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

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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 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.

1. Introduction

Intellectual disability is diagnosed in people with an IQ of less than 70 and deficits in adaptive functioning which are present before 18 years (American Psychiatric Association, 2000). Children with intellectual disability have more challenging behaviours (Baker et al., 2002), more sleep disorders (Richdale et al., 2000) and more psychopathologies than typically developing children (Emerson, 2003). Their mothers also have increased expenses (Parish and Cloud, 2006) perceive more stigma against themselves or their child (Green, 2007) have lower employment levels (Shearn and Todd, 2000) and less informal and family support (Shearn and Todd, 2000) than other mothers. Therefore, it is not surprising that research has identified poorer mental health in mothers of children with intellectual disability compared to the parents of children with no disabilities (Bourke et al., 2008; Emerson et al., 2010; Olsson and Hwang, 2001).

In a previous study (Fairthorne et al., submitted for publication), we found that mothers with an outpatient psychiatric history were about twice as likely to have a child with intellectual disability compared to mothers of children with no intellectual disability. We hypothesised that this might be due to shared genetics of the mother and the child with intellectual disability or prenatal use of medication or life-style factors in women with a psychiatric disorder. In the current paper, we wanted to ascertain whether mothers of a child with intellectual disability and no previous psychiatric history were at increased risk of having a psychiatric disorder after the birth of their child. We reasoned that these comparisons would enable us to discern whether the burden of caring for a child with intellectual disability contributed to the increased rate of psychiatric disorders in their mothers. This being so, better informed services and interventions might be instituted with the aim of reducing the burden of these mothers and improving their mental health.

No previous research has attempted to differentiate whether the excess of psychiatric disorders in mothers of children with intellectual disability after the birth of their child is due to the increased burden of caring, a prior disposition to psychiatric disorders or to

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increased exposure to ante-natal risk factors for intellectual disability in women with a previous psychiatric disorder. Moreover, grouping mothers, according to the level of intellectual disability of their child and according to whether the cause is known would enable the most vulnerable groups of mothers to be identified.

Therefore, according to type and level of intellectual disability, we aimed to:

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

2. Methods

2.1. Study population

The study population consisted of all women who gave birth to a live child in Western Australia (WA) between 1st January 1983 and 31st December 2005 inclusive. We linked de-identified data-sets from four statutory state-based registries and a state-wide disability database (Holman et al., 1999). The *Hospital Morbidity Data System* (HMDS) (Department of Health WA, 2011) provided us with admission dates and ICD-9 and ICD-10 codes for all hospital separations in WA from 1970 to 2010. The *Mental Health Information System* (MHIS) (Department of Health WA, 2011) provided us with 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 us with the birth dates of all children born in WA during the collection period and socio-demographic information which we used to create explanatory variables. The WA Death Registry provided death dates of mothers and children to enable us to adjust the period when women were at risk of a psychiatric disorder due to the burden of care of their child. Using the *Intellectual Disability Exploring Answers* (IDEA) Database (Petterson et al., 2005), we gathered diagnostic information of children born between 1983 and 2005. Personnel from WA's *Data Linkage Unit* (Department of Health WA, 2011) created a unique code for each mother enabling us to link these data-sets. 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, our cohort comprised 277,559 mothers.

2.2. Maternal groups

We excluded mothers of children with ASD from the comparator group because researchers have also found that the mental health of mothers of children with ASD is poorer than that of mothers of typically developing children (Daniels et al., 2008; Montes and Halterman, 2007). Hence our comparator group was all women with a live child born between 1st January 1983 and 31st December 2005 and who had no child diagnosed with intellectual disability or ASD before December 31st, 2010. For comparator mothers, the index child was the first child born during the collection period. We allocated mothers of one or more children with intellectual disability (but not ASD) into one of four case groups. These were labelled *mild–moderate intellectual disability of unknown cause*, *severe intellectual disability of unknown cause*, *Down syndrome* and *intellectual disability of known cause excluding Down syndrome*. For these women, the index child was the eldest child with an intellectual disability. When choosing our case groups, we considered the particular challenges likely to result in differential burdens of care. For example, we separated mothers of children with severe intellectual disability of unknown cause from mothers of children with mild–moderate intellectual disability of unknown cause because children with severe intellectual disability are likely to have a much greater medical burden. Hence, the challenges faced by mothers could be expected to vary. We also separated mothers of children with Down syndrome from other mothers of children with intellectual disability of known cause. This was because the numbers for the former were sufficiently large and because research has identified that these mothers of children are less likely to have poorer mental health outcomes than mothers of children with other intellectual disability of known cause. The inter-relationships of these case groups are illustrated in Fig. 1.

2.3. Psychiatric disorders before the index birth

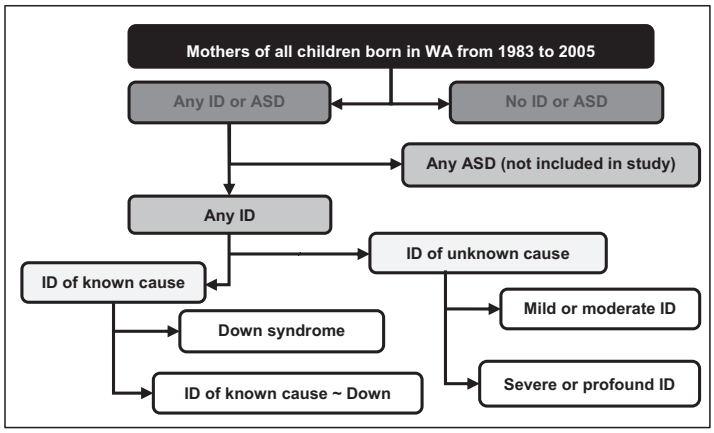
Mothers with a psychiatric disorder before the birth of their index child were excluded from our data-set. These were all women with one or more diagnoses from the eleven blocks of Chapter 5 of the International Statistical Classification of Diseases and Related Health Problems 10th Revision (ICD-10) (World Health Organisation, 2004) or an ICD-9 equivalent code (Supplementary Table 1).

2.4. Explanatory variables

We have previously demonstrated that socio-economic disadvantage, young maternal age and high parity are associated with the risk of mild–moderate intellectual disability of unknown cause (Leonard et al., 2011). Therefore, we included measures of these traits as variables in our model. We calculated a four-level variable for socio-economic status (SES) from the *Index of Relative Socioeconomic Disadvantage* (Australian Bureau of Statistics, 2009) for 2001. Where this was missing, we used the same statistic but for 1996 or 2006 or a similar statistic for 2001 termed 'Statistical local areas' rather than 'Collection Districts'. Maternal age at index birth was categorised as *<20*, *20–35* and *>35 years*. Parity at the time of index birth was categorised as *No previous child*; *One previous child*; *2–3 previous children*; and *>3 previous children*. The birth year of the index child was grouped into bands of 1983–88; 1989–94; 1995–2000 and 2001–5. We did not include births after 2005 as we reasoned that a five year lee-way period was needed for children with mild intellectual disability to have a reasonable opportunity to be diagnosed.

2.5. Psychiatric status

We used seven of the eleven blocks defined in ICD-10 (World Health Organisation, 2004) to categorise psychiatric status after the index birth (Supplementary Table 1). Block 1 (Organic disorders), Block 8 (Mental retardation), Block 9 (Disorders of psychological development) and Block 10 (Behavioural and emotional disorders with onset usually occurring in childhood and adolescence) were omitted because we saw these as unlikely to develop in response to care-giving or because they were life-long disorders. In order to determine the most frequent diagnostic categories, we created variables which 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, we created measures for women with one or more episodes from the block. These women were allocated a score which was equal to the sum of hospital admissions and outpatient contacts which were associated with an



ID, intellectual disability; ASD, autism spectrum disorder; ~ excluding; Down, Down syndrome

Fig. 1. Study population and maternal case groups.

ICD-10 code (or equivalent ICD-9 code) from the particular block. Our final measure of psychiatric status was 'Any psychiatric disorder'. Women were allocated a score which was the sum of all hospital admissions and outpatient contacts for a psychiatric disorder. Each of these measures 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 1
Maternal case groups by socio-demographic traits and index birth year group.

Trait	Comparator group (no intellectual disability or ASD)	Mild–moderate intellectual disability of unknown cause	Severe intellectual disability of unknown cause	Down syndrome	Intellectual disability of known cause excluding Down syndrome	Row totals
Socio-economic status						
Low	61,615	1522	79	109	236	63,561
	23.3%	33.5%	25.3%	23.8%	27.2%	23.5%
Medium	133,512	2353	170	220	453	136,708
	50.5%	51.8%	54.5%	48.0%	52.1%	50.6%
High	69,076	666	63	129	180	70,114
	26.3%	14.7%	20.2%	28.2%	20.7%	25.9%
Missing	7046	88	10	12	20	7176
	2.6%	1.9%	3.1%	2.6%	2.3%	2.6%
Maternal age at the index birth						
<20 years	19,764	519	24	16	71	20,394
	7.3%	11.2%	7.5%	3.4%	8.0%	7.4%
20–34 years	221,229	3668	249	313	691	226,150
	81.6%	79.2%	77.3%	66.6%	77.7%	81.5%
>35 years	32,256	492	49	141	127	31,015
	11.2%	9.6%	15.2%	30.0%	14.3%	11.2%
Parity at the index birth						
No previous child	199,960	1865	132	136	397	202,490
	73.7%	40.3%	50.0%	28.9%	44.7%	73.0%
One previous child	39,687	1372	102	141	256	41,558
	14.6%	29.6%	31.7%	30.0%	28.8%	15.0%
2–3 Previous children	28,218	1135	75	143	196	29,767
	10.4%	24.5%	23.3%	30.4%	22.1%	10.7%
>3 Previous children	3384	257	13	50	40	3744
	1.3%	5.6%	4.0%	10.6%	4.5%	1.4%
Birth year band						
1983–87	80,162	1026	88	106	245	81,627
	29.6%	22.2%	27.3%	22.5%	27.6%	29.4%
1988–93	65,800	1769	128	149	243	68,089
	24.3%	38.2%	39.8%	31.7%	27.3%	24.5%
1994–99	62,578	1227	63	98	224	64,190
	23.1%	26.5%	19.6%	20.9%	25.2%	23.1%
2000–5	62,709	607	43	117	177	63,653
	23.1%	13.1%	14.0%	26.8%	19.1%	22.9%
Total	271,249	4629	322	470	899	277,559
%	97.7%	1.7%	0.1%	0.2%	0.3%	100%

ASD, autism spectrum disorder; SES, socio-economic status.

2.6. Analyses

By maternal case groups, we calculated the incidence rate ratios (IRRs) of psychiatric disorders from each of the most frequently occurring categories, after the birth of the index child, and up to the end of 2010. We adjusted for SES, maternal age, parity and birth year band. We used Negative binomial regression using Stata 12 and report the adjusted IRRs and associated 95% confidence intervals (CI) for each measure of psychiatric status where person time was the 'offset' or denominator.

2.7. Ethics statement

Ethical approval for this study was granted by the WA Department of Health Human Research Ethics Committee (#2011/64).

3. Results

In Table 1, composition of the comparator group of 271,249 (97.7%) mothers and case groups in terms of the socio-demographic variables are shown. As our previous research described, mothers less than 20 years were over-represented in the *mild–moderate intellectual disability of unknown cause* case group (Leonard et al., 2011) and over-represented in the lowest SES group. By frequency, the four primary diagnostic categories were *Alcohol and substance abuse* ($N = 3923$), *Schizoid disorders* ($N = 2228$), *Affective disorders* ($N = 8265$) and *Neurotic disorders* ($N = 8441$). See Table 2.

3.1. Mild–moderate intellectual disability of unknown cause

Mothers of children with mild–moderate intellectual disability of unknown cause, and no previous psychiatric disorder, had significantly higher rates of all categories of psychiatric disorders compared to the mothers of children with no intellectual disability or ASD. These ranged from nearly three and a half times the rate for *Schizoid 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 disorder* [1.80 (95% CI: 1.5, 2.2)] and *Neurotic disorders* [1.80 (95% CI: 1.3, 2.5)] (Table 3, Fig. 2).

3.2. Severe intellectual disability of unknown cause

Mothers of children with severe intellectual disability of unknown cause, 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 (0.6, 6.4)] and about one and a half times the rate of *Schizoid disorders* [1.56 (95% CI: 0.1, 33.3)] as mothers without a child with intellectual disability 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 3, Fig. 3).

3.3. Down syndrome

Mothers of children with Down syndrome, and no previous psychiatric disorder, had reduced rates for all five categories of psychiatric disorders but no measure reached significance. (Table 3, Fig. 4)

3.4. Intellectual disability of known cause excluding Down syndrome

Mothers of children with intellectual disability of known cause excluding Down syndrome and no previous psychiatric disorder

Table 2
Numbers and percentage of mothers with psychiatric episodes after the index birth, by diagnostic category and maternal case group.

Block/category	Comparator group (no intellectual disability & no ASD)	Mild–moderate intellectual disability of unknown cause	Severe intellectual disability of unknown cause	Down syndrome	Intellectual disability of known cause ~ Down syndrome	Row totals
Alcohol and substance abuse	3697	182	8	7	29	3923
	1.4%	3.9%	2.5%	1.5%	3.3%	1.4%
Total episodes/block	18,540	1075	9	32	230	19,886
Schizoid disorders	2093	109	6	3	17	2228
	0.8%	2.4%	1.9%	0.6%	1.9%	0.8%
Total episodes/block	93,315	8347	35	31	2231	103,959
Affective disorders	7881	306	14	13	51	8265
	2.9%	6.6%	4.4%	2.8%	5.7%	3.0%
Total episodes/block	100,977	4286	540	227	484	106,514
Neurotic disorders	8038	326	19	12	46	8441
	3.0%	7.0%	5.9%	2.6%	5.2%	3.0%
Total episodes/block	70,002	3007	179	67	419	73,674
Behaviour disorders	1860	80	1	6	10	1957
	0.7%	1.7%	0.31%	1.3%	1.1%	0.7%
Total episodes/block	9948	321	14	23	15	10,321
Personality disorders	1573	76	4	3	10	1666
	0.6%	1.6%	1.2%	0.6%	1.1%	0.6%
Total episodes/block	21,705	830	13	6	12	22,566
Other disorders	1374	64	4	1	9	1452
	0.5%	1.4%	1.2%	0.2%	1.0%	0.5%
Total episodes/block	5228	211	5	1	24	5469
Any psychiatric disorder	25,818	890	52	50	155	26,965
	9.5%	19.2%	16.2%	10.6%	17.4%	9.7%
Total episodes/category	493,694	24,365	1053	914	4772	524,798
Number of mothers	271,249	4629	322	470	889	277,559
in case group	97.7%	1.7%	0.1%	0.2%	0.3%	100%
Total episodes/maternal	319,715	18,077	795	387	3415	342,389
group	93.4%	5.3%	0.2%	0.1%	1.0%	100%

ASD, autism spectrum disorder; ~, excluding.
Note: some mothers have diagnoses in multiple categories.

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 *Schizoid 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 3, Fig. 5).

4. Discussion

We explored the incidence of primary psychiatric disorders in mothers with no previous psychiatric history, after the birth of their child with intellectual disability, compared to mothers of children with no intellectual disability, no ASD and no psychiatric history whilst adjusting for socio-demographic factors. In this way, we were able to determine if the burden of caring for their child with a disability had contributed to a higher incidence of psychiatric disorders in case mothers.

4.1. Case group similarities

4.1.1. Mild–moderate and severe intellectual disability of unknown cause

Common genetic pathways have been found for intellectual disability and schizophrenia (Mefford et al., 2012). Hence mothers of children with mild–moderate or severe intellectual disability of unknown cause 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 (Tsuang, 2000). Thus, a genetic susceptibility, combined with the added challenges of caring for a child with intellectual disability, may have contributed to the increased incidence of schizophrenia in these mothers. Mothers of children with either mild–moderate or severe intellectual disability of unknown cause have significantly elevated rates of affective disorders compared to mothers of children with no intellectual disability 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 (Lennox et al., 2012).

Table 3
Incidence rate ratios and confidence intervals for blocks/categories of psychiatric disorders after the index birth.

Block/category	Mild or moderate intellectual disability of unknown cause	Down syndrome
Alcohol and substance abuse	2.91 (95% CI: 2.0, 4.3)	0.99 (95% CI: 0.3, 3.4)
Schizoid disorders	3.49 (95% CI: 1.6, 7.5)	0.31 (95% CI: 0.03, 3.6)
Affective disorders	1.98 (95% CI: 1.4, 2.8)	0.59 (95% CI: 0.2, 1.7)
Neurotic disorders	1.80 (95% CI: 1.3, 2.5)	0.68 (95% CI: 0.2, 1.8)
Any psychiatric disorder	1.80 (95% CI: 1.5, 2.2)	0.82 (95% CI: 0.4, 1.6)
	Severe or profound intellectual disability of unknown cause	**Intellectual disability of unknown cause excluding Down syndrome**
Alcohol and substance abuse	0.58 (95% CI: 0.1, 3.2)	2.77 (95% CI: 1.2, 6.5)
Schizoid disorders	1.56 (95% CI: 0.1, 33.3)	2.00 (95% CI: 0.4, 11.3)
Affective disorders	5.12 (95% CI: 1.4, 18.5)	1.12 (95% CI: 0.5, 2.4)
Neurotic disorders	1.98 (95% CI: 0.6, 6.4)	1.73 (95% CI: 0.9, 3.5)
Any psychiatric disorder	0.85 (95% CI: 0.4, 1.9)	1.68 (95% CI: 1.1, 2.7)

4.1.2. Mild–moderate intellectual disability and intellectual disability of known cause excluding Down syndrome

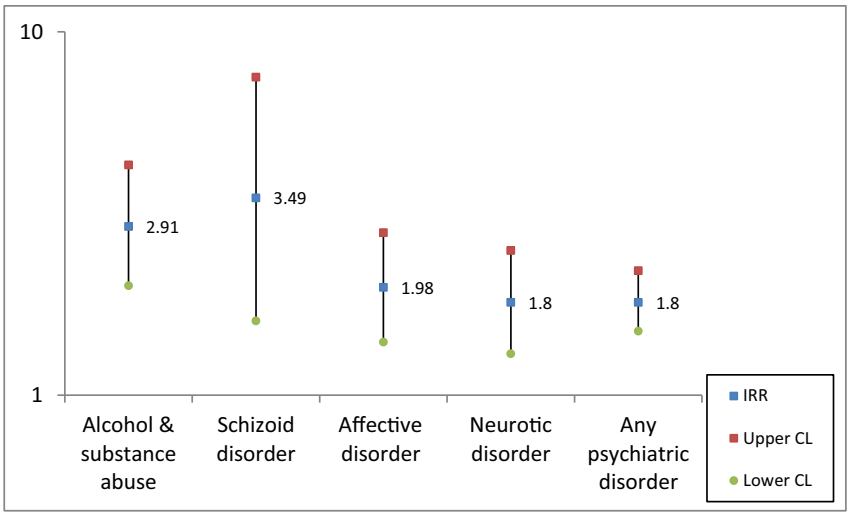
The psychiatric profile of mothers of children with *mild–moderate intellectual disability* and mothers of children with a biomedical cause for their intellectual disability other than Down syndrome were similar (Supplementary Fig. 1). All IRRs were elevated in both groups but higher in the mothers of children with *mild–moderate intellectual disability* 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 sub-groups of mothers in these case-groups would be that the children of the mothers with mild intellectual disability have an undiagnosed *Foetal alcohol spectrum disorder* (FASD) and the children with *intellectual disability excluding 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 (O'Leary, 2004). Finally, the elevated IRRs for schizoid disorders in both groups may be in part due to an interaction of *Schizoid disorders* and *Alcohol and substance abuse* since researchers have identified that cannabis use may impact on the subsequent development of schizophrenia (Van Os et al., 2010).

There are likely to be multiple reasons for the poorer psychiatric health of the mothers of children with mild–moderate intellectual disability of unknown cause compared to that of mothers of children with a known cause other than Down syndrome for their intellectual disability. 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 (Leonard et al., 2004). Further, a 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 (Knott et al., 2012). Parents with a cause for their child's disability are further empowered since 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.

4.2. Other case groups

4.2.1. Severe intellectual disability and mild–moderate intellectual disability

Both mothers of children with severe intellectual disability and mild–moderate intellectual disability had significantly higher rates of affective disorders. Notably, the rate in mothers of children with severe intellectual disability 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 (Morgan et al., 2012) have concluded that mothers with bipolar disorder or unipolar major depression were more likely to have a child with intellectual disability. Using the Beck Depression Inventory (BDI), mothers of children with developmental delay exhibited more depression than mothers of children without these disabilities (Harvey et al., 1997) as did Latina mothers of children with ID compared to Latina mothers of typically developing children (Blacher et al., 1997). However, we found no study which compared the levels in mothers according to the level of the intellectual disability of their child. We believe that we are the first research group to show that mothers of children with severe intellectual disability have higher rates of affective disorders than mothers of children with mild–moderate ID.

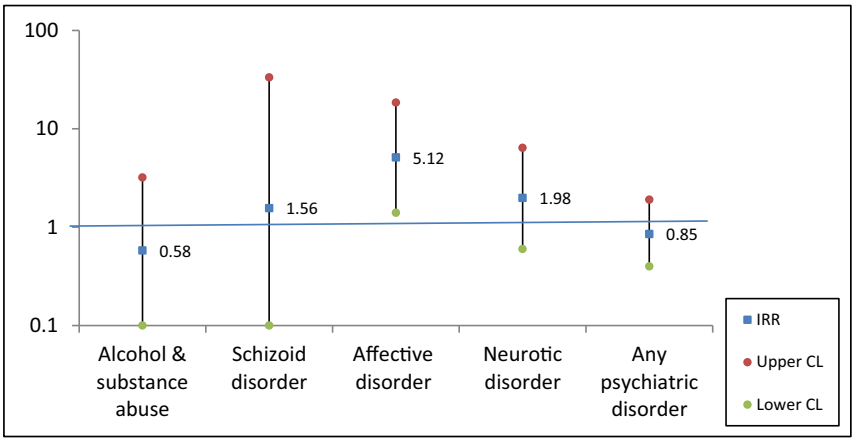


IRR, incidence rate ratio; CL, confidence limit
* IRRs are adjusted for maternal age, SES, parity and index birth year band

Fig. 2. Adjusted* incidence rate ratios, after the index birth, for mothers of children with mild–moderate intellectual disability of unknown cause by block/category.

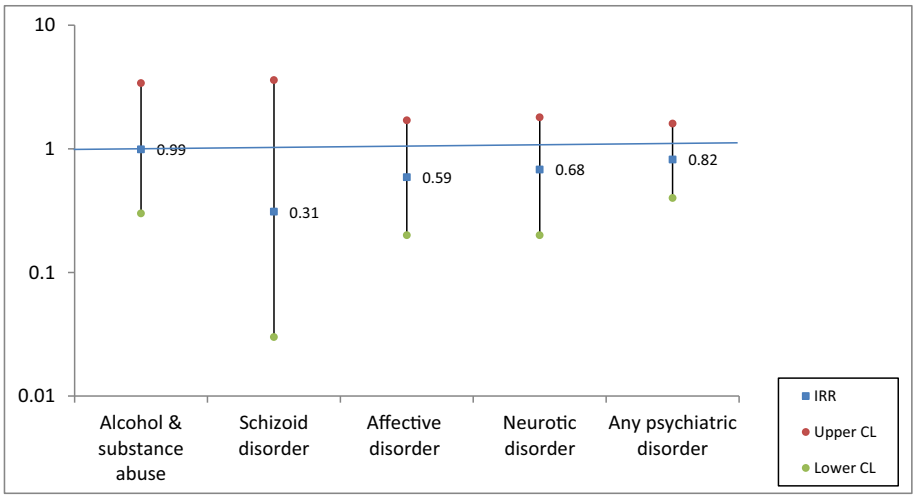
4.2.2. Down syndrome

Mothers with no previous psychiatric disorder prior to the birth of a child with Down syndrome had non-significantly lower rates of psychiatric disorders in all areas compared to other case and comparator mothers. This would be consistent with some 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 intellectual disability (Hodapp et al., 2001; Van Riper et al., 1992). 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 (Bourke et al., 2008; Hedov et al., 2000). Our study was restricted to mothers with no previous psychiatric disorder whereas some of the mothers in the referenced studies (Bourke et al., 2008; Hedov et al., 2000) 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 of Down syndrome. This is often not the case for other forms of intellectual disability. For example, around a half mothers of children with intellectual disability never find out a cause for their child's disability (Leonard and Wen, 2002). Furthermore, unlike some other forms of intellectual disability, 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 (Lenhard et al., 2005). Thirdly, the fact that Down syndrome is not hereditary and is caused by an accident at meiosis means that the mother can bear no responsibility for the occurrence of the disorder in her child. Hence, there can be no maternal guilt associated with the disorder. This is contrasted to maternal guilt and stigmatisation which might be associated with FASD (Chudley et al., 2005) or unjustified, but nevertheless, real guilt where the mother is a carrier in an x-linked disorder such as Fragile X syndrome (James et al., 2006). Finally, the apparent increased resilience of these mothers might be associated with the increased rewards and subjective well-being that mothers of



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

Fig. 3. Adjusted* incidence rate ratios, after the index birth, for mothers of children with severe intellectual disability or unknown cause by block/category.



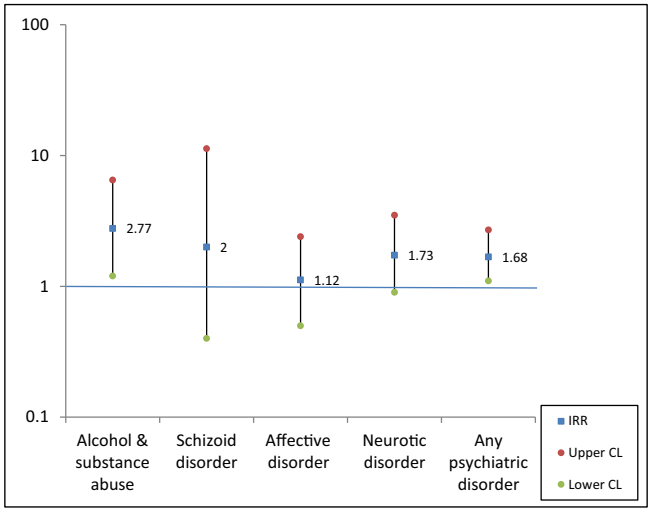
IRR, incidence rate ratio; CL, confidence limit
* IRRs are adjusted for maternal age, SES, parity and index birth year band

Fig. 4. Adjusted* incidence rate ratios, after the index birth, for mothers of children with Down syndrome by block/category and after the index birth.

children with Down syndrome have reported in relation to mothers of children with other developmental disabilities (Corrice and Glidden, 2009).

4.3. Strengths and limitations

The population-based nature of this study was a considerable strength of this study and greatly limited the risk of selection bias. Furthermore, we were able to retrospectively access hospital and outpatient records collected over forty years and, unlike other studies in this area (Bolton et al., 1998; Eriksson et al., 2012; Totsika et al., 2011), our data were independent of maternal recall. The existence of the IDEA data-base allowed us access to categorise intellectual disability by level and cause. The exclusion of all mothers with pre-existing psychiatric disorders enabled us to view emergent psychiatric disorders which were associated with the onset of caring. To our knowledge, no such studies have been published before.



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

Fig. 5. Adjusted* incidence rate ratios, after the index birth, for mothers of children with intellectual disability of known cause excluding Down syndrome, by block/category.

Unfortunately, we had no access to private outpatient data which meant that some mothers with psychiatric disorders would not have been identified in our 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 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 1). However, we adjusted for index parity which would have eliminated the potential bias caused by this inequality.

5. Conclusion and implications

In this study, we excluded mothers who had a hospitalisation or an outpatient contact for a psychiatric disorder in WA before the index birth. We made adjustments for maternal age, parity, socio-economic status and 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 (and attenuated) incidence of psychiatric disorders we identified is mostly due to the burden of caring rather than genetics or pre-existing environmental factors. Hence, we concluded that the burden of caring for a child with *intellectual disability of known cause excluding Down* syndrome and particularly of mild–moderate intellectual disability without a known cause increases the risk of a psychiatric disorder after the birth of their child. We did not find this association for mothers of children with Down syndrome. Exploring the IRRs of psychiatric disorders in these same subgroups of intellectual disability 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 intellectual disability.

Contributors

The study was conceived by Jenny Fairthorne and developed with the assistance of Peter Jacoby and Nick de Klerk. Jenny

Fairthorne wrote the original draft and all authors participated in its subsequent development and approved the final submission.

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Conflicts of interest

None.

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Appendix A. Supplementary data

Supplementary data related to this article can be found at http://dx.doi.org/10.1016/j.jpsychires.2014.11.011.

References

American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 4th ed. 2000. Washington, DC.
Australian Bureau of Statistics. Information paper: an introduction to socio-economic indexes for areas (SEIFA). 2009. Canberra.
Baker B, Blacher J, Crnic K, Edelbrock C. Behavior problems and parenting stress in families of three-year-old children with and without developmental delays. Am J Ment Retard 2002;107:433–44.
Blacher J, Lopez S, Shapiro J, Fusco J. Contributions to depression in Latina mothers with and without children with retardation: implications for caregiving. Fam Relat 1997;46:325–34.
Bolton P, Pickles A, Murphy M, Rutter M. Autism, affective and other psychiatric disorders: patterns of familial aggregation. Psychol Med 1998;28:385–95.
Bourke J, Ricciardo B, Bebbington A, Aiberti K, Jacoby P, Dyke P, et al. Physical and mental health in mothers of children with Down syndrome. J Pediatr 2008;153:320–6.
Chudley A, Conry J, Cook J, Loock C, Rosales T, LeBlanc N. Fetal alcohol spectrum disorder: Canadian guidelines for diagnosis. Can Med Assoc J 2005;172:S1–21.
Corrice A, Glidden L. The Down syndrome advantage: fact or fiction? Am J Intellect Dev Disabil 2009;114:254–68.
Daniels J, Forssen U, Hultman C, Cnattingius S, Savitz D, Feychting M, et al. Parental psychiatric disorders associated with autism spectrum disorders in the offspring. Pediatrics 2008;121:1357–62.
Department of Health WA. What we collect and manage. 2011.
Emerson E. Prevalence of psychiatric disorders in children and adolescents with and without intellectual disability. J Intellect Disabil Res 2003;47:51–8.
Emerson E, McCulloch A, Graham H, Blacher J, Llwellyn G, Hatton C. Socioeconomic circumstances and risk of psychiatric disorders among parents of children with early cognitive delay. Am J Intellect Dev Disabil 2010;115:30–42.
Eriksson M, Westerlund J, Anderlid B, Gillberg C, Fernell E. First-degree relatives of young children with autism spectrum disorders: some gender aspects. Res Dev Disabil 2012;33:1642–8.
Fairthorne J., Hammond G., Bourke J., de Klerk N. and Leonard H., Maternal psychiatric disorder and the risk of intellectual disability or autism spectrum disorder in subsequent offspring, 2014, [submitted for publication].
Green S. We're tired, not sad: benefits and burdens of mothering a child with a disability. Soc Sci Med 2007;64:150–63.
Harvey J, O'Callaghan M, Vines B. Prevalence of maternal depression and its relationship to ADL skills in children with developmental delay. J Paediatr Child Health 1997;33:42–6.
Hedov G, Anneren G, Wikblad K. Self-perceived health in Swedish parents of children with Down's syndrome. Qual Life Res 2000;9:415–22.
Hodapp R, Ly T, Fidler D, Ricci L. Less stress, more rewarding: parenting children with Down syndrome. Parenting 2001;1:317–37.
Holman C, Bass A, Rouse I, Hobbs M. Population-based linkage of health records in Western Australia: development of a health services research linked database. Aust N Z J Public Health 1999;23:453–9.
James C, Hadley D, Holtzman N, Winkelstein J. How does the mode of inheritance of a genetic condition influence families? A study of guilt, blame, stigma, and understanding of inheritance and reproductive risks in families with X-linked and autosomal recessive diseases. Genet Med 2006;8:234–42.
Knott M, Leonard H, Downs J. The diagnostic odyssey to Rett syndrome: the experience of an Australian family. Am J Med Genet 2012;158A:10–2.
Lenhard W, Breitenbach E, Ebert H, Schindelhauer-Deutscher H, Henn W. Psychological benefit of diagnostic certainty for mothers of children with disabilities: lessons from Down syndrome. Am J Med Genet 2005;133A:170–5.
Lennox N, Wong G, Taylor M, Ware R. Family stress and adults with intellectual disability. Intellect Disabil Australasia 2012;33:3–8.
Leonard H, Glasson E, Nassar N, Whitehouse A, Bebbington A, Bourke J, et al. Autism and intellectual disability are differentially related to sociodemographic background at birth. PLoS One 2011;6:e17875.
Leonard H, Slack-Smith L, Phillips T, Richardson S, D'Orsogna L, Mulroy S. How can the internet help parents of children with rare neurologic disorders? J Child Neurol 2004;19:902–7.
Leonard H, Wen X. The epidemiology of mental retardation: challenges and opportunities in the new millennium. Ment Retard Dev Disabil Res Rev 2002;8:117–34.
Mefford H, Batshaw M, Hoffman E. Genomics, intellectual disability, and autism. N Engl J Med 2012;366:733–43.
Montes G, Halterman J. Psychological functioning and coping among mothers of children with autism: a population-based study. Pediatrics 2007;119:1040–6.
Morgan V, Croft M, Valuri G, Zubrick S, Bower C, McNeil T, et al. Intellectual disability and other neuropsychiatric outcomes in high-risk children of mothers with schizophrenia, bipolar disorder and unipolar major depression. Br J Psychiatry 2012;205:282–9.
O'Leary C. Fetal alcohol syndrome: diagnosis, epidemiology, and developmental outcomes. J Paediatr Child Health 2004;40:2–7.
Olsson M, Hwang C. Depression in mothers and fathers of children with intellectual disability. J Intellect Disabil Res 2001;45:535–43.
Parish S, Cloud J. Financial well-being of young children with disabilities and their families. Soc Work 2006;51:223–32.
Petterson B, Leonard H, Bourke J, Sanders R, Chalmers R, Jacoby P, et al. IDEA (intellectual disability exploring answers): a population-based database for intellectual disability in Western Australia. Ann Hum Biol 2005;32:237–43.
Richdale A, Andre F, Gavidia-Payne S, Cotton S. Stress, behaviour, and sleep problems in children with an intellectual disability. J Intellect Dev Disabil 2000;25:147–61.
Shearn J, Todd S. Maternal employment and family responsibilities: the perspectives of mothers of children with intellectual disabilities. J Appl Res Intellect Disabil 2000;13:109–31.
Totsika V, Hastings R, Emerson E, Lancaster G, Berridge D. A population-based investigation of behavioural and emotional problems and maternal mental health: associations with autism spectrum disorder and intellectual disability. J Child Psychol Psychiatry 2011;52:91–9.
Tsuang M. Schizophrenia: genes and environment. Biol Psychiatry 2000;47:210–20.
Van Os J, Kenis G, Rutten B. The environment and schizophrenia. Nature 2010;468:203–12.
Van Riper M, Ryff C, Pridham K. Parental and family well-being in families of children with Down syndrome: a comparative study. Res Nurs Health 1992;15:227–35.
World Health Organisation. International classification of diseases (ICD). 2004.