD. Some of these differences may be reflections of distinct aetiologies for ID and ASD of unknown cause and provide me with directions for future research. As such, primary and secondary prevention strategies may be refined or developed, which will contribute to lower prevalence, reduced levels of severity and better outcomes for affected families.
### Table 8: Socio-demographic associations in the mothers of children with ID or ASD

<table>
<thead>
<tr>
<th>Category</th>
<th>Intellectual disability</th>
<th>Autism spectrum disorder</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mild–moderate ID</td>
<td>Severe ID</td>
</tr>
<tr>
<td>SES</td>
<td>-ve assoc(11, 14, 39)</td>
<td>-ve assoc(12, 35-37, 39)</td>
</tr>
<tr>
<td>SES (Denmark &amp; Sweden)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Education</td>
<td>-ve assoc(11, 12)</td>
<td>-ve assoc(12, 43)</td>
</tr>
<tr>
<td>Marital status at child’s birth</td>
<td>-ve assoc(5)</td>
<td></td>
</tr>
<tr>
<td>(Women with partners)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Paternal/ maternal</td>
<td></td>
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<tr>
<td>Paternal only</td>
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<td>Paternal only</td>
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<td>Paternal /maternal</td>
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<tr>
<td>UK/Denmark/Sweden</td>
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<tr>
<td>Lower parity</td>
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<tr>
<td>Lower parity &amp; age</td>
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**ID**, intellectual disability; **ASD**, Autism spectrum disorder; **Mild–moderate ID**, Mild or moderate ID of unknown cause; **Severe ID**, Severe or profound ID of unknown cause; **SES**, socioeconomic status; **+ve**, positive; **-ve**, negative; **assoc**, association; **DD**, developmental disability; **w/out**, without.

**Note:** **+ve assoc** are shaded green, **-ve assoc**, yellow and **No assoc**, blue.
### Table 9: Immigrant status and race-ethnicity in the mothers of children with ID or ASD

<table>
<thead>
<tr>
<th>Category</th>
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<th>Autism spectrum disorder</th>
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</thead>
<tbody>
<tr>
<td></td>
<td>Mild–moderate ID</td>
<td>Severe ID</td>
</tr>
<tr>
<td>Immigrant status</td>
<td></td>
<td></td>
</tr>
<tr>
<td>(immigrant vs non-immigrant)</td>
<td>-ve assoc(11, 12, 14)</td>
<td>+ve assoc(13, 14)</td>
</tr>
<tr>
<td>Northern Europe &amp; UK</td>
<td></td>
<td></td>
</tr>
<tr>
<td>US</td>
<td>+ve assoc Mexican immigrants in California(43)</td>
<td>+ve assoc in East Asians(14)</td>
</tr>
<tr>
<td>From Asia</td>
<td>-ve assoc for immigrants from Asian in Australia(11, 14) &amp; US(5, 6)</td>
<td>+ve assoc in East Asians(14)</td>
</tr>
<tr>
<td>Race-ethnicity</td>
<td>+ve assoc in Aboriginals(11, 14)</td>
<td>+ve assoc in Aboriginals(11,14)</td>
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<tr>
<td>(non-Caucasian vs Caucasians)</td>
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ID, intellectual disability; ASD, Autism spectrum disorder; Mild–moderate ID, Mild or moderate ID of unknown cause; Severe ID, Severe or profound ID of unknown cause; +ve, positive; -ve, negative; assoc, association; SE, South-eastern; NE, North-eastern.

Note: *+ve assoc* are shaded green, *-ve assoc*, yellow and *No assoc*, blue.
<table>
<thead>
<tr>
<th>Category</th>
<th>Intellectual disability</th>
<th>Autism spectrum disorder</th>
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<tbody>
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<td>Unspecified ID</td>
<td>ASD with ID</td>
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<td>Schizophrenia(80)</td>
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<td>Unipolar major depression(80)</td>
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<td></td>
<td>Bipolar disorder(80)</td>
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<td></td>
<td>ID in mother(80)</td>
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<tr>
<td>Other auto-immune disorders</td>
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<td></td>
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<tr>
<td>Pre-eclampsia &amp; symptoms</td>
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<td>Pre-eclampsia(120)</td>
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<tr>
<td>Other correlates</td>
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<tr>
<td></td>
<td>Renal/urinary conditions (Mild-mod. ID)(113)</td>
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<td></td>
<td>Anaemia (Severe ID)(113)</td>
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<tr>
<td></td>
<td>Trichomoniasis(125) &amp; untreated UTI(126)</td>
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</tr>
<tr>
<td></td>
<td>Asthma (Mild-mod. ID)(113)</td>
<td></td>
</tr>
</tbody>
</table>

ID, intellectual disability; ASD, Autism spectrum disorder; Mild-mod. ID, Mild or moderate ID of unknown cause; Severe ID, Severe or profound ID of unknown cause; Gest, Gestational; DD, developmental disability; ~, not; BAP, Broad Autism Phenotype; abs, antibodies; UTI, urinary tract infection.

Note: Common or related associations of ID and ASD are shaded the same colour.
4.9 References


Chapter 5: Exploring pre-existing differences of mothers of children with ID or ASD

In Chapter 4, I reviewed the literature describing research into the pre-existing differences of mothers of children with intellectual disability (ID) and mothers of children with autism spectrum disorder (ASD) compared to the mothers of typically developing children, before the birth of their children. I considered that my further investigation into three particular areas of difference was most warranted. First, maternal immigration status and race-ethnicity were each described as influencing the odds of having a child with ID or ASD. Moreover, I had found no study that had explored combinations of maternal race-ethnicity, immigration status and region of birth as independent covariates and the risk of ASD in a subsequent child while adjusting for demographic characteristics associated with ASD. Second, pre-existing maternal psychiatric disorder had been associated with ASD but I had found only one paper that examined maternal psychiatric disorder in relation to ID. I wanted to explore ID, ASD and pre-existing psychiatric disorder in a more ordered way than the studies that I had located. Third, I had located research where the authors had detailed the Broad Autism Phenotype (BAP) in mothers of children with ASD. These authors seemed to assume that some mothers had always exhibited this phenotype and that the BAP was immutable. It was as though they had not considered the possibility that years of managing challenging behaviours and sleep disorders in a child with ASD, along with the associated stigma and social isolation, might affect a person’s sociability and preferred past-times. In Part 1 of this chapter, I reported my research into maternal immigration status, race-ethnicity, birthplace and the risk of a child with ASD. Part 2 is devoted to my research that explored the risk of a subsequent child with ID or ASD in mothers with a psychiatric disorder. In the final part, I describe aspects of my qualitative data (Chapter 8) where mothers reported changes in their personality styles and social life after the onset of their child’s disability. I have discussed how such changes would be likely to exacerbate the assessment of their BAP. The abstracts of the submitted manuscripts associated with Parts 1 and 2 comprise Appendices 3 and 4 and a copy of the publication associated with Part 3 is included as Appendix 5.
5.1 Race-ethnicity, immigrant status, birth region and odds of a child with ASD

5.1.1 Introduction

Autism spectrum disorder is a debilitating, lifelong neurodevelopmental disorder characterised by impairments in social interaction, communication, and repetitive, stereotyped behaviours. (1) Intellectual disability is diagnosed in persons with an IQ score <70 and impairments in adaptive functioning that occur before 18 years of age. (1) From 30 to 60% of persons with ASD have comorbid ID. (2, 3) In some countries, the prevalence of diagnosed ASD is reported to be lower among children of mothers from minority ethnic groups. For example, Indigenous mothers in Australia (4) and Canada (5) have a lower prevalence of ASD in their children than Caucasian mothers. In the US, children born to Hispanic and African American mothers have lower rates of diagnosed ASD than children born to Caucasian mothers. (2) Additionally, the severity of identified ASD appears to vary across ethnic groups. For example, the children of Hispanic, Latino, Asian and Black women have been found to have higher rates of ASD with ID in their children than Caucasian women. (6-8)

Many previous studies examining racial–ethnic sub-groups did not explore the immigration status of the women. However, some studies suggested that the prevalence of ASD among the children of immigrant mothers was often different from the prevalence in children of native-born mothers. Several studies have found that the prevalence of ASD in the children of immigrant mothers was higher than the prevalence among children of native-born mothers. (4, 9–13) Additionally, children born to immigrant mothers were more likely to have ASD with ID than ASD without ID. (4, 9) In a Swedish study, (14) immigrant mothers born outside of Europe or North America were three times more likely to have a child with ASD than children of native-born mothers. On the other hand, children of immigrant mothers from Europe and North America had only a slightly increased risk compared to children of native-born mothers. (14) Similarly, a British study reported that the children of immigrant mothers born outside of Europe, and particularly in the Caribbean, had higher rates of ASD than the children of non-immigrant mothers. (13) Although these findings are intriguing, not
all studies reported positive associations between maternal immigration status and ASD. A population-based study of US children reported that children born to immigrant Hispanic mothers had significantly lower rates of ASD than children of non-immigrant Hispanic mothers or Caucasian mothers.(15)

In the studies to date, it is not clear whether the association between maternal immigration status and the risk of having a child with ASD is primarily due to maternal race-ethnicity, immigrant status or associated factors such as the region or birth, the income status of the country, the distance from the parent country, (16) stress, (17, 18) nutrition (19) or other exposures of the mother. Although studies often do not separate race-ethnicity and immigration status, it is recognised that in the general population, mothers from ethnic minority groups often have a lower risk of having a child with ASD than Caucasian women. (4) Thus, the findings from some studies of high ASD rates among the children of immigrant mothers from ethnic minority groups are particularly unexpected. Further, the risk of an immigrant mother having a child with ASD may vary according to her race-ethnicity. For example, children born in the UK,(13) Sweden(16, 20) and US(21) to Black immigrant mothers from Africa, and particularly East Africa,(16, 21, 22) have been found to have higher rates of ASD with ID compared to children of native-born mothers. Higher rates of ASD with ID, were also identified in children born in Western Australia (WA) to mothers born in East Asia(4) compared to Caucasian mothers and in Malmoe, Sweden(20) compared to children of other mothers giving birth in Malmoe. However, other factors besides race-ethnicity and immigration may be involved. In a Swedish study, immigrant mothers born outside of Europe or North America were three times more likely to have a child with ASD whereas mothers from Europe and North America had only a slightly increased risk, compared to mothers born in Sweden.(14) Similarly, a British study reported that immigrant mothers born outside of Europe, and particularly in the Caribbean, had higher rates of ASD in their children than non-immigrant mothers.(13) The prevalence of ASD is increasing (22–23) and in Australia and other Western countries, the rate of immigration often varies, in terms of the number of immigrants and the ethnic composition. (24–26) Hence, year of birth is a potential confounder in these studies. In many of the cited studies, there was no adjustment for the year of birth of the child. (9, 11, 13, 14) I am unaware of any study that investigated the risk of ASD
according to the presence of comorbid ID in the children of mothers from ethnic minority groups according to their immigration status or maternal region of birth while adjusting for the year group of the child’s birth, maternal age, parity and socio-economic status (SES). Therefore, I aimed to compare the risk of ASD with and without ID in mothers according to their race-ethnicity, immigration status and the geographic region of their birthplace while adjusting for these potential confounders. In light of previously cited research,(13, 16, 20, 21) I also aimed to explore the prevalence of ASD with ID in Asian women born in sub-regions of Asia and Black women born in East Africa. Where there was a higher prevalence in a sub-region of either Asia or East Africa, I aimed to explore the prevalence by maternal country of birth.

This study may have three-fold benefits. First, identifying sub-groups of women at altered risk of having a child with ASD with ID and ASD without ID may provide some insight into ASD risk factors related to immigration status. Second, some differences in ASD prevalence among racial–ethnic groups might point to potential differences in ASD ascertainment rather than true differences in ASD and thus identify the need for enhanced screening in some population sub-groups. Third, the identification of an altered prevalence of ASD with or without ID in women of particular ethnicities and from particular countries may provide directions for future research into risk factors for ASD.

5.1.2 Methods

Study population and data collection

My study population consisted of all women with a live-born child in WA between 1 January 1994 and 31 December 2005. The data were from three state-wide registries. The first was the Midwives Notification System (MNS)(27) which provided me with the birthdates of all children born in WA during the collection period, maternal race-, age, parity and SES. The WA Birth Register(27) provided me with maternal country of birth. In WA, attending midwives are required to provide this information for all births and forms are completed in conjunction with the mother. Our second data source was the IDEA Database,(28) which receives diagnostic information on children with ID or ASD that have been referred to the state government organisation, the Disability
Services Commission.(29) Diagnoses of ID were made by clinicians or psychologists using psychometric testing. Diagnoses of ASD were made using DSM-4 criteria with a multidisciplinary panel to assess eligibility.(30) Using the IDEA Database, records of children born between 1994 and 2005 and diagnosed with ASD with and without ID and diagnosed before 31 December 2010 were linked to the corresponding MNS record from their birth. This was enabled by the use of a unique alpha–numeric code that was created for each mother by personnel from WA’s Data Linkage Unit.(27) These codes were used in the dataset in lieu of personal identifiers such as names or residence.

**Maternal groups**

For women with at least one child with ASD, the index child was the eldest child with ASD born during the study period. For the comparator group, Caucasian women who had no child with ASD, the index child was the first-born child during the collection period. Case mothers were grouped into one of two case groups (ASD with ID and ASD without ID) according to the disability of their index child.

**Explanatory variables**

Researchers have demonstrated that SES, maternal age and parity are associated with ASD.(4) The prevalence of ASD(23) and the rate of immigration of people from different regions(27) has varied over time so I included a binary measure of birth year group of the index child. I derived the measure of SES from the Index of Relative Socioeconomic Disadvantage(31) which ranks residential districts according to variables such as low income, lack of a vehicle and low level of education. I used quartile boundaries from the original dataset to define the three levels of SES: Low pertained to the lowest quartile, Medium to the two inner quartiles and High to the top quartile. I calculated maternal age at the index birth and used these ages to define a variable with the levels of Less than 20 years, 20 and less than 30 years, 30 and less than 40 years and 40 years or more. Parity at the time of the index birth was defined by a variable with the levels of 0 previous children, 1 previous child, 2 or 3 previous children and More than 3 previous children. I created a two-level variable for index birth year group. Level 1 was for 1994–99 and Level 2, 2000–05. Even though our
original dataset extended from 1983 to 2010, I excluded all births before 1994 as the
number of births in immigrant mothers of Asian or Black race-ethnicity was small.
Moreover, I did not include births after 2005 as I reasoned that a five-year period was
needed for children with ASD without ID to have a reasonable opportunity to be
diagnosed.

**Region of birth**

I divided the countries of the world into the 11 geographic regions (Table 11). Each
region was the level of a variable pertaining to a mother’s birthplace.

<table>
<thead>
<tr>
<th>Region</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Australia</td>
<td>All Australian states and mainland territories</td>
</tr>
<tr>
<td>Other Australasia</td>
<td>New Zealand, Christmas Island, Niue Island, Norfolk Island and Thursday Island</td>
</tr>
<tr>
<td>United Kingdom</td>
<td>England, Northern Ireland, Scotland or Wales</td>
</tr>
<tr>
<td>Europe</td>
<td>All other European countries</td>
</tr>
<tr>
<td>North America</td>
<td>US, Canada and nearby islands</td>
</tr>
<tr>
<td>South America</td>
<td>South America, Central America and nearby islands</td>
</tr>
<tr>
<td>Central Asia</td>
<td>Includes Cambodia, Thailand and Vietnam</td>
</tr>
<tr>
<td>South Asia</td>
<td>Includes countries in Southern Asia or south-east of this region such as Brunei, Malaysia, Solomon Islands, Samoa and Tonga</td>
</tr>
<tr>
<td>Other Asia</td>
<td>Includes Afghanistan, Iraq, Pakistan and United Arab Emirates (West Asia) China, Japan and Korea, Kazakhstan (East Asia), Mongolia, Russia and Siberia (North Asia)</td>
</tr>
<tr>
<td>East Africa</td>
<td>Includes Ethiopia, Kenya, Uganda, Zimbabwe and nearby islands</td>
</tr>
<tr>
<td>Other Africa</td>
<td>Includes Algeria, Angola, Botswana, Chad, Congo, Gambia, Nigeria, Ivory Coast, Lesotho, Namibia, Rwanda, South Africa Sudan, Zaire and Zambia and nearby islands not included in East Africa region</td>
</tr>
</tbody>
</table>

**Race-ethnicity**

First, I created a binary variable to measure ‘Any race-ethnicity’. Mothers who were not
Caucasian were assigned a level of 1 and Caucasian mothers, 0. Second, I created an
eight-level variable to measure ethnic groups of interest. From information in the MNS
dataset and using the region of birth variable, I created a variable with the levels of
*Caucasian, Indigenous, Asian from South Asia, Asian from Central Asia, Asian from elsewhere, Black from East Africa, Black from elsewhere, and Indian, Maori or Polynesian.* For the women of Asian or Black race-ethnicity, I divided the groups
according to region of birth as I wanted to explore the likelihood of a child with ASD in
women of Asian race-ethnicity from different parts of Asia and Black women from
East Africa. For these groups, I also calculated the prevalence of ASD in their children. In this paper, I have used *Indigenous* as an equivalent to *Aboriginal* and not *non-immigrant*.

**Immigration status**

Using the region of birth variable, I created a binary variable that indicated immigration. All mothers who were born in an Australian state or mainland territory were categorised as non-immigrant and others as immigrant.

**Analyses**

I used multinomial logistic regression where the dependent variable was the ASD case group. The independent variables were one or more of maternal *race*-ethnicity, maternal immigration status and region of maternal birth. I adjusted for maternal age, parity, SES and index birth year group and report the unadjusted and adjusted odds ratios (ORs). STATA 13 was used for all analyses.

**5.1.3 Results**

My dataset contained 134,204 mothers of whom 1,028 mothers had a child with ASD with ID’ and 347 had a child with ASD without ID. Numbers of mothers by *race*-ethnicity, ethnic groups of interest, immigration status and case group are given in Table 12. Notably, all children with ASD born to Black mothers had comorbid ID and a mother born in East Africa (Table 13). Table 14 has number of mothers according to their region on birth and case group. Proportionately, mothers from the UK and North America have more children with ASD without ID than ASD with ID. This situation is reversed in mothers from East Africa, and Central and South Asia.
### Table 12: Number of mothers by demographic characteristics

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Comparator group</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No ASD</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Maternal age at the index birth</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Less than 20 years</td>
<td>11,226 (8.5%)</td>
<td>38 (3.7%)</td>
<td>12 (3.5%)</td>
<td>11,276 (8.4%)</td>
</tr>
<tr>
<td>20 - 29 years</td>
<td>67,079 (50.5%)</td>
<td>426 (41.4%)</td>
<td>154 (44.4%)</td>
<td>67,659 (50.4%)</td>
</tr>
<tr>
<td>30 - 39 years</td>
<td>51,394 (38.7%)</td>
<td>530 (51.6%)</td>
<td>169 (48.7%)</td>
<td>52,093 (38.8%)</td>
</tr>
<tr>
<td>40 years or more</td>
<td>3,130 (2.4%)</td>
<td>34 (3.3%)</td>
<td>12 (3.5%)</td>
<td>3,176 (2.4%)</td>
</tr>
<tr>
<td>Parity at the index birth</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>0 previous child</td>
<td>113,794 (85.7%)</td>
<td>480 (46.7%)</td>
<td>183 (52.7%)</td>
<td>114,457 (85.3%)</td>
</tr>
<tr>
<td>1 previous child</td>
<td>11,696 (8.8%)</td>
<td>345 (33.6%)</td>
<td>105 (30.3%)</td>
<td>12,146 (9.1%)</td>
</tr>
<tr>
<td>2-3 previous children</td>
<td>6,448 (4.9%)</td>
<td>175 (17.0%)</td>
<td>52 (15.0%)</td>
<td>6,675 (5.0%)</td>
</tr>
<tr>
<td>&gt;3 previous children</td>
<td>891 (0.7%)</td>
<td>28 (2.7%)</td>
<td>7 (2.0%)</td>
<td>926 (0.7%)</td>
</tr>
<tr>
<td>SES at the index birth</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low</td>
<td>27,746 (20.9%)</td>
<td>255 (22.8%)</td>
<td>72 (20.8%)</td>
<td>28,073 (20.9%)</td>
</tr>
<tr>
<td>Medium</td>
<td>67,112 (50.5%)</td>
<td>519 (50.5%)</td>
<td>178 (51.3%)</td>
<td>67,809 (50.5%)</td>
</tr>
<tr>
<td>High</td>
<td>34,974 (26.3%)</td>
<td>232 (22.6%)</td>
<td>88 (25.4%)</td>
<td>35,294 (26.3%)</td>
</tr>
<tr>
<td>Missing</td>
<td>2,997 (2.3%)</td>
<td>22 (2.1%)</td>
<td>9 (2.6%)</td>
<td>3,028 (2.3%)</td>
</tr>
<tr>
<td>Index birth year group</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1994-1999</td>
<td>65,460 (49.3%)</td>
<td>452 (44.0%)</td>
<td>313 (90.2%)</td>
<td>66,225 (49.4%)</td>
</tr>
<tr>
<td>2000-2005</td>
<td>67,369 (50.7%)</td>
<td>576 (56.0%)</td>
<td>34 (9.8%)</td>
<td>67,979 (50.6%)</td>
</tr>
<tr>
<td>Total</td>
<td>132,829</td>
<td>1,028</td>
<td>347</td>
<td>134,204</td>
</tr>
</tbody>
</table>

ASD, autism spectrum disorder; ID, intellectual disability; SES, socio-demographic status

### Table 13: Number of mothers by race-ethnicity and birth places of interest

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Comparator group</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Race-ethnicity independent of immigration status</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>118,524 (89.2%)</td>
<td>937 (91.2%)</td>
<td>340 (98.0%)</td>
<td>119,801 (89.3%)</td>
</tr>
<tr>
<td>Any race-ethnicity (not Caucasian)</td>
<td>14,305 (10.8%)</td>
<td>91 (8.9%)</td>
<td>7 (2.0%)</td>
<td>14,403 (10.7%)</td>
</tr>
<tr>
<td>Indigenous</td>
<td>5,368 (4.0%)</td>
<td>23 (2.2%)</td>
<td>2 (0.6%)</td>
<td>5,393 (4.0%)</td>
</tr>
<tr>
<td>Asian</td>
<td>6,517 (4.9%)</td>
<td>56 (5.4%)</td>
<td>5 (1.4%)</td>
<td>6,578 (4.9%)</td>
</tr>
<tr>
<td>Black</td>
<td>594 (0.5%)</td>
<td>7 (0.7%)</td>
<td>0 (0.0%)</td>
<td>601 (0.4%)</td>
</tr>
<tr>
<td>Indian, Maori or Polynesian</td>
<td>1,826 (1.4%)</td>
<td>5 (0.5%)</td>
<td>0 (0.0%)</td>
<td>1,831 (1.4%)</td>
</tr>
<tr>
<td>Racial-ethnic groups of interest</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Asian from South Asia</td>
<td>2,746 (2.1%)</td>
<td>30 (2.9%)</td>
<td>1 (0.3%)</td>
<td>2,777 (2.1%)</td>
</tr>
<tr>
<td>Asian from Central Asia</td>
<td>1,763 (1.3%)</td>
<td>18 (1.8%)</td>
<td>1 (0.3%)</td>
<td>1,782 (1.3%)</td>
</tr>
<tr>
<td>Asian from elsewhere</td>
<td>2,008 (1.5%)</td>
<td>8 (0.8%)</td>
<td>3 (0.9%)</td>
<td>2,021 (1.5%)</td>
</tr>
<tr>
<td>Black from East Africa</td>
<td>246 (0.2%)</td>
<td>7 (0.7%)</td>
<td>0 (0.0%)</td>
<td>253 (0.2%)</td>
</tr>
<tr>
<td>Black from elsewhere</td>
<td>348 (0.3%)</td>
<td>0 (0.0%)</td>
<td>0 (0.0%)</td>
<td>348 (0.3%)</td>
</tr>
<tr>
<td>Immigration status</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Immigrant</td>
<td>36,140 (27.2%)</td>
<td>276 (26.9%)</td>
<td>85 (24.5%)</td>
<td>36,501 (27.2%)</td>
</tr>
<tr>
<td>Non-immigrant</td>
<td>96,689 (72.8%)</td>
<td>752 (73.1%)</td>
<td>262 (75.5%)</td>
<td>97,703 (68.9%)</td>
</tr>
<tr>
<td>Total</td>
<td>132,829 (100%)</td>
<td>1,028 (100%)</td>
<td>347 (100%)</td>
<td>134,204 (100%)</td>
</tr>
</tbody>
</table>

ASD, autism spectrum disorder; ID, intellectual disability
Note: Cells where numbers are too small (<6) for reliable analysis have been shaded.
Table 14: Number of mothers by maternal case group and region of birth

<table>
<thead>
<tr>
<th>Region of birth</th>
<th>Comparator group</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No ASD</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Australia</td>
<td>96,717 (72.8%)</td>
<td>752 (73.2%)</td>
<td>262 (75.5%)</td>
<td>97,731 (72.8%)</td>
</tr>
<tr>
<td>Other parts of Australasia</td>
<td>5,286 (4.0%)</td>
<td>30 (2.9%)</td>
<td>2 (0.6%)</td>
<td>5,318 (4.0%)</td>
</tr>
<tr>
<td>United Kingdom</td>
<td>12,936 (9.7%)</td>
<td>114 (11.1%)</td>
<td>46 (13.3%)</td>
<td>13,096 (9.8%)</td>
</tr>
<tr>
<td>Europe</td>
<td>4,649 (3.5%)</td>
<td>30 (2.9%)</td>
<td>9 (2.6%)</td>
<td>4,688 (3.5%)</td>
</tr>
<tr>
<td>North America</td>
<td>1,201 (0.9%)</td>
<td>7 (0.7%)</td>
<td>5 (1.4%)</td>
<td>1,213 (0.9%)</td>
</tr>
<tr>
<td>South America</td>
<td>394 (0.3%)</td>
<td>1 (0.1%)</td>
<td>1 (0.3%)</td>
<td>396 (0.3%)</td>
</tr>
<tr>
<td>Central Asia</td>
<td>1,869 (1.5%)</td>
<td>21 (2.3%)</td>
<td>1 (0.5%)</td>
<td>1,891 (1.4%)</td>
</tr>
<tr>
<td>South Asia</td>
<td>3,848 (2.9%)</td>
<td>40 (3.9%)</td>
<td>4 (1.2%)</td>
<td>3,892 (2.9%)</td>
</tr>
<tr>
<td>Other Asia</td>
<td>3,232 (2.4%)</td>
<td>12 (1.2%)</td>
<td>7 (2.0%)</td>
<td>3,251 (2.4%)</td>
</tr>
<tr>
<td>East Africa</td>
<td>943 (0.7%)</td>
<td>13 (1.3%)</td>
<td>3 (0.9%)</td>
<td>959 (0.7%)</td>
</tr>
<tr>
<td>Other Africa</td>
<td>1,754 (1.3%)</td>
<td>8 (0.8%)</td>
<td>7 (2.0%)</td>
<td>1,769 (1.3%)</td>
</tr>
<tr>
<td>Total</td>
<td>132,829 (100%)</td>
<td>1,028 (100%)</td>
<td>347 (100%)</td>
<td>134,204 (100%)</td>
</tr>
</tbody>
</table>

ASD, autism spectrum disorder; ID, intellectual disability
Note: Cells where numbers are too small (<6) for reliable analysis have been shaded. (32)

Race-ethnicity

Compared to Caucasian mothers, mothers of minority ethnicities were less likely to have a child with ASD with ID [0.59(95% CI: 0.5, 0.7)] and ASD without ID [0.19(95% CI: 0.1, 0.4)]. Indigenous women had slightly lower but similar odds to mothers of other minority ethnicities of having a child with ASD with ID [0.53(95% CI: 0.3, 0.8)] and ASD without ID [0.14(95% CI: 0.04, 0.6)]. Women of Asian race-ethnicity were 21% less likely to have a child with ASD with ID [0.79(95% CI: 0.6, 1.03)] whereas women of Black race-ethnicity had about half the odds of a child with either ASD with ID [0.51(95% CI: 0.2, 1.1)] (Table 15).
Table 15: Odds of a child with ASD by case group and race-ethnicity

<table>
<thead>
<tr>
<th>Race-ethnicity</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
</tr>
</thead>
<tbody>
<tr>
<td>Caucasians</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Any race-ethnicity (not Caucasian)</td>
<td><strong>0.80 (0.6, 0.998)</strong></td>
<td><strong>0.17 (0.1, 0.4)</strong></td>
</tr>
<tr>
<td>Any race-ethnicity (not Caucasian) adjusted</td>
<td><strong>0.59 (0.5, 0.7)</strong></td>
<td><strong>0.19 (0.1, 0.4)</strong></td>
</tr>
<tr>
<td>Indigenous</td>
<td>0.5 (0.4, 0.8) **</td>
<td>0.13 (0.03, 0.6) **</td>
</tr>
<tr>
<td>Asian</td>
<td>1.09 (0.8, 1.4)</td>
<td>0.27 (0.1, 0.6) **</td>
</tr>
<tr>
<td>Black</td>
<td>1.49 (0.7, 3.2) #</td>
<td>N=0</td>
</tr>
<tr>
<td>Indian, Maori or Polynesian</td>
<td>0.35 (0.1, 0.8) #</td>
<td>N=0</td>
</tr>
</tbody>
</table>

ASD, Autism spectrum disorder; ID, intellectual disability
Φ, unadjusted odds
#, Odds are adjusted for maternal age, parity and SES, each at the time of the index birth and index birth year group.
* p-value <0.05, ** p-value <0.005, ***p-value <0.0005
Note: Cells where numbers are too small (<6) for reliable analysis have been shaded.

**Immigrant status**

As a group, immigrant mothers were less likely both to have a child with ASD with ID and ASD without ID [0.64 (95% CI: 0.6, 0.7), 0.62 (95% CI: 0.5, 0.8)] (Table 16).

**Race-ethnicity and immigrant status**

Compared to Caucasian non-immigrant women, non-Caucasian immigrant women were 40% less likely to have a child with ASD with ID [0.59 (95% CI: 0.5, 0.7)] and around 80% less likely to have a child with ASD without ID [0.19 (95% CI: 0.1, 0.4)]. Indigenous non-immigrant women were around 50% less likely to have a child with ASD with ID and 87% less likely to have a child with ASD without ID [0.50 (95% CI: 0.3, 0.8), 0.13 (95% CI: 0.03, 0.6)]. Reliable ORs were unable to be calculated for other ethnicities (Table 16). Compared to non-immigrant women of any race-ethnicity, Caucasian immigrant women were about 35% less likely to have a child with either ASD with or without ID [0.64 (95% CI: 0.6, 0.7), 0.72 (95% CI: 0.6, 0.9)] and Asian immigrants were around 30% less likely to have a child with ASD with ID and 70% less likely to have a child with ASD without ID [0.70 (95% CI: 0.5, 0.9), 0.29 (95% CI: 0.1, 0.7)]. Black, immigrant women of race-ethnicity were around half as likely to have a child with ASD with ID [0.48 (95% CI: 0.2, 1.03)] (Table 16).
Table 16: Odds of a child with ASD by case group, race-ethnicity and immigration status

<table>
<thead>
<tr>
<th>Race-ethnicity by immigration status</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
</tr>
</thead>
<tbody>
<tr>
<td>Caucasian, non-immigrant</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Indigenous, non-immigrant</td>
<td>0.55(0.4, 0.8)*</td>
<td>0.13(0.03, 0.5)*</td>
</tr>
<tr>
<td>Asian, non-immigrant</td>
<td>0.87(0.2, 3.5)†</td>
<td>N=0</td>
</tr>
<tr>
<td>Indian, Maori or Polynesian, non-immigrant</td>
<td>1.5(0.4, 6.1)†</td>
<td>N=0</td>
</tr>
<tr>
<td>Black, non-immigrant</td>
<td>N=0</td>
<td>N=0</td>
</tr>
<tr>
<td>Non-immigrant</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Immigrant</td>
<td>0.98(0.9, 1.1)†</td>
<td>0.87(0.7, 1.1)†</td>
</tr>
<tr>
<td>Caucasian, immigrant</td>
<td>0.99(0.8, 1.2)†</td>
<td>1.1(0.8, 1.4)†</td>
</tr>
<tr>
<td>Indigenous, immigrant</td>
<td>N=0</td>
<td>N=0</td>
</tr>
<tr>
<td>Asian, immigrant</td>
<td>1.11(0.8, 1.5)†</td>
<td>0.30(0.1, 0.7)*</td>
</tr>
<tr>
<td>Indian, Maori or Polynesian, immigrant</td>
<td>0.23(0.1, 0.7)*</td>
<td>N=0</td>
</tr>
<tr>
<td>Black, immigrant</td>
<td>1.56(0.7, 3.3)†</td>
<td>N=0</td>
</tr>
</tbody>
</table>

ASD, Autism spectrum disorder; ID, intellectual disability; * unadjusted odds
†, Odds are adjusted for maternal age, parity and SES, each at the time of the index birth and index birth year group.

Note: Cells where numbers are too small (<6) for reliable analysis are shaded.
* p-value <0.05, ** p-value <0.005, *** p-value <0.0005

Region of birth

Compared to Australian-born women, women born in *Other parts of Australasia*, North America and South America had less than half the odds of a child with ASD with ID [0.47(95% CI: 0.3, 0.7), 0.48(95% CI: 0.2, 1.03), 0.20(95% CI: 0.03, 1.5)]. Women from the UK had lower odds of a child both with ASD with ID and ASD without ID [0.83(95% CI: 0.7, 1.0), 0.88(95% CI: 0.6, 1.2)], whereas the odds for women from Europe were even lower [0.52(95% CI: 0.4, 0.8), 0.44(95% CI: 0.2, 0.9)]. Women from *Other parts of Asia* and *Other parts of Africa* were more than 70% less likely to have a child with ASD with ID [0.27(95% CI: 0.2, 0.5), 0.28(95% CI: 0.1, 0.6)] but the women for *Other parts of Asia* were less likely to have a child with ASD without ID [0.65(95% CI: 0.3, 1.4)] and the women from *Other parts of Africa* were more likely [1.23(95% CI: 0.6, 2.6)]. Of the birth regions considered, women from Central Asia, South Asia and East Africa had the highest odds of a child with ASD with ID [0.97(95% CI: 0.6, 1.4)].
1.5), 0.87 (95% CI: 0.6, 1.2), 0.98 (95% CI: 0.6, 1.7)] but they were still less likely to have a child with this disability than were Australian-born women (Table 17).

**Racial-ethnic groups of interest**

Compared to other immigrant women, Asian women from Central or South Asia were about 30% more likely to have a child with ASD with ID [1.23 (95% CI: 0.8, 2.1), 1.36 (95% CI: 0.9, 2.0)]. In contrast, women from other regions of Asia had less than half the odds of having a child with ASD with ID [0.41 (95% CI: 0.2, 0.9)]. Black, East African women were more than twice as likely to have a child with ASD with ID [2.24 (95% CI: 1.02, 4.9)] (Table 18). The unadjusted ORs indicated that Asian women from South and Central Asia and Black women from East Africa had a higher prevalence of ASD with ID than Caucasian women (Table 18).

**Table 17: Odds of a child with ASD by case group and birth region**

<table>
<thead>
<tr>
<th>Region of birth</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
</tr>
</thead>
<tbody>
<tr>
<td>Australia</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Other parts of Australasia</td>
<td>0.73 (0.51, 1.1)Φ</td>
<td>0.14 (0.03, 0.6)Φ *</td>
</tr>
<tr>
<td>Other parts of Australasia</td>
<td>0.47 (0.3, 0.7)Φ ***</td>
<td>0.10 (0.02, 0.4)Φ **</td>
</tr>
<tr>
<td>United Kingdom</td>
<td>1.13 (0.93, 1.4)Φ</td>
<td>1.31 (0.96, 1.8)Φ</td>
</tr>
<tr>
<td>United Kingdom</td>
<td>0.83 (0.7, 1.0)Φ</td>
<td>0.88 (0.6, 1.2)Φ</td>
</tr>
<tr>
<td>Europe</td>
<td>0.83 (0.58, 1.2)Φ</td>
<td>0.71 (0.4, 1.4)Φ</td>
</tr>
<tr>
<td>Europe</td>
<td>0.52 (0.4, 0.8)Φ **</td>
<td>0.44 (0.2, 0.9)Φ *</td>
</tr>
<tr>
<td>North America</td>
<td>0.75 (0.36, 1.6)Φ</td>
<td>1.54 (0.6, 3.7)Φ</td>
</tr>
<tr>
<td>North America</td>
<td>0.48 (0.2, 1.03)Φ</td>
<td>1.05 (0.4, 2.6)Φ</td>
</tr>
<tr>
<td>South America</td>
<td>0.33 (0.05, 2.3)Φ</td>
<td>0.94 (0.1, 6.7)Φ</td>
</tr>
<tr>
<td>South America</td>
<td>0.20 (0.03, 1.5)Φ</td>
<td>0.56 (0.1, 4.0)Φ</td>
</tr>
<tr>
<td>Central Asia</td>
<td>1.45 (0.93, 2.2)Φ</td>
<td>0.20 (0.03, 1.4)Φ</td>
</tr>
<tr>
<td>Central Asia</td>
<td>0.97 (0.6, 1.5)Φ</td>
<td>0.21 (0.03, 1.5)Φ</td>
</tr>
<tr>
<td>South Asia</td>
<td>1.34 (0.97, 1.8)Φ</td>
<td>0.38 (0.1, 1.03)Φ</td>
</tr>
<tr>
<td>South Asia</td>
<td>0.87 (0.6, 1.2)Φ</td>
<td>0.36 (0.1, 0.97)Φ *</td>
</tr>
<tr>
<td>Other parts of Asia</td>
<td>0.48 (0.27, 0.8)Φ *</td>
<td>0.80 (0.4, 1.7)Φ</td>
</tr>
<tr>
<td>Other parts of Asia</td>
<td>0.27 (0.2, 0.5)Φ ***</td>
<td>0.65 (0.3, 1.4)Φ</td>
</tr>
<tr>
<td>East Africa</td>
<td>1.77 (1.02, 3.1)Φ *</td>
<td>1.17 (0.4, 3.7)Φ</td>
</tr>
<tr>
<td>East Africa</td>
<td>0.98 (0.6, 1.7)Φ</td>
<td>0.90 (0.3, 2.8)Φ</td>
</tr>
<tr>
<td>Other parts of Africa</td>
<td>0.59 (0.29, 1.2)Φ</td>
<td>1.47 (0.7, 3.1)Φ</td>
</tr>
<tr>
<td>Other parts of Africa</td>
<td>0.28 (0.1, 0.6)Φ ***</td>
<td>1.23 (0.6, 2.6)Φ</td>
</tr>
</tbody>
</table>

ASD, Autism spectrum disorder; ID, intellectual disability
Φ, unadjusted odds
#, ORs are adjusted for maternal age, parity and SES, each at the time of the index birth and index birth year group
* p-value <0.05, ** p-value <0.005, ***p-value <0.0005
### Table 18: Odds of a child with ASD by case group, race-ethnicity and birth region

<table>
<thead>
<tr>
<th>Ethnic group</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
</tr>
</thead>
<tbody>
<tr>
<td>Other immigrant women</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Asian from Central Asia</td>
<td>1.33(0.8, 2.2)Φ</td>
<td>0.20(0.03, 1.4)Φ</td>
</tr>
<tr>
<td></td>
<td>1.23(0.8, 2.1)#</td>
<td>0.28(0.04, 2.0)#</td>
</tr>
<tr>
<td>Asian from South Asia</td>
<td>1.43(0.97, 2.1)Φ</td>
<td>0.13(0.02, 0.9)Φ *</td>
</tr>
<tr>
<td></td>
<td>1.36(0.9, 2.0)#</td>
<td>0.18(0.02, 1.3)#</td>
</tr>
<tr>
<td>Other Asia</td>
<td>0.45(0.2, 1.03)Φ</td>
<td>0.60(0.2, 1.9)Φ</td>
</tr>
<tr>
<td></td>
<td>0.41(0.2, 0.9)#*</td>
<td>0.88(0.3, 2.8)#*</td>
</tr>
<tr>
<td>Black from East Africa</td>
<td>3.71(1.7, 8.0)Φ **</td>
<td>N=0</td>
</tr>
<tr>
<td></td>
<td>2.24(1.02, 4.9)#*</td>
<td></td>
</tr>
</tbody>
</table>

ASD, Autism spectrum disorder; ID, intellectual disability
Φ, unadjusted odds
#, Adjusted for maternal age, parity and SES, each at the time of the index birth and index birth year group
* p-value <0.05, ** p-value <0.005, ***p-value <0.0005
Note: Cells where numbers are too small (<6) for reliable analysis have been shaded.

**Prevalence and country of birth**

I calculated the prevalence of ASD with and without ID in the children of Asian women from Central or South Asia and Black women from East Africa. There were 18 women born in Central Asia who had a child with ASD with ID (and only one who had a child with ASD without ID). For the ASD with ID case group, 14 women were born in Vietnam, three in Thailand and one in Cambodia. Of South Asian women, 30 had a child with ASD with ID and none had a child with ASD without ID. Twelve were born in Malaysia, ten in the Philippines, five in Indonesia, two in Singapore and one in East Timor. For women of Asian race-ethnicity from Central Asia, the prevalence of ASD with ID was 4.4 times that of other immigrant women. Women from Vietnam and Thailand had 4.8 and 3.3 times the prevalence of other immigrant women whereas those from both Cambodia and the entire Central Asian region had 4.4 times the prevalence. Those women from Malaysia, Philippines or any part of South Asia had an increased prevalence of ASD with ID in their children (5.9, 6.4 and 4.8 per 10,000 births). All rates are in Table 19.

Black women from East Africa had nearly 15 times the prevalence of ASD with ID than immigrant women generally. Only women born in Eritrea, Ethiopia, Kenya and Somalia
had a child with ASD with ID. The associated prevalence ranged from 5.3 to 24.2 times that of other immigrant women (Table 19).

Table 19: Number of mothers by race-ethnicity, birth region, case group and ASD prevalence

<table>
<thead>
<tr>
<th>Region &amp; COB by race-ethnicity</th>
<th>No ASD</th>
<th>ASD with ID</th>
<th>ASD w/out ID</th>
<th>Total births</th>
<th>ASD with ID</th>
<th>ASD w/out ID</th>
<th>Prevalence cf to all immigrant women</th>
</tr>
</thead>
<tbody>
<tr>
<td>Women of Asian race-ethnicity from Central Asia</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Vietnam</td>
<td>1,261</td>
<td>14</td>
<td>1</td>
<td>1,276</td>
<td>110</td>
<td>7.8</td>
<td>4.8 times 0.3 times</td>
</tr>
<tr>
<td>Thailand</td>
<td>385</td>
<td>3</td>
<td>0</td>
<td>388</td>
<td>77</td>
<td>0</td>
<td>3.3 times 0</td>
</tr>
<tr>
<td>Cambodia</td>
<td>97</td>
<td>1</td>
<td>0</td>
<td>98</td>
<td>102</td>
<td>0</td>
<td>4.4 times 0</td>
</tr>
<tr>
<td>Central Asia</td>
<td>1,743</td>
<td>18</td>
<td>1</td>
<td>1,762</td>
<td>102</td>
<td>5.7</td>
<td>4.4 times 0.2</td>
</tr>
<tr>
<td>Women of Asian race-ethnicity from South Asia</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Malaysia</td>
<td>879</td>
<td>12</td>
<td>0</td>
<td>891</td>
<td>135</td>
<td>0</td>
<td>5.9 times 0</td>
</tr>
<tr>
<td>Philippines</td>
<td>666</td>
<td>10</td>
<td>0</td>
<td>676</td>
<td>148</td>
<td>0</td>
<td>6.4 times 0</td>
</tr>
<tr>
<td>Indonesia</td>
<td>767</td>
<td>5</td>
<td>0</td>
<td>772</td>
<td>65</td>
<td>0</td>
<td>2.8 times 0</td>
</tr>
<tr>
<td>Singapore</td>
<td>369</td>
<td>2</td>
<td>0</td>
<td>371</td>
<td>54</td>
<td>0</td>
<td>2.3 times 0</td>
</tr>
<tr>
<td>East Timor</td>
<td>25</td>
<td>1</td>
<td>0</td>
<td>26</td>
<td>385</td>
<td>0</td>
<td>16.7 times 0</td>
</tr>
<tr>
<td>South Asia</td>
<td>2,706</td>
<td>30</td>
<td>0</td>
<td>2,736</td>
<td>110</td>
<td>0</td>
<td>4.8 times 0</td>
</tr>
<tr>
<td>Other Asia (Not Central)</td>
<td>1,609</td>
<td>6</td>
<td>3</td>
<td>1,618</td>
<td>37</td>
<td>18.5</td>
<td>1.6 times 0.8</td>
</tr>
<tr>
<td>Black women from East Africa</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Eritrea</td>
<td>17</td>
<td>1</td>
<td>0</td>
<td>18</td>
<td>556</td>
<td>0</td>
<td>24.2 times 0</td>
</tr>
<tr>
<td>Ethiopia</td>
<td>70</td>
<td>4</td>
<td>0</td>
<td>74</td>
<td>541</td>
<td>0</td>
<td>23.5 times 0</td>
</tr>
<tr>
<td>Kenya</td>
<td>30</td>
<td>1</td>
<td>0</td>
<td>31</td>
<td>323</td>
<td>0</td>
<td>14.0 times 0</td>
</tr>
<tr>
<td>Somalia</td>
<td>80</td>
<td>1</td>
<td>0</td>
<td>81</td>
<td>123</td>
<td>0</td>
<td>5.3 times 0</td>
</tr>
<tr>
<td>East Africa</td>
<td>197</td>
<td>7</td>
<td>0</td>
<td>204</td>
<td>343</td>
<td>0</td>
<td>14.9 times 0</td>
</tr>
<tr>
<td>Other Africa</td>
<td>303</td>
<td>0</td>
<td>0</td>
<td>303</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Africa</td>
<td>500</td>
<td>7</td>
<td>0</td>
<td>507</td>
<td>138</td>
<td>0</td>
<td>6 times 0</td>
</tr>
</tbody>
</table>

COB, Country of birth; ASD, Autism spectrum disorder; ID, intellectual disability; cf, compared to; w/out, without

5.1.4 Discussion

I explored maternal race-ethnicity and immigration status as independent risk factors for ASD with and without ID. I also explored the effects of maternal race-ethnicity and region of birth on the risk of a woman having a child with ASD with ID and ASD without ID. Finally, I examined the prevalence of ASD with and without ID by the country of birth in women of Asian race-ethnicity who were born in Central Asia or South Asia and Black women who were born in East Africa or other parts of Africa. As reported by other researchers, mothers of children with ASD were less likely to be of young age and more likely to be more than 40 years.(4) Due to my definition of the index child as the eldest child in comparator mothers and the eldest child with ASD in
case mothers, the association of lower parity was not present in case mothers. The previous association of high SES in mothers of a child with ASD, present in earlier WA datasets,(4) was also not apparent. Primarily, this might be due to our SES variable. In the current study, I used lowest and highest quartiles to define the extremes of SES whereas the previous researchers used sextiles. Other influences might be due to improvements in the accessibility of screening processes for ASD in WA, combined with an increased awareness of ASD in the general population over the last decade. The unequal distribution of mothers of children with ASD without ID between the two year groups is indicative of an under-ascertainment of ASD without ID in the more recent year group.

**Strength and limitations**

My study was enabled by the availability of population-based data on children with ID or ASD and an associated strength is the fact that I used a complete cohort of women and children. The values of important variables such as race-ethnicity, maternal and baby birthdates were collected by medical personnel in conjunction with the mothers and country of birth was provided by the mother when she registered the birth of her child. These were collected before the child was diagnosed with ASD and reduced bias and enhanced the accuracy of our results. A weakness is the small numbers of mothers of some ethnicities. Combined with the small number of children with ASD, this limited the groups I was able to analyse. Another limitation is that the diagnosis of ‘ASD without ID’ appears to be incomplete as the younger children would have been less than eight years old in 2010.

**Race-ethnicity**

As others found, mothers who were of minority ethnicities (not Caucasian) were less likely to have a child with ASD and particularly ASD without ID. For example, three US studies found that mothers of non-Caucasian ethnicities were less likely to have a child with ASD,(33) ASD with ID(34) and ASD without ID.(35) The reason cited for these disparities was poorer access to healthcare and consequent under-ascertainment in the children and I consider that this is also the case for these women in WA. Along with
others,(4, 5) I found that Indigenous women were particularly unlikely to have a child with ASD. In WA, this could be due to lower ascertainment as many of Indigenous people live in remote communities(36) where diagnosis of ASD would be difficult to obtain and of little consequence since there would be few services available.

**Immigrant status and region of birth**

Immigrant women were significantly less likely to have a child with ASD. Previous research exploring immigration as a risk factor for ASD is inconclusive. For example, researchers in California found that immigrant women had a similar or lower risk of a child with ASD than mothers born within the state.(7) On the other hand, US researchers conducting a meta-analysis reported that mothers born outside the country had an increased risk of a child with ASD.(37) Possibly, our lower rates of ASD in the children of immigrant mothers are due to the fact that half (Table 14) of these women were from the UK, Europe or North America and hence are likely to be English speaking and healthy,(38) which might decrease their likelihood of having a child with ASD.(39) Further, families with an existing child with a disability are generally precluded from immigrating to Australia.(40) Thus, those successful in immigrating would have reduced odds of a disability in any subsequent children.

**Race-ethnicity and immigrant status**

Non-Caucasian, non-immigrant women were less likely to have a child with either ASD with ID or ASD without ID. Ninety-eight % of Indigenous women were non-immigrant. This explains why the odds of a child with ASD with and without ID are almost the same (and just as low) for Indigenous women by race-ethnicity only and Indigenous non-immigrant women. Immigrant women of Caucasian, Asian or Black race-ethnicity were from 30 to 70% less likely to have a child with ASD with ID, which differs from the results of previous studies.(4–6, 20) All but one of these studies(4) related to immigration to Europe or the US. Hence, reasons for the difference might be different immigration policies and/or different conditions in Australia. Unlike here, the other West Australian study(4) found higher ORs for ASD with ID in immigrant mothers from certain parts of Asia. This might be because I included only Asian mothers that
were born in Asia, adjusted for different variables and my cohort was from a more recent period: 1994–2005 v. 1984–99.

This might be because I included only Asian mothers that were born in Asia, adjusted for different variables, grouped country of birth differently and my cohort was from a more recent period: 1994–2005 v. 1984–99.

**Ethnic groups of interest**

Asian women from South or Central Asia had a higher prevalence of children with ASD with ID. A research group in Eastern Australia and another in WA reported that mothers born in South-east or North-east Asia had the highest risk of a child with ASD.(4, 11) A Swedish study(20) also reported higher odds in women from East Asia and a Californian study(6) reported about four times the risk of ASD with ID in women of Asian race-ethnicity compared to Caucasian women. My results included only women of Asian race-ethnicity whereas all but the Californian study may have included women of other ethnicities. The higher prevalence of ASD with ID in immigrant Asian women was not exacerbated by a higher overall SES in Asian women from Central Asia as these women had a lower overall SES (Table 20). However, Asian immigrant women from South Asia had a slightly higher average SES compared to other women in WA (Table 20). Another possibility might be an unknown risk factor for ASD with ID that is associated with emigrating from particular regions of Asia. For example, data has shown that immigrants have more severe allergic diseases than native-born residents and that most often this is associated with asthma and rhinitis.(41) It is important to note that it was only Asian women from Central and South Asia that had a higher prevalence of ASD with ID. Asian women from other parts of Asia had less than half the odds of ASD of other immigrant women. This might indicate effect modification where different countries of origin have different effects on the risk of ASD with ID in women of Asian race-ethnicity.

Before adjustment, Black women from East Africa were more than three and a half times as likely to have a child with ASD with ID as Caucasian mothers. These women were of lower average SES than Caucasian mothers, so this may relate to an unknown
risk factor for ASD with ID in these women. Other studies have reported increased rates of ASD with ID in women from sub-Saharan Africa(13, 20) and particularly countries in East Africa.(16, 21, 42).

### Table 20: SES by maternal region of birth and race-ethnicity

<table>
<thead>
<tr>
<th>SES at time of index birth</th>
<th>Asian from Central Asia</th>
<th>Asian from South Asia</th>
<th>Asian from Other Asia</th>
<th>Black from East Africa</th>
<th>Non-immigrant Caucasian</th>
</tr>
</thead>
<tbody>
<tr>
<td>Low</td>
<td>599 (33.6%)</td>
<td>444 (16.0%)</td>
<td>352 (17.4%)</td>
<td>85 (33.6%)</td>
<td>18,560 (20.2%)</td>
</tr>
<tr>
<td>Medium</td>
<td>920 (52.2%)</td>
<td>1,288 (46.4%)</td>
<td>1,031 (51.1%)</td>
<td>138 (54.6%)</td>
<td>47,791 (52.0%)</td>
</tr>
<tr>
<td>High</td>
<td>235 (13.6%)</td>
<td>998 (35.8%)</td>
<td>618 (30.6%)</td>
<td>28 (11.1%)</td>
<td>23,417 (25.5%)</td>
</tr>
<tr>
<td>Missing</td>
<td>18 (1.0%)</td>
<td>47 (1.7%)</td>
<td>18 (0.9%)</td>
<td>2 (0.8%)</td>
<td>2,176 (2.4%)</td>
</tr>
<tr>
<td>Total</td>
<td>1,782 (100%)</td>
<td>2,777 (100%)</td>
<td>3,251 (100%)</td>
<td>253 (100%)</td>
<td>91,944 (100%)</td>
</tr>
</tbody>
</table>

SES, socioeconomic status

**Prevalence and country of birth**

Compared to the group of all immigrant women, I found a higher prevalence of ASD with ID in the children of mothers from Central and South Asia and particularly those from Vietnam (N=14), where the prevalence was five times that of other immigrant women. Similarly there was a higher prevalence in the children of women born in Central Asia and particularly those from Malaysia and the Philippines, where around six times the prevalence was noted. A higher prevalence in women from Asia was noted by others although race-ethnicity was not considered. For example, a US study reported four times the odds of ASD with ID in the children of Asian women.(6) When examining maternal immigration from Asia, three studies reported increased odds of a child with ASD with ID. The unadjusted ORs here indicated a slightly higher prevalence of ASD with ID and a much lower prevalence of ASD without ID in women of Asian race-ethnicity. However, after adjustment the ORs for ASD with ID were attenuated to <1, indicating that one or more of maternal age, parity, SES and index birth year group may be accounting for the higher prevalence.

A higher prevalence of ASD with ID in the children of mothers from East Africa was indicated by their increased unadjusted odds of having a child with ASD with ID compared to immigrant women overall. After adjustment, the ORs remained larger,
which may indicate that other factors are affecting the prevalence. The increased prevalence in East Africa was nearly 15 times that of immigrant women overall. I noted that all East African women came from a cluster of countries that are located in the eastern most part of East Africa (Figure 5). The higher prevalence in the current study of ASD with ID in Black women from Africa was also found by others in the UK,(13) Sweden,(20) and Northern US.(21) Some researchers(42) suggested that the higher rates might be the result of the lower amounts of sunlight experienced at the high latitudes in these countries. My results do not support this hypothesis since the latitude of WA ranges from 15 to 35º, which is considerably lower than central latitudes of the UK (52º), Sweden (64º), and Northern US (45º). Notably, in my study, it was only Black women from East Africa who had a child with ASD. The prevalence of ASD without ID in Black mothers is 0% v. 0.2% in the larger group of immigrant mothers. Others have reported higher rates of ASD with ID and low rates of ASD without ID in immigrant women from Africa(13) and sub-Saharan Africa(20) and Black women from Uganda and Somalia in East Africa.(16, 21, 42) I found no reports of higher rates of ASD in Black immigrant women from other regions of Africa. I suggest that Black immigrant women from a localised region of East Africa (Figure 5), experience a risk factor for ASD with ID in subsequent children that is associated with their immigration. I also suggest that women of other ethnicities from this region and Black women from other parts of Africa are either not exposed or not sensitive to this risk factor. As others have suggested,(7) this is likely due to a gene–environment interaction with the specific environmental factor(s) not yet to be identified. I encourage more work in this area.
5.1.5 Summary and implications

Both race-ethnicity and immigrant status in isolation were associated with a lower risk of ASD with and without ID, compared to non-immigrant Caucasian women. There was a similar result when race-ethnicity was considered by immigration status. I suggested that this is due to social factors. Compared to Caucasian women, women of Asian race-ethnicity from South or Central Asia and Black women from East Africa had a higher prevalence of ASD with ID in their children. Adjusting for socio-demographic factors accounted for the increased prevalence in the Asian women but not the women from East Africa. Black women, from East Africa had nearly a 15-fold increase of ASD with ID in their children compared to other immigrant women. I suggest this is due to a gene–environment interaction in these women. This may relate to obstetrics, environmental exposures or other factors.

Research is needed to identify specific risk factors for ASD prior to immigration in Black women from countries in the identified high-risk region in East Africa. There is also a need to explore neonatal, perinatal and obstetric complications in Black mothers who have had a child with ASD as such complications have been associated with the risk of having a child with ASD and may be more likely in these women.(43) Ascertainment of ASD in marginalised populations such as Indigenous children appears poor. In light of the improved outcomes in children with ASD who receive early
intervention. Improved methods of ascertainment and access to services in these populations are indicated. Our findings also point to the need to separate maternal race-ethnicity from immigration status when examining factors that may contribute to the development of ASD with and without ID in the children of these mothers.
5.2 Maternal psychiatric disorder and the risk of ID or ASD in subsequent offspring

5.2.1 Introduction

The relationship between intellectual disability (ID) and autism spectrum disorder (ASD) is poorly understood.(45) People with ID have impairments in cognitive and adaptive functioning(1) and those with ASD have impairments in social interaction, communication, behaviours and interests.(1) In Western Australia (WA), the prevalence of ID is 1.4%(46) and is more common than ASD, which affects only 0.3%.(46)

The prevalence of ASD is increasing,(30) but no such trend has been reported for ID. Both conditions have genetic and environmental aetiologies,(47) with around half of ID having a clear biomedical cause,(48) comprising either genetic aetiologies, such as Down syndrome, or environmental aetiologies, such as fetal alcohol syndrome and head injury. In contrast, the aetiology of ASD is mostly unknown, although a genetic basis involving many genes and gene–environment interactions has been implicated.(48) Clinical similarities between ID and ASD include delayed milestones, language impairments(45) and comorbidities such as epilepsy.(49) Both disorders share genetic factors with schizophrenia.(50) Intellectual disability and ASD commonly coexist, with about 60% of persons with ASD also having ID.(51) This reinforces the notion that both share common genetic and environmental causes.(45)

Often, parents of children with ID or ASD face behavioural and sleep issues with their children(52), along with societal stigma(53) social isolation,(53) financial(54) and employment difficulties.(54) Therefore, the poorer mental health repeatedly reported for parents is assumed to be a consequence of the burden of caring. For example, compared to parents of typically developing children, one study identified more depression in the mothers caring for their children with ID or ASD.(55) Another study(56) compared mental health in mothers caring for their children with Down syndrome with population normative values and identified significantly poorer self-perceptions in these women. A population-based study(57) reported that maternal emotional disorder was more prevalent in mothers caring for their child with ASD compared to both mothers of
children with ID and mothers of typically developing children. However, in all of these studies, the mothers’ mental health prior to the birth was not investigated and thus the poorer health may have been pre-existing. A Danish record linkage study found that pre-existing parental psychiatric disorders were associated with increased rates of ASD in the offspring. Therefore, it is feasible that psychiatric conditions are already pre-existing and more likely to be associated with ID or ASD in the offspring due to genetic or prenatal environmental factors.

I found no research that examined pre-existing psychiatric disorders in mothers of children with ID while taking account of the cause or severity of the ID. Further, I found no research that compared pre-existing psychiatric disorders in parents of children with ASD with and without ID. These comparisons might provide an insight into aetiology. Different levels and types of ID have different aetiologies and some researchers consider that ASD with and without ID have at least some different aetiologies.

For these reasons, I aimed to answer three questions. First, do mothers who receive mental health services before the birth of a child have increased odds of a child with ID or ASD? Second, for these women, do the odds change according to the level and type of ID in the child or according to whether the ASD is associated with ID? Third, for these women, are the odds affected by the particular type of psychiatric diagnosis?

5.2.2 Methods

Study population

I included all mothers of a live child born in WA between 1983 and 1999 inclusive. Data were obtained from two statutory, state-wide databases and a disability database. The Mental Health Information System (MHIS) provided public outpatient contacts for all women from 1969 to 1999. The Midwives Notification System (MNS) provided maternal socio-demographic characteristics and dates for all births in WA during the collection period. The Intellectual Disability Exploring Answers (IDEA) Database provided diagnostic information on children with ID or ASD born from
1983 to 1999 and up to the end of 2010. The Data Linkage Unit for WA created a unique code for each woman, enabling me to link the datasets. (27)

**Maternal groups and index child**

Case mothers were assigned to a group according to the disability of their index child. I formed the six case groups of *Mild or moderate ID of unknown cause* (Mild–moderate ID), *Severe or profound ID of unknown cause* (Severe ID), *Down syndrome*, *ID of known cause (not Down syndrome)*, *ASD with ID* and *ASD without ID*. Five composite case groups were formed by combining various core case groups. Mothers were assigned to either a case or comparator group according to the status of their index child. In mothers of children with ID or ASD, the index child was their eldest child with a disability. Mothers in the comparator group had no children with ID or ASD and their index child was the eldest child born during the study period. The comparator group, five composite and six core case groups and their inter-relationships were shown in Figure 4 (Chapter 3).

**Explanatory variables**

As previous research shows that socio-economic status (SES), maternal age and parity are associated with ID and ASD, (4) these were included in my model. Due to the increasing prevalence of ASD during the collection period, an index birth year variable was also included and years were grouped into the following year bands: 1983–87, 1988–91, 1992–95 and 1996–99. Socioeconomic status was measured by a three-level variable calculated from the *Index of Relative Socioeconomic Disadvantage*. (31) Here ‘low’ pertained to the most disadvantaged quartile of scores, ‘medium’ to the inner two quartiles and ‘high’ to the most advantaged quartile, with the quartiles determined from my original dataset. Maternal age at the index birth was calculated by the difference between the mother’s birth year and the birth year of the index child. I stratified this variable as follows to limit within-group confounding: *less than 20 years*, *20 to 29 years*, *30 to 39 years* and *more than 40 years*. Parity was measured at the time of the index birth and the associated variable had the four levels of 0 (no previous child), 1 (one previous child), 2-3 and >3.
Psychiatric status

I used a woman’s clinical outpatient contacts with MHIS prior to the birth of her index child to assess her psychiatric status. First, I created a variable representing the existence of any psychiatric contact with the levels of ‘Yes’ for those with one or more contacts and ‘No’ for others. Second, each contact was grouped into one of seven diagnostic categories created within the dataset and according to the ICD code ascribed to a patient’s first contact (Table 21). Seven more binary measures of psychiatric status were created. Women with one or more contacts for a category were given the level of ‘Yes’ for this category and all other women, ‘No’. In this way, I had measures of the existence of a diagnosis for each mother for each of the seven categories of Substance abuse disorders, Schizophrenia spectrum disorders, Affective disorders, Obsessive–compulsive/personality disorders, Developmental disorders, Childhood/adolescent disorders, and Other mental health diagnoses.

Analyses

I performed multinomial logistic regressions where mothers of children with no ID or ASD were the comparator group. Sample size permitting, I report the adjusted ORs and associated 95% CI for the eight variables measuring psychiatric status. I used STATA 11.2 to perform the analyses.
Table 21: Composition of diagnostic categories

<table>
<thead>
<tr>
<th>Categories from the original dataset</th>
<th>Categories for analyses</th>
<th>Associated ICD-9 codes</th>
<th>Associated ICD-10 codes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Substance abuse disorders</td>
<td></td>
<td>291.0, 291.2, 291.8, 291.9, 292.0, 292.8, 292.9, 294.0, 303.0, 303.9, 304.0–304.9, 305.0, 305.2–305.9</td>
<td>F10–F19, F55</td>
</tr>
<tr>
<td>Mood disorders</td>
<td></td>
<td>296.0–296.9, 311, 316</td>
<td>F30–F39, F54</td>
</tr>
<tr>
<td>Anxiety disorders</td>
<td>Affective disorders</td>
<td>300.0, 300.2, 300.4, 300.5, 300.8, 300.9</td>
<td>F20.4, F34.1, F40–F41, F45.0, F45.1, F48</td>
</tr>
<tr>
<td>Stress &amp; adjustment disorders</td>
<td></td>
<td>308.0–308.4, 309</td>
<td>F43, F94.0</td>
</tr>
<tr>
<td>Obsessive-compulsive disorders</td>
<td>Obsessive-compulsive/personality disorders</td>
<td></td>
<td>F42</td>
</tr>
<tr>
<td>Personality disorders</td>
<td></td>
<td>12.4, 312.8, 312.9</td>
<td>F60, F61, F62, F68, F69, F91, F92</td>
</tr>
<tr>
<td>Mental retardation, psychological development disorders</td>
<td>Developmental disorders</td>
<td>299.0, 299.1, 299.8, 299.9, 315, 317–319</td>
<td>F70, F71, F72, F73, F78, F79 F80–F84, F88–F89</td>
</tr>
<tr>
<td>Childhood &amp; adolescent disorders</td>
<td></td>
<td>307, 312, 313, 314, 330.8</td>
<td>F45.4, F45.8, F63, F84.2, F90, F91.1, F91.2, F93, F94.1, F94.2, F94.8, F94.9, F95, F98.0, F98.1, F98.4, F98.5, F98.6, F98.8, F98.9</td>
</tr>
<tr>
<td>Other mental health conditions</td>
<td></td>
<td>290.0–290.4, 298.0, 298.9 291.1, 293, 294.1, 294.8, 294.9, 298.9, 300.10, 310.0–310.2, 310.8, 310.9</td>
<td>F00–F07, F09, F10.6, F44, F68.0</td>
</tr>
<tr>
<td>Somatoform disorders</td>
<td></td>
<td>300.6, 300.7</td>
<td>F45.2, F48.1</td>
</tr>
<tr>
<td>Eating disorders</td>
<td></td>
<td>307.1, 307.5</td>
<td>F54.9, F50, F92.8, F98.3</td>
</tr>
<tr>
<td>Behavioural syndromes</td>
<td>Other mental health diagnoses</td>
<td>305.1, 306, 307.4, 316.0, 316.9</td>
<td>F17.0, F17.1, F45.3, F45.9, F51, F52.5, F53.0, F53.1, F53.8, F53.9, F59</td>
</tr>
<tr>
<td>Sexual disorders</td>
<td></td>
<td>302</td>
<td>F52, F64, F65, F66</td>
</tr>
<tr>
<td>Other related mental health conditions</td>
<td></td>
<td>331.0, 368.16, 648.4, 655.45, 655.53, 780.1, 780.5, 784.6, 785.50, 799.9–799.8</td>
<td>G30.0, G47, O35.4, O35.5, O99.3, R44, R45, R48</td>
</tr>
<tr>
<td>Unspecified mental health conditions</td>
<td></td>
<td>799.9</td>
<td>F99</td>
</tr>
</tbody>
</table>

5.2.3 Results

My dataset contained 213,656 mothers, 9,339 (4.4%) of whom had a psychiatric outpatient record. My comparator group of mothers of children with no ID or ASD contained a total of 207,827 mothers (Table 22). Composition of maternal case groups by SES, age and parity, all at the time of the index birth, and the year group of the index birth are also provided (Table 22). As shown previously,(4) the relationships between SES, maternal age and parity for mothers of children with ID of unknown cause and mothers of children with ASD were different. Low SES, young age (<20 years) and high parity were more prevalent in the mothers of children with ID of unknown cause, whereas high SES, being over 40 years and low parity were more prevalent in the
mothers of children with ASD. For any ASD, the number of children born with subsequent diagnoses increased with successive year groups.
### Table 22: Case groups by SES, age, parity and birth year group as percentages

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Comparator group</th>
<th>Case groups</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No ID or ASD</td>
<td>Mild-mod ID</td>
</tr>
<tr>
<td>Socioeconomic status</td>
<td>Low</td>
<td>20.1</td>
</tr>
<tr>
<td></td>
<td>Medium</td>
<td>40.8</td>
</tr>
<tr>
<td></td>
<td>High</td>
<td>20.5</td>
</tr>
<tr>
<td></td>
<td>Missing</td>
<td>18.4</td>
</tr>
<tr>
<td>Maternal age at index birth in years</td>
<td>&lt; 20</td>
<td>6.9</td>
</tr>
<tr>
<td></td>
<td>20-29</td>
<td>58.5</td>
</tr>
<tr>
<td></td>
<td>30-39</td>
<td>32.9</td>
</tr>
<tr>
<td></td>
<td>≥ 40</td>
<td>1.6</td>
</tr>
<tr>
<td>Parity at index birth</td>
<td>0</td>
<td>70.2</td>
</tr>
<tr>
<td></td>
<td>1</td>
<td>16.3</td>
</tr>
<tr>
<td></td>
<td>2-3</td>
<td>12.2</td>
</tr>
<tr>
<td></td>
<td>&gt;3</td>
<td>1.5</td>
</tr>
<tr>
<td>Index birth year group</td>
<td>1983-87</td>
<td>37.0</td>
</tr>
<tr>
<td></td>
<td>1988-91</td>
<td>21.8</td>
</tr>
<tr>
<td></td>
<td>1992-95</td>
<td>20.5</td>
</tr>
<tr>
<td></td>
<td>1996-99</td>
<td>20.7</td>
</tr>
<tr>
<td>% of total</td>
<td>97.3</td>
<td>1.7</td>
</tr>
<tr>
<td>Total</td>
<td>207,827</td>
<td>3,606</td>
</tr>
</tbody>
</table>

ASD, Autism spectrum disorder; ID, intellectual disability; Mild-mod ID, mild or moderate of unknown cause; Severe ID, Severe or profound ID of unknown cause; ~, not; Down, Down syndrome
Table 23: Number of women by category and case group prior to the index child’s birth

<table>
<thead>
<tr>
<th>Category</th>
<th>Comparator group</th>
<th>Maternal case group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No ID or ASD</td>
<td>Mild-mod ID</td>
</tr>
<tr>
<td>Any psychiatric contact</td>
<td>8,803</td>
<td>358</td>
</tr>
<tr>
<td>Substance abuse disorders</td>
<td>95</td>
<td>5</td>
</tr>
<tr>
<td>Schiz disorders</td>
<td>208</td>
<td>8</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>3,995</td>
<td>147</td>
</tr>
<tr>
<td>Obsess-compulsive &amp; personality disorders</td>
<td>562</td>
<td>29</td>
</tr>
<tr>
<td>Developmental disorders</td>
<td>34</td>
<td>12</td>
</tr>
<tr>
<td>Child &amp; adolescent disorders</td>
<td>1,339</td>
<td>55</td>
</tr>
<tr>
<td>Other mental health diagnoses</td>
<td>2,571</td>
<td>102</td>
</tr>
<tr>
<td>No psychiatric diagnosis</td>
<td>199,024</td>
<td>3,248</td>
</tr>
<tr>
<td>Total</td>
<td>207,827</td>
<td>3,606</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Category</th>
<th>Comparator group</th>
<th>Maternal case group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mild or moderate ID of unknown cause</td>
<td>Severe ID</td>
</tr>
<tr>
<td>Any psychiatric contact</td>
<td>12</td>
<td>370</td>
</tr>
<tr>
<td>Substance abuse disorders</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Schiz disorders</td>
<td>9</td>
<td>1</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>151</td>
<td>30</td>
</tr>
<tr>
<td>Obsess-compulsive &amp; personality disorders</td>
<td>29</td>
<td>4</td>
</tr>
<tr>
<td>Developmental disorders</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Child &amp; adolescent disorders</td>
<td>58</td>
<td>4</td>
</tr>
<tr>
<td>Other mental health diagnoses</td>
<td>13</td>
<td>5</td>
</tr>
<tr>
<td>No psychiatric diagnosis</td>
<td>3490</td>
<td>333</td>
</tr>
<tr>
<td>Total</td>
<td>3,860</td>
<td>572</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Category</th>
<th>Comparator group</th>
<th>Maternal case group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Any ID with ID</td>
<td>ASD without ID</td>
</tr>
<tr>
<td>Any psychiatric contact</td>
<td>51</td>
<td>41</td>
</tr>
<tr>
<td>Substance abuse disorders</td>
<td>2</td>
<td>7</td>
</tr>
<tr>
<td>Schiz disorders</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>37</td>
<td>37</td>
</tr>
<tr>
<td>Obsess-compulsive &amp; personality disorders</td>
<td>5</td>
<td>3</td>
</tr>
<tr>
<td>Developmental disorders</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Child &amp; adolescent disorders</td>
<td>7</td>
<td>3</td>
</tr>
<tr>
<td>Other mental health diagnoses</td>
<td>18</td>
<td>12</td>
</tr>
<tr>
<td>No psychiatric diagnosis</td>
<td>854</td>
<td>344</td>
</tr>
<tr>
<td>Total</td>
<td>926</td>
<td>4,786</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Category</th>
<th>Comparator group</th>
<th>Maternal case group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Any ASD with ID</td>
<td>Any ASD without ID</td>
</tr>
<tr>
<td>Any psychiatric contact</td>
<td>442</td>
<td>41</td>
</tr>
<tr>
<td>Substance abuse disorders</td>
<td>7</td>
<td>0</td>
</tr>
<tr>
<td>Schiz disorders</td>
<td>12</td>
<td>2</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>188</td>
<td>22</td>
</tr>
<tr>
<td>Obsess-compulsive &amp; personality disorders</td>
<td>34</td>
<td>3</td>
</tr>
<tr>
<td>Developmental disorders</td>
<td>12</td>
<td>1</td>
</tr>
<tr>
<td>Child &amp; adolescent disorders</td>
<td>65</td>
<td>6</td>
</tr>
<tr>
<td>Other mental health diagnoses</td>
<td>124</td>
<td>8</td>
</tr>
<tr>
<td>No psychiatric diagnosis</td>
<td>124</td>
<td>8</td>
</tr>
<tr>
<td>Total</td>
<td>926</td>
<td>4,786</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Category</th>
<th>Comparator group</th>
<th>Maternal case group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Any ID or ASD</td>
<td>TOTAL</td>
</tr>
<tr>
<td>Any psychiatric contact</td>
<td>536</td>
<td>9,339</td>
</tr>
<tr>
<td>Substance abuse disorders</td>
<td>8</td>
<td>103</td>
</tr>
<tr>
<td>Schiz disorders</td>
<td>17</td>
<td>225</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>230</td>
<td>4,225</td>
</tr>
<tr>
<td>Obsess-compulsive &amp; personality disorders</td>
<td>39</td>
<td>601</td>
</tr>
<tr>
<td>Developmental disorders</td>
<td>13</td>
<td>47</td>
</tr>
<tr>
<td>Child &amp; adolescent disorders</td>
<td>79</td>
<td>1,418</td>
</tr>
<tr>
<td>Other mental health diagnoses</td>
<td>151</td>
<td>2,722</td>
</tr>
<tr>
<td>No psychiatric diagnosis</td>
<td>5293</td>
<td>204,317</td>
</tr>
<tr>
<td>Total</td>
<td>5,829</td>
<td>213,656</td>
</tr>
</tbody>
</table>

ID, intellectual disability; ASD, Autism spectrum disorder; Mild-mod; Mild or moderate ID of unknown cause; Severe ID, Severe or profound ID of unknown cause; dis, disorders; Obsess, Obsessive; Schiz, Schizophrenia spectrum disorders.

Note 1: Women may have a diagnosis for more than one diagnostic category.
Note 2: Numbers too small (<6) for analysis are shaded.
**Any outpatient contact**

Table 23 displays the number of women with a psychiatric contact before the birth of their index child by maternal case group and the associated adjusted ORs are provided in Figure 6. Compared to mothers with no previous psychiatric contact, those with a psychiatric contact were more than two and a half times as likely to have a child with ASD with ID [2.53(95% CI: 1.8, 3.5)], more than twice as likely to have a child with mild-moderate ID [2.21(95% CI: 2.0, 2.5)], *ID of unknown cause* [2.14(95% CI: 1.9, 2.4)], ASD without ID [2.13(95% CI: 1.6, 2.8)], *any ID* [2.07(95% CI: 1.9, 2.3)], *any ASD* [2.07(95% CI: 1.7, 2.6)] or ‘any ID or ASD’ [2.06(95% CI: 1.9, 2.3)] and nearly twice as likely to have a child with *ID of known cause* [1.80(95% CI: 1.4, 2.3)] or *ID of known cause (not Down syndrome)* [1.80(95% CI: 1.35, 2.4)]. Further, mothers with a pre-existing outpatient contact had elevated odds of a child with Down syndrome [1.30(95% CI: 0.8, 2.0)] and severe ID [1.08(95% CI: 0.6, 1.9)] (Figure 6, Table 24).

**Figure 6: Odds of a child with ID or ASD for mothers with a previous psychiatric contact**

<table>
<thead>
<tr>
<th>Condition</th>
<th>OR (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>No ID and no autism</td>
<td>1(1,1)</td>
</tr>
<tr>
<td>Autism with ID</td>
<td>2.53(1.8,3.5)</td>
</tr>
<tr>
<td>Mild ID or unknown cause</td>
<td>2.21(2.0,2.5)</td>
</tr>
<tr>
<td>Autism without ID</td>
<td>2.14(1.7,2.6)</td>
</tr>
<tr>
<td>Any ID</td>
<td>2.13(1.9,2.3)</td>
</tr>
<tr>
<td>Any ASD</td>
<td>2.07(1.9,2.3)</td>
</tr>
<tr>
<td>'any ID or ASD'</td>
<td>2.06(1.9,2.3)</td>
</tr>
<tr>
<td>ID of known cause</td>
<td>1.80(1.4,2.3)</td>
</tr>
<tr>
<td>ID of known cause (not Down syndrome)</td>
<td>1.80(1.35,2.4)</td>
</tr>
<tr>
<td>Down syndrome</td>
<td>1.30(0.8,2.0)</td>
</tr>
<tr>
<td>Severe ID (unknown cause)</td>
<td>1.08(0.6,1.9)</td>
</tr>
</tbody>
</table>

OR, odds ratio; CL, confidence limits; ID, Intellectual disability; Autism, Autism spectrum disorder; Mild, Mild or moderate; Down, Down syndrome, Severe, Severe or profound

Note: ORs are adjusted for SES, maternal age, parity and index birth year group.
Table 24: Odds by diagnostic category and case group

<table>
<thead>
<tr>
<th>Comparator group</th>
<th>Category</th>
<th>Maternal case group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No ID or ASD</td>
<td>Mild-mod ID</td>
</tr>
<tr>
<td>Any psychiatric contact</td>
<td>1.0</td>
<td>2.21(2.0, 2.5)</td>
</tr>
<tr>
<td>Substance abuse disorder</td>
<td>1.0</td>
<td>2.12(0.8, 5.4)</td>
</tr>
<tr>
<td>Schiz disorders</td>
<td>1.0</td>
<td>2.10(1.0, 4.3)</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>1.0</td>
<td>1.97(1.7, 2.3)</td>
</tr>
<tr>
<td>Obsessive-compulsive/personality disorders</td>
<td>1.0</td>
<td>2.62(1.8, 3.9)</td>
</tr>
<tr>
<td>Developmental disorders</td>
<td>1.0</td>
<td>22.0(10.8, 45)</td>
</tr>
<tr>
<td>Childhood/adolescent disorders</td>
<td>1.0</td>
<td>2.20(1.7, 2.9)</td>
</tr>
<tr>
<td>Other mental health diagnoses</td>
<td>1.0</td>
<td>1.95(1.6, 2.4)</td>
</tr>
</tbody>
</table>

ID, intellectual disability; Mild-mod ID, Mild or moderate ID of unknown cause; Severe, Severe or profound ID of unknown cause; ASD, autism spectrum disorder; ~, not; Down, Down syndrome, Schiz, Schizophrenia spectrum.

Note 1: Odds ratios are adjusted for SES, maternal age, parity and index birth year group.
Note 2: Significant ORs are shaded.
**Diagnostic categories**

In Table 23, the number of women with a psychiatric contact by diagnostic category and case group are given. My comparator group for these analyses was the group of mothers of children with no ID and no ASD who had no diagnosis from the category in question. I considered cells that contained numbers of less than six to be too small for reliable analysis\(^{32}\) and hence I report no ORs for these cells. All reported ORs and CI are adjusted for SES, maternal age, parity and index birth year group.

Women with a diagnosis of *Substance abuse disorders* were more than twice as likely to have a child with *any ID or ASD* \([2.26(95\% \text{ CI}: 1.1, 4.8)]\) (Figure 7). Women with a diagnosis of *Schizophrenia spectrum disorders* were more than three times as likely to have a child with *any ASD* \([3.31(95\% \text{ CI}: 1.2, 9.0)]\) and more than two and a half times as likely to have a child with *any ID or ASD* \([2.66(95\% \text{ CI}: 1.6, 4.4)]\) or *any ID* \([2.53(95\% \text{ CI}: 1.4, 4.5)]\). Additionally, women with a diagnosis of *Schizophrenia spectrum disorders* were more than twice as likely to have a child with mild-moderate ID or ID of unknown cause (Figure 8).

**Figure 7: Odds of a child with ID or ASD for mothers with substance abuse disorders**

<table>
<thead>
<tr>
<th>Condition</th>
<th>Odds Ratio (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>No ID and no autism</td>
<td>1.00</td>
</tr>
<tr>
<td>Any ID or ASD</td>
<td>2.26 (1.1, 4.8)</td>
</tr>
<tr>
<td>Mild ID of unknown cause</td>
<td>2.12</td>
</tr>
<tr>
<td>ID of unknown cause</td>
<td>1.99</td>
</tr>
<tr>
<td>Any ID</td>
<td>1.95</td>
</tr>
</tbody>
</table>

OR, odds ratio; CL, confidence limits; ID, Intellectual disability; Autism, Autism spectrum disorder
Note: ORs are adjusted for SES, maternal age, parity and index birth year group.
Women with a diagnosis for *Affective disorders* were nearly three times as likely to have a subsequent child with ASD with ID [2.82(95% CI: 1.8, 4.3)] and nearly two and a half times as likely to have a child with ‘any ASD’ [2.45(95% CI: 1.8, 3.3)]. Further, women with a diagnosis for *Affective disorders* were around twice as likely to have a child with *ID of known cause (not Down syndrome)* [2.23(95% CI: 1.5, 3.2)], mild–moderate ID [95% CI: 1.97(1.7, 2.3)], ‘any ID or ASD’ [1.9(95% CI: 1.7, 2.2)] or *ID of unknown cause* [1.89(95% CI: 1.7, 2.2)]. All ORs are in Figure 9.

Those women with a diagnosis for *Obsessive–compulsive/personality disorders* were about two and a half times as likely to have a child with mild-moderate ID [2.62(95% CI: 1.8, 3.9)], ID of unknown cause [2.45(95% CI: 1.7, 3.6)] or any ID [2.35(95% CI: 1.7, 3.4)] and about twice as likely to have a subsequent child with ‘any ID or ASD’ [2.21(95% CI: 1.16, 3.1)] or ID of known cause [1.99(95% CI: 0.9, 4.5)]. All ORs are in Figure 10.
Figure 9: Odds of a child with ID or ASD, in mothers with affective disorders

OR, odds ratio; CL, confidence limits; ID, Intellectual disability; ASD, Autism spectrum disorder; Down, Down syndrome
Note: ORs adjusted for SES, maternal age, parity and index birth year group

Figure 10: Odds of a child with ID or ASD in mothers with obsessive-compulsive or personality disorders

ASD, Autism spectrum disorder; ID, Intellectual disability; OR, odds ratio; CL, confidence limits; Mild-moderate, Mild or moderate ID of unknown cause
Note: ORs are adjusted for SES, maternal age, parity and index birth year group

Women with previous outpatient contacts for Developmental disorders were about 20 times as likely to have a child with mild–moderate ID [22.04(95% CI: 10.8, 45.0)] or ID of unknown cause [20.57(95% CI: 11.0, 41.9)]. These women also had significant
increases in the odds of a child with any ID [16.9 (95% CI: 8.4, 34.1)] or any ID or ASD [14.8 (95% CI: 7.5, 29.1)]. All ORs are displayed in Figure 11.

Women with previous outpatient contacts for Childhood/adolescent disorders were more than twice as likely to have a child with ID of known cause [2.32 (95% CI: 1.3, 4.1)], any ID [2.21 (95% CI: 1.7, 2.8)], mild–moderate ID [2.2 (95% CI: 1.7, 2.9)] or any ID or ASD [2.08 (95% CI: 1.6, 2.6)] (Figure 12).

**Figure 11: Odds of a child with ID or ASD in mothers with developmental disorders**

ID, Intellectual disability; ASD, Autism spectrum disorder; Mild-moderate, Mild or moderate ID of unknown cause; Note: ORs are adjusted for SES, maternal age, parity and index birth year group.
Figure 12: Odds of a child with ID or ASD in mothers with Childhood or adolescent disorders

ID, Intellectual disability; ASD, autism spectrum disorder; Mild-moderate, Mild or moderate ID of unknown cause
Note: ORs are adjusted for SES, maternal age, parity and index birth year group

Having a previous contact for Other mental health diagnoses rendered women more than twice as likely to have a child with ASD with ID [2.4(95% CI: 0.6, 3.8)], and nearly twice as likely to have a child with one of mild–moderate ID [1.95(95% CI: 1.6, 2.4)], ID of unknown cause [1.91(95% CI: 1.6, 2.3)] or ‘any ID or ASD’ [1.80(95% CI: 1.5, 2.1)]. Complete results are in Figure 13.

Figure 13: Odds of a child with ID or ASD in mothers with Other mental health diagnoses

ID, Intellectual disability; ASD, Autism spectrum disorder; Mild-moderate, Mild or moderate ID of unknown cause; Down, Down syndrome
Note: ORs adjusted for SES, maternal age, parity and index birth year group
ID of known cause (not Down syndrome)

Women with any outpatient contact had nearly twice the odds of a child with ID of known cause (not Down syndrome). Further, all diagnostic categories for this case group had elevated odds from around one and a half up to nearly two and a quarter. In the ID of known cause (not Down syndrome) case group, 62% of causes for ID in this group are genetic (including de novo mutations) or chromosomal (Table 25).

Table 25: Causes of ID of known cause (not Down syndrome)

<table>
<thead>
<tr>
<th>Basis of ID*</th>
<th>Number</th>
<th>Percentage</th>
</tr>
</thead>
<tbody>
<tr>
<td>Defects of central nervous system &amp; other congenital defects</td>
<td>71</td>
<td>17</td>
</tr>
<tr>
<td>Genetic: X-linked, autosomal and chromosomal</td>
<td>263</td>
<td>62</td>
</tr>
<tr>
<td>Neonatal and post-neonatal events</td>
<td>47</td>
<td>11</td>
</tr>
<tr>
<td>Teratogens: chemical and infectious</td>
<td>40</td>
<td>10</td>
</tr>
<tr>
<td>TOTAL</td>
<td>421</td>
<td>100</td>
</tr>
</tbody>
</table>

*Medical diagnosis categories based on the Heber classification. Metabolic causes have been incorporated into the genetic category.(60)

5.2.4 Discussion

This study investigated the association between pre-existing maternal outpatient psychiatric disorders and ID or ASD in subsequent children. Mothers with any pre-existing contact were from nearly two to two and a half times more likely to have a child with ID (other than Down syndrome and severe ID) or ASD than mothers with no such history. The risk of a child with ID or ASD was elevated for all mothers with contacts from each of the diagnostic categories.

Seven studies were identified that investigated psychiatric disorders using hospitalisation and/or outpatient data in parents of children with ASD(12, 61-64) and another focused on children with ID (including ID with ASD).(64) Of these, three used only inpatient data(61-63) and four used both inpatient and outpatient data.(12, 58, 64, 65) I used only outpatient data since this provided an opportunity to investigate psychiatric disorders that did not warrant hospitalisation. Only two of these seven studies separately analysed pre-existing psychiatric disorders in mothers(12, 62) but
each of these looked only at the risk of ASD. A third study(58) looked at parental psychiatric disorder prior to the diagnosis of a child with ASD and a fourth study(65) looked at the risk of ID in the children of mothers with severe, lifelong psychiatric disorders. My study stands alone because I looked at a range of pre-existing psychiatric disorders in mothers and the risk of both ID and ASD in their children where the ID was differentiated by level and cause and the ASD according to whether or not it was associated with ID. I found that a woman with a psychiatric contact before the birth of her child had more than twice the odds of having a child with ASD with ID, ASD without ID or any ASD. This matches with the findings of a Swedish study(62) that also found that mothers with a psychiatric contact before the birth had more than twice the odds of having a child with ASD. However, I showed that the odds were higher for having a child with ASD with ID than for ASD without ID. This suggests that the aetiological factors underlying these disorders may differ.

Developmental disabilities are more likely in children whose mothers were exposed to alcohol and/or recreational drugs prenatally. Using record linkage, researchers in WA attributed around 2.5% of non-genetic ID to heavy prenatal alcohol exposure.(66) In relation to recreational drugs, a study published in 1992(67) found that of 70 children with prenatal cocaine exposure, more than 10% were diagnosed with ASD. My analyses showed that mothers of children with a pre-existing diagnosis of substance abuse were about twice as likely to have a child with any ID or ASD, mild–moderate ID or ID of known cause (not Down syndrome).

In a previous population study, mothers with lifelong schizophrenia were more than three times as likely to have a child with ID.(65) Results from a case–control study(64) using registry data from Sweden and Israel indicated that parents with schizophrenia had nearly triple the odds of having a child with ASD with ID, double the odds of having a child with ASD without ID and about two and a half times the odds of having a child with any ASD. Other research groups reported that mothers with schizophrenia(61, 62) had from two to two and a half times the odds of having a child with ASD and parents with a schizophrenia-like psychosis had about about three and a half times the odds of a child with ASD compared to those without these conditions. (58) None of these results took into account the timing of the diagnosis in
relation to the birth of the child with ASD. The relationships I found for mothers with 
pre-existing *Schizophrenia spectrum disorders* were similar with elevated odds from 
just over two and up to nearly three and a third for a child with ID or ASD. I also noted 
that, in comparison with mothers of children with no ID and no ASD, mothers with a 
previous contact for *Schizophrenia spectrum disorders* had higher odds of having a 
child with ASD than having a child with ID. Further, in mothers with a previous contact 
for this diagnostic category, the odds found here were much the same as those of other 
studies that made no allowance for the timing of the birth of the child with ASD. This 
could indicate that caring for a child with ASD does not increase the maternal 
prevalence of schizophrenia.

My study grouped bipolar disorder with other affective disorders in the category 
*Affective disorders*. I found that pre-existing maternal affective disorders were 
significantly associated with nearly three times the odds of having a child with ASD 
with ID, two and a half times the odds of having a child with *any ASD* and around 
twice the odds of having a child with *ID of unknown cause (not Down syndrome)* and 
ASD without ID. As found here, an international population study(64) reported 
significantly elevated rates of parental bipolar disorder, but comparisons are difficult as 
in that study the diagnostic category was narrower and paternal diagnoses and 
diagnoses after the birth were also included. A previous WA population study(65) 
found that women with bipolar disorder were more than three times as likely to have a 
child with ID and around four times as likely to have a child with rare genetic 
syndromes. My case group, *ID of known cause (not Down syndrome)* includes rare 
genetic syndromes. Compared to the current study, the higher odds in the previous 
study(65) for having a child with a rare genetic syndrome might be because only 
severe, lifelong psychiatric disorders were explored and bipolar disorder was examined 
separately from other affective disorders.

Previous research has also associated ASD with parental schizophrenia(58, 62) and 
affective disorder(58, 61, 62) but again no allowance was made for the timing of the 
birth of the child with ASD. Of these studies, the Danish study,(58) reported that 
parents with either schizophrenia or an affective disorder, before the diagnosis of a child 
with ASD, had about three times the odds of having a child with ASD. These are similar
to my results showing that mothers with pre-existing schizophrenia had about three times the odds of a child with ‘any ASD’ and mothers with pre-existing affective disorder had about twice the odds of ‘any ASD’.

I found that mothers with a pre-existing affective disorder had significantly increased odds of from around one and a half to nearly two and a quarter for all types of ID apart from Down syndrome. Similarly, mothers with lifelong depression had around three times the odds of ID in their children.(65) The lower odds found here are likely to result from my inclusion of less severe forms of affective disorder in this diagnostic category.

A research group using data from Swedish registries(62) made no allowance for the timing of the birth of the child with ASD in mothers with diagnoses of schizophrenia or affective disorder. They reported that mothers with a diagnosis of schizophrenia had nearly twice the odds of having a child with ASD and mothers with a diagnosis of affective disorder had nearly one and a quarter the odds of having a child with ASD. The odds in my study for having a child with ‘any ASD’ are more than three times higher for mothers with a pre-existing diagnosis of Schizophrenia spectrum disorders and mothers with a pre-existing diagnosis of Affective disorders had nearly two and a half times the odds of a child with ASD. Here, I can state unequivocally that these increased odds are not due to the burden of caring, so the higher odds for both pre-existing Schizophrenia spectrum disorders and Affective disorders are unexpected. Perhaps the higher rates for affective disorder are due to the inclusion of bipolar disorder in this category. Further, I was able to differentiate between ASD with ID and ASD without ID. Mothers with a pre-existing affective disorder were nearly three times as likely to have a child with ASD with ID. In contrast, these mothers had slightly less than twice the odds of having a child with ASD without ID. Again, this could be indicative of different causes of these two forms of ASD.

Using interviews, researchers probed the psychiatric history of the family of 99 persons with ASD.(68) They suggested that parental obsessive–compulsive disorders indicated a liability to ASD in the offspring. Using hospitalisation data, Scandinavian researchers reported that personality disorders were more common among mothers of children with ASD.(62) Both these results pertain to diagnoses after the birth of a child with ASD. I
estimated that mothers with a pre-existing diagnosis pertaining to Obsessive–compulsive/personality disorders were significantly more likely to have a child with 'mild–moderate ID’, ‘ID of unknown cause’, ‘any ID’ and ‘any ID or ASD’. I believe that these are the first results that suggest a woman with a diagnosis of Obsessive–compulsive/personality disorders is more likely to have a child with ID.

Women with previous outpatient contacts for developmental disorders had from 14 to 22 times the risk of a child with ID or ASD. Of the 22 mothers with contacts for developmental disorders with a subsequent child with ‘mild–moderate ID’, 10 had a primary diagnosis of ID and one of ASD. Other researchers(65) also found increased odds of ID in the offspring of mothers with ID and one study reported that a mother with ASD had given birth to a child who developed ASD.(62) This association could be caused by both genetic and environmental factors. For example, mothers with mild–moderate ID may have an undiagnosed X-linked mental retardation syndrome (including Fragile X syndrome) and hence would be likely to have an affected child.(69) Women with ID have also been shown to have higher rates of adverse birth and pregnancy outcomes, including pre-eclampsia and babies of low birth weight, than those without developmental disorders.(70) These conditions are risk factors for ID(71) and ASD(72) in the offspring.

I found that a diagnosis for Childhood/adolescent disorders significantly increased the odds of a woman having a child with ID or ASD. Others(61) found that these diagnoses significantly increased the risk of having a child with ASD and particularly Asperger syndrome.

The higher rates identified here of psychiatric contacts in mothers of children with ID or ASD are certainly not due to a burden of care. This association could be mediated by the common genetics of psychiatric disorders and ASD.(61) In addition, chromosomal causes of ID and de novo mutations might be more likely in mothers with a psychiatric disorder due to their increased stress and the association of increased stress with accelerated ovarian ageing.(73) In the ID of known cause (not Down syndrome) case group where a majority of causes are either genetic or chromosomal, these might be factors that contribute to the higher ORs in mothers with a pre-existing psychiatric disorder.
Another possible cause is differential prenatal medication use in women with a psychiatric disorder compared to other women. Researchers have found that the prenatal use of medication such as anti-depressants,\(^{(63)}\) psycho-actives\(^{(63)}\) and anti-epileptics\(^{(74)}\) are associated with a higher risk of ASD. An underlying mechanism might be medications crossing the placental barrier and affecting foetal brain development. Further, women with a psychiatric disorder have lower levels of self-care than other women\(^{(75)}\) These might be manifest in terms of higher smoking rates and increased alcohol consumption compared to other women. These behaviours during pregnancy increase the risk of ID or ASD in the offspring.\(^{(47,75)}\) Finally, another contributing factor might be higher level of obstetric complications\(^{(43)}\)

**Strengths and limitations**

A considerable strength of this study is the large cohort and the lack of selection that thereby minimizes any bias. Further, I had the advantage of being able to retrospectively access mental health data from outpatient registry records collected over 30 years. Unlike many other studies in this area\(^{(57, 63, 68)}\) my data are independent of maternal recall. Further, I was able to include accurate values of important variables such as diagnoses of ID and ASD, birthdates, dates of psychiatric appointments, parity, SES and diagnostic codes as they were all measured and transcribed by professionals such as clinicians, nurses and paramedical personnel. My study utilised reliable birth and contact dates from the years preceding the index birth. Hence, mothers are unaffected by the burden of caring for their children. This is the first study to investigate pre-existing psychiatric disorders in mothers of children with different levels and types of ID or ASD.

This study was limited by small numbers in some case groups, particularly the severe ID and Down syndrome groups. Further, private outpatient data, paternal date of birth and information on prescribed medication were unavailable. The absence of private outpatient data may have resulted in an under-ascertainment of psychiatric disorders treated in a non-residential setting. Access to paternal date of birth would have enabled me to adjust for this potential confounder.\(^{(4)}\) Information on prescribed medication
would have provided some indication of the likely prenatal exposures in case groups. There is also a small chance that some women with no outpatient contact may have in fact had either or both of inpatient admissions or private outpatient contacts and would have been included in the comparator group. However, this would have attenuated my ORs.

The definition of index child was different for the comparator group and the case groups. The index child of comparator mothers was their eldest child born during 1983–99. For 70% of comparator mothers, the index child was their eldest child. On the other hand, for case mothers only around 40% of index children were the eldest child. (Table 22) However, potential bias resulting from this differential selection was adjusted for by the use of index parity as an explanatory variable.

5.2.5 Conclusion and implications

My research adds to existing evidence that the rate of pre-existing psychiatric disorders in mothers of children with ASD is higher than that of mothers in the general population. Importantly, it also provides evidence about the relationship of pre-existing maternal psychiatric disorders and ID in subsequent offspring. Further research exploring the effect on pregnancy outcomes of medications prescribed to women with psychiatric disorders is warranted.
5.3 Is the Broad Autism Phenotype in mothers of children with ASD exacerbated by the challenges of caring for their children?

Seventy years ago, the child psychiatrist Leo Kanner described mothers of children with autism spectrum disorder (ASD) as distant and aloof. This concept of milder autistic traits in the relatives of children with ASD has developed considerably in the 21st century. Some researchers believe that collectively, these milder autistic traits are the phenotypic expression of a genetic predisposition to ASD and have named them the Broad Autism Phenotype (BAP). They have suggested that components of the BAP in relatives of people with ASD might be used as a complementary tool to studies exploring the genetics and other biological aspects of ASD. One questionnaire devised to assess the BAP in adults is the Autism Spectrum Quotient (AQ) which consists of 50 statements. Respondents are asked to indicate on a four-point scale how well that statement applies to them (strongly agree, agree, disagree, strongly disagree). A score of 1 is given to a response indicating an autistic-like trait (strongly agree/agree or strongly disagree/disagree) and 0 to a response not indicative of an autistic-like trait. Researchers have consistently found that parents of children with ASD score more highly than control parents, particularly in the social skills and communication sub-scales.

However, the idea that the BAP among parents of children with ASD reflects a genetic liability for the disorder does not take into account the challenges that arise as a result of rearing a child with a disability. Research has consistently identified the competing demands faced by parents of children with ASD. There is more supervision and less sleep, more expenses, and lower incomes. Challenges include coping with stigma and managing violent and/or self-harming behaviours in their children. Do the additional demands of parenting a child with ASD lead to behavioural and personality changes similar to the BAP that are being taken to represent a genetic liability for ASD?
I individually interviewed 16 mothers of children with ASD and comorbid ID (age range of children: 11–24 years) about their quality of life (QoL).(84) Although not seeking to explore the BAP, I noted that my hermeneutic phenomenological analysis(85) revealed that seven of the 16 mothers reported personality or behavioural changes since the onset of their child’s ASD, particularly in the area of social enjoyment and relationships. For example, when asked, ‘How has your daughter’s disability impacted on your social relationships?’ One mother explained: ‘I find that my personality has changed too. I used to love fun and loved to get with people and have a great time whereas now I just …’. She also reported that she used to enjoy planning for the future but now she did not bother. Another mother, when asked of her working status, detailed the enormous effort of raising her daughter and went on to say that her daughter’s ASD had put ‘bad grooves’ in her personality.

The attitudes of mothers towards social relationships were also found to be fundamentally altered following their child’s diagnosis of ASD. Mothers were asked: ‘Have you ever felt included or excluded because of your child’s disability?’ In response, some mothers spoke of the difficulty of retaining old friends and how it was almost impossible to instigate friendships. On retaining old friends, one mother expounded: ‘You can’t really go out with your friends anymore ... You’d have to run after your child. You can’t just sit down and say have a coffee. You’re always looking out the side there, wondering where she is’.

On instigating friendships, another mother explained: ‘You don’t visit people. You don’t have friends’.

Also on friendships, two mothers spoke of how they had neither the time nor energy to make new friends. The first mother reported: ‘I didn’t have a social life ... It takes effort to make friends. You have to reciprocate ... There was nothing that I could do apart from care for my children’.

The second mother said, ‘I haven’t had the time, the energy or the inclination to go and make friends’.
In response to the same question, a third mother described how she had lost the desire to be included. She reflected:

I’ve also lost interest in going out. I’m tired and so I just don’t have the energy to do the sort of partying that I was involved with before … I became incredibly socially isolated because the moment I tried to have a conversation my child would run away’.

Four of these comments align with items of the ‘Social Skill’ sub-scale of the AQ (Table 26).

Table 26: Alignment of the Autism Spectrum Quotient in items with personality changes after the child’s diagnosis

<table>
<thead>
<tr>
<th>Autism Spectrum Quotient item</th>
<th>Mothers’ comment</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. I prefer to do things on my own rather than with others.</td>
<td>I haven’t had the time, the energy or the inclination to go and make friends.</td>
</tr>
<tr>
<td>11. I find social situations easy (reverse scored).</td>
<td>I became incredibly socially isolated because the moment I tried to have a conversation my child would run away.</td>
</tr>
<tr>
<td>22. I find it hard to make new friends.</td>
<td>You can’t really go out with your friends anymore... You’d have to run after your child. You can’t just sit down and say have a coffee. You’re always looking out the side there, wondering where she is.</td>
</tr>
<tr>
<td>44. I enjoy social situations</td>
<td>I’ve also lost interest in going out.</td>
</tr>
</tbody>
</table>

My qualitative analyses did not identify similarities between mother responses and items pertaining to other scales on the AQ, such as ‘Attention to Detail’, ‘Attention Switching’ and ‘Communication and Imagination’. However, I noted that ‘false positives’ on even one scale can have substantial influence on the interpretation of study data. For example, if a mother endorsed all items presented in Table 26, she would end up with a score of 4 for the Social Skills sub-scale, which is a score that has been taken by previous studies to be indicative of the BAP in parents.(77) However, rather than solely reflecting a genetic liability for ASD, the nature and context of the comments provided by the mothers indicate that this score may be more indicative of the difficulties with social relationships that accompany caring for a child with ASD.

Compare Kanner’s description of mothers of children with ASD in his clinic to the following insight provided by a similar mother, in a similar situation, many years later.
This mother had made the decision to be very unemotional when talking to professionals about her son and acknowledged the following coping mechanism:

If I let myself get emotional with one of these people and with one of their questions, I thought the flood-gates are going to open and I’m never going to be able to get myself back from it.

Echoing Kanner, her husband reported ‘You come across as very cold-hearted’.

These preliminary data urge at least a degree of caution when interpreting the scores of the AQ and other BAP assessments, particularly in the domains of social engagement and enjoyment. Behaviours consistent with the BAP may not solely represent a genetic liability for ASD, and scores on these assessments may be inflated by an environmental reaction to having a child with ASD.

5.4 References


Chapter 6: Early mortality and primary causes of death in mothers of children with ID or ASD

In Chapter 5, I explored pre-existing differences in mothers of children with intellectual disability (ID) or autism spectrum disorder (ASD). Identifying that mothers with a pre-existing psychiatric disorder had about twice the chance of having a child with ID or ASD than other mothers (Section 5.2) motivated me to explore maternal health in a broader sense. The study reported in this chapter of the mortality of mothers of children with ID or ASD is the result. The abstract and authors of the paper are in Appendix 6.

6.1 Introduction

Poorer health, particularly mental health, has consistently been documented in the mothers of children with ID(1, 2) or ASD.(3–5) The degree of health impairment in mothers has been shown to vary according to the type of their child’s disability, to some extent. For example, in studies that compared the mental health of mothers of children with different developmental disabilities, mothers of children with Down syndrome had the least impaired health and mothers of those with ASD, the most impaired.(6–8)

About half of people with ASD have comorbid ID. Whether or not the ASD is associated with comorbid ID may also have an effect on maternal health. Moreover, due to the greater independence of their children, mothers of children with mild or moderate ID of unknown cause (mild–moderate ID) are likely to have different challenges than mothers of children with severe or profound ID of unknown cause (severe ID).

Earlier chapters identified that mothers with a previous psychiatric disorder were about twice as likely to have a child with ID or ASD than mothers without a psychiatric disorder(9) and we know that people with psychiatric disorders have higher mortality rates.(10) Hence, I was also interested in investigating whether the existence of a psychiatric disorder, either before or after the birth of the child, affected mortality. Diagnosis of ASD is increasing.(11) This, combined with the progressive closing of residential facilities for people with disabilities in developed nations,(12) is likely to
have resulted in many more mothers now caring at home for their children with ID or ASD.

The examination of mortality rates and identification of any causes of early death that are more common in particular groups of mothers of children with ID or ASD would enable services and interventions to be directed to those whose health is most at risk. In this way, any increased mortality might be reduced, along with the corresponding emotional and financial burden to affected families and economic burden to the community.

Therefore, for the study period, I aimed to:

1. Estimate the survival rates in mothers of children with ID or ASD and in those whose children have neither ID or ASD
2. Compare the risk of death in mothers of children from different sub-groups of ID and ASD with that of mothers whose children have neither ID or ASD
3. Examine the extent to which a psychiatric disorder can explain any observed differences in mortality rates
4. Identify the primary causes of death in mothers of children with ID or ASD and estimate the risks compared with mothers whose children have neither ID nor ASD.

6.2 Methods

6.2.1 Study population

The study population comprised all women who gave birth to a live child in Western Australia (WA) in the years from 1983 to 2005. The de-identified data were obtained from five state-wide sources. The first was the Midwives Notification System (MNS) and from here I obtained mothers’ socioeconomic status (SES) and the birthdates of mothers and their babies. Second, children’s diagnostic information pertaining to the presence of ID (including type and level) or ASD (including whether associated with ID) was provided by the IDEA database. Third, from the State Mortality Registry, I accessed dates and cause of death by ICD-9 or 10 codes of all mothers in the study population that had died from 1983 to 2010. Last, in order to explore the effect of a psychiatric disorder on mortality, I accessed datasets from the MHIS and the Hospital
Morbidity Data System (HMDS). I linked all datasets by using a unique alpha–numeric identifier created for each mother by WA’s Data Linkage Unit.(14)

### 6.2.2 Maternal groups and the index child

 Mothers were assigned to a group according to the disability of their index child. The maternal groups used for comparisons and the definition of the index child were described previously (Section 3.2).

### 6.2.3 Analyses

 Using Kaplan–Meier analysis,(15) I calculated the survival rates of mothers of children with any ID, any ASD and no ID or ASD. All survival curves were tested for differences using the log-rank test for equality of survivor functions.

 I tested the Proportional Hazards Assumption(16) with the dependent variable for the six core case groups and where failure was death by any cause and each of the primary cause of death categories. No hazard curve varied significantly from the baseline curve with respect to proportionality. Hence, I concluded that the data were suitable for Cox regression analysis.

 Maternal age and SES are both related to the risk of ID,(17) ASD(17) and death(18); hence I adjusted for these potential confounders. I created a three-level variable for age at the index birth: Less than 20 years, 20 to 35 years and 35 years or more.

 Socioeconomic status was measured by a three-level variable calculated from the Indices of Relative Socioeconomic Disadvantage(19) for 2001, which uses residence grouped by the unit termed ‘collection district’. For mothers where this was not available, I used the same index but with measures from 1996 or 2006 or a similar index for 2001, which used ‘statistical local areas’, although these are larger than collection districts. In my variable, ‘low’ pertained to the most disadvantaged quartile of scores, ‘medium’ to the inner two quartiles and ‘high’ to the most advantaged quartile, where I determined the quartiles from the study population. Due to our different definitions of
the index child which relate to parity, I also adjusted for parity which was measured using a four level variable.

Using Cox regression with ‘Death by any cause’ as the event of interest and time measured as the number of years since the index birth, I calculated the hazard ratios (HRs) for death by maternal case group compared to the comparison group. The period at risk extended from the date of the index birth until death or 31 December 2010, whichever came first. I calculated unadjusted HRs and HRs adjusted for each of maternal age at the index birth and SES.

### 6.2.4 Effect of a psychiatric disorder

I created a binary variable with the level of ‘Yes’ for women who had had an outpatient clinical psychiatric contact or a hospital admission in relation to a psychiatric diagnosis in WA during their lifetime. Using the Kaplan–Meier method, I compared the survival rates in mothers according to the existence of both a psychiatric disorder and the disability status of the index child. I also calculated the HRs for ‘Death by any cause’ for each of the three case groups and compared to mothers of children with no ID or ASD and no psychiatric disorder.

### 6.2.5 Cause of death

I grouped the causes of death into the 12 categories of Infections or parasites, Cancer, Diabetes, Cardiovascular diseases, Respiratory diseases, Pneumonia and influenza, Digestive diseases, Kidney diseases, Pregnancy complications, Misadventure, Other causes, Genetic and congenital disorders and Mental disorders. The first ten of these were formed from the 38 groups of the ICD-10 Cause of death codes.(20) The remaining two, Genetic and congenital disorders and Mental disorders were added as death was associated with these disorders for 59 mothers. The ICD codes pertaining to each diagnostic category are in Table 27. I performed Cox regression analyses where ‘failure’ was death attributed to each of the three largest causes of death categories. In turn, I used each of the three largest case groups—mothers of children with ID; mothers
of children with ID or ASD and mothers of children with ASD—as independent variables. Again, the period at risk extended until death from any cause or the end of the study period, whichever occurred first. The base level was the comparison group and I adjusted for maternal age at index birth and SES. STATA 12 was used for all analyses.
Chapter 6
Table 27: Cause of death code by diagnostic category
Category
Infections/parasites
Cancer

Diabetes
Cardiovascular
diseases

Respiratory diseases
& pneumonia
Digestive diseases
Kidney diseases
Pregnancy
complications
Misadventure

Genetic/ congenital
disorders
Mental disorders
Other causes

ICD-9 code
1369, 3239, 3240, 3241, 3249, 3249, 3409
1419, 1479, 1489, 1510, 1519, 1529, 1531, 1533,
1535-1537, 1539-1542, 1550, 1551, 1570, 1571,
1579, 1580, 1590, 1623, 1625, 1629, 1706, 1715,
1719, 1723, 1726, 1727, 1729, 1742, 1744, 1748,
1749, 1809, 1819, 1830, 1889, 1890, 1910-1912,
2002, 2008, 2019, 2028, 2030, 2040, 2050, 2051
2500, 2773, 362
3941, 3949, 3969, 3970, 3989, 4049, 4109, 4140,
4149, 4151, 4160, 4209, 421, 4210, 422, 4229, 4240,
4249, 4251, 4254, 4275, 4279, 4280, 4281, 429, 4309,
4319, 4321, 4329, 4331, 4369, 4371, 4410, 4411,
4472, 4478, 7100
2770, 463, 4819, 4859, 4869, 4939, 4949, 4969, 5159,
5163, 5168
389, 5301, 5314, 5350, 5532, 5679, 5698, 5712, 5713,
5715, 5718, 5733, 5770, 705
5820, 5822, 5859, 5869, 5939
6339, 6370, 6419, 6541, 6651, 6653, 6661, 6670,
6688, 6709, 6731, 6740, 6741
8051, 8100, 8120, 8121, 8139, 8147, 8150, 8151,
8159-8161, 8169, 8181, 8190, 8210, 8219, 8227,
8250, 8251, 8413, 8415, 8500, 8502, 8505, 8509,
8520, 8532, 8540, 8583, 8588, 8589, 8641, 8658,
8703, 8768, 8829, 8889, 8903, 8912, 8969, 9102,
9104, 9108, 9119, 9138, 9169, 9203, 9239, 9289,
9290, 9299, 9500, 9502- 9505, 9509, 9520, 9521,
9530, 9549, 9552, 9569, 9570, 9580, 9589, 9600,
9639, 9652, 9654, 9669, 9680, 9682, 9688, 9689,
9803, 9888
3310, 3318, 3352, 7424, 7469, 7472, 7595, 7598
2918, 2940, 3039, 3040, 3041, 3047, 3048, 3049,
3050, 3071
2127, 2270, 2609, 2780, 2790, 2791, 3453, 3459,
4511, 6144, 7101, 7109, 7140, 785, 7982, 7989, 799,
7998, 7999

ICD-10 code
A169, A391, A400, A403, A415, A419, A810, B169, B171, B182, B199, B200, B207, B227, B238, B24, B332, B449, B674, B942
C029, C07, C089, C099, C109, C140, C159, C161, C162, C169, C179, C180-C182, C184-C189, C19, C20, C210, C220, C221, C229, C23, C240,
C241, C250, C251, C259, C260, C261, C269, C310, C329, C340, C341-C343, C349, C37, C402, C419, C433, C434, C435, C436, C437, C439,
C442, C444, C449, C450, C459, C479, C480, C482, C494, C495, C499, C500, C501, C502, C503, C504, C505, C508, C509, C52, C530, C539,
C541, C55, C56, C64, C66, C679, C693, C700, C710-C713, C718- C720, C725, C729, C73, C740, C741, C749, C750, C755, C759, C762, C763,
C767, C786, C80, C812, C819, C822, C830, C833- C835, C837, C845, C851, C859, C900, C902, C910, C911, C919-C921, C924, C925, C959,
C97
I050, I059, I080, I089, I091, I099, I110, I119, I120,
I219, I250, I251, I255, I258, I259, I269, I270, I272,
J09, J110, J13, J152, J154, J159, J180, J181, J189, J42, J439, J440, J448, J449, J459, J46, J47, J690, J841, J849, J850, J851, J869, J969, J984,
J988
K047, K221, K254, K296, K318, K37, K409, K439, K519, K529, K550, K559, K564, K625, K631, K638, K650, K701, K703, K704, K709, K729,
K746, K754, K767, K769, K802, K819, K829, K830, K85, K852, K859, K869, K922
N039, N179, N180, N189, N19
O149, O721, O754, O85, O861
W34, W65, W650, W655, W67, W670, W69, W698, W708, W738, W74, W748, W750, W76, W760, W808, W809, W879, W990, X000,
X009, X038, X040, X06, X09, X310, X399, X400, X41, X410, X412, X419, X42, X420, X429, X44, X440, X445, X448, X449, X45, X450, X468,
X59, X599, X600, X604, X609, X61, X610, X615, X620, X63, X639, X64, X640, X644, X649, X67, X670, X674, X678, X679, X69, X70, X700,
X704, X705, X708, X709, X71, X710, X718, X730, X740, X76, X780, X782, X80, X802, X804, X814, X815, X818, X82, X824, X84, X910, X94,
X940, X944, X95, X99, X990, X994, X998, X999, Y00, Y000, Y008, Y034, Y04, Y040, Y069, Y08, Y09, Y090, Y098, Y099, Y140, Y148, Y158,
Y190, Y208, Y260, Y31, Y324, Y330, Y340, Y348, Y450, Y600, Y830, Y836, Y848, Y850, Y86

Q070, Q209, Q211, Q213, Q218, Q249, Q251, Q254, Q273, Q279
No equivalent ICD-10 codes
D350, D373, D379, D383, D430, D432, D469, D593, D619, D649, D65, D688, D689, D693, D696, D735, D849, D869, E055, E668, E669,
E752, E780, E785, E840, E849, E854, E859, F03, F100- F102, F109, F112, F162, F191, F192, F199, F329, G060, G061, G10, G122, G239,
G319, G35, G409, G419, G713, G903, G931, G934, G939, G961, I802, I81, I828, L031, L930, M311, M313, M321, M329, M331, M348,
M349, N110, N12, N139, N390, N719, N800, N832, Q282, R568, R98, R99, W790, W795, X458

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6.3 Results

6.3.1 Study population

The original dataset contained the records of 300,123 mothers and of these 3,693 (1.2%) had died before 2011. Their mean age of death was 42 years and their ages at death ranged from 16 to 74 years. The maternal age at the index birth and SES of these women are given by case groups in Table 28.

6.3.2 Survival and death by any cause

Five to 27 years after the birth of their index child, the survival of mothers of children with no ID and no ASD were around 98%, followed by 96% for mothers of children with ASD and 95% for mothers of children with ID. Mothers in both the ID (log-rank p-value = 0.00005) and ASD (log-rank p-value = 0.0436) case groups had significantly poorer survival than comparison mothers (Figure 14).

Before adjustment, all core case groups had greater risk of death by any cause. During the study period, mothers of children with ID of known cause (not Down syndrome) [2.31(95% CI: 1.6, 3.3)] and mothers of children with mild-moderate ID [2.29(95% CI: 1.9, 2.7)] had the highest risk of death by any cause. Mothers of children with Down syndrome [1.36(95% CI: 0.7, 2.7)] and mothers of children with severe ID [1.31(95% CI: 0.6, 2.9)] had the lowest risk of case mothers. In the final model, I adjusted for both maternal age at the index birth and SES. All HRs were slightly attenuated and remained >1. Hazard ratios for the mothers of children with ID of known cause (not Down syndrome) [2.27(95% CI: 1.6, 3.3)], mothers of children with mild-moderate ID [2.24(95% CI: 1.9, 2.6)], and mothers of children with ASD with ID [1.71(95% CI: 1.02, 2.8)] were significant (Figure 15).

Effect of a psychiatric disorder

I compared the cumulative survival of mothers with and without a psychiatric disorder and according to the disability status of their index child. Twenty-five years after the index birth, the cumulative survival of the comparison group and three case groups, and
the p-values associated with comparing the cumulative survival to that of the comparator group, in descending order, were as follows: Mothers with no psychiatric disorder and no child with ID or ASD had the highest cumulative survival of about 98.5% (p-value<0.00005); mothers with a psychiatric disorder and no child with ID or ASD had a cumulative survival of 95% (p-value<0.00005); and mothers with no psychiatric disorder and a child with ID or ASD had a cumulative survival of about 90% (p-value<0.00005) (Figure 16). Compared with mothers who had no psychiatric disorder and no child with ID or ASD, mothers with both a psychiatric disorder and a child with ID or ASD had about six and a half times the risk of death whereas those with a psychiatric disorder and no children with ID or ASD had about four times the risk of death. Mothers with no psychiatric disorder but a child with ID or ASD had a 52% increased risk of death.

**Figure 14: Kaplan–Meier survival rates of mothers of children with no ID or ASD, mothers of children with ID and mothers of children with ASD**

ID, intellectual disability; ASD, autism spectrum disorder
## Table 28: Characteristics of the study population by case group

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Comparator Group</th>
<th>Mild–moderate ID</th>
<th>Severe ID</th>
<th>Down syndrome</th>
<th>ID of known cause (not Down)</th>
<th>Any ID</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
<th>Any ASD</th>
<th>Any ID or ASD</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Maternal age at the index birth</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Less than 20 years</td>
<td>N=22,042</td>
<td>N=607</td>
<td>N=26</td>
<td>N=17</td>
<td>N=89</td>
<td>N=739</td>
<td>N=52</td>
<td>N=28</td>
<td>N=80</td>
<td>N=819</td>
<td>N=22,861</td>
</tr>
<tr>
<td></td>
<td>7.6%</td>
<td>10.7%</td>
<td>2.9%</td>
<td>7.9%</td>
<td>9.6%</td>
<td>3.7%</td>
<td>4.4%</td>
<td>3.9%</td>
<td>8.4%</td>
<td>7.6%</td>
<td></td>
</tr>
<tr>
<td>20 - 34 years</td>
<td>N=235,644</td>
<td>N=4,476</td>
<td>N=283</td>
<td>N=365</td>
<td>N=869</td>
<td>N=5,993</td>
<td>N=1,080</td>
<td>N=494</td>
<td>N=1,574</td>
<td>N=7,567</td>
<td>N=243,211</td>
</tr>
<tr>
<td></td>
<td>81.2%</td>
<td>79.0%</td>
<td>77.7%</td>
<td>62.7%</td>
<td>77.5%</td>
<td>76.7%</td>
<td>78.0%</td>
<td>77.1%</td>
<td>77.4%</td>
<td>81.0%</td>
<td></td>
</tr>
<tr>
<td>35 years or more</td>
<td>N=32,662</td>
<td>N=584</td>
<td>N=55</td>
<td>N=200</td>
<td>N=163</td>
<td>N=1,002</td>
<td>N=276</td>
<td>N=111</td>
<td>N=387</td>
<td>N=1,389</td>
<td>N=34,051</td>
</tr>
<tr>
<td></td>
<td>11.2%</td>
<td>10.3%</td>
<td>15.1%</td>
<td>34.4%</td>
<td>14.5%</td>
<td>13.0%</td>
<td>19.6%</td>
<td>17.5%</td>
<td>19.0%</td>
<td>14.2%</td>
<td>11.3%</td>
</tr>
<tr>
<td>Socioeconomic status</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low</td>
<td>N=64,570</td>
<td>N=2,297</td>
<td>N=129</td>
<td>N=115</td>
<td>N=374</td>
<td>N=2,915</td>
<td>N=351</td>
<td>N=144</td>
<td>N=495</td>
<td>N=3,410</td>
<td>N=68,080</td>
</tr>
<tr>
<td></td>
<td>22.2%</td>
<td>42.3%</td>
<td>35.4%</td>
<td>19.8%</td>
<td>33.4%</td>
<td>37.7%</td>
<td>24.9%</td>
<td>22.8%</td>
<td>24.3%</td>
<td>34.9%</td>
<td>22.7%</td>
</tr>
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<td>N=305</td>
<td>N=522</td>
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<td>N=1,026</td>
<td>N=4,486</td>
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</tr>
<tr>
<td></td>
<td>49.8%</td>
<td>43.5%</td>
<td>45.9%</td>
<td>52.4%</td>
<td>46.6%</td>
<td>44.7%</td>
<td>50.1%</td>
<td>50.6%</td>
<td>50.3%</td>
<td>45.9%</td>
<td>49.7%</td>
</tr>
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<td>N=156</td>
<td>N=319</td>
<td>N=1,218</td>
<td>N=319</td>
<td>N=150</td>
<td>N=469</td>
<td>N=1,687</td>
<td>N=73,936</td>
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<tr>
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<td>24.9%</td>
<td>12.1%</td>
<td>15.4%</td>
<td>25.1%</td>
<td>22.7%</td>
<td>15.7%</td>
<td>22.7%</td>
<td>23.7%</td>
<td>23.0%</td>
<td>17.3%</td>
<td>21.4%</td>
</tr>
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<td>117</td>
<td>12</td>
<td>27</td>
<td>32</td>
<td>188</td>
<td>N=19</td>
<td>N=32</td>
<td>N=51</td>
<td>N=239</td>
<td>N=9,011</td>
</tr>
<tr>
<td></td>
<td>3.0%</td>
<td>2.1%</td>
<td>3.3%</td>
<td>2.4%</td>
<td>2.3%</td>
<td>2.4%</td>
<td>2.3%</td>
<td>3.0%</td>
<td>2.5%</td>
<td>2.4%</td>
<td>3.0%</td>
</tr>
<tr>
<td>Existence of a psychiatric disorder</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Yes</td>
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<td>N=2,018</td>
<td>N=113</td>
<td>N=120</td>
<td>N=387</td>
<td>N=2,638</td>
<td>N=365</td>
<td>N=184</td>
<td>N=549</td>
<td>N=3,187</td>
<td>N=53,772</td>
</tr>
<tr>
<td></td>
<td>17.4%</td>
<td>35.6%</td>
<td>31.0%</td>
<td>20.6%</td>
<td>34.5%</td>
<td>34.1%</td>
<td>25.9%</td>
<td>29.1%</td>
<td>26.9%</td>
<td>32.6%</td>
<td>17.9%</td>
</tr>
<tr>
<td>TOTAL</td>
<td>290,348</td>
<td>5,667</td>
<td>364</td>
<td>582</td>
<td>1,121</td>
<td>7,354</td>
<td>1,408</td>
<td>663</td>
<td>2,041</td>
<td>9,775</td>
<td>300,123</td>
</tr>
</tbody>
</table>

ID, intellectual disability; ASD, autism spectrum disorder; Mild–moderate ID, mild or moderate intellectual disability of unknown cause; Severe ID, Severe or profound intellectual disability of unknown cause; Down, Down syndrome
Figure 15: Hazard ratios for death by maternal case group

ID, intellectual disability; ASD, autism spectrum disorder; Mild-moderate ID, Mild or moderate intellectual disability of unknown cause; Severe ID, Severe or profound intellectual disability of unknown cause; ID known cause (not Down), Intellectual disability of known cause (not Down syndrome); CL, confidence limits

Note: Hazard ratios are adjusted for SES, maternal age, parity and index birth year group.

6.3.3 Cause of death

The three primary causes of death in the study cohort were cancer (N = 1,452), misadventure (N = 843) and cardiovascular diseases (N = 433). The ICD-9 and 10 codes for each ‘cause of death’ category are in Table 27. Elevated HRs for death due to cancer ranged from 1.41(95% CI: 1.1, 1.8) for mothers of children with ID, and 1.54(95% CI: 0.8, 2.9) for mothers of children with ASD. The HRs for dying of a cardiovascular disease were 2.60(95% CI: 1.8, 3.9) in mothers of children with ID and 2.52(95% CI: 1.7, 3.7) in mothers of children with ID or ASD. For death by misadventure, HRs were significantly increased for mothers of children with ID or ASD [1.75(95% CI: 1.3, 2.4)] and ID [1.95(95% CI: 1.4, 2.7)]. All HRs and numbers of mothers by the case groups of ID, ASD and either ID or ASD are shown for the primary causes of death in Table 29. Analyses were not performed in cells with numbers of less than six as results would have been unreliable.(89)
Chapter 6

6.4 Discussion

6.4.1 Risk of death

Compared with mothers of no child with ID or ASD, mothers from each case group had an increased risk of death during the study period. When I adjusted for maternal age at the index birth and SES, all HRs were reduced but remained elevated, indicating that all case groups had an increased risk of death independent of their age at the index birth, parity and SES.

Significant HRs for death from any cause were found in mothers of children with ID of known cause (not Down syndrome), mothers of children with mild–moderate ID and mothers of children with ASD with ID. This suggests that these mothers were the most vulnerable to death of all the case groups during the study period.

The higher risk of death in the mothers of children with ID of known cause (not Down syndrome) may be due to the fact that some of these mothers have a genetic disorder,
such as neurofibromatosis or Fragile X syndrome that was inherited by their child. (21) Associated comorbidities such as arterial, vascular and malignant neoplasms with neurofibromatosis, (22, 23) and anxiety, social phobia, and depression in pre-mutation carriers of Fragile X, (24) could have been a contributing factor to the early death of these mothers.

Children with mild–moderate ID may have an undiagnosed, inherited cause for their disability. In mothers of such children, researchers in the US identified various associated medical conditions such as hypertension, diabetes and thyroid disease. (25) These may have contributed to their mothers’ early deaths. Further, WA researchers previously found that mothers of children with mild–moderate ID had an increased risk of epilepsy and asthma, which may have also affected their mortality. (21) The higher mortality of mothers of children with ASD might be mediated by their higher risk of hospitalisation for a psychiatric disorder (26) or their increased stress. (27) Some maternal conditions such as diabetes and epilepsy also increase the risk of ASD in subsequent offspring. For example, women with diabetes were found to have nearly three times the risk, and women with epilepsy, around four times the risk of a subsequent child with ASD. (21) The higher risk of death in these mothers could relate to the increased prevalence of these conditions. Both smoking (28) and obesity (29) increase the mortality risk and could be mediators of the increased risks identified in mothers of children with ID or ASD. Increased smoking is associated with higher levels of stress (30), and obesity with less exercise. (31) Mothers of children with ID or ASD have been shown to experience greater stress (32, 33) and one might expect that these time-poor women (34) also exercise less.

Researchers report that mothers of children with Down syndrome have less stress and increased subjective well-being than mothers of children with other forms of ID or ASD. (6, 35, 36) Further, mothers of children with Down syndrome would not have health issues that are genetically related to their children’s disability. Consistent with these factors, our results indicated that these mothers had the lowest risk of death during the study period of all case groups.
6.4.2 Effect of a psychiatric disorder

Psychiatric disorders were more prevalent in all case groups than in the comparison group. The increased prevalence ranged from 18% higher in mothers of children with Down syndrome to about 50% higher in mothers of children with ASD with ID, to more than double in mothers of children with mild-moderate ID (Table 28). Compared to mothers with no psychiatric disorder and no child with ID or ASD, I found that the poorest survival was in mothers with both a psychiatric disorder and a child with ID or ASD. We also showed that having a psychiatric disorder had greater effect on mortality than having a child with ID or ASD.

Figure 16: Kaplan-Meier survival rates of mothers according to psychiatric history and the disability status of the index child

![Kaplan-Meier survival rates graph]

psych, psychiatric; dis, disability; ID, intellectual disability; ASD, autism spectrum disorder

6.4.3 Cause of death

Research has identified that cancer and stress are positively correlated(37) and hence the higher risk of death from cancer might be mediated through the higher levels of stress experienced by these mothers. However, there is also the possibility that these mothers have higher mortality from cancer but not a higher incidence. This could be a result of lower levels of self-care in these women resulting in reduced participation in cancer screenings for breast, cervical and bowel cancer. This might be due to the
increased time constraints experienced by mothers in the care of their children with ID or ASD.

As with cancer, the increased risk of death due to cardiovascular diseases in mothers of children with ID or ASD might be caused by elevated stress levels compared to mothers of children without these disabilities as stress is also associated with an increased risk of cardiovascular disease.(38) Other contributing factors might be that women with psychiatric disorders have lower levels of self-care, including higher levels of smoking,(39) a risk factor for cardiovascular disease.(40) Further, research has documented increased rates of this disease in people with a psychiatric illness.(10) In my study, the Misadventure category includes all causes of death in the dataset related to homicide, suicide or accident. One might hypothesise that mothers of children with ID or ASD are more vulnerable to misadventure because, due to the care of their child with a disability, they have more challenges in their everyday lives,(41) more depression,(42) and less sleep,(43, 44)—known risk factors for accidents(45) and suicide.(46)

6.4.4 Strengths and weaknesses

Western Australia’s ID database, IDEA, made my study possible. Its main strength is the utilisation of data from a complete cohort with linkage of all relevant information. Further, my study investigates maternal death objectively and does not rely on the recall of family members for dates or causes of death. These two factors reduce bias and enhance the accuracy of my results. One weakness is the smaller numbers of children with ASD, which severely limits analyses for this group in most areas. Another is that the categorisation of ID may be incomplete; for example, some children may have had their condition diagnosed subsequent to registration with the database. This would result in their mothers being wrongly allocated to the ID of unknown cause case groups instead of the ID of known cause (not Down syndrome) case group. A final weakness is that the comparison group would have included a small number of mothers of children with other disabilities such as blindness and cystic fibrosis. All that is known of this group is that their children have neither ID nor ASD. This fact would have attenuated the results.
6.5 Summary

All maternal case groups had increased risk of death during the study period. Mothers with no psychiatric disorder and a child with ID or ASD were one and a half times as likely to die whereas mothers with a psychiatric disorder and no child with ID or ASD were more than four times as likely to die as mothers with no psychiatric disorder and no child with ID or ASD. This suggests that a mother having a psychiatric disorder has a higher risk of mortality than if she had a child with ID or ASD. Cancer, cardiovascular disease and misadventure were the three primary causes of death in case mothers. Mothers of children with either ID or ASD were 35–40% more likely to die of cancer during the study period than mothers of children without these disabilities. They were also two and a half times more likely to die from cardiovascular disease, and nearly twice as likely to die as a result of misadventure as comparison mothers. I hypothesise that these increased hazards may be related to the increased stress of raising a child with these disabilities.

6.6 Future implications

Apart from the Cancer category, small numbers in the case groups of mothers of children with ASD prohibited any analyses. Pooling these data with corresponding data from ASD registries from elsewhere might enable a greater understanding of factors increasing the mortality rates in mothers of children with ASD. In this way, informed services and preventions might be developed with the aim of improving the health and survival of these women.

6.7 References

Chapter 7: Onset of maternal psychiatric disorders after the birth of a child with ID or ASD

The previous chapter described the earlier mortality of mothers of children with Intellectual disability (ID) or autism spectrum disorder (ASD) and their primary causes of death. I also found that mothers with a psychiatric disorder had an increased risk of death compared to other mothers and more so if they also had a child with ID or ASD. These results led me to explore the rate of psychiatric disorders in mothers after the birth of a child with ID or ASD. My broad aim was to assess the burden of care in these mothers according to the sub-type of their child’s disability. In the second study of Chapter 5, I found that mothers with a pre-existing psychiatric disorder were about twice as likely to have a child with ID or ASD. In view of this result, I decided that it was important to consider only mothers without a pre-existing psychiatric disorder.

This chapter has three parts. In the first part, I compare the odds of a psychiatric disorder in mothers after the birth of a child with ID compared to other mothers. Part 2 details my study pertaining to the odds of a psychiatric disorder in mothers after the birth of a child with ASD compared to other mothers. Part 3 provides a comparison of psychiatric disorders in mothers of children with ID and mothers of children with ASD. Appendices 7 and 8 provide the abstracts and list of authors for each related paper.

7.1 Onset of maternal psychiatric disorders after the birth of a child with ID

7.1.1 Introduction

Intellectual disability is diagnosed in people with an IQ of less than 70 and deficits in adaptive functioning that are present before 18 years of age.(1) Children with ID have more challenging behaviours,(2) more sleep disorders(3) and more psychopathologies(4) than typically developing children. Their mothers also have increased expenses,(5) perceive more stigma against themselves or their child,(6) have
lower employment levels,(7) and less informal and family support(7) than other mothers. Therefore, it is not surprising that research has identified poorer mental health in mothers of children with ID compared to the parents of children with no disabilities.(8–10).

In my study described in Chapter 5 (Section 5.2), I found that mothers with an outpatient psychiatric history were about twice as likely to have a child with ID compared to mothers of children with no ID. I hypothesised that this might be due to shared genetics of the mother and the child with ID, prenatal use of medication or lifestyle factors in women with a psychiatric disorder. In this paper, I wanted to ascertain whether mothers of a child with ID and no previous psychiatric history were at increased risk of having a psychiatric disorder after the birth of their child. These comparisons would enable me to discern whether the burden of caring for a child with ID contributed to the increased rate of psychiatric disorders in the mothers. If this were the case, better informed services and interventions might be instituted with the aim of reducing the burden and improving maternal mental health. No previous research has attempted to differentiate whether the excess of psychiatric disorders in mothers of children with ID after the birth of their child is due to the increased burden of caring, a prior disposition to psychiatric disorders or to an increased exposure to antenatal risk factors for ID in women with a previous psychiatric disorder. Moreover, grouping mothers according to the level of ID of their child and according to whether the cause is known would enable the most vulnerable groups of mothers to be identified. The abstract and authors of the paper arising from this chapter comprise Appendices 7 and 8.

According to type and level of ID, I aimed to:

1. Compare the incidence of any psychiatric diagnosis in mothers after the birth of a child with ID compared to mothers with no child with ID or autism spectrum disorder (ASD) where mothers had no record of a psychiatric disorder before the birth
2. Compare the incidence of the most frequent psychiatric diagnostic categories, in mothers after the birth of a child with ID compared to mothers with no child with ID or ASD and where mothers had no record of a psychiatric disorder before the birth.
7.1.2 Methods

Study population

The study population consisted of all women who gave birth to a live child in Western Australia (WA) between 1 January 1983 and 31 December 2005 inclusive. I linked de-identified datasets from four statutory state-based registries and a state-wide disability database. The *Hospital Morbidity Data System* (HMDS)(11) provided admission dates and ICD-9 or ICD-10 codes for all hospital separations in WA from 1970 to 2010. The *Mental Health Information System* (MHIS)(11) provided appointment dates and the associated ICD-9 and ICD-10 codes for all public outpatient mental health contacts in WA from 1970 to 2010. The *Midwives Notification System* (MNS) provided me with the birthdates of all children born in WA during the collection period and socio-demographic information that I used to create explanatory variables. The WA Death Registry provided death dates of mothers and children, which allowed me to adjust for the period when women were at risk of a psychiatric disorder due to the burden of caring for their child. Using the *Intellectual Disability Exploring Answers* (IDEA) Database,(12) I gathered diagnostic information about children born between 1983 and 2005. Personnel from WA’s *Data Linkage Unit*(11) created a unique code for each mother, enabling the linkage of these datasets.

Maternal groups

I excluded mothers of children with ASD from the comparator group because researchers have reported that the mental health of mothers of children with ASD is poorer than that of mothers of typically developing children.(13, 14) Hence the comparator group was all women with a live child born between 1 January 1983 and 31 December 2005 and who had no child diagnosed with ID or ASD before 31 December 2010 and no record of a psychiatric disorder. For comparator mothers, the index child was the first child born during the collection period. I allocated mothers of a child with ID (but not ASD) into one of four case groups. These were labelled *mild or moderate ID of unknown cause* (mild–moderate ID), *severe or profound ID of unknown cause* (severe ID), *Down syndrome* and *ID of known cause (not Down syndrome)*. For these women, the index child was the eldest child with ID. When choosing case groups, I took
into account the particular challenges likely to result in differential burdens of care. For example; I separated mothers of children with severe ID from mothers of children with mild–moderate ID because children with severe ID are likely to have a much greater medical burden. Hence, the challenges faced by mothers could be expected to vary. The inter-relationships of these case groups are illustrated in Figure 17.

**Figure 17: Study population and maternal case groups**

![Diagram showing the relationships between different conditions and case groups](image)

ID, intellectual disability; ASD, autism spectrum disorder; ~, not; Down, Down syndrome; Mild–moderate ID, Mild or moderate intellectual disability of unknown cause; Severe ID, severe or profound intellectual disability of unknown cause

**Psychiatric history**

Mothers with a psychiatric disorder before the birth of their index child were excluded from the dataset. Hence, I excluded women with one or more diagnoses from the eleven blocks of Chapter 5 of the International Statistical Classification of Diseases and Related Health Problems 10th Revision (ICD-10)(15) or an ICD-9 equivalent code (Table 29).
Chapter 7

**Explanatory variables**

Researchers in WA(16) have previously demonstrated that socioeconomic disadvantage, young maternal age and high parity were associated with the risk of mild–moderate ID. Therefore, I included measures of these traits as independent variables in my model. I created a three-level variable for SES from the *Index of Relative Socioeconomic Disadvantage*(17) for 2001. I calculated maternal age at the index birth and used these ages to define a three-level variable that included *Less than 20 years, 20 to 34 years* and *35 years or older*. Parity at the time of index birth was defined by a four-level variable. The birth year of the index child was grouped into bands of 1983–88; 1989–94; 1995–2000 and 2001–05. I did not include births after 2005 as I reasoned that a five-year-leeway period was needed for children with milder levels of disability to have a reasonable opportunity to be diagnosed.

**Psychiatric status**

I used seven of the 11 blocks defined in ICD-10(15) to categorise psychiatric status after the index birth (Table 31). Block 1 (Organic mental disorders), Block 8 (Mental retardation), Block 9 (Disorders of psychological development) and Block 10 (Behavioural disorders) were omitted because I saw these as unlikely to develop in response to caregiving or because they were lifelong disorders. Excluded categories have been highlighted in Table 30. In order to determine the most frequent diagnostic categories, I created variables that counted the number of women with one or more diagnoses in each of the seven blocks of interest. For blocks with higher numbers of affected mothers, I created measures for women with one or more episodes from the block. These women were allocated a score that was equal to the sum of hospital admissions and outpatient contacts, which were associated with an ICD-10 code (or equivalent ICD-9 code) from the particular block. My final measure of psychiatric status was *any psychiatric disorder*. Women were allocated a score equal to the number of hospital admissions and outpatient contacts for a psychiatric disorder. Each measure was offset by ‘exposure’, which was the number of years from the index birth to either maternal death, death of the index child or the end of the study period, whichever was first.
### Table 30: Diagnostic categories and the associated ICD-9 and ICD-10 codes

<table>
<thead>
<tr>
<th>ICD-10 Block Description</th>
<th>ICD-9</th>
<th>ICD-10</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Organic mental disorders</td>
<td>290.0, 290.10, 290.11, 290.40, 293.0, 293.82, 293.89, 294.09, 294.1, 294.8, 294.9, 310.1, 310.2, 310.8, 310.9,</td>
<td>F00-F09</td>
</tr>
<tr>
<td>Organic, including symptomatic, mental disorders</td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. Alcohol and substance abuse</td>
<td>292.0, 292.83, 292.89, 292.9, 304.00, 304.10, 304.20, 304.30, 304.40, 304.50, 304.60, 304.80, 305.1, 305.20, 305.30, 305.40, 305.50, 305.60, 305.70, 305.90</td>
<td>F10-F19</td>
</tr>
<tr>
<td>Mental &amp; behavioural disorders due to alcohol /substance use</td>
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<td></td>
</tr>
<tr>
<td>Schizophrenia, schizotypal &amp; delusional disorders</td>
<td>297.1, 297.3, 297.8, 297.9, 298.3, 298.8, 298.9</td>
<td></td>
</tr>
<tr>
<td>Disorders in which the fundamental disturbance is a change in affect to depression</td>
<td></td>
<td></td>
</tr>
<tr>
<td>5. Neurotic disorders</td>
<td>300.00, 300.01, 300.02, 300.09, 300.11, 300.12, 300.13, 300.14, 300.15, 300.16, 300.19, 300.20, 300.21, 300.22, 300.23, 300.29, 300.3, 300.4, 300.5, 300.6, 300.7</td>
<td>F40-F48</td>
</tr>
<tr>
<td>Neurotic stress-related and somatoform disorders</td>
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<td></td>
</tr>
<tr>
<td>Behavioural syndromes associated with physiological/physical factors</td>
<td>Not F54 &amp; F55</td>
<td></td>
</tr>
<tr>
<td>7. Personality disorders</td>
<td>301, 312.4, 312.8, 312.9</td>
<td>F60-F69</td>
</tr>
<tr>
<td>Adult personality/behaviour disorders</td>
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<td></td>
</tr>
<tr>
<td>8. Mental retardation</td>
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<td>F70-F79</td>
</tr>
<tr>
<td>9. Disorders of psychological development</td>
<td>299.00, 299.10, 299.80, 299.90</td>
<td>F80-F89</td>
</tr>
<tr>
<td>315.00, 315.09, 315.1, 315.2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Behaviour/emotional disorders with onset in childhood/adolescence</td>
<td></td>
<td></td>
</tr>
<tr>
<td>11. Other psychiatric disorders</td>
<td>331.0, 368.16, 648.4, 655.45, 655.53, 780.1, 780.5, 784.6, 785.50, 799</td>
<td>F99, G30, G47, O35.4, R44, R45, R48</td>
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<td>Unspecified mental disorders</td>
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<td></td>
</tr>
</tbody>
</table>

Note: Excluded blocks have been shaded.

### Analyses

By maternal case groups, I calculated the incidence rate ratios (IRRs) of psychiatric disorders for the most frequently occurring categories, after the birth of the index child, and up to the end of 2010. I adjusted for SES, maternal age, parity and birth year band. I used negative binomial regression using STATA 13 and report the adjusted IRRs and associated 95% confidence intervals (CIs) for each measure of psychiatric status.

### 7.1.3 Results

After removing 20,583 (6.9%) mothers with a psychiatric disorder prior to the birth of their index child and all mothers and babies who had died on the date of the index birth, the cohort comprised 277,559 mothers. In Table 32, composition of the comparator group of 271,249 (97.7%) mothers and case groups in terms of the socio-demographic variables are shown. As previous research described,(16) mothers less than 20 years
were over-represented in the ‘mild–moderate ID of unknown cause case group’ (16) and over-represented in the lowest SES group. By frequency, the four primary diagnostic categories were Alcohol and substance abuse (N = 3,923), Schizophrenia spectrum disorders (N = 2,228), Affective disorders (N = 8,265) and Neurotic disorders (N = 8,441). See Table 32.

**Mild–moderate intellectual disability**

Mothers of children with mild–moderate ID, and no previous psychiatric disorder had significantly higher rates of all categories of psychiatric disorders compared to the mothers of children with no ID or ASD. These ranged from nearly three and a half times the rate for Schizophrenia spectrum disorders [3.49(95% CI: 1.6, 7.5)], nearly three times the rate for Alcohol and substance abuse disorders [2.91(95% CI: 2.0, 4.3)] and nearly twice the rate for Affective disorders [1.98(95% CI: 1.4, 2.8)], Any psychiatric diagnosis [1.80(95% CI: 1.5, 2.2)] and Neurotic disorders [1.80(95% CI: 1.3, 2.5)] (Table 33, Figure 18).

**Severe intellectual disability**

Mothers of children with severe ID and no previous psychiatric disorder had more than five times the rate of Affective disorders [5.12(95% CI: 1.4, 18.5)], nearly twice the rate of Neurotic disorders [1.98(95% CI: 0.6, 6.4)] and about one and a half times the rate of Schizophrenia spectrum disorders [1.56(95% CI: 0.1, 3.3)] as mothers without a child with ID or ASD and without a previous psychiatric disorder. These case mothers had reduced rates of Alcohol and substance abuse [0.58(95% CI: 0.1, 3.2)] and Any psychiatric disorder [0.85(95% CI: 0.4, 1.9)] (Table 33, Figure 19).
### Table 291: Maternal case groups by socio-demographic traits and index birth year

<table>
<thead>
<tr>
<th>Trait</th>
<th>Comparator group</th>
<th>Mild–mod ID</th>
<th>Severe ID</th>
<th>Down syndrome</th>
<th>ID (not Down syndrome)</th>
<th>Row total</th>
</tr>
</thead>
<tbody>
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<td>Low</td>
<td>61,615 (23.3%)</td>
<td>1,522 (33.5%)</td>
<td>79 (25.3%)</td>
<td>109 (23.8%)</td>
<td>236 (27.2%)</td>
<td>63,561 (23.5%)</td>
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<tr>
<td>Medium</td>
<td>133,512 (50.5%)</td>
<td>2,353 (51.8%)</td>
<td>170 (54.5%)</td>
<td>220 (48.0%)</td>
<td>453 (52.1%)</td>
<td>136,708 (50.6%)</td>
</tr>
<tr>
<td>High</td>
<td>69,076 (26.3%)</td>
<td>666 (14.7%)</td>
<td>63 (20.2%)</td>
<td>129 (28.2%)</td>
<td>180 (20.7%)</td>
<td>70,114 (25.9%)</td>
</tr>
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<td>7,046 (2.6%)</td>
<td>88 (1.9%)</td>
<td>10 (3.1%)</td>
<td>12 (2.6%)</td>
<td>20 (2.3%)</td>
<td>7,176 (2.6%)</td>
</tr>
<tr>
<td><strong>Maternal age at the index birth</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 20 years</td>
<td>19,764 (7.3%)</td>
<td>519 (11.2%)</td>
<td>24 (7.5%)</td>
<td>16 (3.4%)</td>
<td>71 (8.0%)</td>
<td>20,394 (7.4%)</td>
</tr>
<tr>
<td>20–34 years</td>
<td>221,229 (81.6%)</td>
<td>3,668 (79.2%)</td>
<td>249 (77.3%)</td>
<td>313 (66.6%)</td>
<td>691 (77.7%)</td>
<td>226,150 (81.5%)</td>
</tr>
<tr>
<td>&gt;35 years</td>
<td>32,256 (11.2%)</td>
<td>492 (9.6%)</td>
<td>49 (15.2%)</td>
<td>141 (30.0%)</td>
<td>127 (14.3%)</td>
<td>31,015 (11.2%)</td>
</tr>
<tr>
<td><strong>Parity at the index birth</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No previous child</td>
<td>199,960 (73.7%)</td>
<td>1,865 (40.3%)</td>
<td>132 (50.0%)</td>
<td>136 (28.9%)</td>
<td>397 (44.7%)</td>
<td>202,490 (73.0%)</td>
</tr>
<tr>
<td>1 previous child</td>
<td>39,687 (14.6%)</td>
<td>1,372 (29.6%)</td>
<td>102 (31.7%)</td>
<td>141 (30.0%)</td>
<td>256 (28.8%)</td>
<td>41,558 (15.0%)</td>
</tr>
<tr>
<td>2–3 previous children</td>
<td>28,218 (10.4%)</td>
<td>1,135 (24.5%)</td>
<td>75 (23.3%)</td>
<td>143 (30.4%)</td>
<td>196 (22.1%)</td>
<td>29,767 (10.7%)</td>
</tr>
<tr>
<td>&gt;3 previous children</td>
<td>3,384 (1.3%)</td>
<td>257 (5.6%)</td>
<td>13 (4.0%)</td>
<td>50 (10.6%)</td>
<td>40 (4.5%)</td>
<td>3,744 (1.4%)</td>
</tr>
<tr>
<td><strong>Birth year band</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1983–87</td>
<td>80,162 (29.6%)</td>
<td>1,026 (22.2%)</td>
<td>88 (27.3%)</td>
<td>106 (22.5%)</td>
<td>245 (27.6%)</td>
<td>81,627 (29.4%)</td>
</tr>
<tr>
<td>1988–93</td>
<td>65,800 (24.3%)</td>
<td>1,769 (38.2%)</td>
<td>128 (39.8%)</td>
<td>149 (31.7%)</td>
<td>243 (27.3%)</td>
<td>68,089 (24.5%)</td>
</tr>
<tr>
<td>1994–99</td>
<td>62,578 (23.1%)</td>
<td>1,227 (26.5%)</td>
<td>63 (19.6%)</td>
<td>98 (20.9%)</td>
<td>224 (25.2%)</td>
<td>64,190 (23.1%)</td>
</tr>
<tr>
<td>2000–05</td>
<td>62,709 (23.1%)</td>
<td>607 (13.1%)</td>
<td>43 (14.0%)</td>
<td>117 (26.8%)</td>
<td>177 (19.1%)</td>
<td>63,653 (22.9%)</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td>271,249 (97.7%)</td>
<td>4,629 (1.7%)</td>
<td>322 (0.1%)</td>
<td>470 (0.2%)</td>
<td>899 (0.3%)</td>
<td>277,559 (100%)</td>
</tr>
</tbody>
</table>

ID, intellectual disability; Mild–mod, Mild or moderate ID of unknown cause; Severe ID, Severe of profound ID of unknown cause; ASD, ID not Down, ID of known cause (not Down syndrome)
Table 302: Maternal psychiatric episodes by category and number in case group

<table>
<thead>
<tr>
<th>BLOCK/CATEGORY</th>
<th>Comparator group (No ID or ASD)</th>
<th>Mild–mod ID</th>
<th>Severe ID</th>
<th>Down syndrome</th>
<th>ID (not Down)</th>
<th>TOTAL</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alcohol &amp; substance abuse</td>
<td>3,697 (1.4%)</td>
<td>182 (3.9%)</td>
<td>8 (2.5%)</td>
<td>7 (1.5%)</td>
<td>29 (3.3%)</td>
<td>3,923 (1.4%)</td>
</tr>
<tr>
<td>Episodes/block</td>
<td>18,540</td>
<td>1075</td>
<td>9</td>
<td>32</td>
<td>230</td>
<td>19,886</td>
</tr>
<tr>
<td>Schizophrenia spectrum disorders</td>
<td>2,093 (0.8%)</td>
<td>109 (2.4%)</td>
<td>6 (1.9%)</td>
<td>3 (0.6%)</td>
<td>17 (1.9%)</td>
<td>2,228 (0.8%)</td>
</tr>
<tr>
<td>Episodes/block</td>
<td>93,315</td>
<td>8,347</td>
<td>35</td>
<td>31</td>
<td>230</td>
<td>103,959</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>7,881 (2.9%)</td>
<td>306 (6.6%)</td>
<td>14 (4.4%)</td>
<td>13 (2.8%)</td>
<td>10 (5.7%)</td>
<td>8,265 (3.0%)</td>
</tr>
<tr>
<td>Episodes/block</td>
<td>70,002</td>
<td>3,007</td>
<td>179</td>
<td>67</td>
<td>419</td>
<td>73,674</td>
</tr>
<tr>
<td>Neurotic disorders</td>
<td>8,038 (3.0%)</td>
<td>326 (7.0%)</td>
<td>19 (5.9%)</td>
<td>12 (2.6%)</td>
<td>46 (5.2%)</td>
<td>8,441 (3.0%)</td>
</tr>
<tr>
<td>Episodes/block</td>
<td>100,977</td>
<td>4,286</td>
<td>540</td>
<td>227</td>
<td>484</td>
<td>106,514</td>
</tr>
<tr>
<td>Personality disorders</td>
<td>1,573 (0.6%)</td>
<td>76 (1.6%)</td>
<td>4 (1.2%)</td>
<td>3 (0.6%)</td>
<td>10 (1.1%)</td>
<td>1,666 (0.6%)</td>
</tr>
<tr>
<td>Episodes/block</td>
<td>9,948</td>
<td>321</td>
<td>14</td>
<td>23</td>
<td>15</td>
<td>10,321</td>
</tr>
<tr>
<td>Other disorders</td>
<td>1,374 (0.5%)</td>
<td>64 (1.4%)</td>
<td>4 (1.2%)</td>
<td>1 (0.2%)</td>
<td>9 (1.0%)</td>
<td>1,452 (0.5%)</td>
</tr>
<tr>
<td>Episodes/block</td>
<td>21,705</td>
<td>830</td>
<td>13</td>
<td>6</td>
<td>12</td>
<td>22,566</td>
</tr>
<tr>
<td>Any psychiatric disorder</td>
<td>25,818 (9.5%)</td>
<td>890 (19.2%)</td>
<td>52 (16.2%)</td>
<td>50 (10.6%)</td>
<td>155 (17.4%)</td>
<td>26,965 (9.7%)</td>
</tr>
<tr>
<td>Episodes/category</td>
<td>493,694</td>
<td>24,365</td>
<td>1,053</td>
<td>914</td>
<td>4,772</td>
<td>524,798</td>
</tr>
<tr>
<td>Number of mothers in case group</td>
<td>271,249 (97.7%)</td>
<td>4,629 (1.7%)</td>
<td>322 (0.1%)</td>
<td>470 (0.2%)</td>
<td>889 (0.3%)</td>
<td>277,559 (100%)</td>
</tr>
<tr>
<td>Total episodes/group</td>
<td>319,715 (93.4%)</td>
<td>18,077 (5.3%)</td>
<td>795 (0.2%)</td>
<td>387 (0.1%)</td>
<td>3,415 (1.0%)</td>
<td>342,389 (100%)</td>
</tr>
</tbody>
</table>

Note: Some mothers have diagnoses in multiple categories
Table 313: Incidence rate ratios by block/category and case group

<table>
<thead>
<tr>
<th>Block/category</th>
<th>Mild–mild ID of unknown cause</th>
<th>Down syndrome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alcohol and substance abuse</td>
<td>3.45 (95% CI: 2.3, 5.1)***</td>
<td>1.13 (95% CI: 0.3, 4.0)†</td>
</tr>
<tr>
<td></td>
<td>2.91 (95% CI: 2.0, 4.3)***</td>
<td>0.99 (95% CI: 0.3, 3.4)†</td>
</tr>
<tr>
<td>Schizophrenia spectrum disorders</td>
<td>4.62 (95% CI: 2.2, 9.7)***</td>
<td>0.20 (95% CI: 0.02, 2.0)†</td>
</tr>
<tr>
<td></td>
<td>3.49 (95% CI: 1.6, 7.5)***</td>
<td>0.31 (95% CI: 0.03, 3.6)†</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>2.47 (95% CI: 1.8, 3.4)***</td>
<td>2.1 (95% CI: 0.7, 5.8)†</td>
</tr>
<tr>
<td></td>
<td>1.98 (95% CI: 1.4, 2.8)***</td>
<td>0.59 (95% CI: 0.2, 1.7)†</td>
</tr>
<tr>
<td>Neurotic disorders</td>
<td>2.40 (95% CI: 1.8, 3.2)***</td>
<td>0.72 (95% CI: 0.3, 1.9)†</td>
</tr>
<tr>
<td></td>
<td>1.80 (95% CI: 1.3, 2.5)***</td>
<td>0.68 (95% CI: 0.2, 1.8)†</td>
</tr>
<tr>
<td>Any psychiatric disorder</td>
<td>2.77 (95% CI: 2.3, 3.3)***</td>
<td>1.27 (95% CI: 0.7, 2.3)†</td>
</tr>
<tr>
<td></td>
<td>1.80 (95% CI: 1.5, 2.2)***</td>
<td>0.82 (95% CI: 0.4, 1.6)†</td>
</tr>
</tbody>
</table>

Severe ID of unknown cause | ID of known cause (not Down syndrome)

| Alcohol and substance abuse           | 0.49 (95% CI: 0.1, 2.5)†   | 3.49 (95% CI: 1.4, 8.5)†   |
|                                       | 0.58 (95% CI: 0.1, 3.2)†   | 2.77 (95% CI: 1.2, 6.5)†   |
| Schizophrenia spectrum disorders      | 0.36 (95% CI: 0.02, 5.9)†   | 5.89 (95% CI: 1.1, 31.3)C*  |
|                                       | 1.56 (95% CI: 0.1, 33.3)†  | 2.00 (95% CI: 0.4, 11.3)†  |
| Affective disorders                   | 4.0 (95% CI: 1.2, 13.7)†   | 1.4 (95% CI: 0.7, 2.9)C*   |
|                                       | 5.12 (95% CI: 1.4, 18.5)†  | 1.12 (95% CI: 0.5, 2.4)†   |
| Neurotic disorders                    | 2.20 (95% CI: 0.7, 6.8)†   | 1.73 (95% CI: 0.9, 3.4)†   |
|                                       | 1.98 (95% CI: 0.6, 6.4)†   | 1.73 (95% CI: 0.9, 3.5)†   |
| Any psychiatric disorder             | 1.70 (95% CI: 0.8, 3.5)†   | 2.71 (95% CI: 1.8, 4.2)*** |
|                                       | 0.85 (95% CI: 0.4, 1.9)†   | 1.68 (95% CI: 1.1, 2.7)†   |

Mild-moderate, Mild or moderate; ID, Intellectual disability; Severe, Severe or profound; C, crude IRR; A, adjusted IRR.

Note: IRRs are adjusted for SES, maternal age, parity and index birth year group.

* p-value <0.05, ** p-value <0.005, *** p-value <0.0005

Figure 18: IRRs for a psychiatric disorder by block/category of mothers of children with mild-moderate ID

IRR, incidence rate ratio; CL, confidence limit

Note: IRRs are adjusted for maternal age, SES, parity and index birth year band
Figure 19: Incidence rate ratios for a psychiatric disorder by block/category of mothers of children with severe ID

![Diagram showing incidence rate ratios for psychiatric disorders by block/category.]

IRR, incidence rate ratio; CL, confidence limit
Note: IRRs are adjusted for maternal age, SES, parity and index birth year band

**Down syndrome**

Mothers of children with Down syndrome and no previous psychiatric disorder had reduced IRRs for all categories of psychiatric disorders but none reached significance (Table 33, Figure 20).

Figure 20: Incidence rate ratios for a psychiatric disorder by block/category of mothers of children with Down syndrome

![Diagram showing incidence rate ratios for psychiatric disorders by block/category.]

IRR, incidence rate ratio; CL, confidence limit
Note: IRRs are adjusted for maternal age, SES, parity and index birth year band
Mothers of children with ID of known cause (not Down syndrome) and no previous psychiatric disorder had higher rates of all categories of psychiatric disorders after the birth of their index child. These mothers had more than two and a half times the rate of Alcohol and substance abuse [2.77(95% CI: 1.2, 6.5)], and about twice the rate of Schizophrenia spectrum disorders [2.00(95% CI: 0.4, 11.3)]. The rates of Affective disorders, Neurotic disorders and Any psychiatric disorder were elevated though not significant (Table 33, Figure 21).

7.1.4 Discussion

I explored the incidence of primary psychiatric disorders in mothers with no previous psychiatric history and after the birth of their child with ID, compared to mothers of children with no ID, no ASD and no psychiatric history whilst adjusting for socio-demographic factors. In this way, I was able to determine if the burden of caring for their child with a disability had contributed to a higher incidence of psychiatric disorders.
**Case group similarities**

**Mild–moderate and severe intellectual disability**

Common genetic pathways have been found for ID and schizophrenia.(18) Hence, mothers of children with mild–moderate or severe ID may be more likely to have a genetic propensity for schizophrenia than other mothers. Add to this genetic propensity an environmental trigger (such as stress), and the phenotype of schizophrenia might result.(19) Thus, a genetic susceptibility, combined with the added challenges of caring for a child with ID, may have contributed to the increased incidence of schizophrenia in these mothers. Mothers of children with either mild–moderate or severe ID have significantly elevated rates of affective disorders compared to mothers of children with no ID or ASD. In these mothers, both self-report and validated questionnaires have attributed this poorer mental health to the burden of caring for their child with a disability.(20)

**Mild–moderate ID and ID of known cause (not Down syndrome)**

The psychiatric profile of mothers of children with mild–moderate ID and mothers of children with *ID of known cause (not Down syndrome)* were similar (Figure 22). All IRRs were elevated in both groups but higher in the mothers of children with mild–moderate ID than in the other group. These similarities might be explained in terms of the relationship between these two groups. Possibly, some of the mothers with diagnoses in this category had undiagnosed alcohol or substance abuse problems during their pregnancy. Hence, the only difference between some mothers in these case groups would be that the children of the mothers with mild–moderate ID have an undiagnosed *Fetal alcohol spectrum disorder* (FASD) and the children with *ID of known cause (not Down syndrome)* have a diagnosed FASD. The diagnosis of FASD can be problematic as it may rely on maternal self-report of alcohol consumption and is considered to be under-diagnosed in Australia.(21) Finally, the elevated IRRs for *Schizophrenia spectrum disorders* in both groups may be in part due to an interaction of the incidence rate of these disorders and that of *Alcohol and substance abuse*. This may have resulted cannabis use influencing the subsequent development of schizophrenia.(22)
There are likely to be multiple reasons for the poorer psychiatric health of the mothers of children with mild–moderate ID compared to that of mothers of children with a known cause other than Down syndrome for their ID. Mothers in the second group have a causal diagnosis for their children’s condition. This may have the advantage of putting a mother in contact with a relevant support group and other mothers of children with the same condition. Further, knowing the cause has the distinct advantage of providing parents with information about potential treatments, ongoing research in the area and a likely prognosis for their child. Parents with a known cause for their child’s disability are further empowered as they are informed in relation to the likelihood of re-occurrences of the condition in future offspring and in the offspring of their typically developing children.

**Other case groups**

**Severe ID and mild–moderate ID**

Mothers of children with either severe ID or mild–moderate ID had significantly higher rates of affective disorders. Notably, the rate in mothers of children with severe ID was more than twice that of mothers of children with mild–moderate ID. In this study, the category *Affective disorders* included bipolar and depressive disorders. Other researchers have concluded that mothers with bipolar disorder or unipolar major depression were more likely to have a child with ID. Using the Beck Depression Inventory (BDI), mothers of children with developmental delay exhibited more depression than mothers of children without these disabilities, as did Latina mothers of children with ID compared to Latina mothers of typically developing children. However, I found no study that compared the levels in mothers according to the level of the ID of their child. I believe that this is the first research that suggests mothers of children with severe ID have higher rates of affective disorders than mothers of children with mild–moderate ID.
Figure 22: IRRs of psychiatric disorders in mothers of children with mild-moderate ID and mothers of children with ID of known cause (not Down syndrome)

IRR, incidence rate ratios; Mild-moderate ID, Mild or moderate intellectual disability of unknown cause; ID ~Down, Intellectual disability of known cause (not Down syndrome)
Mothers with no previous psychiatric disorder prior to the birth of a child with Down syndrome had lower rates of psychiatric disorders in all areas compared to other case and comparator mothers. This is consistent with previous research that suggested that the mental health of mothers of children with Down syndrome was less impaired than those of mothers with other forms of ID (28, 29). On the other hand, others have assessed that the mental health of mothers of children with Down syndrome is poorer than that of mothers of typically developing children (8, 30). This study was restricted to mothers with no previous psychiatric disorder whereas some of the mothers in the referenced studies (8, 30) may have had a pre-existing psychiatric disorder, contributing to adverse mental health outcomes after the birth. This, along with their smaller sample sizes, may account for the differing conclusions.

Reasons for the better psychiatric health of the mothers of children with Down syndrome compared to other case mothers might relate to the relative ease and the early timing of a diagnosis. This is often not the case for other forms of ID. For example, around half of mothers of children with ID never find out a cause for their child’s disability (31). Further, unlike some other forms of ID, mothers of children with Down syndrome would have had the opportunity to receive support from a well-established organisation at the time of initial diagnosis and subsequently (32). Third, the fact that Down syndrome is not hereditary and is caused by an accident at meiosis means that the mother is less likely to feel guilty about the occurrence of the disorder in her child. This is contrasted to maternal guilt and stigmatisation that might be associated with FASD (33) or unjustified, but nevertheless, real guilt where the mother is a carrier in an X-linked disorder such as Fragile X syndrome (34). Finally, the apparent increased resilience of these mothers might be associated with the increased rewards and subjective well-being that mothers of children with Down syndrome have reported in relation to mothers of children with other developmental disabilities (35).
7.1.5 Strengths and limitations

The population-based nature of this study was a considerable strength and greatly limited the risk of selection bias. Further, I was able to retrospectively access hospital and outpatient records collected over 40 years and, unlike those of other studies in this area, (36–38) my data were independent of maternal recall. The existence of the IDEA database allowed me access to ID diagnoses that were categorised by level and cause. The exclusion of all mothers with pre-existing psychiatric disorders enabled me to view emergent psychiatric disorders that were associated with the onset of caring. To my knowledge, this has not previously been done.

Unfortunately, I had no access to private outpatient data, which meant that some mothers with psychiatric disorders would not have been identified in my study. This would have attenuated the IRRs for mothers in case groups. A small number of immigrant mothers and mothers from interstate may have been wrongly assessed as having no previous psychiatric disorder due to their records not being in state registries. This would also have attenuated the IRRs for mothers in case groups. The allocation of the ‘index child’ was necessarily different for comparator and case mothers. For comparator mothers, the index child was their first child born from 1983 to 2005. This resulted in the index child being the eldest for 74% of comparator mothers but only the eldest for about 40% of case mothers (not Down syndrome) and 29% for mothers of children with Down syndrome (Table 32). However, I adjusted for index parity, which would have reduced the potential bias caused by this inequality.

7.1.6 Conclusion and implications

In this study, I excluded mothers who had a hospitalisation or an outpatient contact for a psychiatric disorder in WA before the index birth. I made adjustments for maternal age, parity, SES and the year band of the index birth, all of which might have been related to the odds of a subsequent psychiatric disorder. Therefore, it is reasonable to conclude that the elevated (or attenuated) incidence of psychiatric disorders I identified is mostly due to the burden of caring rather than genetics or pre-existing environmental factors. Hence, I concluded that the burden of caring for a child with ID of known cause (not
Down syndrome) and particularly of mild–moderate ID without a known cause increases the risk of a psychiatric disorder after the birth of their child. I did not find this association for mothers of children with Down syndrome. Exploring the IRRs of psychiatric disorders in these same sub-groups of ID but in mothers with previous psychiatric disorders might provide evidence of groups of mothers who are particularly vulnerable after the birth of their child with ID.
7.2 Onset of maternal psychiatric disorders after the birth of a child with ASD

7.2.1 Introduction

Autism spectrum disorder is a severe, lifelong neurobiological disorder in people that exhibit deficits in the areas of sociability, communication and behaviour.(1) Persons with IQs <70 and impairments in adaptive functioning presenting before 18 years are diagnosed with ID.(1) Up to 60% of children with ASD have been reported to have comorbid ID.(16, 39) Researchers have described the mental health of mothers of children with ASD as poorer than mothers of typically developing children(13, 14) and poorer than that of mothers of children with other developmental disabilities.(40, 41) However, children with ASD both with and without comorbid ID are likely to provide quite different challenges for their caregivers. A significant proportion of those with ASD with ID are non-verbal(42) and most require high levels of support throughout their lives.(43) In contrast, those with ASD and no ID often attend mainstream schools(44) and many are able to live independently as adults.(43) However, no research was identified that investigated whether the presence of comorbid ID in the affected children altered the relationship with maternal mental health. Over the last 50 years, many residential institutions for people with ID have been closed.(45) This, combined with the increasing prevalence of ASD,(46) means that many more mothers are caring for their children with ASD in their family home. The results reported in Section 5.2 found that mothers with a pre-existing psychiatric disorder were at increased risk of having a child with ASD or ID. However, my previous research also showed that having a child with ID or ASD can also adversely affect maternal mental health. Hence, for mothers with no previous psychiatric history I wanted to compare the onset of psychiatric disorders in those with children with ASD (taking into account the presence or absence of comorbid ID) and in those whose children had neither ID nor ASD. In this way, an informed measure of the relative effect of caring for their children on maternal mental health and the relative vulnerability of different groups might be determined. Hence, better informed services and interventions might be developed with the aim of targeting the most vulnerable mothers.
Hence, focusing on mothers with no previous psychiatric history, my aims were to:

1. Compare the incidence of a psychiatric diagnosis after the birth of a child with ASD, according to the presence of comorbid ID, to that in mothers of children without such disabilities
2. Compare the incidence of the most frequent diagnostic categories of psychiatric disorders in the same groups of mothers.

### 7.2.2 Methods

#### Study population

The study population was described in 7.1.2.

#### Maternal groups

All records of mothers of children with ID but without ASD were excluded so that the comparator group comprised all mothers of children born during the study period who had not been diagnosed with ID or ASD before 1 January 2011. In these mothers, I defined their *index child* as their eldest child who was born during the study period. For case mothers, the index child was the eldest child diagnosed with ASD before 1 January 2011. Case mothers were divided into two case groups where the first was all mothers whose index child was diagnosed with ASD with ID. The second case group comprised all mothers whose index child was diagnosed with ASD but without ID. The inter-relationships between the comparator and case groups are shown in Figure 23.
Psychiatric history

Mothers with a psychiatric disorder before the birth of their index child were excluded from the dataset. The associated methods are described under this same heading in 7.1.2.

Explanatory variables

Researchers have previously demonstrated that socioeconomic advantage, increased maternal age and low parity are associated with the risk of ASD in WA.(16) The explanatory variables were described before in 7.1.2 under this heading.

Psychiatric status

Psychiatric status was described under this same heading in 7.1.2.

Analyses

A description of the analyses is in 7.1.2 under this heading.
7.2.3 Results

In Table 34, number and percentage of mothers are given by case group, socio-demographic traits and index birth year band. Socioeconomic status was distributed fairly uniformly throughout the two ASD case groups. Mothers from the comparator group were more likely to be of lower parity than mothers from either of the ASD case groups. Older mothers were more likely to have a child with ASD, both with and without ID. The number of children born and subsequently diagnosed with ASD with ID increased with subsequent year bands. This was not the case for ASD without ID where the most recent year band had the smallest number of children born who later were given this diagnosis. Women were most frequently allocated to the four blocks: Alcohol and substance abuse; Schizophrenia spectrum disorders; Affective disorders; and Neurotic disorders (Table 35).

Table 324: Maternal characteristics by case group

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Comparator group No ID or ASD</th>
<th>Case group 1 ASD with ID</th>
<th>Case group 1 ASD without ID</th>
<th>Row totals</th>
</tr>
</thead>
<tbody>
<tr>
<td>Socioeconomic status at the index birth</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low</td>
<td>61,615 (22.7%)</td>
<td>254 (22.0%)</td>
<td>104 (19.2%)</td>
<td>61,973 (22.7%)</td>
</tr>
<tr>
<td>Medium</td>
<td>133,512 (49.2%)</td>
<td>634 (54.8%)</td>
<td>305 (56.3%)</td>
<td>134,451 (49.3%)</td>
</tr>
<tr>
<td>High</td>
<td>69,076 (25.5%)</td>
<td>244 (21.1%)</td>
<td>121 (22.3%)</td>
<td>69,441 (25.4%)</td>
</tr>
<tr>
<td>Missing</td>
<td>7,046 (2.6%)</td>
<td>24 (2.1%)</td>
<td>12 (2.2%)</td>
<td>7,082 (2.6%)</td>
</tr>
<tr>
<td>Parity at the index birth</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No previous children</td>
<td>199,960 (73.7%)</td>
<td>553 (47.8%)</td>
<td>289 (53.3%)</td>
<td>200,802 (73.6%)</td>
</tr>
<tr>
<td>1 previous child</td>
<td>39,687 (14.6%)</td>
<td>393 (34.0%)</td>
<td>159 (29.3%)</td>
<td>40,239 (14.7%)</td>
</tr>
<tr>
<td>2-3 previous children</td>
<td>28,218 (10.4%)</td>
<td>187 (16.2%)</td>
<td>79 (14.6%)</td>
<td>28,484 (10.4%)</td>
</tr>
<tr>
<td>&gt;3 previous children</td>
<td>3,384 (1.3%)</td>
<td>23 (2.0%)</td>
<td>15 (2.8%)</td>
<td>3,422 (1.3%)</td>
</tr>
<tr>
<td>Maternal age at the index birth</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 20 years</td>
<td>19,764 (7.3%)</td>
<td>42 (3.6%)</td>
<td>23 (4.2%)</td>
<td>19,829 (7.3%)</td>
</tr>
<tr>
<td>20-29 years</td>
<td>160,675 (59.2%)</td>
<td>554 (47.9%)</td>
<td>266 (49.1%)</td>
<td>161,495 (59.2%)</td>
</tr>
<tr>
<td>30-39 years</td>
<td>81,495 (30.0%)</td>
<td>488 (42.2%)</td>
<td>223 (41.1%)</td>
<td>82,206 (30.1%)</td>
</tr>
<tr>
<td>&gt; 40 years</td>
<td>9,315 (3.4%)</td>
<td>72 (6.2%)</td>
<td>30 (5.5%)</td>
<td>9,417 (3.5%)</td>
</tr>
<tr>
<td>Index birth year band</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1983-7</td>
<td>80,162 (29.6%)</td>
<td>56 (4.8%)</td>
<td>51 (9.4%)</td>
<td>80,269 (29.4%)</td>
</tr>
<tr>
<td>1988-93</td>
<td>65,800 (24.3%)</td>
<td>183 (15.8%)</td>
<td>188 (34.7%)</td>
<td>66,171 (24.2%)</td>
</tr>
<tr>
<td>1994-99</td>
<td>62,578 (23.1%)</td>
<td>413 (35.7%)</td>
<td>276 (50.9%)</td>
<td>63,267 (23.2%)</td>
</tr>
<tr>
<td>2000-5</td>
<td>62,709 (23.1%)</td>
<td>504 (43.6%)</td>
<td>27 (5.0%)</td>
<td>63,240 (23.2%)</td>
</tr>
<tr>
<td>Total</td>
<td>271,249 (100%)</td>
<td>1,156 (100%)</td>
<td>542 (100%)</td>
<td>272,947 (100%)</td>
</tr>
</tbody>
</table>

ID, intellectual disability; ASD, autism spectrum disorder
All comparisons are for mothers without a pre-existing psychiatric disorder. Compared to mothers of no child with ID or ASD, mothers of children with ASD with ID had a 17% [1.17(95% CI: 0.7, 2.1)] increased incidence of a psychiatric disorder compared to a 140% increased incidence [2.40(95% CI: 1.1, 5.3)] in mothers of children with ASD without ID. Mothers of children with ASD with ID had nearly four times the incidence of Schizophrenia spectrum disorders [3.75(95% CI: 0.8, 17.8)] and elevated IRRs for Affective disorders [1.36(95% CI: 0.7, 2.7)] and Neurotic disorders [1.11(95% CI: 0.6, 2.1)]. Compared to mothers of children with ASD with ID, mothers of children with ASD without ID had less than a third the risk of Schizophrenia spectrum disorders [1.21(95% CI: 0.1, 11.7)] but about three times the incidence of Affective disorders [2.88(95% CI: 1.1, 7.7)] and Neurotic disorders [3.19(95% CI: 1.3, 7.8)]. However, mothers of children with ASD with ID had a reduced IRR for Alcohol and substance abuse [0.27(95% CI: 0.1, 0.8)] and mothers of children with ASD without ID had about the same incidence as other mothers [1.01(95% CI: 0.3, 3.4)]. All results are in Table 36 and Figure 24.

Table 335: Psychiatric episodes by block/category and case group

<table>
<thead>
<tr>
<th>BLOCK/CATEGORY</th>
<th>Comparator group</th>
<th>Case group 1</th>
<th>Case group 2</th>
<th>TOTAL</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No ASD or ID</td>
<td>ASD with ID</td>
<td>ASD without ID</td>
<td></td>
</tr>
<tr>
<td>Alcohol and substance abuse</td>
<td>3,697 (1.4%)</td>
<td>6 (0.5%)</td>
<td>10 (1.9%)</td>
<td>3,713 (1.4%)</td>
</tr>
<tr>
<td>Schizophrenia spectrum disorders</td>
<td>2,093 (0.8%)</td>
<td>11 (1%)</td>
<td>9 (1.7%)</td>
<td>2,113 (0.8%)</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>7,881 (2.9%)</td>
<td>50 (4.3%)</td>
<td>36 (6.7%)</td>
<td>7,967 (2.9%)</td>
</tr>
<tr>
<td>Neurotic disorders</td>
<td>8,038 (3.0%)</td>
<td>84 (4.7%)</td>
<td>41 (7.6%)</td>
<td>8,133 (3.0%)</td>
</tr>
<tr>
<td>Behaviour disorders</td>
<td>1,860 (0.7%)</td>
<td>15 (1.3%)</td>
<td>6 (1.1%)</td>
<td>1,881 (0.7%)</td>
</tr>
<tr>
<td>Personality disorders</td>
<td>1,573 (0.6 %)</td>
<td>7 (0.6%)</td>
<td>10 (1.9%)</td>
<td>1,590 (0.6 %)</td>
</tr>
<tr>
<td>Other disorders</td>
<td>1,374 (0.5%)</td>
<td>6 (0.5%)</td>
<td>10 (1.9%)</td>
<td>1,390 (0.5%)</td>
</tr>
<tr>
<td>Any psychiatric disorder</td>
<td>25,818 (9.5%)</td>
<td>168 (14.5%)</td>
<td>97 (17.9%)</td>
<td>26,083 (9.6%)</td>
</tr>
</tbody>
</table>

ASD, autism spectrum disorder; ID, intellectual disability
### Table 36: Incidence rate ratios of mothers by psychiatric category and case group

<table>
<thead>
<tr>
<th>Block/category</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alcohol and substance abuse</td>
<td>0.29(0.1, 0.7)Φ*</td>
<td>0.78(0.2, 2.7)Φ</td>
</tr>
<tr>
<td></td>
<td>0.27(0.1, 0.8)#*</td>
<td>1.01(0.3, 3.4)#</td>
</tr>
<tr>
<td>Schizophrenia spectrum disorders</td>
<td>1.66(0.4, 7.4)Φ</td>
<td>1.52(0.2, 13.5)Φ</td>
</tr>
<tr>
<td></td>
<td>3.75(0.8, 17.8)#</td>
<td>1.21(0.1, 11.7)#</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>1.73(0.9, 3.4)Φ</td>
<td>4.23(1.6, 11.0)Φ*</td>
</tr>
<tr>
<td></td>
<td>1.36(0.7, 2.7)#</td>
<td>2.88(1.1, 7.7)#*</td>
</tr>
<tr>
<td>Neurotic disorders</td>
<td>1.1(0.7, 2.4)Φ</td>
<td>4.5(1.9, 10.7)Φ**</td>
</tr>
<tr>
<td></td>
<td>1.11(0.6, 2.1)#</td>
<td>3.19(1.3, 7.8)#*</td>
</tr>
<tr>
<td>Any psychiatric disorder</td>
<td>1.36(0.8, 2.3)Φ</td>
<td>3.29(1.5, 7.0)Φ*</td>
</tr>
<tr>
<td></td>
<td>1.17(0.7, 2.1)#</td>
<td>2.4(1.1, 5.3)#*</td>
</tr>
</tbody>
</table>

ASD, autism spectrum disorder; ID, intellectual disability
Significant IRRs have been shaded
Φ, Unadjusted IRRs
#, IRRs have been adjusted for SES, maternal age and parity all at the time of the index birth and index birth year band
*p-value <0.05, **p-value <0.005, ***p-value <0.0005

### Figure 24: Incident rate ratios for psychiatric disorders by case group and block/category

ASD, autism spectrum disorder; ID, intellectual disability; Alc/subst, Alcohol and substance abuse; dis, disorder; Affect; Affective; Neuro, Neurotic; Psych, Psychiatric.
Note: IRRs are adjusted for SES, parity and maternal age, all at the index birth and Index birth year band.
7.2.4 Discussion

In this study, mothers of children with ASD did not tend to be of higher SES as noted by other researchers. (16) This may be because I included only women without a pre-existing psychiatric disorder. Researchers have noted that mothers of children with ASD have lower parity than other mothers. (16) This was not the case in this study. Here, mothers from the comparator group were more likely to be of lower parity than mothers from either of the ASD case groups because the index child was the eldest child born during the study period. In contrast, the index child for case mothers was the eldest child with a disability born during the study period. As found in other studies, (16) older maternal age was associated with both ASD with and without ID. The number of children born and subsequently diagnosed with ASD increased with successive Index birth year bands (Table 34) with one exception. Due to the documented increase in the diagnostic prevalence of ASD, (46) I expected increases with time. The exception was for the case group: ASD without ID where the number of children born between 2000 and 2005 was only 27 whereas in the preceding band, 279 children had been diagnosed. I consider this to be because some children in this study would not have been diagnosed as the youngest would have been only six years old in 2011 when the last records of diagnoses were obtained. The average age for diagnosis for Asperger syndrome, a large sub-group of ASD without ID, is seven years. (47) This is not the case for ASD with ID where the average age for diagnosis is about three years. (48)

Mothers of children with ASD without ID had a significantly elevated incidence of psychiatric disorders and particularly affective and neurotic disorders. The elevated incidences for these disorders ranged from nearly twice to about three times the incidence of comparator mothers. In mothers of children with ASD with ID, there was a reduced risk of Alcohol and substance abuse than for comparator mothers. However, what is most notable is the fact that these two groups are presenting as quite distinct in terms of their risk of psychiatric disorders after the birth of their index child.

This study was a large population-based, retrospective cohort study (N = 272,947), which reduced its risk of selection bias. In addition, access to 40 years of retrospective medical records eliminated the bias associated with personal or family recall. Thus I was able to exclude mothers with a psychiatric history prior to the birth of their index
child and identify psychiatric disorders with onset subsequent to the child’s birth. I accessed both inpatient and outpatient data, which enabled me to include the records of women with a psychiatric disorder that did not warrant hospitalisation. In contrast, some other studies relied on maternal recall,(13, 40, 49, 50) had comparatively small sample sizes(40, 50) or analysed only hospitalisation data.(14, 51) Unlike many other studies,(13, 14, 40, 50, 52–54) this one also benefitted from information on whether or not ASD was associated with comorbid ID.

A limitation of this study was the unavailability of private outpatient data in WA. This meant that outpatient data were not available for women who consulted their family doctor or private psychiatrist for treatment of a psychiatric disorder. Due to the increased cost of the latter option, such women may have been more likely to be from a higher SES background. My adjustment for maternal SES would have reduced any resulting bias.

My records are state based. Hence, a small proportion of immigrant or interstate mothers who had psychiatric disorders diagnosed elsewhere may have been wrongly categorised. Therefore I could have a small number of women in the comparison group that had in fact had a previous psychiatric disorder, which would have attenuated my results. Necessarily, the index child was defined differently for case and comparator mothers with the end result that the index child was the first born for about 70% of comparator mothers. For case mothers, the index child was the first born for about 40% of mothers. However, biases that may have arisen from this discrepancy would have been eliminated by my adjustment for parity.

In a similar way to this study, one other research group assessed the odds of the occurrence of a psychiatric disorder in mothers of children with any ASD with no previous psychiatric history.(14) This study used diagnoses from hospitalisation records and the researchers estimated that these mothers were at increased risk of a psychiatric disorder with onset after the diagnosis of their child. However, my results are more informative because I found that this risk was particularly increased for mothers of children with ASD without ID. Reasons for the increased vulnerability of the mothers of children with ASD without ID might be because these mothers have a greater genetic predisposition. To some degree at least, these two forms of ASD may have different
aetiologies, in keeping with the contrasting risk profiles previously identified. Other reasons might be the later diagnosis in children with greater cognitive abilities, resulting in increased stress, self-blame and stigma for the mothers.

In WA, and possibly elsewhere, families with a child with ASD without ID usually receive less funding than those with a child with ASD with ID. For example, in Australia, the Helping children with autism funding package applies to children from 0 to 6 years. The later diagnosis of children with ASD and no ID means that some families may miss out on funding altogether. This might further contribute to the maternal burden.

Mothers of children with ASD without ID had more than twice the incidence of affective disorders compared to other mothers of children with ASD and nearly three times the rate for comparator mothers. My category of Affective disorders included depressive and bipolar disorders. Other researchers have identified higher rates of affective disorders and depressive symptoms in parents of children with ASD though no allowance was made for the timing in relation to the index birth, the existence of a previous psychiatric disorder or the presence of comorbid ID in the offspring. This higher rate of depressive disorders in parents of children with ASD could be a consequence of increased sleep deprivation and more challenging child behaviours. There is also evidence that bipolar disorder and ASD share common genetic factors. Hence, the higher incidence of affective disorders might be in part due to a genetic predisposition in mothers of children with ASD (specifically those without comorbid ID) coupled with environmental triggers associated with the burden of care.

Mothers of children with ASD without ID had more than three times the rate of neurotic disorders of both mothers of children with ASD with ID, and comparator mothers. Researchers using population data from Finland found an elevated incidence of maternal neurotic disorders but no difference between similar groups (mothers of children with childhood autism and mothers of children with Asperger syndrome). However they did not take account whether or not mothers had a previous psychiatric disorder. As with affective disorder, these results may reflect a greater genetic susceptibility, a differential burden of care or a combination of these in mothers of
children with ASD without ID compared to both other mothers of children with ASD and comparator mothers.

I found that mothers of children with ASD with ID had a lower incidence of alcohol and substance abuse than comparator mothers. In contrast, others, using population data from Sweden(14) and Finland,(51) found elevated odds of alcohol and substance abuse in parents of children with childhood autism, though the ORs were not significant. However, unlike the Swedish and Finish studies, I only included mothers in my analyses and males have been identified as having higher levels of alcohol and substance abuse than females.(58) Moreover, my exclusion of mothers with a previous psychiatric disorder would seem likely to explain the lower rates in case mothers in the current study. A prevalence study(53) of alcoholism in families with children affected by ASD involved 167 families ascertained through a service provider. Mothers of children with ASD were found to be twice as likely to be alcoholic. However, again comparisons with this study are difficult since the rates described were lifetime rates, were obtained through family report and were not adjusted for SES or maternal age.

Researchers have identified common biological pathways for ASD and schizophrenia(57) As expected, I found elevated rates of schizophrenia in each case group. Moreover, I found more than three times the rate in the mothers of children with ASD with ID compared to mothers of children with ASD without ID (Table 35) though neither of the IRRs were significant, possibly due to the small numbers in each case group (N = 11, N = 9). Nevertheless, this provides further evidence that the presence or absence of comorbid ID should be taken into account in aetiology studies.(59)

### 7.2.5 Conclusion and implications

Mothers with no psychiatric history and a child with ASD, and particularly those of a child without ID, were more likely to have a psychiatric disorder with an onset after the birth of their child than other mothers with no previous psychiatric history. Moreover, the incidence rates of mothers of children with ASD with and without ID with the same psychiatric disorder were different, indicating different aetiologies for these conditions. Increased support and interventions are indicated, particularly for the mothers of children with ASD without ID.
7.3 Comparison of results and overall conclusion

The IRRs from the studies described in Sections 7.1 and 7.2 are directly comparable as the measures of psychiatric status, explanatory variables and the comparator groups are the same for each study. Both sets of adjusted IRRs are included in Table 37. Mothers of children with mild–moderate ID have the greatest incidence rate of psychiatric disorders of all case groups of mothers of children with ID or ASD for all four of the primary blocks and for any psychiatric disorder. The second most affected group is the mothers of children with ASD without ID who had elevated rates in all blocks and for any psychiatric disorder. Significant IRRs were for affective disorders, neurotic disorders and any psychiatric disorder where the rates were between two and three times that of mothers of children with no ID or ASD. Mothers of children with _ID of known cause (not Down syndrome)_ had the next highest incidence of psychiatric disorders after the birth of their index child with significant increases in incidence rate for _Alcohol and substance abuse_ of nearly three times and more than one and a half times for any psychiatric disorder. Elevated IRRs were present for the remaining blocks. The next group in terms of increased incidence rates was mothers of children with ASD with ID who had elevated IRRs (though not significant) in all blocks and for any psychiatric disorder. Mothers of children with severe ID had more than five times the rate of affective disorders but this may have been an artefact due to the small number (N = 14). Incidence rate ratios for other case groups were not significant and some were elevated and others attenuated. Mothers of children with Down syndrome had attenuated but insignificant IRRs for all blocks and the category ‘any psychiatric disorder’. This indicates that these mothers have the least impaired mental health of any case group of mothers of children with ID or ASD and are no different from that of mothers of children with no ID or ASD. In terms of the existing research, these two studies support the better mental health of mothers of children with Down syndrome compared to mothers of other case groups of ID and ASD. However, my results are at odds with many studies that have reported that mothers of children with ASD have poorer mental health than mothers of children with ID.
Table 347: Incidence rate ratios of mothers by category and case group

<table>
<thead>
<tr>
<th>Block/category</th>
<th>Mild–mod ID</th>
<th>Severe ID</th>
<th>Down syndrome</th>
<th>ID (not Down syndrome)</th>
<th>ASD with ID</th>
<th>ASD without ID</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alcohol &amp; subst abuse</td>
<td>2.91(2.0, 4.3)**</td>
<td>0.58(0.1, 3.2)</td>
<td>0.99(0.3, 3.4)</td>
<td>2.77(1.2, 6.5)*</td>
<td>0.27(0.1, 0.8)*</td>
<td>1.01(0.3, 3.4)</td>
</tr>
<tr>
<td>Schizophrenia spectrum disorders</td>
<td>3.49(1.6, 7.5)**</td>
<td>1.56(0.1, 33.3)</td>
<td>0.31(0.03, 3.6)</td>
<td>2.00(0.4, 11.3)</td>
<td>3.75(0.8, 17.8)</td>
<td>1.21(0.1, 11.7)</td>
</tr>
<tr>
<td>Affective disorders</td>
<td>1.98(1.4, 2.8)***</td>
<td>5.12(1.4, 18.5)***</td>
<td>0.59(0.2, 1.7)</td>
<td>1.12(0.5, 2.4)</td>
<td>1.36(0.7, 2.7)</td>
<td>2.88(1.1, 7.7)*</td>
</tr>
<tr>
<td>Neurotic disorders</td>
<td>1.80(1.3, 2.5)***</td>
<td>1.98(0.6, 6.4)</td>
<td>0.68(0.2, 1.8)</td>
<td>1.73(0.9, 3.5)</td>
<td>1.11(0.6, 2.1)</td>
<td>3.19(1.3, 7.8)*</td>
</tr>
<tr>
<td>Any psychiatric disorder</td>
<td>1.80(1.5, 2.2)***</td>
<td>0.85(0.4, 1.3)</td>
<td>0.82(0.4, 1.6)</td>
<td>1.68(1.1, 2.7)*</td>
<td>1.17(0.7, 2.1)</td>
<td>2.4(1.1, 5.3)*</td>
</tr>
</tbody>
</table>

ID, intellectual disability; Mild-mod ID, Mild or moderate ID of unknown cause; Severe ID, Severe of profound ID of unknown cause; ASD, ID (not Down syndrome), ID of known cause (not Down syndrome); subst, substance; ID, intellectual disability; Severe, severe or profound ID of unknown cause

Note 1: Significant IRRs are shaded
Note 2: IRRs are adjusted for SES, maternal age, parity and index birth year group

* p-value <0.05, ** p-value <0.005, ***p-value <0.0005

7.4 References

Chapter 8: Experiences impacting the quality of life of mothers of children with ASD with ID

In Chapter 5, I reported that mothers with a pre-existing psychiatric disorder were more than twice as likely to have a child with intellectual disability (ID) or autism spectrum disorder (ASD). Intellectual disability is characterised by an IQ of <70 and limitations in adaptive skill that are manifest before 18 years of age. (1) Around 70% of persons with ASD also have ID or ASD. (2) As a result of the outcomes of the research reported in Chapter 6, I concluded that mothers of children with ID or ASD had a higher mortality than other mothers during the study period. My exploration of psychiatric disorders, with onset after the birth of a child with ID (Chapter 7), affirmed higher rates in all maternal case groups except for mothers of children with Down syndrome and mothers of children with severe or profound ID of unknown cause (severe ID). Compared to mothers of children with no ID or ASD, mothers of children with Down syndrome had lower odds (though not significantly so) for all groups of psychiatric disorders. I concluded that the mental health of these mothers was as good as the mental health of mothers of children with no ID or ASD. Small numbers in the severe ID case group meant that I could draw no conclusion. In the remaining mothers of children with ID, significant odds ratios for a psychiatric condition with onset after the birth indicated that their mental health was poorer than other mothers after the birth of their child. In a corresponding study but with the mothers of children with ASD, I concluded that mothers of children with ASD, and particularly ASD without ID, had poorer mental health after the birth of their child. With the evidence from these four studies, I was now very keen to meet some of these mothers and ask them about the impact of their child’s disability on their quality of life (QoL). This chapter reports on a qualitative study in which I interviewed 16 mothers of children with ASD with ID. Data pertaining to self-perceived personality or behavioural changes was reported previously in Section 5.3. The first page of the publication resulting from the broader study reported in this chapter has been included as Appendix 9.
Quality of life (QoL) is a concept describing overall well-being, and results from a complex interaction of health, independence, relationships, goals and standards in the context of a person’s environment. (1) Intellectual disability is characterised by an IQ of <70 and limitations in adaptive skill that are manifest before 18 years. (2) Around 70% of persons with ASD also have ID. (3) Autism spectrum disorder represents a group of severe, chronic, lifelong, neurodevelopmental disorders that are diagnosed by impairments in the areas of Social interaction, Communication and Repetitive behaviours or interests. (4)

Since the 1980s, the prevalence of an autism diagnosis has been increasing both internationally (5) and in Australia. (6) Parents of children with ASD have been reported to have poorer physical and mental health (7) and QoL (8) than other parents. Further, mothers of children with ASD have been shown to have more compromised health than fathers, (8) mothers of healthy children (9) and mothers of children with other developmental disabilities. (10)

Reasons for the impaired QoL of mothers of children with ASD are likely to be complex. Some have postulated a genetic basis for the increased prevalence of mental disorders. (11) However, there are many external factors that may explain why mothers might have a lower QoL. Children with ASD have more challenging behaviours (12) than healthy children and challenging behaviours are associated with lower maternal QoL. (13) Other factors include poor levels of social (14) and family support (15) and an overall perception of stigma against their children. (16) Note that all of these risk factors are potentially modifiable.

Mothers with a lower QoL are more likely to relinquish the care of their child (17) and the public costs of caring for a person with ASD are up to 8.5 times that of a person with no disability. (18) Some research suggests that the healthy siblings of children with ASD have more behavioural problems than siblings of children with no disabilities (19) whereas others have described both advantages and disadvantages to the siblings of children with developmental disability disorders. (20) Either way, these healthy children benefit from the improved well-being of their mothers. (21)
The effects on mothers of raising a child with ASD with ID compared to those of raising a child with ASD without ID or a child with just ID are likely to be different. For example, children with ASD without ID, due to their higher level of functioning, have more independence in the community. Thus they are more exposed to bullying and accidents. Therefore, in contrast to other groups, the factors impinging on the QoL of mothers of children with both ASD and ID are likely to be different.

Exploring the experiences affecting the QoL of mothers of children with ASD with ID is a response to an important public health issue. It is also a first step in the development of evidence-based interventions and services to improve mothers’ QoL and consequently their ability to care for all of their children. This could reduce the need for the community provision of expensive full-time residential care for persons with ASD with ID as well as ad hoc services for their mothers and potentially disadvantaged siblings. Therefore, the aim of this study was to identify factors perceived by mothers of children with ASD with ID affecting their QoL.

8.2 Methods

8.2.1 Research design

In this qualitative phenomenological study, I was guided by the hermeneutic principles of Van Manen(22) and Heidegger as described by Gadamer.(23) Methodologically, hermeneutic phenomenology is appropriate to explore the essential features of lived experience affecting QoL for mothers of children with ASD with ID, as characterised by an IQ <70 and limitations in adaptive skill that are manifest before 18 years.(1). Around 70% of persons with ASD also have ID.(2) within a social and contextualised world.(22)

8.2.2 Sampling and recruitment

Sampling was purposeful, and through the Autism Association of Western Australia (AAWA), I recruited 16 mothers of 11–24-year-old children with ASD with ID from suburban Perth. The child’s diagnosis of ASD was confirmed as this is necessary for a
family membership of AAWA. The presence of ID was reported by the mother and checked by the first researcher using the type of school that the child attended (or had attended). I considered that mothers of 11–24 year olds would have had many experiences affecting their QoL. Participants were also required to have fluent English to enable a detailed and rich understanding of lived experience to be elicited through interview.

8.2.3 Data collection and analysis

Interviews lasted from 40–120 minutes and were one to one and semi-structured, and addressed the experiences of the mothers in context. Interviews were audio recorded and transcribed verbatim; at transcription, pseudonyms were assigned to mothers and other potentially identifying items to preserve confidentiality. Expletives were moderated. Data were analysed by giving full consideration to the parts and the whole through a reflective back-and-forth process between analysis and writing akin to Heidegger’s hermeneutic circle.(23) As such, I read transcripts in their entirety several times, and this was followed by a close, line-by-line reading to identify significant statements or horizons of experiences pertaining to QoL. Significant statements were then clustered to form units of meaning. The software package NVivo10 (QSR International) facilitated storage, management, analysis and interrogation of data.

I discussed the units of meaning with my collaborator Dr Colleen Fisher to ensure they were essential to the experience of impacted QoL and further abstracted them to form themes. This ensured a rigorous analysis of the data. Hence, I extracted multiple meanings of the mothers’ QoL experiences(22) and was able to elucidate the universal essence of the factors influencing this.(24) Rigour was further enhanced by the maintenance of an audit trail of decisions made during data collection and analysis.

8.3 Results and discussion

8.3.1 Participants and their children

Of the 17 study children, 11 were boys and 6 were girls. Nine of them had functional language. Using the traits of functional language, type of school attended and mothers’
descriptions, I categorised the child’s level of functioning within this disability group. Seven were assessed as low functioning, four medium and six high. Although these distributions are not reflective of proportions in the general population, these children functioned across the range of levels found in persons with ASD with ID. Characteristics of the 16 mothers and their 17 children are displayed in Table 38.

### Table 358: Characteristics of mothers and their children with ASD with ID

<table>
<thead>
<tr>
<th>Mother</th>
<th>Age</th>
<th>Marital status</th>
<th>Child with ASD &amp; ID</th>
<th>Mother employed</th>
<th>Child's age/gender</th>
<th>No. of children with ASD</th>
<th>Child verbal</th>
<th>Child's functioning level</th>
<th>Child at home</th>
</tr>
</thead>
<tbody>
<tr>
<td>Amelie</td>
<td>54</td>
<td>Divorced</td>
<td>Guillaume</td>
<td>Yes</td>
<td>20 ♂</td>
<td>1</td>
<td>No</td>
<td>Low</td>
<td>No</td>
</tr>
<tr>
<td>Catherine</td>
<td>46</td>
<td>Divorced</td>
<td>Michaela</td>
<td>Yes</td>
<td>13 ♀</td>
<td>2</td>
<td>No</td>
<td>Low</td>
<td>Yes</td>
</tr>
<tr>
<td>Christa</td>
<td>50</td>
<td>Married</td>
<td>Caterina</td>
<td>Yes</td>
<td>24 ♂</td>
<td>2</td>
<td>No</td>
<td>Low</td>
<td>No</td>
</tr>
<tr>
<td>Coby</td>
<td>53</td>
<td>Married</td>
<td>Samson</td>
<td>Yes</td>
<td>17 ♀</td>
<td>1</td>
<td>No</td>
<td>Low</td>
<td>No</td>
</tr>
<tr>
<td>Elise</td>
<td>45</td>
<td>Divorced</td>
<td>Jack</td>
<td>No</td>
<td>11 ♂</td>
<td>1</td>
<td>Yes</td>
<td>High</td>
<td>Yes</td>
</tr>
<tr>
<td>Jane</td>
<td>49</td>
<td>Single</td>
<td>Faith</td>
<td>No</td>
<td>11 ♀</td>
<td>1</td>
<td>Yes</td>
<td>High</td>
<td>Yes</td>
</tr>
<tr>
<td>Joanne</td>
<td>50</td>
<td>Married</td>
<td>Aidan, Caitlin</td>
<td>Yes</td>
<td>14 ♂, 12 ♀</td>
<td>2</td>
<td>Yes, no</td>
<td>Medium, low</td>
<td>Yes, yes</td>
</tr>
<tr>
<td>Kirsty</td>
<td>48</td>
<td>Single</td>
<td>Scarlett</td>
<td>Yes</td>
<td>11 ♀</td>
<td>1</td>
<td>No</td>
<td>Low</td>
<td>Yes</td>
</tr>
<tr>
<td>Kylie</td>
<td>42</td>
<td>Married</td>
<td>Justin</td>
<td>No</td>
<td>19 ♂</td>
<td>1</td>
<td>Yes</td>
<td>High</td>
<td>Yes</td>
</tr>
<tr>
<td>Nadia</td>
<td>55</td>
<td>Remarried</td>
<td>Craig</td>
<td>No</td>
<td>19 ♂</td>
<td>1</td>
<td>No</td>
<td>Low</td>
<td>No</td>
</tr>
<tr>
<td>Patrice</td>
<td>50</td>
<td>Married</td>
<td>Dimitria</td>
<td>No</td>
<td>17 ♂</td>
<td>1</td>
<td>Yes</td>
<td>Medium</td>
<td>Yes</td>
</tr>
<tr>
<td>Philippa</td>
<td>48</td>
<td>Married</td>
<td>Angus</td>
<td>Yes</td>
<td>20 ♂</td>
<td>1</td>
<td>Yes</td>
<td>Medium</td>
<td>No</td>
</tr>
<tr>
<td>Roslyn</td>
<td>63</td>
<td>Divorced</td>
<td>Edward</td>
<td>No</td>
<td>17 ♂</td>
<td>1*</td>
<td>No</td>
<td>Medium</td>
<td>Yes</td>
</tr>
<tr>
<td>Sharon</td>
<td>50</td>
<td>Remarried</td>
<td>David</td>
<td>Yes</td>
<td>19 ♂</td>
<td>1</td>
<td>Yes</td>
<td>High</td>
<td>Yes</td>
</tr>
<tr>
<td>Tracy</td>
<td>42</td>
<td>Married</td>
<td>Jake</td>
<td>Yes</td>
<td>11 ♂</td>
<td>1</td>
<td>Yes</td>
<td>High</td>
<td>Yes</td>
</tr>
<tr>
<td>Victoria</td>
<td>42</td>
<td>Married</td>
<td>Finlay</td>
<td>No</td>
<td>11 ♀</td>
<td>1</td>
<td>Yes</td>
<td>High</td>
<td>Yes</td>
</tr>
</tbody>
</table>

ID, intellectual disability; ♂, male; ♀, female; ASD, autism spectrum disorder  
* Includes 1 grandchild with ASD  
* Includes a child with ASD without ID

I identified three multidimensional themes as essential to the experience of living with a child with ASD with ID and affecting the mothers’ QoL. These were Living with the traits of the child’s disability; Adapting to a new lifestyle; and Surviving challenges and savouring rewards. Despite their being intertwined in complex ways, I present these themes discretely for clarity (Figure 25).

#### 8.3.2 Living with the traits of the child’s disability

Living with the traits of the disability of a child with ASD with ID provides immediate challenges to parents that over the longer term can be very taxing. Compared to healthy children and children with other disabilities, children with ASD with ID present more challenges to mothers in most areas of child rearing. These include toilet training,(25) life skills,(26) socially appropriate behaviours,(27) challenging behaviours,(28) sleep
Challenging behaviours

At interview, mothers described challenging behaviours in their children, including running away, screaming, tantrums, self-harm, violence and sleep issues. These behaviours had negatively affected their QoL.
Running away

A child’s propensity to run away had major effects on QoL and generated stress over safety concerns, foiled attempts to secure their properties and by limiting their lifestyles. For example, Nadia’s son Craig went missing because a visitor left the gate open. She reported:

He was gone and he’s a quick mover so I phoned the police. A detective came and picked me up in a police car. They even had the chopper out. Craig has no road sense at all. This was real panic stuff.

Elise recounted how Jack used to just ‘disappear’: ‘He (Jack) disappeared down the shops ... crossed the main road. I just about freaked out’.

Screaming and tantrums

Mothers reported that screaming and tantrums occurred when the child did not get his/her own way, when routine was interrupted and sometimes for no ascertainable reason. These behaviours reduced QoL because of stress, limitation of lifestyle or criticisms from bystanders. Coby recalled a day when she was shopping with four-year-old Samson:

I strapped him into his stroller … There was a queue. In the end, he was so crazed, he was standing up strapped to his stroller and I kept saying, ‘Sam, sit down’. He just got my hair and just was yanking. I whacked down on his arms this time because I had had a gutful … And there was a guy with his perfect twins in their perfect pram sitting there. And he said, ‘I don’t believe what I just saw’. I was like with scratches, bleeding … And he said, ‘You’re a dinosaur lady. Who does that to their kid?’

Kirsty recounted how Scarlett’s tantrums ruled their lives:

She was so distressed at the Wiggles finishing that it took me over an hour to get her out of the Entertainment Centre … She almost pushed me over the balcony … We had all of management around me with walkie talkies trying to get her out of the arena. They got her a wheelchair and they strapped her in.
They were stopping traffic so that I could go and get my car to come in a hurry to get her into the car. And the whole time she’s kicking and screaming.

Kylie recalled: ‘If Justin wasn’t used to the thought of going to a particular place, he’d kick up, tantrum and scream, ‘Not going’. So we’d end up not going’.

**Self-harm and violence**

Self-harm and aggression has been identified in about half of people with ASD.(31) This can range from mild, such as gentle head banging, to severe, which might include self-biting and violent head banging.(32) Christa, Joanne and Nadia reported varying degrees of head banging in their children. Christa described how Caterina would scream while banging her head on the wall. Joanne recalled how Caitlin used to head butt a wall until her head bled, and then play with the blood. Nadia told of the time Craig hit his head so hard against a wall that the plaster on the other side fell off. Apart from concerns regarding injury, these behaviours lower a mother’s QoL by limiting her child’s involvement in school and community activities.(33)

A quarter of the mothers in this study reported extreme violence in their child, which placed the physical safety of those around them at risk. Amelie told of Guillaume’s violence at six years: ‘He smashed the whole house, the windows everything. I had the police …’

Philippa reported:
Angus wasn’t violent until he was about ten or eleven and it was hormonal, puberty. He got very violent. He would bite, scratch, pull your hair out by the handful, smash furniture and smash windows.

The mothers who experienced extreme violence found it the most intolerable of their children’s challenging behaviours. Aggressive behaviours such as self-harm and violence have the greatest negative impact on caregivers and are the strongest predictor of parental stress.(34) Thus, though parents find that extreme violence is the most challenging disability trait, reports indicate there are fewer resources for them in terms of available treatments or residential facilities for their children.(35)
Sleep issues

A majority of mothers reported extended periods of sleep deprivation and its negative impact on QoL. For instance, Elise reported that Jack would never settle to sleep. Similarly, Victoria described how she and her husband ‘lived off very little sleep’. They coped with Finlay’s night-time wakefulness by sleeping in shifts, with one sleeping from 7.30 pm to midnight and the other taking over Finlay’s supervision after that.

Other challenging behaviours

Destructive behaviours were problematic because they isolated the family socially, increased workload and expenses and were upsetting. For example, Nadia said of Craig, ‘He destroyed the beautiful things in our lives. I bought a rose to plant and he destroyed it’.

Out in the community, Patrice’s daughter, Dimitria, would demand her mother’s attention by making louder and louder noises. This would severely limited Patrice’s ability to informally chat with friends and others. Socially unacceptable behaviours such as this are stressful and ultimately isolating. Tracy reported that Jake (11 years old) liked young girls of six or seven and added, ‘This is to do with his sexuality. In parks, he’ll just go up and start playing and try to give them piggy backs’.

Consequently, on family outings, Tracy and her husband were unable to relax and interactions with others were limited because they had to constantly monitor Jake’s behaviour.

Diagnostic and education issues

Nearly half of our participants described diagnostic issues. Amelie reported that she ‘died inside’ when the doctor said to her three times that there was no hope for her then three-year-old son Guillaume. Some mothers had to push for a diagnosis. Coby
described how the paediatrician would not diagnose Samson because he was only 17 months old. To secure a diagnosis, she would just go and sit in the waiting room of his practice. Coby would show copies of reports to the paediatrician whenever he emerged and tell him that these provided evidence that her son had ASD. Within a month, Coby had the diagnosis and she said, ‘I think ASD was a great diagnosis for him (Samson). It meant that we had a starting point (for research, therapies and services)’.

Prior to a diagnosis of ASD in their child, parents have expressed feelings of guilt, helplessness and ignorance. Some wait years and with a diagnosis they are both relieved and distressed. Commonly, parents are instructed to wait and see how their child develops. This is stressful and upsetting, especially if they are aware that early intervention might improve the prognosis of their child.

Despite a large body of research identifying suitable educational practice for children with ASD, little has been adopted by schools in the US. In Canada, the education system for children with ASD is difficult to access and inadequately resourced. Mothers in this study described difficulties that they found affected their QoL. In Western Australia, students with ASD are catered for in separate ‘Special Schools’, ‘Education Support Units’ within mainstream schools and in mainstream classes with an aide. Christa said:

I don’t think Special Schools really know how to cater for autism … When she (Caterina) started screaming, they would just lock her up in a room. My husband went there (school) a couple of times and told them off. He could hear her screaming from the street.

When Dimitria was around six, Patrice recalled how frustrated and disrespected she felt at the teacher’s inappropriate response when Dimitria put her hands down her pants in front of the class and then proceeded to put her hands in her mouth, ‘She sent me a letter! That was a real low in my life’.

8.3.3 Adapting to a new lifestyle

All mothers described lifestyle changes: caregiving increased, expenses escalated, many fought for services and some experienced bullying or witnessed the bullying of their
child. Mothers grieving their child’s ASD became increasingly isolated and some felt consumed by the disability. Dreams had been destroyed and future plans were now uncertain. Mothers developed a range of coping strategies.

More demands, less leisure

The increased caregiving demands and subsequent reduced leisure of parents of children with ASD is identified in the literature. In this study, all mothers battled to adjust to this change. For example, four mothers learned Applied Behaviour Analysis (ABA), which uses positive reinforcements to improve children’s behaviour. For children with ASD, this involves a one-to-one relationship with a therapist who conducts repeated trials. For example, the therapist lines up different coloured cups on a table and instructs the child to ‘Give me red’. Successful completion of tasks results in intermittent rewards for the child such as a sweet or a glimpse of a flashing light. This is time consuming for a mother because often she has multiple roles in her child’s ABA program, which might run for up to 40 hours a week. For instance, the mother might be one or more of therapist, programme developer, trainer and employer of other therapists, shift manager and paymaster.

The four mothers, who ran a programme in their home reported their increase in workload. Coby said, ‘You can’t do it (run an ABA program). I burnt myself stupid. I was so thin and just didn’t eat. I was awake all night, never slept. I was a bloody mess’.

The negative maternal effect of an ABA programme reported by these four mothers reflects published research findings. Further, the words of Coby humanise their finding that programme intensity was inversely correlated with maternal well-being. Mothers described the extra supervision required for their child. To supervise Faith in a playground, Jane would have to be constantly vigilant and always prepared to instantly chase and restrain her. She contrasted this to the more sociable and relaxing scenario for parents of healthy children by saying, ‘Most other parents sit down and talk while their kids play’.
Mothers were regularly called to their child’s school. Kylie noted, ‘I was always called up to the school (kindergarten) a couple of days a week. He (Justin) was not doing what he was told and he would just run amuck … at primary school it got worse’.

There was no leisure for some of the mothers, even with grown children. Christa’s children were Fabia (25), Caterina (24 with ASD with ID) and Anna (19 with ASD without ID). Her response when asked about leisure was, ‘What leisure? There was no spare time when Caterina was home and there’s no leisure time now either’.

**Financial impacts**

Families with a child with developmental disabilities have increased financial stress(43) with increased costs due to therapeutic and medical expenses.(44) Nadia recalled the high cost of early intervention, therapists and psychiatrists. Roslyn commented, ‘It’s killed us financially because of all the money’s been spent on the therapy … and there’s no extra money ever for anything much’.

**Fighting for support**

Adapting to lives with their children with ASD with ID often involved battles with personnel whose employers were allocating supports. Sometimes this took many hours over months or years. This, combined with the ignorance, rudeness and lack of respect displayed by some staff can have a major and negative effect on parents’ QoL.(39) Jane harassed the principal at Faith’s school to request more aide support from the Education Department. Two mothers won additional support because they persisted with appeals against decisions made by government departments or sought the support of people in high positions. It took Victoria two years to receive therapy from a government agency. Weekly requests by phone to the agency made no difference. Then, Victoria met the state premier and asked him why older children were not allocated services. He urged her to contact him if this continued. She did and within a week, the agency had found a place.

Joanne had three children with a disability. Her first had a physical disability and her second and third had ASD with ID. Her third appeal at a social security claims tribunal
for a Carer’s Pension was rejected because the family’s needs were not considered profound. At the fourth, Joanne and her husband finally gathered the strength to tell of their trials publicly, and they won.

**Bullying**

Bullying affected the QoL of the mothers of higher- and medium-functioning children. Jack’s mother was distraught when a child put a video on *YouTube* disrespecting Jack and his family. Kylie reported that her son did not like being in mainstream classes because the other students always picked on him and called him an ‘idiot kid’. Roslyn described how a mainstream classmate had taught Edward to masturbate under the desk at the back of the classroom. Notably, bullying was not discussed by mothers of low-functioning children, perhaps because their children were more closely supervised. Although other researchers report that children with ASD are at an increased risk of bullying,(45, 46) this study has added depth to this finding through identifying that children’s experiences might be influenced by their level of functioning.

**Grief**

Mostly, mothers related their grief to a loss. They described losing their expected healthy child to ASD. Joanne explained, ‘It’s like grieving the death of the children we should have had. And for many, many years I struggled with it’.

Roslyn constantly grieved for the loss of Edward’s future. Parents of a child with ID also describe their ongoing grief for the loss of the expected healthy child.(47)

**Isolation**

Parents of children with ASD isolate themselves socially because of their child’s disruptive behaviours.(39) Isolation also strongly affected these mothers’ QoL and increased with time while living with their child. Due to Craig’s self-harming and aggressive behaviours, Nadia no longer went out socially, visited people or had friends. Patrice, Jane, Christa and Nadia described a ‘shrinking’ world, with Patrice
commenting, ‘You feel like you’re becoming a hermit in a way because it (coping with Dimitria) just gets too hard as well. It just gets way too hard, even to go to church’.

**Consumed by disability**

Some parents of children with ASD miss a ‘normal’ way of life because of the heavy load associated with their child’s care. Likewise, the majority of mothers in this study alluded to a lifestyle that had evolved in response to their child’s disability. Jane believed that there was more pressure on the parents of children with ASD than that on the parents of children with Down syndrome (say) because there were numerous books and reports of children recovering from ASD. In contrast, with Down syndrome, the chromosomal abnormality essential for diagnosis, was always going to be there, regardless of therapies tried. Jane explained, ‘There are books like the Catherine Maurice one which says, ‘You can recover your child. So you’ve got this enormous pressure to do like 40 hours of therapy (a week)’.

For others, the enormous focus on their child’s disability had practical origins due to the caregiving burden. Nadia and Elise described how their sons consumed their lives. Nadia considered that Craig had taken over her whole being. Elise explained how a family holiday at a local resort was cut short because Jack screamed continually and added, ‘He took up my whole, entire life’.

**Coping strategies**

All mothers developed strategies to accommodate their new demands. Some, such as Catherine and Amelie chose escape mechanisms. Catherine confided, ‘I’m an alcoholic and a drug user. It’s probably the only thing that gets me through. And I do. I come back here (home) at night and get drunk’.

Amelie viewed home as a haven and rationalised, ‘We couldn’t go out because of Guillaume’s wanting to touch the DVD, press the things … so it was safety at home’.

Others adopted positive coping strategies. In the community, when people stared, Philippa coped by explaining pleasantly, ‘That’s autism. It’s just what he does’.
Out shopping, Victoria’s son was often distraught. In order to stifle the potential comments of gawking bystanders, she would say ‘Have you got a problem with my child? Ask me about autism’.

Many mothers coped by planning their life around their child’s disability. For example, Philippa planned family events closely around 20-year-old Angus, and Nadia bought a house that met Craig’s needs and not necessarily those of others in the family.

Coby recounted another positive coping mechanism. She made the decision to be very unemotional when talking to professionals about her son and, as previously noted, acknowledged, ‘If I let myself get emotional with one of these people and with one of their questions, I thought the flood-gates are going to open and I’m never going to be able to get myself back from it’. I found no research recognising Coby’s method of coping. Her mechanism of shifting to cold affect, with little visible emotion, might be largely unrecognised by researchers and clinicians. Instead, it might be seen as intrinsic to the mother’s personality.

**Future plans**

Mothers’ futures were extensively altered by their children’s disability. More than half considered that they would eventually have their own lives. For example, Amelie said ‘I need to have a dream … It’s changed but I can still have a life for myself’.

Others considered the focus of their lives would always be their child with ASD with ID. Sixty-three-year-old Roslyn had two children with ASD and is a primary caregiver of her three-year-old grandson with ASD. She said, ‘Well, my life really has been my children and keeping my home together and I don’t see that that’s going to change’.

Catherine conveyed acceptance: ‘I don’t worry about my life anymore. What’s done is done’.

These findings are new. Although research was found examining life plans for people with ASD,(48) the perspective of mothers’ futures was unexplored.
8.3.4 Surviving challenges and savouring rewards

Many mothers recognised that they were battling to survive the challenges. As in warfare there were casualties that here were represented as poorer health, relinquished employment, lowered self-esteem, guilt, perceived personality change and altered relationships. With survival, mothers experienced rewards associated with their child’s disability and because of their paucity, these rewards were savoured.

Health

Mothers of children with ASD have poorer overall well-being compared to mothers of healthy children. Our data provided insight into the lived experience underlying the findings of previous research. Mothers described mental health issues that included depression, breakdowns and attempted suicide. Most considered that their poorer mental health was a direct result of the stress of parenting their child.

Jane recognised that she had a mental health issue and described an altered state in which she felt ‘apart from her body’. Catherine reported a nervous breakdown. Other reports of stress and depression were common, with Amelie recalling, ‘With Guillaume, it (daily life) was so stressful. We just survived’.

Sharon related depression to her general health and concluded:

Depression, big depression … To try and stay positive is the hard part … And when you’re depressed, when you’re down on yourself, anything can attack you … Your complete health (mental and physical) goes down the tube.

This association of mental and physical health is described extensively in the literature. Some mothers had thoughts of ending their lives. Joanne related:

I was suicidal most of the time, even going to work every day … I thought, I could just go and drive my car into the ocean … and I felt like that most days because there was no end to this misery.
Only Amelie acknowledged a suicide attempt. With no money or care for her son, she rang a community help line, ended up in hospital and was supplied with a government-owned home and care for her son.

Physical health issues can result from chronic stressors.(21) Mothers reported a range of physical health issues and some considered the genesis was related to the stress of caregiving. Coby underwent a hysterectomy and Nadia described severe migraines, high blood pressure and cholesterol, arthritis and a hip replacement. Philippa had suffered breast cancer and had very high levels of the stress hormone cortisol that she attributed to the stress associated with caring for her child. She explained, ‘I’ve had breast cancer. It’s (breast cancer) definitely linked because my cortisol levels were really high … There is a causal link with cancer and elevated levels of cortisol’.

**Employment**

Outside employment can be protective in supporting the QoL of mothers of children with developmental disabilities.(51) Likewise, some mothers in this study recognised that employment aided their survival because it provided respite from their children and opportunities to interact in the outside world. Joanne said, ‘I got a job two days a week at Dudley Park Hospital, just to keep myself sane’.

However, it was often difficult for mothers to work. Philippa remembered saying to herself:

You can’t do this (work as a lawyer) with Angus. You have to stop. I’m so glad I got it (breast cancer) … I stopped and you have to because you’re going to have a nervous breakdown (if you keep working).

Coby explained the impossibility of reconciling her caregiver and employment responsibilities because employers would be unlikely to accept her saying (for example) ‘Sorry, I’ve got to go now, Samson’s had a seizure’.

Sharon recalled how her return to part-time work was aborted because the child care centre did not cope with David’s behaviour. In line with these mothers’ comments, researchers cite some of the causes of lower employment of mothers of children with
ASD as the difficulties associated with accommodating all child-care issues, carer and employee responsibilities.(52)

**Self-esteem and personality**

Parenting a child with ASD can obscure a parent’s other sources of identity.(53) In such a way, Joanne felt that she had lost her personal identity due to having children with special needs. Mothers acknowledged their lowered self-esteem and many attributed this to guilt. Catherine said, ‘What did I do wrong? ... I blamed myself initially (for my two children’s autism)’.

Personal guilt has been linked to lower self-esteem(54) and increasing levels of guilt are associated with higher levels of anxiety in mothers of children with ASD.(55) Nearly half of the mothers interviewed here perceived personality changes since the onset of their child’s autism. Amelie reported how she gradually lost the opportunity to function independently as Guillaume’s autism developed. Jane believed that her daughter’s autism had put ‘bad grooves’ in her personality. Catherine recalled how she used to enjoy planning for the future but now she didn’t bother, adding ‘I find that my personality has changed too. I used to love fun and love to get with people and have a great time’.

This study is the first to report self-perceived personality changes in the mothers of children with autism. However, there are reports of milder autistic traits related to anxiety and sociability in the first-degree relatives of autistic probands.(56) Perhaps, because of a lack of contact with parents caring for a child with ASD, this association is attributed entirely to genetics with no allowance for the pressure and constraints of caregiving.

**Relationships**

Mothers described the negative effects of their child’s disability on family relationships, their healthy children and friendships. In the community with their child, many suffered ignorant and negative judgements. In combination, these effects reduced the mothers’ QoL.
Partner

Some mothers described their spouse’s rejection of their child with ASD with ID. Nadia reported that her former husband, David, rejected Craig completely because of his autism. She said, ‘After the marriage breakdown, he (David) would come over with Christmas presents for the other two (children) but nothing for Craig’.

Several mothers reported less severe rejections, and possibly in relation to the difficult domestic situation rather than the child per se. Amelie’s former husband would physically and emotionally isolate himself in his study, and ignore his wife and daughter’s cries for help.

Those whose marriages disintegrated after the birth of their children with ASD with ID believed that the disability was a major contributing factor. Elise explained it like this: ‘Jack (son with ASD with ID) took away any time I had to give to my other children and husband, any quality time. That was it! It (the marriage) was over’.

These findings provide new insight into the effect of ASD with ID on the relationship between parents and the difficulty of some fathers in adapting to a life with their child with a disability. In some, this could be related to their belief that their wives caused their sons’ autism. In this regard, Amelie said, ‘His father (her son’s), the whole family, went against me. Everybody said that I was the problem’.

In a similar vein, Sharon recalled her husband saying that she was the cause of her son’s problems.

This is the first research to describe paternal reactions to their children with ASD with ID and resultant family situations. I found only one recent report(57) of a husband that blamed his wife for their child’s autism. Descriptions from parents of a child with attention deficit hyperactivity disorder (ADHD) reflect a similar perspective to that described by mothers in this study. For example, some fathers of boys with ADHD perceived that their wives’ mothering styles might be encouraging behavioural problems. In a similar way to some partners of women in this study, fathers left their wives to tackle the child’s problems alone.(58)
Other children

Consistent with previous research,(19) mothers in this study indicated disadvantages to their children without disability. This was another source of stress that again affected the mothers’ QoL. Mothers relayed how their children with ASD with ID were usually the focus of attention, leaving their siblings with little. For example, Guillaume’s ABA therapists were in the home for dozens of hours a week. They were totally focused on Guillaume and lavished him with attention and praise. In comparison, his younger sister, Rebecca, had little praise and had to teach herself many life skills such as toileting and eating with a spoon. Coby described how Brittany fretted because her younger brother had ABA and she would wail, ‘Why can’t I have ABA? I want ABA’.

Philippa used to cue people into Angus’s condition by saying that Angus was special because he had autism. When, his older brother, Samuel was about seven, he said to his mother despondently, ‘You never tell anybody that I’m special’.

Other mothers described how their healthy children saw their siblings with ASD with ID as being the mothers’ favourites. Kylie’s healthy children accused her of favouring Justin. Similarly, Tracy reported Jake’s younger siblings as complaining that Jake got ‘so much’.

Limitations to family activities were problematic, particularly for siblings. Nadia described how Craig’s autism affected the family’s QoL: ‘We go to the beach. Craig loses it, so we have to pack up and come home. So we don’t go to the beach anymore’.

Similarly, Roslyn recalled how they could not go to the movies or to dinner because of Edward’s inability to cope and summarised by saying, ‘It (Edward’s disability) has impacted every … every part of our family life’.

Special arrangements were necessary for friends of healthy siblings to visit. In relation to Dimitria’s older sister and boyfriends, Patrice said:

When young boys started coming here (to see Dimitria’s sister), I’d know to keep Dimitria, sort of away, type of thing because it is embarrassing, having to
explain (Dimitria’s behaviour) while Dimitria’s jumping around and her breasts are flying all over the place.

Of Edward, Roslyn said, ‘He used to take his clothes off in front of visitors and they (his sisters) were very embarrassed and upset with his behaviour’.

Of the effect on Samuel of Angus’s emergent violence, Philippa said, ‘Samuel would walk in the front gate (after school) and not know what conditions were behind the front door. Things might be fine or he might find his mother under attack’.

Possibly, children who experienced violence from their sibling with ASD with ID were the most traumatised. Amelie and Nadia spoke of how their child with a disability had attacked his siblings. Amelie remembered how Guillaume assaulted Rebecca repeatedly: ‘She was always bruised, beaten, scratched … It was as though he wanted to kill her’.

Nadia described a horrific incident when Craig was about seven and his brother, Rory, four. Craig knocked a carer down and then he went to strangle Rory: ‘Without a doubt, he (Craig) was trying to kill him, honestly kill him’.

Guillaume’s younger sister, Rebecca is now 17 and has made multiple suicide attempts and recently spent eight weeks in a psychiatric clinic. Coby’s daughter Brittany is 21 and her mother reported that she has been troubled with behavioural issues throughout secondary school. Nadia’s son, Rory, is 17 and has oppositional defiance disorder. Any links to their childhood traumas can only be surmised. Other studies have reported mothers’ descriptions of the negative effects on their other children in terms of the resultant pressure on these children to grow up too quickly, their reduced opportunities to socialise and their reduced attention from parents.(59) In contrast, in families with a child with Down or Rett syndrome, parents attributed increased tolerance, compassion and appreciation to their other children(60).
**Extended family and friends**

The attitudes of extended family varied from supportive; well-meaning but ignorant; estranged; or judgemental and damning. Almost half of the mothers in this study benefitted from helpful and supportive family members. Such family support is associated with improved outcomes in parents of children with a disability. Kirsty reported that, apart from a sister, her ‘family was cool with Scarlett’ and they provided help by babysitting. For many years, Sharon’s mother helped her regularly with her son. Mothers also reported ignorant but well-meaning relatives that had problems coping with aspects of the disability. For example, Patrice described her parents-in-law as believing that ‘If I take her (Dimitria) to church and she gets communion and that, it’s all going to go away’.

Philippa’s mother-in-law was a former schoolteacher and lived in England. Shortly after Angus’s diagnosis, she said: ‘I’ve shown a photograph of Angus to my neighbour and she’s a psychologist and she said there’s nothing wrong with him’.

Sometimes extended family members sever connections because of a child’s autism. This was Catherine’s experience and she reported: ‘My brother and his wife, never come near me … It’s (my children’s autism) embarrassing (for them)’.

Finally, other mothers described judgemental members of their husbands’ families that blamed them for their children’s autism. Amelie had the horrific experience of her husband’s entire family blaming her for her son’s autism. Christa’s self-esteem had been negatively affected by her mother-in-law. She related how her mother-in-law would visit and say, ‘Your child should be like this. She should be toilet trained … It’s your side of the family … She said I wasn’t “mothering properly”.

Historically, mothers have been blamed for their child’s autism. From some mothers’ perspectives, this practice is continuing. Improved outcomes in mothers of children with autism are positively correlated with both increased and perceived friendship support.
Mothers in this study experienced difficulty with retaining old friends and described the
difficulty of instigating new friendships. For instance, Joanne spoke of her friends at the
time of her children’s diagnoses, saying ‘Some of the friends we made, we lost. We
were never told the reason but we assumed they might have been scared off in case we
asked them for help’.

Amelie and Kirsty had neither the time nor energy to make new friends. Amelie said, ‘I
didn’t have a social life … It takes effort to make friends. You have to reciprocate …
There was nothing that I could do apart from care for my children’.

Kirsty explained how she had neither the time, energy nor the inclination to make
friends. The reduction of social support for mothers of children with ASD since the
development of their children’s disability is described in the literature. These mothers
have less social support than either mothers of children with ID or mothers of healthy
children,(64) and the effect on their QoL is likely to be considerable.(63)

**Community**

Unlike Down syndrome, children with autism are not recognisable by physical traits.
Usually, people in the community are unaware that a child with ASD has a problem
until he or she behaves badly. The insensitive reactions of bystanders would likely be
caused by ignorance, and parents describe this ignorance as isolating.(39) Such
reactions include damning comments about the mother’s parenting skills, inappropriate
suggestions about dealing with the behaviour or blatant staring. A majority of mothers
described upsetting experiences with their children in the community. Coby recalled
negative comments directed at her when she had behaviour management problems with
Samson. People would walk past her and say, ‘Jesus!’ in disgust. Of the wider
community and her daughter’s autism, Christa said:

They don’t understand … I took her (Caterina, 12) to Bindajyne Shopping
Centre … This kid had an ice-cream and we were passing. She grabbed this
ice-cream from this kid. The mother came up to me and said, ‘Your kid stole
my kid’s ice-cream. Hasn’t your kid got any manners?’ I said, ‘Excuse me but
my daughter has autism’. She said, ‘I don’t care’. Then I said, ‘Look, I’ll pay
for the ice-cream’. The husband came up and said, ‘Don’t worry about it’. …
The other mother went on, ‘She should educate her child more’.

Jane was upset by what she described as:

The total lack of fricking empathy and understanding in the community and
the constant trying to explain and feeling like you are a mad person and the
exhaustion and people expecting you to do more.

Some mothers suggested that negative interactions in the wider community most
affected their QoL. This might be a result of the persistent belief that parents,
particularly mothers, cause their children’s challenging behaviours.(65)

**Rewards**

In spite of challenges, most mothers identified one or more personal rewards associated
with their child’s disability. Some spoke of their love for their child. Of Jack, Elise declared, ‘I love him. I love the spunk in his personality’.

Some mothers consider that their personal development had been enhanced by caring
for their child.(66) Along these lines, Amelie confided, ‘I feel blessed that I had
Guillaume. I’m a better person’.

Others described joy from their child’s development. For instance, Catherine said, ‘Now
Michaela has eye contact which is really nice; especially when they look you in the eye
and they are so sweet’.

Many described the exceptional people whom they had met because of their child’s
disability. Roslyn spoke in superlatives of Edward’s two classroom aides who had
enabled him to attend his school’s annual Year 11 Dinner Dance. She said, ‘Two ladies
in the Ed (Education) Support Unit, Mrs Robertson and Mrs Boyle, his two favourites,
picked him up from home and brought him back. They’re the most wonderful women
on earth’. 
Other mothers related their increased appreciation of simple things. For example, Patrice recalled, ‘Having Dimitria has made me appreciate the simple things in life … like appreciating the naivety and the cheekiness of little kids sometimes’.

Finally, mothers translated the possible benefits to others from their participation in this study into their own personal satisfaction.

### 8.3.5 Limitations and recommendations

Mothers might have omitted factors affecting their QoL due to a desire to protect their privacy, embarrassment or a belief that it was not relevant. Those struggling to cope might have been unable to respond to our invitation to participate. Hence, the demonstrated effects on QoL could be incomplete and attenuated. Diagnostic and educational issues, family and community attitudes might vary in other countries. Also, all participants spoke fluent English and lived in the metropolitan area of a city of nearly two million people. Mothers in rural areas and mothers from culturally and linguistically diverse backgrounds might perceive different impacts on their QoL.

Research is indicated to inform governments and agencies of evidence-based services and interventions likely to improve the overall QoL of mothers of children with ASD with ID. Modifiable risk factors, such as community and family ignorance and coping strategies affecting these women’s QoL might be targeted by interventions or services. For example, communities and extended families might be informed of autistic behaviours and parents’ difficulties in their management. Conversely, parents might be provided with positive options to deal with ignorant and/or critical members of their families and the community. Finally, research is recommended to explore the emotional and physical protection of siblings of children with ASD and violent behaviour.

### 8.3.6 Conclusion

In this study, mothers of children with ASD with ID experienced a reduced QoL. Few areas of their lives had been spared from the pervasive and negative effects of their children’s disability. Living with disability traits, adapting to changed lifestyles and
surviving the challenges left many with a lifestyle with little resemblance to its precursor. For some, careers were lost, marriages broken, social networks destroyed and health damaged. Nevertheless, most mothers reported rewards associated with their children with a disability. Some continued to see no future apart from their child. Others were able, or planned to emerge from this challenging period and build a future not wholly centred around their children with ASD with ID.

8.4 References
Chapter 9: Discussion

In this closing chapter, I review my main findings by manuscript, relate these findings to my four aims and discuss the implications for further research. I conclude by summarising my findings and their relationship to the achievement of my aims. Part 1 has nine sub-sections and each is devoted to a paper within this thesis. For each paper, I systematically discuss the findings, their contribution to my aims and their implications for further research. Within Part 2, I provide an overview of the principal findings of my research and the achievement of my four aims.

9.1 The findings, contributions and implications of my research

The three overarching aims and fourth primary aim of my research were:

1. **Pre-existing differences**
   To explore pre-existing differences of mothers of children with intellectual disability (ID) or autism spectrum disorder (ASD) compared to mothers of typically developing children

2. **Health compared to mothers of typically developing children**
   To explore the health of mothers of children with ID or ASD compared to the health of mothers of typically developing children;

3. **Health by the child’s sub-type of ID or ASD**
   To explore the health of mothers of children with a sub-type of ID or ASD compared to the health of mothers of children with a different sub-type of ID or ASD

4. **Self-perceived quality of life of mothers of children with ID or ASD**
   To interview mothers of a child with a sub-type of ID or ASD regarding factors affecting their quality of life (QoL)

The main findings of each paper, its resulting contribution to the aims and the implication for future research now follow.
9.1.1 Health and its correlates in the mothers of children with ID or ASD—a review

This study provided an overview of the current state of the research. I found that mothers of children with ID or ASD generally have poorer self-reported health and QoL and a higher prevalence of psychiatric disorders. This overview also enabled me to identify the limitations of the existing research and the opportunities to add to the existing knowledge base. Specifically, I was alerted to the importance of separating the periods before and after the birth of the child with a disability and the fact that the majority of studies did not take into account the severity of ID, whether the cause was known or whether the ASD was associated with comorbid ID. Further, I was aware that many of the studies may have been subject to collection and recall bias. This knowledge enabled me to direct my research so that, as far as possible, I could overcome these limitations. In such a way, this review indirectly contributed to my achievement of the overarching aims.

9.1.2 Pre-existing differences of mothers of children with ID or ASD—a review

As a result of this review, I became aware that there are contrasting associations of maternal socio-demographic status (SES), age, parity, immigrant status and race-ethnicity with ID and ASD. This had a direct bearing on my subsequent research as I now realised the importance of adjusting for certain of these factors when examining the maternal covariates of ID or ASD in the offspring. There were also reports of a relationship between maternal immigrant status and race-ethnicity and ID and ASD in the child. My interest was further fuelled by the fact that I had found no study that considered race-ethnicity and immigration status independently. This interest led to my research in Chapter 5, which investigated the association between maternal race-ethnicity, immigration, country of birth and ASD in the offspring in Western Australia (WA). Finally, I found that there was little research on pre-existing maternal psychiatric disorder and subsequent ID in the offspring. Further, a few studies had examined this phenomenon in the parents of children with ASD but none had compared the prevalence in the mothers of children with ASD with and without ID. This newly acquired knowledge provided the impetus for the three papers on maternal psychiatric disorder
with onset either before or after the birth of a child with ID or ASD (Chapters 4 and 7). Hence, this review contributed to my overall aims by alerting me to areas that had yet to be researched or that begged further research with a modified approach.

9.1.3 Race-ethnicity, immigrant status, birth region and the odds of having a child with ASD

Compared to Caucasian women, non-Caucasian women were less likely (about 60% as likely) to have a child with ASD with ID and even less likely (about 20% as likely) to have a child with ASD without ID. Compared to non-immigrant women, immigrant women were about 60% as likely to have a child with either ASD with ID or ASD without ID. However, when examining both race-ethnicity and birth region, Black women from East Africa had more than twice the odds of a child with ASD with ID, though numbers were small. It was important to note that all of these East African women came from a cluster of countries in the east of East Africa and that no other Black woman had a child with ASD (N=613). These findings furthered my exploration of pre-existing differences in the mothers of children with ID or ASD.

Non-Caucasian (especially Indigenous) and immigrant women had lower odds of having a child with ASD with ID and even lower odds of a child with ASD without ID. However, Asian women from South or Central Asia and Black women from East Africa had an increased prevalence of ASD with ID compared to other immigrant women, though numbers were small. It was important to note that all of these East African women came from a cluster of countries in the east of East Africa and that no other Black woman had a child with ASD (N=613). These findings furthered my exploration of pre-existing differences in the mothers of children with ID or ASD.

My research is unfinished. I now need to examine the risk of having a child with ID by diagnostic sub-group in these women. It is also important to find out why Black African women from East Africa and Asian women from certain parts of Asia have higher rates of ASD with ID so that preventative measures can be implemented. The numbers of these immigrant women in Australia are increasing, which will facilitate this research. Possibilities for further research include an examination of:

- Obstetric disorders in Black African and Asian women
The timing of maternal immigration and the risk of having a subsequent child with ID or ASD

ASD in Indigenous children

Diagnosing ASD without ID in the children of Australian immigrants.

9.1.4 Psychiatric disorder and ID or ASD in subsequent offspring

After analysing data from outpatient consultations, I concluded that mothers with pre-existing psychiatric disorders had about twice the odds of a child with ID or ASD than other mothers. Further, these odds were similar for mothers of children with ID and mothers of children with ASD. However, the odds varied between mothers of children with different sub-types of ID or ASD. Such findings have direct bearing on all three of my overarching aims.

The Australian Pharmaceutical Benefits Scheme is a national database that contains records of all medications prescribed to Australian citizens and permanent residents. This affords the opportunity of linking this information, for the period of gestation in women, to investigate a suspected teratogen in relation to the odds of a subsequent child with a disability. For example, in WA, researchers linked a group of medications dispensed to pregnant women who later had a child with a birth anomaly. In a similar way, one could examine whether taking particular prescribed medications during pregnancy increased the risk of ID or ASD in the offspring.

My research into pre-existing maternal psychiatric disorders used only outpatient records. Further insight into this situation could be achieved by exploring the onset of maternal psychiatric disorders in mothers with a subsequent child with ID or ASD using both hospitalisation and outpatient records.
9.1.5 Is the Broad Autism Phenotype in mothers of children with ASD exacerbated by the challenges of caring for their children?

Some researchers regard the Broad Autism Phenotype (Section 2.4) as a pre-existing trait of mothers of children with ASD. In this paper, I challenged this idea using data I had collected by interviewing 16 mothers of children with ASD (Chapter 8). This was a further contribution to my aim of exploring pre-existing differences in the mothers of children with ID or ASD.

This research led to a hypothesis that is difficult to test because parents of children with ASD are not identifiable until after their child’s diagnosis. A suitable longitudinal study could be a large-scale ‘birth cohort’ study that administers the AQ (Section 5.3) to score a parent’s BAP at points during pregnancy and throughout the child’s life. I would predict a significant interaction between scores of parents of children with ASD and parents of typically developing children when examined during their child’s infancy (before diagnosis) and at various times after diagnosis, such as during childhood, adolescence and finally when parents no longer co-reside with their child. Given the infrequency of ASD occurrence, such a design might be prohibitively expensive. A second option could be to utilise a large birth cohort study such as the Avon Longitudinal Study of Parents and Children (3) and Generation R9 (4). Researchers could recruit two groups of parents: full-time carers cohabiting with their child, and former carers no longer cohabiting with their child. A between-groups comparison of AQ scores would provide useful data for hypothesis testing.

In my research, mothers of children with ASD have demonstrated poorer health (Chapters 2, 6 and 7), particularly mental health (Chapters 2 and 7). Differences have been demonstrated between mothers of children with ASD with ID and without ID (Chapters 6 and 7). Further research could be designed that explored the relationship of aspects of health and particular traits within the Broad Autism Phenotype. This could be done by administering the Short Form (36-item) Health Survey and the AQ concurrently to mothers of children with ASD, with and without ID. Any significant correlations between components of the AQ and poorer aspects of health might be used to subsequently identify mothers of children with either ASD with ID or ASD without
ID who were at higher risk of particular types of health problems. In this way, services or workshops might be offered to such women with the aim of assisting these to maintain or improve their health.

9.1.6 Early mortality and causes of death in mothers of children with ID or ASD

I found that mothers of children with ID or ASD had an increased risk of death during the study period. Further, the risk of death varied by maternal case group with mothers of children with mild or moderate ID of unknown cause (mild–moderate ID) having the highest rate of death and mothers of children with severe or profound ID, the lowest. Moreover, case group mothers with both a psychiatric disorder and a child with ID or ASD had about six and a half times the risk of death for other mothers. Mothers of children with either ID or ASD were between 35 and 40% more likely to die of cancer during the study period than mothers of children without these disabilities. Therefore, this paper has explored the health of mothers of children with ID or ASD by examining mortality rates and primary causes of death by case group and compared to the mothers of children with no ID or ASD. In this way, this paper has contributed to the second and third of my overarching aims.

Apart from the Cancer category, small numbers in the case groups of mothers of children with ASD prohibited any analyses of death due to a particular cause. Further studies, pooling WA data and data from autism registries elsewhere, might enable more extensive analyses of the primary causes of death in the mothers of children with ASD. In my study, I was unable to ascertain whether the increase in mortality was due to a higher prevalence of the disease or a higher mortality due to the disease. Using existing WA data, I could compare prevalence and mortality rates due to cancer in mothers of children with ID or ASD. Using data from other autism and hospitalisation databases, such as are available in Scandinavia, more extensive analyses of the primary causes of death for the ASD with ID and ASD without ID case groups might be possible. In this way, informed services and preventions might be developed with the aim of improving health and increasing the longevity of these mothers.
9.1.7 Onset of maternal psychiatric disorders after the birth of a child with ID

Women with a child with ID have more psychiatric disorders after the birth of their child than do other mothers. However, it is unclear if this is because they have more psychiatric disorders before the child’s birth or if the increase is related to the burden of caring for the child. I aimed to calculate the rate of new psychiatric disorders in women after the birth of their eldest child with ID born between 1983 and 2005 and compare these with rates in women with a child with no ID or ASD born during the same period. By linking data from Western Australian population-based registries on births, deaths and mental health, I selected women with no psychiatric history before the birth of their child and compared rates of psychiatric disorders for women with and without a child with ID. Negative binomial regression using STATA 12 was used for analysis. Mothers of children with mild–moderate ID of unknown cause had around two to three and a half times the rate of psychiatric disorders of mothers of children without ID or ASD. The incidence rates of mothers of children with ID of known cause excluding Down syndrome were similar but attenuated, ranging from around one to two and a half times the rate of comparator mothers. For mothers of children with Down syndrome, reduced IRRs were estimated for all diagnostic categories.

In this study, I examined rates of onset of psychiatric disorders after the birth of their child in mothers with no previous psychiatric history. There were three main findings. First, mothers of children with mild–moderate ID had around two to three and a half times the rate of psychiatric disorders of mothers of children without ID or ASD. Second, the IRRs of mothers of children with ID of known cause excluding Down syndrome were similar but attenuated, ranging from around one to two and a half times the rate of comparator mothers. Third, for mothers of children with Down syndrome, reduced IRRs were estimated for all diagnostic categories. These findings relate to Aims 2 and 3.

Women with psychiatric disorders and a child with ID or ASD had six and a half times the risk of death during the study period (Chapter 6). Mothers of children with Down syndrome and no previous psychiatric disorder had better mental health after the birth of their child than did mothers of children with no ID or ASD. One possible reason for
their better mental health might be the quality and extent of the support provided by agencies that assist families with a child with Down syndrome in WA. This being so, it would be useful to explore the risk of further mental health issues in mothers with a pre-existing psychiatric disorder after the birth of a child with Down syndrome. It would seem likely that these women are particularly vulnerable. Due to small numbers it might be necessary to access international data for such a study. In the same way, exploring the IRRs of psychiatric disorders in mothers of children from other sub-groups of ID but in mothers with previous psychiatric disorders might provide evidence of groups of mothers who are particularly vulnerable after the birth of their child with ID.

9.1.8 Onset of maternal psychiatric disorders after the birth of a child with ASD

As with the previous paper, I considered only mothers with no psychiatric history. Apart from Alcohol and substance abuse, mothers of children with ASD had higher incidences of all categories of psychiatric disorders than mothers of children without ASD or ID. Further, compared to mothers of children with ASD with ID, mothers of children with ASD without ID had increased incidences for all categories apart from Schizophrenia spectrum disorders. Again, these findings relate to Aims 2 and 3.

9.1.9 Experiences impacting the QoL of mothers of children with ASD with ID

It must be noted that this study was completed before the study described in 9.1.8 began. Had the chronology been different, I would likely have interviewed mothers of children with ASD without ID instead due to their higher rates of psychiatric disorders. In the light of the increased vulnerability of mothers of children with ASD without ID, a similar qualitative study would undoubtedly provide insight into the mental health of these women. Furthermore, comparisons between this study and the one reported in this thesis might provide useful information into factors impacting the health of mothers of child with ASD generally. In such a way, informed interventions and supports might be generated.

Since the onset of their child’s autism, mothers reported that challenging behaviours, the increased demands and isolation all lowered their QoL. Surviving had negative
impacts on their health and relationships, but a majority described rewards associated with their children. In this study, I listened to mothers of children with ASD with ID and their descriptions of how their child’s disability had changed their lives. This first-hand insight provided another dimension to my understanding of why the health of mothers of children with ASD with ID might be impaired (Aim 1).

Three of the mothers interviewed had two children with ASD, which left me wondering if the odds of ASD in half and full siblings had changed over time. A 2013 study used Danish registry data to explore these odds and concluded that in contrast to prevalence data, there was no time trend of the within-family recurrence risk of ASD. Other conclusions were that the higher recurrence risk in full siblings indicated a genetic component of ASD and the higher risk in maternal half siblings indicated a possible association with pregnancy and obstetric factors. A replication study using WA data might provide supplementary or additional information relating to the aetiology of ASD.

9.2 Comparing the health of mothers by case group

9.2.1 Methods

My research in Chapters 5, 6 and 7 resulted in measures of aspects of maternal health by case group using ORs or incident rate ratios (IRRs) with reference to a comparison group. The study in Chapter 5 related to pre-existing health and those in Chapters 6 and 7 related to health after the birth of the index child. In order to summarise my results, I felt that it would be meaningful to rank maternal case groups using these adjusted ORs and IRRs. This is because, in each of the studies, the comparator group was the mothers of children with ID or ASD and the explanatory variables were the same. Further, the study populations were the same in the last three studies and similar in the first study but where births were truncated at the end of 1999 rather than 2005.

From the first study of psychiatric disorder (Chapter 5), I used the ORs to provide a ranking of pre-existing mental health (Figure 2). The IRRs for the existence of one or more psychiatric disorders, according to maternal case group combined with the HRs for death during the study period (Chapter 6), were averaged by case group. The ORs from Section 5.2 were used to rank mothers by their estimated pre-existing health
status. The HRs for death in Chapter 6 and the IRRs from Chapter 7 were summed by case group, averaged and then used to estimate the health of mothers after the birth of their index child. Again, the comparator group was the mothers of children with no ID or ASD. Due to the small numbers, the Severe ID case group was omitted. Hence, I was able to rank case groups according to estimates of pre-existing mental health and also according to measures of health after the onset of caring. The rankings for pre-existing health relate only to mental health, whereas rankings after the birth are more representative of overall health because mortality and psychiatric disorders were used in the calculation. It must be emphasised that these rankings are preliminary only, due to the limited data. Larger numbers indicate poorer health and the comparator group remains mothers of children with no ID and ASD with a ranking of 1.

9.2.2 Results

All case groups had elevated odds of pre-existing mental health issues compared to mothers of children with no ID or ASD. While having 30% increased odds of a pre-existing mental health issue, these odds for mothers of children with Down syndrome were not significant. Mothers of children with ASD with ID had the most impaired mental health and were 153% more likely, followed by mothers of children with mild or moderate ID of unknown cause (mild–moderate ID) (121% more likely) and mothers of children with ASD without ID (113% more likely). The resulting ranking of pre-existing mental health is shown in Figure 25.

Figure 25: Pre-existing mental health by case group

* The comparator group is the mothers of children with no ID or ASD with a measure of 1

The overall assessment of health after the onset of caring indicated that mothers of children with Down syndrome had the least impaired health and that they were 15%
more likely to experience health issues compared to comparator mothers. In order and by increasing health impairment, mothers of children with ASD with ID were next (43%), then mothers of children with ASD without ID (92%), mothers of children with ID of known cause not Down syndrome (98%) and finally mothers of children with mild–moderate ID had the most impaired health (102%). The ranking is illustrated in Figure 27.

**Figure 26: Health after the birth of the index child**

ID, intellectual disability; ~, not; ASD, autism spectrum disorder; Mild-moderate, mild or moderate ID of unknown cause

Note: The comparator group is the mothers of children with no ID or ASD with a measure of 1.

### 9.2.3 Strengths and limitations

A considerable strength of the epidemiological studies within this thesis is that these studies were population-based. Further, I had complete data for all but one covariate, SES where 2.6% of values were missing. In relation to psychiatric disorders and cause of death, I did not rely on maternal or family recall but instead used records of clinical diagnoses of disorders. A weakness is my use of the *Index of Relative Socioeconomic Disadvantage* as a proxy for SES. This is an area-level measure and such measures have not been found to be good proxies for individual measures such as income.(6) However, our use of an area level measure for SES represented the best use of available information since the only potential individual proxy, education was missing for 18.4% of mothers in the data-set. Furthermore, the weakness of my measure for SES was possibly exacerbated since a significant proportion of the mothers (27%) included in the epidemiological studies were immigrant. Accurately measuring the SES of immigrant populations is even more problematic since educated immigrants may be employed in unskilled professions and living in temporary accommodation which may not reflect the assessed SES of their eventual residence.(7) Population-based data, such as those used in this thesis, are collected for administrative, not research purposes. As a result, some measures (such as the date of a child’s diagnosis) which would have been useful have
not been collected. In addition, other measures, such as race-ethnicity are less precise than one which would have been designed for research purposes.

9.2.4 Discussion and conclusion

**Overarching aims**

Mothers of children with Down syndrome were ranked as having both the least impaired pre-existing mental health and the least impaired mental health after the onset of caring. Compared to other case mothers, there was no evidence of increased mortality in this group. As discussed in Chapter 5, I am unaware of any research that has studied the health of mothers of children with Down syndrome prior to the birth of their child. However, the ranking I have demonstrated is consistent with research reporting that their health subsequent to their child’s birth is less impaired than that of other mothers of children with ID or ASD. I found no study that compared the pre-existing health of mothers of children with ASD without ID to mothers of children with ASD with ID. My finding that the mental health of the mothers of the higher-functioning children is less impaired than that of the mothers of the lower-functioning children is probably a first report in this area. After the onset of caring for their children, this finding was reversed, with the mothers of children with ASD without ID now showing more impaired health. This is not inconsistent with the three papers found in this area. Two reported no difference between the groups and the other reported higher levels of emotional disorder in the mothers of children with ASD with ID compared to the mothers of children with ASD with ID. Apart from the mothers of children with Down syndrome, most previous research did not compare mothers according to the sub-type of their child’s ID. In fact, only one study was found and here researchers reported that that the mothers of children with ID of unknown cause had poorer aspects of mental health than the mothers of children with Down syndrome. Our rankings are consistent with this study since mothers of children with mild–moderate ID (of unknown cause) exhibited both poorer pre-existing health and the most impaired health after the onset of caring compared to other mothers. Further research to explore the health of these mothers is required.
Fourth aim

My qualitative study provided the opportunity for me to witness mothers’ descriptions of the factors that affected their QoL.

9.3 Overview and final conclusion

My findings were varied. First, when compared to Caucasian women, I identified that women of minority ethnicities had a lower risk of a child with ASD with ID and particularly ASD without ID. In a similar way, compared to Australian-born women, immigrant women had a about a 40% lower risk of a child with ASD with ID and ASD without ID. Mothers of Asian race-ethnicity from Central and South Asia had about 30% increased odds of having a child with ASD with ID while Black mothers from the east of East Africa had more than twice the odds of a child with ASD.

My second group of findings pertained to my remaining studies using linked data. Here, I identified that mothers of children with Down syndrome were no more likely to have a pre-existing psychiatric disorder than comparator mothers. In other mothers of children with ID or ASD, pre-existing psychiatric disorders were about twice as likely. Moreover, five to 27 years after the birth of their child, mothers of children with ID or ASD were at increased risk of an early death. At highest risk were mothers of children with ID of known cause (not Down syndrome) and mothers of children with mild-moderate ID who had more than double the odds of death. These were followed by mothers of children with ASD who had about 1.5 times the odds, though these failed results to reach significance. Again, mothers of children with Down syndrome had the lowest risk of death of all case mothers and their odds, whilst elevated, were not significant. After the birth of their child, mothers of children with Down syndrome were no more likely to have a psychiatric disorder than other mothers. By contrast, after the birth of their child, mothers of children with ASD without ID had nearly two and a half times the odds of a psychiatric disorder and mothers of children with ID (but not Down syndrome) were more than 1.5 times as likely.
My third group of findings stemmed from my interview data. The **Broad Autism Phenotype** in mothers of children with ASD might be exacerbated by the challenges of caring for their children. This is important because some researchers envisage using measures of the Broad Autism Phenotype as a supplementary tool in genetic studies. Finally, mothers of children with ASD with ID perceived a poorer QoL after the onset of their child’s disorder.

As a group, mothers of children with ASD are less likely to be immigrant or of minority ethnicities. After the birth of their index child, mothers of children with ID or ASD have a higher risk of death than other mothers. Apart from mothers of children with Down syndrome, these mothers also have an increased risk of a psychiatric disorder after the birth. Their increased risk of a psychiatric disorder before the birth indicates that this poorer health has its origins before the birth of their children. Analysis and consideration of my qualitative data emphasised the importance of using qualitative data in conjunction with quantitative data, to provide worthy ideas for future quantitative research.

I have achieved each of my overarching aims in multiple ways. Pre-existing differences (Aim 1) were explored in relation to combinations of race-ethnicity, immigration and region of birth and in terms of the onset of a psychiatric disorder. The health of the mothers of children with ID or ASD was compared to the health of mothers of typically developing children in Chapters 5, 6 and 7 (Aim 2). Aim 3 was achieved when I compared maternal health by case group in Chapters 4, 5 and 7. Finally, I interviewed mothers of children and have reported factors affecting their QoL in Chapter 8 (Aim 4).

### 9.4 References

Appendix 1: Health and its correlates in the mothers of children with ID and ASD

Title
Health and its correlates in the mothers of children with intellectual disability and autism spectrum disorder: a review

Authors
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Abstract

Background
Mothers of children with intellectual disability (ID) and autism spectrum disorder (ASD) have poorer health and this is correlated with child and maternal characteristics.

Objectives
We aimed to compare the health of mothers of children with ID or ASD to that of other mothers and to each other, according to the disability sub-type of their child. We also aimed to identify the stronger correlates of this poorer health.

Methods
We searched databases using search terms related to ID and ASD, carers and health. Papers were retained that met seven criteria and sorted into two groups. First, we set aside all papers that compared the health of mothers of children with ID or ASD to other mothers or by the sub-group of the child’s disability. Second, we set aside all papers exploring the correlates of their poorer health.
Results
We retained 58 papers and an additional 22 papers detailing the correlates of poorer health. Research has consistently identified poorer health in mothers of children with ID or ASD. Further, research has suggested that mothers of children with Down syndrome had the least impaired and mothers of children with ASD had the most impaired health. We used the stronger correlates with poorer health to explain some of the observed intergroup health disparities.

Conclusion
The stronger correlates with poorer health in mothers of children with ID or ASD provide valuable information for service providers in their planning of services and interventions that have the aim of assisting mothers to improve their health.
Appendices

Appendix 2: Pre-existing differences of mothers of children with ID or ASD

Chapter 18

Pre-Existing Differences in Mothers of Children with Autism Spectrum Disorder and/or Intellectual Disability: A Review

Jenny Fairthorne, Amanda Langridge, Jenny Bourke and Helen Leonard

Additional information is available at the end of the chapter http://dx.doi.org/10.5772/54488

Introduction

The autism spectrum disorders (ASD) represent a group of severe and chronic neuro-developmental disorders often simply referred to as autism. [1] Using the criteria provided by the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, ASD are diagnosed by impairments within the three strands of DSM-4: social interaction, communication and repetitive behaviours or interests. [2] The aetiology of autism is complex. [3] Research has implicated a strong genetic basis [4-7] involving multiple genes [5, 7, 8] and possible gene-environment interactions. [9-13] Advances in chromosomal microarray analysis and gene sequencing technologies have improved diagnoses and suggest that aetiologies of ASD will continue to be uncovered. [9] In addition, a child presenting with autistic symptoms may be found to have a certain genetic mutation which accounts for their true underlying biological diagnosis. For example, a diagnosis of Rett syndrome would be confirmed when a girl with ASD and intellectual disability was found to have a mutation of the MECP2 gene on the X-chromosome. [14] Children with ASD and intellectual disability have been found to have an expansion of the FMR1 gene confirming a diagnosis of Fragile X syndrome. [15]

Autism and intellectual disability commonly coexist with 30-80% of persons with ASD reported as also having ID. [16, 17] Currently, the relationship between ASD and comorbid ID is poorly understood. [18] However, it is known that phenotypically, persons with these disorders can be grouped into the three categories of ASD without ID, ASD with ID and ID only. [18] Intellectual disability (ID) is characterized by an intelligence quotient (IQ) of less than 70 which is associated with limitations in at least two areas of adaptive skill and which
Appendix 3: Race-ethnicity, immigrant status, birth region and the odds of a child with ASD

Title
The impact of maternal race-ethnicity, immigrant status and country of birth on the risk of a child with autism spectrum disorder in Western Australia

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Abstract

Background
The risk of having a child with autism spectrum disorder (ASD) varies in immigrant women according to their race-ethnicity and region of birth. Conversely, the same risk in women of minority ethnicities may vary according to their immigrant status. Previous studies document that Asian women, immigrant women from Asia and Black women from East Africa are more likely to have a child with ASD and comorbid intellectual disability (ID) than other women. Understanding which socio-demographic sub-groups of women are at increased risk of having a child with ASD might provide insight into risk factors for ASD in Western Australia and more generally.

Aims
First, we aimed to investigate the risk of ASD both with and without comorbid ID in the children of mothers from ethnic minority groups and in immigrant mothers. Second, we aimed to investigate this same risk in mothers according to their region of birth. Finally, we aimed to investigate the risk of ASD with ID and ASD without ID in Asian women that were from Asia and Black women from East Africa. Our large sample allowed us to perform a more
comprehensive assessment than previous studies that investigated race-ethnicity and immigration status more generally.

**Methods**

We linked state-wide databases and examined the risk of ASD with and without ID in children born in Western Australia (WA) from 1 January 1994 to 31 December 2005. We examined ASD prevalence according to maternal race-ethnicity, immigration status and region of birth, and for those born in Asia or East Africa, country of birth. Case mothers were grouped as ASD with ID and ASD without ID according to the disability status of their eldest child with ASD. Comparator mothers were those that had no child with ASD or ID and their index child was the eldest child. We used multinomial linear regression and adjusted for maternal age, socioeconomic status, parity and the year group of the index birth. STATA 13 was used for all analyses.

**Results and discussion**

When investigating race-ethnicity alone and compared to Caucasian women, we found that non-Caucasian women (especially Indigenous women) had lower odds of having a child with ASD with ID and even lower odds of a child with ASD without ID. Similarly, as one group, and when compared to non-immigrant women, immigrant women were less likely to have a child with ASD with ID and particularly ASD without ID. Further, non-Caucasian immigrant women were less likely to have a child with either ASD with or without ID. However, Asian women from South or Central Asia had about four and a half times the prevalence of ASD with ID in their children whereas Black women from East Africa had nearly 15 times the prevalence of ASD with ID. Further, we noted that all of these East African women came from a cluster of countries in the east of East Africa. No other Black African woman in the sample had a child with ASD.

**Conclusions and implications**

In WA, immigrant women of some minority ethnicities, and particularly Black women from East Africa, are at increased risk of a child with ASD with ID. Further research is needed to identify exposures to risk factors for ASD with ID in these women.
Appendix 4: Pre-existing maternal psychiatric disorder and the risk of ID or ASD

Title
Maternal psychiatric disorder and the risk of intellectual disability or autism spectrum disorder in subsequent offspring

Authors
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Abstract

Background
Psychiatric disorders are more common in the mothers of children with intellectual disability (ID) or autism spectrum disorder (ASD).

Objective
To assess the relationship between women’s outpatient psychiatric contacts and subsequent offspring with ID or ASD

Method
We linked three Western Australian population-based registers. The relationship between maternal outpatient psychiatric contacts before the birth and the odds of ID or ASD in a subsequent child were investigated using multinomial logistic regression.
Results

Women with previous outpatient psychiatric contacts were more likely to have a child with ASD with ID [2.5(95% CI: 1.8, 3.5)], mild or moderate ID of unknown cause [2.2(95% CI: 2.0, 2.5)], ID of unknown cause [2.14(95% CI: 1.91, 2.39)], ‘ASD without ID’ [2.1(95% CI: 1.6, 2.8)] or ‘any form of ID [OR, 2.1(95% CI: 1.9, 2.3)].

Conclusion

The likelihood of a child with ID of ASD increased for women with pre-existing psychiatric contacts.
Appendices

Appendix 5: The Broad Autism Phenotype and the burden of caring

Is the broad autism phenotype in mothers of children with autism spectrum disorder exacerbated by the challenges of caring for their children?

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Abstract

Study purpose: This study raises the hypothesis that the additional demands of parenting a child with autism spectrum disorder (ASD) may lead to behavioural and personality changes consistent with the Broader Autism Phenotype (BAP).

Background: In a previous study, 16 mothers of children with ASD were interviewed about their quality of life. A number of mothers indicated that they believed the additional demands of parenting a child with ASD led to changes in their behaviour and personality. These changes are of particular interest in relation to the BAP, which refers to the presence of mild autistic traits in an individual. Researchers have typically used the existence of a BAP to indicate genetic liability for ASD. However, it is possible that behavioural and personality changes in response to parenting a child with ASD may be skewing scores on measures of the BAP, such as the Autism-Spectrum Quotient (AQ).

Presentation of hypothesis: This qualitative study of parental interviews provided a preliminary examination of whether behaviours consistent with the BAP may have been exacerbated by the challenges of raising a child with ASD.

Testing methods: Qualitative analyses from the previous study revealed that seven of the 16 mothers reported behavioural and personality changes since the onset of their child’s ASD. We examine these behaviours in relation to the Autism-Spectrum Quotient and provided two potential designs for future studies to examine whether BAP-like behaviours may be exacerbated by parenting demands.

Implications of the hypothesis: A degree of caution is needed when researchers interpret measures of the BAP in parents who are full-time carers of their child with ASD. Some scores indicative of this phenotype may not solely represent a genetic liability for ASD. Longitudinal studies that explore the BAP among parents of children with ASD before, during and after the onset of caring will shed light on this complex research area.

Keywords: Autism, phenotype, mothers, autism spectrum disorders, genetic
Appendices

Appendix 6: Early mortality and causes of death in mothers of children with ID or ASD

Title
Early mortality and primary causes of death in mothers of children with intellectual disability or autism spectrum disorder: a retrospective cohort study

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Abstract

Introduction
Mothers of children with intellectual disability or autism spectrum disorder (ASD) have poorer health than other mothers; yet no research has explored whether this poorer health is reflected in mortality rates or whether certain causes of death are more likely. We aimed to calculate the hazard ratios (HRs) for death and for the primary causes of death in mothers of children with intellectual disability or ASD compared to other mothers.

Methods
The study population comprised all mothers of live-born children in Western Australia from 1983 to 2005. We accessed state-wide databases that enabled linking of socio-demographic details, birthdates, diagnoses of intellectual disability or ASD in the children and dates and causes of death for all mothers who had died prior to 2011. Using Cox regression with death by any cause and death by each of the three primary causes as the event of interest, we calculated HRs for death for mothers of children intellectual disability or ASD compared to other mothers.
Results and discussion
During the study period, mothers of children with intellectual disability or ASD had more than twice the risk of death. Mothers of children with intellectual disability were 40% more likely to die of cancer; 150% more likely to die of cardiovascular disease and nearly 200% more likely to die from misadventure than other mothers. Due to small numbers, only HRs for cancer were calculated for mothers of children with ASD. These mothers were around 50% more likely to die from cancer than other mothers. Possible causes and implications of our results are discussed.

Conclusion
Similar studies, pooling data from registries elsewhere, would improve our understanding of factors increasing the mortality of mothers of children with intellectual disability or ASD. This would allow the implementation of informed services and interventions to improve these mothers’ longevity.
Appendix 7: Onset of psychiatric disorders after the birth of a child with ID: a retrospective cohort study

TITLE: Onset of maternal psychiatric disorders after the birth of a child with intellectual disability: a retrospective cohort study

ABSTRACT: Mothers of a child with intellectual disability (ID) have more psychiatric disorders after the birth of their child than other mothers. However, it is unclear if this is because they have more psychiatric disorders before the birth or if the increase is related to the burden of caring for the child. We aimed to calculate the rate of new psychiatric disorders in mothers after the birth of their eldest child with ID born between 1983 and 2005 and to compare these with rates in women with a child with no ID or autism spectrum disorder (ASD) born during the same period. By linking data from Western Australian population-based registries, we selected women with no psychiatric history who survived the birth of their live-born child (N= 277,559) and compared rates of psychiatric disorders for women with a child with ID and women without a child with intellectual disability or ASD. Negative binomial regression with STATA 12 was used for all analyses. Mothers of children with mild-moderate ID of unknown cause had around two to three and a half times the rate of psychiatric disorders of mothers of children without ID or ASD. Mothers of children with Down syndrome and no pre-existing psychiatric disorder showed resilience and had no impairments in their mental health. Interventions and services are needed for mothers of other children with ID to improve their mental health. Further research is implicated to explore the mental health of mothers of children with ID and a pre-existing psychiatric disorder.

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Appendix 8: Onset of maternal psychiatric disorders after the birth of a child with ASD

Title
Onset of maternal psychiatric disorders after the birth of a child with autism spectrum disorder: A retrospective cohort study

Authors
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Abstract

Background
Mothers of a child with autism spectrum disorder (ASD) have more psychiatric disorders after the birth of their child. This might be because they have more psychiatric disorders before the birth; alternatively, the increase could be related to the burden of caring for their child.

Aims
We aimed to calculate the incidence of a psychiatric diagnosis in women with no psychiatric history after the birth of their eldest child with ASD compared to women with no child with ASD or intellectual disability (ID) and no psychiatric history.

Methods
By linking datasets from Western Australian population-based registries, we calculated the incidence of a psychiatric disorder in mothers of children with ASD and compared to mothers of children with no ASD or intellectual disability. Negative binomial regression using STATA 13 was used for all analyses.
Results

Apart from Alcohol and substance abuse, mothers of children with ASD had higher incidences of all categories of psychiatric disorders than other mothers.

Conclusions and implications

The higher incidence of psychiatric disorders in mothers of children with ASD and no psychiatric history compared to similar mothers with no child with ASD or ID might be due to a genetic predisposition coupled with an environmental trigger provided by the challenges of raising their children with ASD. In addition, the increased burden borne by these mothers might result in a higher incidence of psychiatric disorders in mothers that are not genetically disposed.
Experiences Impacting the Quality of Life of Mothers of Children with Autism and Intellectual Disability*

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In order to identify factors affecting the quality of life of mothers of children with both autism and intellectual disability, the first author interviewed sixteen mothers of affected 11-24 year olds and transcribed audio recordings of the interviews. The first two authors analysed the resulting texts using a hermeneutical phenomenological approach. Mothers described living with their child’s challenging behaviors, adapting to the increased demands and resultant isolation as lowering their quality of life. They described how surviving had strong negative impacts on their health and relationships. A majority of mothers described rewards associated with their children.

Keywords: quality of life, autism, intellectual disability, mothers, qualitative

Introduction

Quality of life is a concept describing overall well-being and results from a complex interaction of health, independence, relationships, goals and standards in the context of a person’s environment (Centre for Participant Reported Outcomes, 2007). Autism represents a group of severe, chronic, lifelong, neurodevelopmental disorders which are diagnosed by impairments in the areas of social interaction, communication, and repetitive behaviors or interests (American Psychiatric Association, 2002). Intellectual disability is characterized by an intelligence quotient of less than 70 and limitations in adaptive skill which are manifest before 18 years (Leonard & Wen, 2002). About 70% of persons with autism also have intellectual disability (Leonard et al., 2011).

Since the 1980s, the prevalence of autism diagnosis has been increasing both internationally (Centers for Disease Control and Prevention, 2012) and in Australia (Nassar et al., 2009). Parents of children with autism have been reported to have poorer physical and mental health (Mugno, Ruta, D’Arrigo, & Mazzone, 2007) and quality of life (Allik, Larsson, & Smedje, 2006) than other parents. Furthermore, mothers of children with autism have been shown to have more compromised health than fathers (Allik et al., 2006), mothers of healthy children (Totsika, Hastings, Emerson, Lancaster, & Berridge, 2011) and the mothers of children with other developmental disabilities (Griffith, Hastings, Nash, & Hill, 2010).

Reasons for the impaired quality of life of mothers of children with autism are likely to be complex. Some have postulated a genetic basis for the increased prevalence of mental disorders (Bolton, Pickles, Murphy, & Rutter, 1998). However, there are many external factors why mothers might have a lower quality of life. Children with autism have more challenging behaviors (Matson & Rivet, 2008) than healthy children and challenging behaviors are associated with lower maternal quality of life (Bourke‐Taylor, Law, Howie, & Pallant, 2010).

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