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Exploring mental health outcomes in parents of children with spina bifida in Sweden: a population-based study

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Abstract

Background

Spina bifida (SB) is a neural tube defect causing disability. Qualitative and non-population-based studies show that parents of children with disabilities experience stress, depression, and anxiety. This study assessed whether having a child with SB is associated with higher risks of parents receiving diagnoses of mood, anxiety, or sleep disorders, and medications prescribed for these conditions.

Methods

This population-based cohort study included 682 parents of children with SB and a matched group of 4,205 controls. Logistic regression was used to analyze the association between having a child with SB and parental mental health outcomes; a sub-analysis explored the effects of SB-related factors. Survival analysis was used to assess the timing of occurrence of the outcomes.

Results

No significant associations between having a child with SB and parental mental health outcomes were found. However, mothers of children with SB, with lower education, and with a mental health condition at baseline are more likely to present mental health outcomes. In parents of children with SB, the developmental stage of the child was associated with a reduced risk of mood disorders (OR = 0.64, p = 0.050) in early childhood, antidepressant use in infancy (OR = 0.80, p = 0.049), early childhood (OR = 0.77, p = 0.044), and middle childhood (OR = 0.60, p < 0.001), anxiolytic use in middle childhood (OR = 0.71, p = 0.011), and sedatives use in early (OR = 0.73, p = 0.037) and middle childhood (OR = 0.46, p < 0.001). The severity of SB and comorbidities were associated with parental mental health. Survival analysis showed no significant differences between the two groups.

Conclusion

Parents of children with SB, during certain developmental stages or with SB-related comorbidities, should be monitored to prevent adverse mental health outcomes. Prevention strategies and support are recommended.

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Introduction

Spina bifida (SB) is a congenital neural tube defect caused by an incomplete closure of the spinal cord. The birth prevalence of the condition has varied over time and regions, with some countries and regions experiencing a higher prevalence of SB, such as Algeria, Nigeria, Jordan, and Northern China (1, 2). Changes in incidence depend on several factors, including prenatal screening and prevention methods such as folic acid fortification in certain food staples and prenatal vitamin intake (1, 3). The incidence of SB has decreased over the years in Western countries, however, the prevalence among the adult population has grown due to higher survival rates (4).

There are several types of SB, that are associated with different levels of severity and are usually classified in open and closed (also called SB occulta) (5). The three most frequent types include myelomeningocele (MMC), meningocele, and SB occulta (6). MMC is the commonest and most severe type of open SB, and it involves extrusion of the meninges, cerebrospinal fluid, and nerves and causes more functional and neurological limitations and disabilities (5, 6). Meningocele is a type of open SB that involves spinal elements but without affecting the nerves, and therefore generally causes minor disabilities (5). Finally, SB occulta is the mildest type of SB, where a gap in the spine is present but no opening, nerve, or cord damage is present; this type of SB usually does not cause disability and does not require any specific intervention (6, 7). In general, the higher the lesion is, the more significant the impairments will be, as all the nerves below the damage will be affected (1).

The extent of symptoms and the impact on quality of life depend on several factors, both condition-specific, such as type of SB, level of lesion, presence of hydrocephalus and related neurological conditions (e.g., Arnold Chiari type II malformation), surgical interventions, comorbidities, and the development of secondary conditions, as well as more social factors, such as socio-economic status (SES) (8). A major complication of SB is hydrocephalus, an abnormal accumulation of cerebrospinal fluid that can increase the risk of morbidity and mortality (9). The number of newborns that develop hydrocephalus is around 80%, with variations dependent on pre- or post-natal repair (10). Generally, those undergoing fetal surgery are less likely to have hydrocephalus compared to those who close the lesion after birth (11). Secondary conditions, such as bladder and bowel incontinence, pain, spinal curvature anomalies, infections, and comorbidities, such as epilepsy and cognitive

impairment, are frequent in people with SB (12, 13). Studies also show a higher incidence of depression, anxiety, insomnia, and other mental health conditions in people with SB compared to their peers (14, 15), which have been found to be associated with health/condition-specific, demographic, and SES factors (16, 17). Secondary conditions and comorbidities can have a strong impact on the quality of life of people with SB, causing higher rates of hospitalization for preventable conditions and limitations in social activities and participation. Despite improvements in early medical and surgical treatments of individuals with SB, people with the condition have been shown to experience shorter life expectancy, up to 40% lower, compared to their peers varying based on personal and contextual factors (8, 18, 19). Adolescence, with the transition from pediatric to adult care, is usually one of the most vulnerable stages, as it becomes harder to be followed in a multidisciplinary setting – largely due to a lack of qualified adult providers –, which can lead to a higher risk of complications and non-adherence to treatment (20). People with SB can also be more likely to encounter social stigma and isolation (12). These distressing factors can lead to increased rates of mental health issues (21).

In the context of disability, parents of children with chronic health conditions or disabilities are also confronted with many short and long-term challenges related to the health and well-being of their children and their perceived vulnerability including medical treatments, medical emergencies, surgeries, lack of or delayed independence, social stigma, and uncertainty about future expectations (22, 23). Mental health-related symptoms can influence the parents' functional capacity, emotional aspects, and overall mental health (24). Families have to provide basic care for different aspects of their children’s lives such as development, nutrition, and personal hygiene (23, 24). While this may be true for all parents, this role of caregiving continues for longer in parents of children with SB. Parents cover the roles of caregivers and have multiple obligations that can be overwhelming and demanding (25, 26). Although the role of caregiving often is rewarding, it can also have negative consequences on the parents, who can experience emotional, physical, social, and financial burdens. High levels of stress are common in informal caregivers, usually parents, of children with disabilities, and they may lack the ability, resources, or time to cope with it. This results in parents having a higher risk of depression, anxiety, anger, and insomnia, which in turn can reduce the quality of the care given to children (27, 28). Furthermore, parents may have to change their routines and social habits, which can increase loneliness and depression and contribute to couple dissatisfaction (29, 30).

While Sweden has laws that alleviate the financial burden for people with disabilities and provide additional benefits if needed, research from other contexts has shown that having a child with disability can contribute to financial distress for families (31, 32). Data from Australia, a country with a universal healthcare system that is administered differently from Sweden's, indicate that having a child with disability is also associated with a higher financial burden due to both increased expenses and difficulties in adjusting employment to accommodate caregiving responsibilities, often leading to a reduction in household income (33, 34). As most children with SB – in particular those with MMC and hydrocephalus – present executive dysfunction and therefore have difficulties in planning, decision-making, and regulation (35), worries about the children's future and their ability to function in daily life also contribute to parental concerns, adding to the usual pressure experienced by parents during the transition to independence phases (29). To further exacerbate the issue, in certain contexts, it may be difficult for parents, to get support from community programs, which decreases feelings of self-efficacy and preparedness. This has been found in research studies from Australia and South Africa (26, 34) and may or may not be relevant in the Swedish context. Nevertheless, parents often report inadequate support and dissatisfaction with the services available to aid them in their caregiving responsibilities, highlighting many unmet needs in this population (26). Difficulties in getting help and support from family and friends, who might feel uncomfortable or insecure when it comes to taking care of children with disabilities or providing emotional and psychological support, is also present, thus increasing the parents' sense of isolation (36). They may also experience loneliness and feel like they do not belong in social circuits because of their different parenthood experience (37).

Several factors are involved in the association between mental health and having a child with disabilities, both related to functional state and context (38). Factors that influence depression and anxiety in parents are, for example, the need and resources for continuing re/habilitation care, the child's in/ability to walk, the presence of hydrocephalus and the shunt management, the severity of the condition, and the oftentimes numerous surgeries (36, 39, 40). Control of continency and concerns over clean and intermittent catheterization (CIC) are also important functional factors for children with SB and their caregivers, as it can lead to trauma, low self-esteem, and have negative effects on social interactions (36, 41).

SES and environmental factors can also mediate the association between having a child with SB and negative mental health outcomes in the parents. In fact, parents from lower-income households are shown to experience higher levels of stress and have less access to assistive

technology, re/habilitation services, catheterization, and family support (22, 36). Parents have reported financial aid to be one of the most needed elements to improve the care and management of children with disabilities (23). Parents' education, employment status, marital status, and race have also been found to be associated with parental mental health outcomes (22, 36, 39). Specifically, having a foreign background can lead to more challenges in accessing and navigating the healthcare and social systems. Furthermore, SB seems to be more prevalent among people with foreign backgrounds in Sweden, possibly due to higher survival influenced by the "healthy migrant effect", that is migrants often have a better health status than the population of their native country (19, 42). In fact, as observed in the national follow-up program and registry for individuals with SB – MMCUP –, most of the children with MMC in Sweden today were born abroad or have parents who were born abroad. The age of the child and the parents can also influence the level of stress in parents, with inconsistent findings showing both an increase and decrease in parental stress as the children become older (22, 39). Furthermore, expectations of what developmental milestones their child should achieve over time might be another stressor for parents of children with disabilities such as SB. For example, as children become older, parents typically expect them to become more independent and may develop stress if they do not follow the "typical" developmental trajectories (22, 43, 44). The realization that they may need continued attention or assistance from parents or caregivers may also exacerbate stress (22).

Several studies have investigated the impact of children's disabilities on their parents, to understand whether these families are more or less resilient compared to parents who do not have the experience of raising a child with a disability. Resilience is a positive and adaptive response to difficult conditions, contributing to better coping with negative or distressing situations (45, 46). The development of resilience depends on different risk and protective factors, and the person's internal resources such as self-efficacy or optimism (47). Overall, different levels of resilience can influence the perception that parents have of the challenges associated with having a child with disability, and it can decrease their risk of developing adverse mental health outcomes (45). Some studies have suggested that families of children with disability can exhibit more resilience and unity between parents, especially during the adolescent transition phase, and that parents overall can still have a positive view of their lives (24, 36, 43). Nevertheless, most studies have shown that parents of children with disabilities are more likely to experience higher levels of stress, depression, and anxiety which can lead to lower quality of life (23, 36). A study from China by Xia et al. has reported that 33.6% and

36.1% of parents of children with disabilities have experienced depression and anxiety, at levels higher than those observed in the general population (23). A qualitative study conducted in Uganda (36) indicates that over half of the parents of children with disabilities interviewed were on the 90th percentile of the stress measurement scale used. Furthermore, most studies have focused on maternal adjustment to disability, however, as the caregiving role of fathers has increased in recent decades, the burden of care is often shared between mothers and fathers and can affect their well-being in different ways (22, 48). While mothers usually experience a higher risk of poor mental health, depression, and anxiety, broader research will be needed (48).

Negative mental health outcomes and parental stress may influence both family relations and health outcomes in children. Parental perception of child vulnerability (PPCV) refers to the parents' belief and fear that their child may be more susceptible to illness or early death, and it is common in parents of children who have a disability or a chronic illness (49). High levels of anxiety and depression might increase the perceived vulnerability of children (22). Findings on PPCV vary by age – with Driscoll et al. reporting that, as children grow older, parents perceive higher vulnerability during adolescence (22), whereas Malm-Buatsi et al. finding the highest vulnerability perception during preschool ages (39). However, they all report a negative influence on parental and children's outcomes. High levels of parenting stress are also associated with a lower quality of life in children and might serve as an accurate proxy for the child's quality of life (50). Mental health conditions in parents are associated with the well-being of the child and can lead to behavioral changes and a higher risk of developing adverse mental health conditions in children (51). Furthermore, despite contributing to the development of resilience in the family, parenting stress can influence family cohesion and conflicts in different degrees, depending, for example, on age groups (with less cohesion during pre-adolescence)(43).

Most of the existing studies on the mental health of parents of children with disabilities do not specifically focus on parents of children with SB but rather explore various types of disabilities. While some experiences may be overlapping, this fails to capture the complexity and variety present in SB, which is a highly complex disability. To the author's knowledge, there are no population-based studies on mental health in parents of children with SB, as most of the reviewed literature is qualitative or has used surveys. Furthermore, the Swedish universal healthcare system and the social welfare state present important contextual

differences, as more support systems, both for practical management of the disability and for easing financial distress, are present (31). Hence, findings from existing studies may not be applicable to parents of children with SB in Sweden.

Aim of the study and research question

This study has the objective of expanding the current knowledge by providing a wide analysis of mental health outcomes in parents of children with SB accounting for socio-economic and disability-related factors. The overall aim of this study was to assess the prevalence of mood disorders., anxiety disorders, and sleep disorders as well as the most commonly prescribed medications to treat these conditions – namely anti-depressants, anxiolytics, and sedatives and compare these rates to those of a matched control group. Moreover, we assessed how certain disability-specific factors were associated with these mental health conditions and the medications used to treat these conditions. Specifically, we focused on three research questions:

1. Do parents of children with SB have a higher risk of developing negative mental health outcomes (e.g., mood disorders, anxiety disorders, and sleep disorders)?
2. Are specific characteristics of SB symptoms and severity associated with a higher risk of negative mental health outcomes in parents of children with SB?
3. Do the associations change in different children's developmental stages?

We hypothesized that parents of children with SB would be more likely to have negative mental health outcomes compared to parents of children without SB and that this would be reflected by the International Classification of Disorders (ICD) and Anatomical Therapeutic Chemical (ATC) codes indicating mental health conditions and dispensed medications to treat such conditions. Moreover, within the SB group, we expected parents of children who have more severe forms of SB and comorbidities to be more likely to present the outcomes.

Methods

Study design and setting

The present study was a retrospective cohort population-based study set in Sweden. The data included came from the Swedish CPNorth database (52). In this thesis, data from the Statistics Sweden's Longitudinal Integrated Database for Health Insurance and Labor Market Studies,

the patient's register, the pharmaceutical register, the medical birth register, and the national quality and follow-up register for SB (MMCUP) were merged and included. The study period was from 2001 to 2015.

Study population

The exposure in this study was a binary variable identified as having a child with SB as identified by their ICD-10 diagnosis. Participants were parents of children (defined as individuals under 18 years) with SB identified in the CPNorth database by their ICD-10 codes and children who do not have SB (or cerebral palsy). A control group is included that is matched in a proportion of 5:1 by sex, birth year, and municipality. Codes used to identify SB are Q05.0 to Q05.9 (unspecified: Q05, Q05.4, Q05.9; cervical: Q05.0, Q05.5; thoracic: Q05.1, Q05.6; lumbar: Q05.2, Q05.7; sacral: Q05.3, Q05.8).

Diagnostic codes in national registers may be subjected to overestimations and misdiagnoses (53), and therefore specific exclusion criteria were used according to the following: individuals with diagnoses considered incompatible with SB (persistent cloaca: Q437; anencephaly: Q00; congenital absence, atresia, and stenosis of large intestine: Q42; other congenital malformations of nervous system-Arnold Chiari: Q07), individuals with other spinal diagnoses that lack a specific SB diagnosis (with codes Q05.1–8), and individuals with SB diagnosis only from the medical birth register but not from the national patient register (unless SB is listed as a cause of death). The exclusion criteria were developed by a senior pediatric neurologist with extensive knowledge of SB. Only parents with data from at least two years before the birth of the child and one year after were included to avoid information bias.

Study variables

Demographic variables included age, sex, foreign background (binary variable coded as Swedish or not Swedish), education (binary variable coded as mandatory education or less, and secondary education or higher), and parity (binary variable coded as birth of parity < 1 and > 1). Additional variables related to the *condition-specific/health of the children with SB* included: type of SB, level of lesion, presence of hydrocephalus, intellectual disability, and epilepsy. The type of SB was classified as open, closed, or suspected; the levels of lesion were classified in accordance with the ICD-10 Q05 codes as cervical, thoracic, lumbar, sacral, and unspecified; presence of hydrocephalus and lifetime experience of epilepsy were binary variables (yes/no), and finally, the F70 codes from the ICD-10 for intellectual disability

were used: mild, moderate, severe, and profound. The codes used to identify SB-related conditions were: G91.0-G91.9, Q03.0-Q03.9, Q05.0-Q05.4 for hydrocephalus, G40.0-G41.9 for epilepsy, and F70.0-F79.9 for intellectual disability (mild: F70, moderate: F71, severe: F72, profound: F73, other/unspecified: F74, F79).

Children were also divided in age groups, according to their developmental stage, for further analysis, and groups included were neonatal period and infancy (<= 1 year), toddler (1-2 years), early childhood (2-5 years), middle childhood (6-11 years), and early adolescence (12-18 years) (54). The categories chosen were developed by the United States National Institute of Child Health and Human Development (NICHD) based on research from several pediatric organizations with the only variation being the merging of the neonatal and infancy periods to avoid small samples (54).

While the specific etiology of mental illness is still unknown, it is attributable to a mix of genetic and environmental factors in different proportions, in this study the decision was made to study the occurrence of mental health disorders that are more influenced by environmental factors and traumatic events (i.e., not focusing on the most heritable types of psychiatric conditions) (55, 56). Furthermore, the dispensation of medications used to treat these conditions, namely antidepressants, anxiolytics, and sedatives, which often overlap will also be investigated (54). Psychotherapy, another common treatment, is not the scope of this specific study. Therefore, seven mental health outcomes of interest were included, all of them were binary variables and were coded as 1 if the individual had at least one diagnosis or prescription in the registers. Those were: the presence of mood disorders, anxiety disorders, sleep disorders, prescription (and dispensation) of antidepressants, anxiolytics, and sedatives, and the presence of any of those six (referred to as “any outcome”).

Those were identified using the corresponding ICD-10 for diagnoses and ATC codes for medication. Codes used to identify the outcomes are as follows: F30.0-F39.9 for mood disorders, F41.0-42.9, F93.0-F93.9, F06.4 for anxiety disorders, F51.0-F51.9, G47.0-G47.9 for sleep disorders, N06A for antidepressants, N05B for anxiolytics, and N05C for sedatives.

Statistical analysis

Descriptive statistics were used to analyze the parents’ and children’s characteristics, combined and by group (parent of child with SB or parent of child without SB (or cerebral

palsy)). Multiple logistic regression models were conducted to assess the associations between mental health conditions (i.e., mood disorder, anxiety disorder, or sleep disorder) and medications dispensed (i.e., antidepressants, anxiolytics, sedatives) and having a child with SB. Interaction factors - namely the presence of the same mental health outcome (diagnosis or medication) at baseline, sex of the parent, and parental education - were also included in the models. All crude models were then adjusted for socio-demographic factors (age, sex of the child, sex of the parent, foreign background, parental education, parity) and for presence of the same mental health condition at baseline, i.e., before the birth of the child. The models were subsequently run dividing participants according to their NICHD developmental stage category (54). To assess the impact of different SB-related factors on mental health diagnoses and medications, the group consisting of parents of children with SB were further analyzed as a subset of the sample. Type of SB, level of lesion, presence of hydrocephalus, epilepsy, and intellectual disability were included to assess how they were associated with mood disorders, anxiety disorders, and sleep disorders, and the medications dispensed that were included. This was done both as unadjusted and adjusted regression models and according to the children's developmental stage.

Finally, survival analyses were conducted. Participants were followed over time starting from the year of birth of the child and until the occurrence of the outcome of interest (diagnosis of mental health condition or medication dispensed), death, or end of follow-up. Kaplan-Meier curves and log-rank tests were used to describe survival, followed by Cox Proportional Hazard tests.

Data management and analysis were conducted using Stata (StataCorp. 2021. Stata Statistical Software: Release 17. College Station, TX: StataCorp LLC). Statistical significance was set at $p < 0.05$ and 95% confidence intervals (C.I.).

Ethical considerations

Ethical approval for Swedish projects in the CPNorth program was obtained from the Regional Ethics Board in Lund. Furthermore, national registries have strict rules regarding access, storage, and use of the data (52). The data extracted for this study are confidential and each person is identified with a code. Furthermore, the study focuses on sensitive topics, and caution was used in analyzing the data and interpreting the findings, so as to not stigmatize participants or contribute to increasing misconceptions about disability and mental health. As

many parents were studied for the study, cultural sensitivity was used to not further marginalize portions of the population. Findings are reported at group levels.

Results

Descriptive statistics

A total of 4,897 parents were included in the study, of which 682 had a child with SB and 4,205 did not. The characteristics of the sample are shown in Table 1.1. Demographic characteristics of their children were also extracted and are shown in Table 1.2. Children with SB presented heterogeneous characteristics and symptoms. The majority (65.57%) had open SB and lumbar-sacral level of lesion (63.44%). Furthermore, most children in both groups did not present a diagnosis of hydrocephalus, epilepsy, or intellectual disabilities. Medical characteristics of the children with SB are presented in Table 1.3.

The outcomes of interest (diagnoses of mood disorders, anxiety disorders, sleep disorders, and medications) are presented in Table 2.1. A majority of the parents in both groups did not have any formal diagnosis of mental health conditions registered with an ICD-10 code, neither before nor after the birth of their child. For parents of children with SB, the number of mental health conditions after the birth of their child increased from 13.73% at baseline to 31.36% after the birth, an increase of 128.36%. The corresponding numbers for parents of children who did not have SB were 10.87% at baseline and 27.49% after the birth of the control child, an increase of 152.90%. In total, the most common mental health condition present before birth was anxiety (3.76% of the total sample). The most common medication dispensed prior to the birth of the child was antidepressants (5.82% of the total sample) closely followed by anxiolytics (4.12% of the total sample) while the medications dispensed after the birth of the child were 18.81% for antidepressants, 14.09% for anxiolytics, and 12.56% for sedatives. The prevalence of all three mental health condition categories included in the study, as well as the medications under study, increased after birth, albeit in different proportions, regardless of case-control status. A Pearson's chi2 test was performed to assess differences between the prevalence of mental health disorders in the two groups and showed a statistically significant difference in the prevalence of sedative use before birth and any of the outcomes both before and after the birth of the child, as shown in Table 2.1.

The before-after birth diagnosis of mood disorders, anxiety disorders, or sleep disorders or dispensed medications (divided into four categories according to combinations of the

condition's presence before and after the birth of the child) were investigated. Overall, 20.93% of all parents, 23.12% of parents of children with SB and 20.57% of the controls, developed at least one mental health condition or had medications dispensed after the birth of the child while not having had any of these diagnoses/medications dispensed before the birth of the child. Only 4.17% of the whole sample, 5.49% of parents of children with SB and 3.95% of the controls, had an improvement in their mental health status, meaning that they had a diagnosis of mood-, anxiety, sleep disorder or dispensed medications of antidepressants, anxiolytics, or sedatives before the birth of the child but not after. The rest of the sample had no shift in diagnosed mental health conditions during the study period. A Pearson's chi2 test showed that there was a statistically significant difference in the distribution of the two groups in the four categories for "any outcome" ($p = 0.026$) and for anxiety ($p = 0.046$). Proportions are shown in Table 2.2.

Logistic regression models

Logistic regression models were used to test the associations between exposure and the seven outcomes. Three models were fitted: unadjusted, adjusted for demographic factors, and adjusted for demographic factors and mental health condition at baseline. The results of those analyses are presented below in separate sections according to the outcome.

Mental health diagnoses

1. Mood disorders

In both the unadjusted and adjusted models, the odds of developing mood disorders were not statistically significantly associated with having a child with SB. ORs for the three models are shown in Table 3. The results show a statistically significant interaction between having a child with SB and sex of the parent, education level, and diagnosis of mood disorder at baseline. For mothers, participants with lower education, and with a mood disorder diagnosis at baseline, the ORs of developing a mood disorder after the birth of a child with SB were respectively 1.84 ($p = 0.014$), 2.23 ($p = 0.001$), and 23.82 ($p < 0.001$).

2. Anxiety disorders

In both the unadjusted and adjusted models, the odds of developing anxiety disorders were not statistically significantly associated with having a child with SB. ORs for the three models are shown in Table 3. The results show a statistically significant interaction between having a child with SB and sex of the parent, education level, and diagnosis of an anxiety disorder at baseline. For mothers, participants with lower education, and with an anxiety disorder

diagnosis at baseline, the ORs of developing an anxiety disorder after the birth of a child with SB were respectively 2.60, 2.76, and 8.39 ($p < 0.001$ for all interactions).

3. Sleep disorders

In both the unadjusted and adjusted models, the odds of developing sleep disorders were not significantly associated with having a child with SB. ORs for the three models are shown in Table 3. The results did not show a statistically significant interaction between having a child with SB and sex of the parent, education level, or diagnosis of sleep disorders at baseline.

Medications dispensed

4. Antidepressants

In both the unadjusted and adjusted models, the odds of being prescribed antidepressants were not statistically significantly associated with having a child with SB. ORs for the three models are shown in Table 4. The results show a statistically significant interaction between having a child with SB and sex of the parent, education level, and prescription of antidepressants at baseline. For mothers, participants with lower education, and with a prescription of antidepressants at baseline, the ORs of being prescribed an antidepressant after the birth of a child with SB were respectively 2.21, 2.01, and 8.55 ($p < 0.001$).

5. Anxiolytics

In both the unadjusted and adjusted models, the odds of being prescribed anxiolytics were not significantly associated with having a child with SB. ORs for the three models are shown in Table 4. The results show a statistically significant interaction between having a child with SB and sex of the parent, education level, and prescription of anxiolytics at baseline. For mothers, participants with lower education, and with a prescription of anxiolytics at baseline, the ORs of being prescribed an anxiolytic after the birth of a child with SB were respectively 1.87, 1.59, and 4.52 ($p < 0.001$).

6. Sedatives

In both the unadjusted and adjusted models, the odds of being prescribed sedatives were not significantly associated with having a child with SB. ORs for the three models are shown in Table 4. The results show a statistically significant interaction between having a child with SB and sex of the parent, education level, and prescription of sedatives at baseline. For mothers, participants with lower education, and with a prescription of sedatives at baseline, the ORs of being prescribed a sedative after the birth of a child with SB were respectively 1.64 ($p = 0.001$), 1.52 ($p < 0.001$), and 6.58 ($p < 0.001$).

7. Any of the outcomes

In the unadjusted model, the odds of developing mood disorders, anxiety disorders, sleep disorders, and/or being prescribed one of the related medications in parents of children with SB was higher compared to that of parents of children who did not have SB (ORs = 1.20, p= 0.036). However, after adjusting for demographic variables and mood disorder, anxiety disorder, sleep disorder, and related prescriptions at baseline the association between having a child with SB and the outcome became statistically non-significant. ORs for the three models are shown in Table 5. Furthermore, interactions between having a child with SB and diagnosis at baseline, sex of the parent, and education levels were assessed. The results show a statistically significant interaction between having a child with SB and all the interaction factors. For women, participants with lower education, and with conditions at baseline, the ORs of developing any of the outcomes studied after the birth of a child with SB were 2.27, 2.06, and 5.00 respectively ($p < 0.001$).

Next, we analyzed parental mental health diagnoses and the three types of dispensed medications for these conditions by the developmental stage of all children. Results for the unadjusted models can be found in Appendix 1. Only some of the statistically significant associations were retained when these models were adjusted for all demographics and mental health conditions at baseline. Those were antidepressants in infancy (OR = 0.80, p = 0.049), early childhood (OR = 0.77, p = 0.044), and middle childhood (OR = 0.60, $p < 0.001$), sedatives (OR = 0.46, $p < 0.001$) in middle childhood, and any of the outcomes (OR = 0.76, p = 0.012) in middle childhood. After adjusting the models, some new associations between having a child with SB and mental health outcomes were found, specifically mood disorders (OR = 0.64, p = 0.050) in early childhood, anxiolytics (OR = 0.71, p = 0.011) in middle childhood, and sedatives (OR = 0.73, p = 0.037) in early childhood. There was not enough data to conduct the adjusted model for early adolescents.

Furthermore, we tested the associations between SB-related factors, namely type of SB, level of lesion, presence of hydrocephalus, epilepsy, and intellectual disability, and mental health outcomes in parents of children with SB only. Foreign background and the parents' initial health status, including mental health conditions and medications dispensed, were included as control variables for these outcomes. The only factor with a statistically significant association was intellectual disability, which was associated with higher odds of sleep disorders among parents of children with SB (OR = 5.09, p = 0.046). The analysis was also

conducted by the developmental stage of the child. Having a mental health condition at baseline was associated with all diagnoses ($p < 0.001$). Certain SB-specific factors were seen to be associated with the mental health outcomes in parents of children with SB. Mood disorders (OR = 0.64, p = 0.031) and anxiety disorders (OR = 0.66, p = 0.019) were associated with hydrocephalus in middle childhood. Sleep disorders were associated with intellectual disability in infancy (OR = 5.08, p = 0.005), toddlers (OR = 4.62, p = 0.008), and early childhood (OR = 4.72, p = 0.008), and with hydrocephalus in early (OR = 0.30, p = 0.033) and middle childhood (OR = 0.30, p = 0.001). Antidepressant use was associated with intellectual disability (OR = 0.53, $p < 0.001$) and epilepsy (OR = 1.70, p = 0.037) in middle childhood. Anxiolytic use was associated with intellectual disability (OR = 0.69, p = 0.047), and hydrocephalus (OR = 0.77, p = 0.044) in middle childhood. Sedative use was associated with hydrocephalus (OR = 0.74, p = 0.019) and level of lesion (OR = 1.16, p = 0.001) in middle childhood. Finally, the prevalence of any of the outcomes studied was associated with intellectual disability (OR = 0.72, p = 0.017) and epilepsy (OR = 1.62, p = 0.044) in middle childhood. Complete observations in early adolescence were too few to produce reliable finding.

Survival analysis

Kaplan Meier survival models were produced for each outcome and are shown in Figure 1. Based on the log-rank test conducted on all the outcomes, there is insufficient evidence to reject the null hypothesis, meaning that there is no difference in survival between cases and controls. The same conclusion was supported by the Cox proportional hazard tests.

Discussion

In this study, we assessed the associations between mood disorders, anxiety disorders, sleep disorders, and three different kinds of dispensed medications commonly used to treat the mental health disorders under study and having a child with SB. We also assessed the likelihood of having any of the six outcomes under study and having a child with SB. Furthermore, because the associations with the seven outcome variables might differ based on disability-specific factors of the child, we also included some of the most important SB-related variables. As parental mental health impacts the well-being of both the parents and their children, it is important to understand how different factors can affect these relationships.

In concordance with much of the current literature, the unadjusted results showed associations between having a child with SB and for the parents to develop mental health problems or to be more likely to be dispensed medications commonly used to treat mood-, anxiety-, or sleep disorders. However, after adjusting the models for possible confounders and effect modifiers, these associations were no longer statistically significant. Importantly, when looking at the specific mental health outcomes (i.e., mood disorders, anxiety disorders, or sleep disorders), no statistically significant associations between any of these diagnoses and having a child with SB were found. This stands in stark contrast with the majority of the current literature (23, 50), which generally shows an increased risk of depression and anxiety in parents of children with SB. Nevertheless, studies have shown that there is a substantial variability in the way children's health problems influence mental health conditions in parents, and many of these studies report on self-reported depressive symptomatology, anxiety, or sleep problems rather than clinical mental health conditions diagnosed by a healthcare professional as was the case in the current study (57).

Our findings suggest that the extent to which having a child with SB is associated with parental mental health outcomes is moderate by several factors. The well-established association between being female and reporting more adverse mental health outcome (58) is present also in parents of children with SB in our study. This association is observed in all outcomes except for sleep disorders, and it suggests that mental health in parents (mothers) of children with SB follows the same patterns that are present in the general population, where mothers are more likely to experience mental health conditions. Furthermore, in families of children with chronic illnesses, traditional family roles are more likely to be followed as they facilitate the handling of health-related demands (e.g., numerous different kinds of healthcare appointments, fitting of assistive technology if needed). Possibly, the fact that the idea of parental competency tends to be more embedded in traditional women's roles, mothers might be more exposed to risk factors of parenting distress and may feel less competent than fathers (57). If it were to be the case that mothers, in general, do spend more time with their child, it is also possible that they are more keenly aware of SB-related concerns, such as dangerous side effects and the presence of executive dysfunctions. However, these suggestions warrant further research. Level of education also moderates the association between having a child with SB and the presence of most of the mental health outcomes and the dispensed medications under study. Both mothers and fathers with an education level of attending mandatory school or less (i.e., lower level of education) are more likely to develop all mental

health outcomes under study except for sleep disorders. This is in line with existing literature on the topic, as least as far as for mothers (59). Furthermore, the presence of one of these mental health conditions before the birth of the child is the most predictive risk factor for experiencing mood-, anxiety-, sleep disorders or being dispensed antidepressants, anxiolytics, or sedatives after the birth of the child. This is in accordance with evidence suggesting that having a psychological/psychiatric condition is the strongest predictor for risk of developing additional mental health conditions in the future (60). Even in parents of children with SB, pre-existing psychological well-being can predict the way they perceive their child's vulnerability and their adjustment to having a child with a disability, more than the child's health condition per se (22). Overall, parents' personalities and psychosocial traits can influence their parenting style and their resilience towards traumatic events and should be better explored in future research (57).

Parity also seems to be associated with a slightly higher risk of being prescribed certain medications, which seems to be different from what has been noted in other study populations, where having more children has been positively associated with maternal mental health (61). This may be due to the higher pressure of having to take care of a child with a disability that tends to be quite time-consuming with many medical appointments, possible discussions with school, assessing for pressure sores, and carrying out CIC throughout the day and of other children at the same time.

When analyzing the associations between having a child with SB and the outcomes of interest according to the child's developmental stages, there were more substantial differences between parents of children with SB and their peers. After adjusting for demographics and the presence of mental health conditions at baseline, some statistically significant associations remained significant but with parents of children with SB being less vulnerable to mental health outcomes than their counterparts during certain developmental stages of their children. Specifically, those were mood disorders during early childhood (2-5 years), prescription of antidepressants during infancy (up to 1 year), early and middle childhood, prescription of anxiolytics during middle childhood, prescription of sedatives during early and middle childhood, and presence of any of the mental health conditions during the child's middle childhood (6-11 years). These findings likely depend on multiple factors. A study by Malm-Buatsi et al. has shown that having younger children with SB is associated with higher levels of parental stress and an increased perception of the child's vulnerability (39). Even though

most parents of children in the Western world are aware that their child will be born with SB, stress and negative emotions can be highly prevalent during the early years as parents adjust to caring for a child with complex medical needs and how to care for the child (62). However, compared to parents of children not born with a complex disability, in some cases there might also be an element of a sense of relief – possibly a gratefulness – that perhaps things are progressing better than anticipated. Depending on severity and comorbidities, children with SB often undergo many surgeries, and it is possible that these cluster around certain developmental stages, which can be both stressful but also offer a sense of relief if the surgery is deemed successful. This might partially help explain the fact that the lower risk is present when children are in middle childhood. These are hypotheses that warrant further study. On the other hand, a 2017 study by Driscoll et al. (22) reported higher levels of perceived child vulnerability in parents of older children. Parents may expect their child to become more independent despite their disability and may become stressed if these expectations of "typical" milestones of development are not met (22). This is in contrast to what was found in this study, and it may need to be further explored in future research. The fact that there are odds ratios below 1 for having a child with SB and being diagnosed with parental mental health conditions during the child's older years (i.e., they are less likely to develop the outcomes), could also be attributable to the fact that late childhood and early adolescence represent an important transition period, where "typically" parents and children usually experience changes in their relations and mood (63). This tendency may be lower for children with SB as children might not go through the "typical" independence phase, or oftentimes at a different rate, than other children go through, and they remain more dependent on their parents. Furthermore, despite studies having highlighted the difficulties during the child-to-adult transition period for children with SB, in which follow-up and continuity of care are associated with multiple challenges and the heightened pressure placed on the family, our results would show that it does not cause increased levels of mental health outcomes (64, 65). This may be due to parents becoming accustomed to their child's disability and to a better ability to navigate the healthcare system. It could also come as a relief not to have to attend habilitation clinics or multidisciplinary clinics the same way as during early childhood.

Differences in parental mental health outcomes were also observed within the SB group, likely due to the fact that the severity of SB can differ substantially as can the co-occurrence of comorbid conditions and secondary conditions. In the complete sample, statistical

significance is present only between intellectual disability of the child and mental health outcomes of the parents. Meanwhile, analyses grouped according to the child's developmental stage showed greater differences. Most of the associations between SB-related conditions and parental mental health outcomes were seen during the child's middle childhood and could be connected, as discussed previously, to challenges in the children's milestones and independence (44). In children with more comorbid conditions, such as epilepsy, or who were reported to have more indicators of severe SB, such as a higher level of lesion, some of the parental mental health outcomes were more likely to occur. In contrast, some of the more severe conditions associated with SB such as hydrocephalus, seem to be associated with a lower risk of developing mental health outcomes. For example, hydrocephalus was associated with a lower prevalence of sleep disorders in early childhood, and a lower prevalence of mood disorders, anxiety disorders, sleep disorders, anxiolytic use, and sedative use in middle childhood. A similar finding was noted with intellectual disability which, despite having been associated with sleep disorders in infancy, toddlers, and early childhood, decreased the risk of anxiolytic use in middle childhood. There are several explanations for these findings. First, these parents may have developed higher resilience earlier in their child's life as they had to manage severe health conditions and potential risks for their children's lives and well-being (16). This might make them less prone to developing mental health conditions during the transition phase. Higher severity of the children's conditions may also eliminate the difficulties of the transition phase as it could be easier to guarantee continuity of care to those with more severe forms of SB. These children may also be less likely to become completely independent, meaning that the worry connected to this developmental stage could be delayed.

Finally, foreign background was associated with a lower risk of mood disorders in infancy and childhood, antidepressant use in infancy, and anxiety in middle childhood but as people from foreign backgrounds can experience more challenges in accessing healthcare and navigating the system, this type of experience, perhaps together with better family support systems or religious beliefs, could also contribute to resilience. Nevertheless, higher stigma could be present in other cultures and make it more difficult to report mental health challenges. People with foreign backgrounds could also have more trouble getting access to this type of healthcare and therefore to an official diagnosis and/or prescription (66). Finally, our study did not show any significant difference in the timing of the occurrence of mental health conditions.

Overall, this study presents some important differences from what is seen in the existing literature. as no major differences in mental health outcomes between parents of children with SB and without were found. These findings are also coherent with the disability paradox – which can be summarized as the notion that many people with serious disabilities still report a good quality of life – despite the external idea that quality of life with disabilities is necessarily inferior (67). In other words, people who do not have disabilities assume that those who do must have worse qualities of life. This might potentially be applied to parents as well. Nevertheless, as living with a disability might be posed with many challenges and barriers for children and parents alike, it is positive and reassuring that parents of children with SB seem to report levels of mental health illness similar to their counterparts who do not have children with SB or cerebral palsy. It is still possible that they are more likely to report depressive symptomatology, anxiety, and sleeping difficulties. However, these symptoms do not seem to be of such a severe level that they have been diagnosed by a health professional or received pharmacological treatment for it. Context is also relevant as, in the case of a universal healthcare system like the one in Sweden, it may be somewhat less challenging to provide care for a child with disability, as this may not necessarily be associated with a big economic burden and stressors related to financial burden. In Sweden, people with disabilities and their families can receive social benefits, personal assistance, and support, which decreases the burden of care (31). This can help explain the difference with the current literature, as all studies reviewed, to the author's knowledge, come from countries outside of Scandinavia and with different healthcare and welfare systems. Previous studies on mental health in parents of children with SB also used different methodologies, with none using clinical register-based information. Using self-reported information or surveys can result in different results compared to what was found in the present study. Finally, it is important to differentiate between statistically significant and clinically significant results, with certain findings being statistically significant but not presenting clinical significance, due to large samples, repeated analyses, and presence of systematic errors or bias.

On the other hand, these findings can underestimate the real presence of mental health issues. While this is true for both groups, it may present more challenges for parents of children with SB, as they already have the burden of care for their children, thus making it harder to get a diagnosis and/or treatment for themselves. Therefore, this study should be generalized with caution, especially in contexts that are substantially different from Sweden.

Strengths and Limitations

This study presents several limitations. First, the National Patient Register does not include data from primary care. Furthermore, it collects to record population statistics and was not specifically designed to be used in research. Specifically, information on SB or mental health diagnoses may be less accurate than it would be if we had gathered the data for research purposes. However, doing so would mean that the data would most likely not be population-based. Furthermore, many different providers and staff are involved in registering data in national registers, and it is possible that the criteria used by different professionals in different parts of Sweden may differ and that data may not be homogenous. This can be reflected in both records about SB and in mental health disorders, which can also be classified with different levels of accuracy, depending on the experience of the reporter and the type of test or method used to assess the condition. As mental health outcomes in parents may vary based on the severity of SB, our findings may not be accurate if records do not report the severity or level of SB. A similar issue can also happen if a person moves to Sweden after receiving a previous diagnosis in another country: if someone was diagnosed with a mental health condition before but only later diagnoses made in Sweden were included in the analyses, findings may be biased. While this applies to both groups, the proportion of parents of children with disabilities who have a foreign background is higher.

Moreover, our outcomes only include official ICD-10 diagnoses and ACT codes for medication and do not assess subclinical mental health conditions experienced by the parents. However, that was not the purpose of the study, and is not a limitation per se. Furthermore, we only studied pharmacological interventions and we did not have data on psychotherapy. It is quite possible that the first line of treatment is cognitive behavioral therapy or some other type of therapy, however, that information was not available to us.

Furthermore, antidepressants, anxiolytics, and sedatives may be used more broadly to treat other types of conditions and may not be indicative of mood-, anxiety-, or sleep disorders. For instance, benzodiazepines can be used both as a sedative and an anxiolytic. The ICD and ACT codes for the outcomes were divided into broad groups; therefore, we do not have information on the specific sub-diagnosis the individual may have received. In other words, we could not decipher the exact condition that the person presented, and how severe it was. For example, anxiety disorders include a range of conditions such as generalized anxiety disorder and post-traumatic-stress-disorder, which, despite being in the same group, have very different

etiologies and symptoms. Furthermore, as some records only included an unspecified SB diagnosis, specific levels of lesions could not be assessed in all children with SB.

Nevertheless, this study has several strengths. As it was a register-based population study, it was possible to include all the individuals with SB present in Sweden who met the inclusion criteria. This reduces the risk of selection bias and makes the results more generalizable. It can also increase variability and heterogeneity in the group, both in outcomes and risk factors. The study cohort spanned over a long time period, which may make the results more reliable. Finally, our study presents unique findings that can contribute to knowledge on the topic given that medically diagnosed conditions and medications dispensed were studied rather than self-reported data which may be prone to recall bias. The few studies that have focused on mental health in parents of children with SB have mainly focused on mothers while our study includes both mothers and fathers in similar proportions. The study uses diagnoses and prescriptions as outcomes instead of assessing general distress, which may make the results more clinically accurate. Finally, we explored the impact of having a child with SB on sleep disorders and sedatives as well, which, to our knowledge, has not been done before.

Conclusion

Despite showing some associations between having a child with SB and developing adverse parental mental health conditions, our findings are somewhat different from what has been reported in the literature thus far. While this is positive and might be a relief for parents-, or parents to be-, who will care for a child with SB, mental health concerns for parents of children with SB should still not be underestimated. Parents who present certain characteristics who are likely to increase risk, such as mothers, parents with lower education levels, with a mental health condition at baseline, and those having children with more severe SB, must be appropriately monitored and followed up. Further research is needed to understand variations in mental health outcomes in different contexts and understand the impact of different socio-economic and health-related factors. For example, financial aspects may be relevant even in a universal healthcare context like Sweden’s and it may be interesting to explore them in future research. The impact of community and social circles should also be taken into consideration when looking at how to improve the well-being of parents. Support for the children and their families should be provided, when needed, as adverse mental health conditions in parents can influence the quality of life and well-being of both them and the children. Specifically, for children with SB, a deeper understanding of the conditions that

impact mental health and quality of life in parents can inform policymakers on possible prevention strategies. Prevention strategies and support should be provided to children and their families through collaborative relations among parents, services, and children.

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Tables and Figures

Table 1 – Descriptive statistics of the sample

Table 1.1- Parents' demographic information

	Parents of children without SB or CP n = 4,205	**Parents of children with SB n = 692**
Gender		
Male	2,106 (50.08)	347 (50.14)
Female	2,099 (49.92)	345 (49.86)
Age at baseline (mean ± SD)	32.32 ± 5.88	32.09 ± 6.14
Year of birth		
Born before 1964	222 (5.28)	31 (4.34)
Born between 1965-1980	3,018 (71.77)	496 (71.68)
Born between 1981-1999	965 (22.95)	165 (23.84)
Area of birth*		
Sweden	3,444 (81.90)	528 (76.30)
Rest of Europe	313 (7.44)	59 (8.52)
Africa	107 (2.54)	37 (5.35)
Asia and Oceania	275 (6.54)	54 (7.80)
North America	28 (0.67)	3 (0.43)
South America	38 (0.90)	10 (1.45)
Missing	0	1 (0.14)
Education*		
Mandatory or less	521 (12.39)	130 (18.79)
Secondary and higher	3,661 (87.06)	525 (75.87)
Missing [a]	23 (0.55)	37 (5.35)
Parity of the mother*		
First child (parity = 0)	1,196 (28.44)	177 (25.58)
Younger child (parity > 1)	1,616 (38.43)	239 (34.54)
Missing [a]	1,393 (33.13)	276 (39.88)

[a] *missing data were reported when present*

** statistically significant differences ($p < 0.05$) in proportion between the two groups*

Table 1.2 - children's demographic information

Variable	Children without SB n = 4,205	Children with SB n = 692
Gender		
Male	2,020 (48.04)	336 (48.55)
Female	2,185 (51.96)	356 (51.45)
Year of birth		
2003	538 (12.79)	98 (14.16)
2004	433 (10.30)	71 (10.26)
2005	349 (8.30)	54 (7.80)
2006	411 (9.77)	71 (10.26)
2007	287 (6.83)	51 (7.37)
2008	407 (9.68)	68 (9.83)
2009	397 (9.44)	62 (8.96)
2010	268 (6.37)	44 (6.36)
2011	274 (6.52)	46 (6.65)
2012	316 (7.51)	52 (7.51)
2013	292 (6.94)	38 (5.49)
2014	233 (5.54)	37 (5.35)
Foreign background*		
Yes	502 (11.94)	149 (21.53)
No	3,703 (88.06)	543 (78.47)
Area of birth*		
Sweden	4,194 (99.74)	654 (94.51)
Europe	5 (0.12)	8 (1.17)
Africa	0	1 (0.14)
Asia	4 (0.10)	29 (4.19)
South America	2 (0.05)	0
Parental education*		
Mandatory or less	185 (4.40)	51 (7.37)
Secondary and higher	3946 (94.27)	561 (81.08)
Missing [a]	56 (1.33)	50 (7.23)

[a] *missing data were reported when present*

** statistically significant differences ($p < 0.05$) in proportion between the two groups*

Table 1.3 - characteristics of individuals with SB

Variable	
Type of SB	
Open	139 (65.57)
Covered	63 (29.72)
Suspected	10 (4.72)
Missing	480 (69.36)
Specific SB diagnosis	
Q05 - Spina bifida	18 (2.60)
Q05.0 Cervical SB with hydrocephalus	11 (1.59)
Q05.1 Thoracic SB with hydrocephalus	43 (6.21)
Q05.2 Lumbar SB with hydrocephalus	163 (23.55)
Q05.3 Sacral SB with hydrocephalus	31 (4.48)
Q05.4 Unspecified SB with hydrocephalus	17 (2.46)
Q05.5 Cervical SB without hydrocephalus	6 (0.87)
Q05.6 Thoracic SB without hydrocephalus	23 (3.32)
Q05.7 Lumbar SB without hydrocephalus	119 (17.20)
Q05.8 Sacral SB without hydrocephalus	126 (18.21)
Q05.9 Unspecified SB	135 (19.51)
Presence of hydrocephalus	
Yes	317 (45.81)
No	375 (54.19)
Shunt operation (out of people who have hydrocephalus)	
Yes	187 (58.99)
No	130 (41.01)
Lifetime epilepsy	
Yes	36 (5.20)
No	656 (94.80)
Intellectual disability	
No disability	655 (94.65)
Mild	27 (3.90)
Moderate	6 (0.87)

Severe	4 (0.58)

Table 2 – Mental health outcomes in the sample

Table 2.1 – parent's mental health outcomes diagnoses and medications

Variable	**Parents of children without SB or CP n = 4,205**	**Parents of children with SB n =692**
Mood disorders (before birth)		
Mood disorder	85 (2.02)	20 (2.89)
No mood disorder	4,120 (97.98)	672 (97.11)
Mood disorders (after birth)		
Mood disorder	207 (4.92)	37 (5.35)
No mood disorder	3,998 (95.08)	655 (94.65)
Anxiety disorders (before birth)		
Anxiety disorder	149 (3.54)	35 (5.06)
No anxiety disorder	4,056 (96.46)	657 (94.94)
Anxiety disorders (after birth)		
Anxiety disorder	270 (6.42)	56 (8.09)
No anxiety disorder	3,935 (93.58)	636 (91.91)
Sleep disorders (before birth)		
Sleep disorder	26 (0.62)	7 (1.01)
No sleep disorder	4,179 (99.38)	685 (98.99)
Sleep disorders (after birth)		
Sleep disorder	79 (1.88)	17 (2.46)
No sleep disorder	4,126 (98.12)	675 (97.54)
Antidepressants (before birth)		
Antidepressant use	235 (5.59)	50 (7.23)
No antidepressant use	3,970 (94.41)	642 (92.77)
Antidepressants (after birth)		
Antidepressant use	775 (18.43)	146 (21.10)
No antidepressant use	3,430 (81.57)	546 (78.90)
Anxiolytics (before birth)		
Anxiolytic use	167 (3.97)	35 (5.06)

No anxyolitic use	4,038 (96.03)	657 (94.94)
Anxiolytics (after birth)		
Anxiolytic use	588 (13.98)	102 (14.74)
No anxyolitic use	3,617 (86.02)	590 (85.26)
Sedatives (before birth) *		
Sedative use	162 (3.85)	39 (5.64)
No sedative use	4,043 (96.15)	653 (94.36)
Sedatives (after birth)		
Sedative use	521 (12.39)	94 (13.58)
No sedative use	3,684 (87.61)	598 (86.42)
Any mental health condition (before birth) *		
Any mental health outcome	457 (10.87)	95 (13.73)
No mental health outcome	3,748 (89.13)	597 (86.27)
Any mental health condition (after birth) *		
Any mental health outcome	1,156 (27.49)	217 (31.36)
No mental health outcome	3,049 (72.51)	475 (68.64)

** significant difference ($p < 0.05$) in proportion between exposed and unexposed*

Table 2.2 – comparison between mental health outcomes before and after the birth of the child

Variable	**Parents of children without SB or CP n = 4,205**	**Parents of children with SB n =692**
Mood disorders		
No – No	3,954 (94.03)	645 (93.21)
No – Yes	166 (3.95)	27 (3.90)
Yes – Yes	41 (0.98)	10 (1.45)
Yes – No	44 (1.05)	10 (1.45)
Anxiety disorders *		
No – No	3,846 (91.46)	612 (88.44)
No – Yes	210 (4.99)	45 (6.50)
Yes – Yes	60 (1.43)	11 (1.59)

Yes – No	89 (2.12)	24 (3.47)
Sleep disorders		
No – No	4,108 (97.69)	668 (96.53)
No – Yes	71 (1.69)	17 (2.46)
Yes – Yes	8 (0.19)	0
Yes – No	18 (0.43)	7 (1.01)
Antidepressants		
No – No	3,334 (79.29)	527 (76.16)
No – Yes	636 (15.12)	115 (16.62)
Yes – Yes	139 (3.31)	31 (4.48)
Yes – No	96 (1.28)	19 (2.75)
Anxiolytics		
No – No	3,517 (83.64)	567 (81.94)
No – Yes	521 (12.39)	90 (13.01)
Yes – Yes	67 (1.59)	12 (1.73)
Yes – No	100 (2.38)	23 (3.32)
Sedatives		
No – No	3,595 (85.49)	573 (82.80)
No – Yes	448 (10.65)	80 (11.56)
Yes – Yes	73 (1.74)	14 (3.61)
Yes – No	89 (2.21)	25 (3.61)
Any mental health		
outcome*	2,883 (68.56)	437 (63.15)
No – No	865 (20.57)	160 (23.12)
No – Yes	291 (6.92)	57 (8.24)
Yes – Yes	166 (3.95)	38 (5.49)
Yes – No		

** significant difference ($p < 0.05$) in proportion between exposed and unexposed*

Data in this table is presented in four categories:

- *No-no: individuals who did not have a mental health outcome before the birth of the child and did not develop one afterwards.*
- *No-yes: individuals who did not have a mental health outcome before the birth of the child but developed one afterwards.*

- *Yes-yes: individuals who had a mental health outcome before the birth of the child and still had one afterwards.*
- *Yes-no: individuals who had a mental health outcome before the birth of the child and but did not have one afterwards.*

Table 3 – Logistic regression models for mental health diagnoses

The following tables present OR and associated 95% confidence intervals (C.I.) from logistic regression models investigating the association between having a child with SB and presence of mood disorders, anxiety disorder, and sleep disorders. The model includes the exposure variable and adds potential effect modifiers, first socio-demographics and then mental health condition at baseline.

Statistical significance ($p < 0.05$) is shown in bold.

	OR (95 % C.I.)								
	Mood disorders			**Anxiety disorders**			**Sleep disorders**		
	Model 1	**Model 2**	**Model 3**	**Model 1**	**Model 2**	**Model 3**	**Model 1**	**Model 2**	**Model 3**
Child with SB - Yes - No				1.28 (0.95 – 1.73) 1.00	1.34 (0.91 – 1.96) 1.00	1.26 (0.84 – 1.88) 1.00	1.32 (0.77 – 2.24) 1.00	1.51 (0.75 – 3.04) 1.00	1.46 (0.71 – 2.99) 1.00
Age (continuos)	1.18 (0.97 – 1.44)	0.87 (0.65 – 1.16)	0.80 (0.59 – 1.09)		1.00 (0.98 – 1.03)	1.00 (0.97 – 1.03)		1.03 (0.98 – 1.08)	1.01 (0.97 – 1.06)
Sex of the parent - Female - Male	[1]	**1.00**	**1.00**		**1.95 (1.45 – 2.64)** **1.00**	**1.75 (1.28 – 2.39)** **1.00**		0.63 (0.35 – 1.12) 1.00	0.61 (0.34 – 1.10) 1.00
Sex of the child - Female - Male		1.02 (1.00 – 1.03)	1.01 (0.99 – 1.03)		0.89 (0.67 – 1.18) 1.00	0.88 (0.66 – 1.18) 1.00		1.28 (0.75 – 2.19) 1.00	1.37 (0.79 – 2.38) 1.00
Foreign background - Non-Swedish - Swedish		2.24 (1.84 – 2.74)	2.03 (1.64 – 2.50)		0.87 (0.57 – 1.33) 1.00	0.90 (0.57 – 1.39) 1.00		0.84 (0.37 – 1.91) 1.00	0.95 (0.41 – 2.19) 1.00
Parental education - Mandatory or less - Secondary or higher		0.99 (0.82 – 1.19)	**0.97 (0.79 – 1.18)**		**1.75 (1.28 – 2.38)** **1.00**	**1.56 (1.13 – 2.15)** **1.00**		1.59 (0.90 – 2.83) 1.00	1.36 (0.75 – 2.45) 1.00
Parity - One child - More than two children		0.84 (0.63 – 1.12)	0.88 (0.65 – 1.19)		**1.15 (1.02 – 1.30)** **1.00**	1.12 (0.99 – 1.28) 1.00		1.01 (0.79 – 1.29) 1.00	0.99 (0.77 – 1.28) 1.00
Mood disorder at baseline - Yes - No		1.58 (1.29 – 1.94)	**1.57 (1.27 – 1.95)**			**9.21 (6.34 – 13.36)** **1.00**			**21.06 (8.08 – 54.92)** **1.00**

Table 4 – Logistic regression models for dispensed medications

The following tables present OR and associated 95% confidence intervals (C.I.) from logistic regression models investigating the association between having a child with SB and having a prescription for antidepressants, anxiolytics, and sedatives. The model includes the exposure variable and adds potential effect modifiers, first socio-demographics and then mental health condition at baseline.

Statistical significance ($p < 0.05$) is shown in bold.

	OR (95 % C.I.)								
	Prescription od antidepressants			**Prescription of anxiolytics**			**Prescription of sedatives**		
	Model 1	**Model 2**	**Model 3**	**Model 1**	**Model 2**	**Model 3**	**Model 1**	**Model 2**	**Model 3**
Child with SB Yes No	 1.18 (0.97 – 1.44) 1.00	 0.87 (0.65 – 1.16) 1.00	 0.80 (0.59 – 1.09) 1.00	 1.06 (0.85 – 1.34) 1.00	 0.90 (0.66 – 1.24) 1.00	 0.86 (0.62 – 1.20) 1.00	 1.11 (0.88 – 1.41) 1.00	 0.89 (0.63 – 1.26) 1.00	 0.82 (0.57 – 1-18) 1.00
Age (continuos)		1.02 (1.00 – 1.03)	1.01 (0.99 – 1.03)		1.01 (0.99 – 1.03)	1.01 (0.99 – 1.03)		**1.03 (1.01 – 1.05)**	**1.02 (1.00 – 1.05)**
Sex of the parent - Female - Male	 	 **2.24 (1.84 – 2.74)** **1.00**	 **2.03 (1.64 – 2.50)** **1.00**	 	 **1.80 (1.44 – 2.25)** **1.00**	 **1.76 (1.40 – 2.21)** **1.00**	 	 **1.79 (1.41 – 2.27)** **1.00**	 **1.75 (1.37 – 2.23)** **1.00**
Sex of the child - Female - Male	 	 0.99 (0.82 – 1.19) 1.00	 0.97 (0.79 – 1.18) 1.00	 	 1.01 (0.82 – 1.25) 1.00	 0.99 (0.80 – 1.23) 1.00	 	 0.90 (0.71 – 1.12) 1.00	 0.90 (0.72 – 1.14) 1.00
Foreign background - Non-Swedish - Swedish	 	 0.84 (0.63 – 1.12) 1.00	 0.88 (0.65 – 1.19) 1.00	 	 1.00 (0.73 – 1.36) 1.00	 1.03 (0.75 – 1.42) 1.00	 	 0.87 (0.61 – 1.23) 1.00	 0.84 (0.58 – 1.20) 1.00
Parental education - Mandatory or less - Secondary or higher	 	 **1.58 (1.29 – 1.94)** **1.00**	 **1.57 (1.27 – 1.95)** **1.00**	 	 **1.78 (1.42 – 2.24)** **1.00**	 **1.81 (1.43 – 2.29)** **1.00**	 	 **1.82 (1.43 – 2.32)** **1.00**	 **1.80 (1.40 – 2.31)** **1.00**
Parity - One child - More than two children	 	 **1.12 (1.02 – 1.22)** **1.00**	 **1.12 (1.02 – 1-23)** **1.00**	 	 **1.13 (1.02 – 1.24)** **1.00**	 **1.11 (1.01 – 1.22)** **1.00**	 	 1.02 (0.92 – 1.13) 1.00	 1.02 (0.91 – 1.14) 1.00
Mood disorder at baseline - Yes - No	 	 	 **8.56 (6.53 – 11.22)** **1.00**	 	 	 **5.07 (3.69 – 9.96)** **1.00**	 	 	 **7.38 (5.35 – 10.16)** **1.00**

Table 5 – logistic regression model for any mental health outcome

The following table presents OR and associated 95% confidence intervals (C.I.) from a logistic regression model investigating the association between having a child with SB and having any of the mental health outcomes of interest. The model includes the exposure variable and adds potential effect modifiers, first socio-demographics and then mental health condition at baseline.

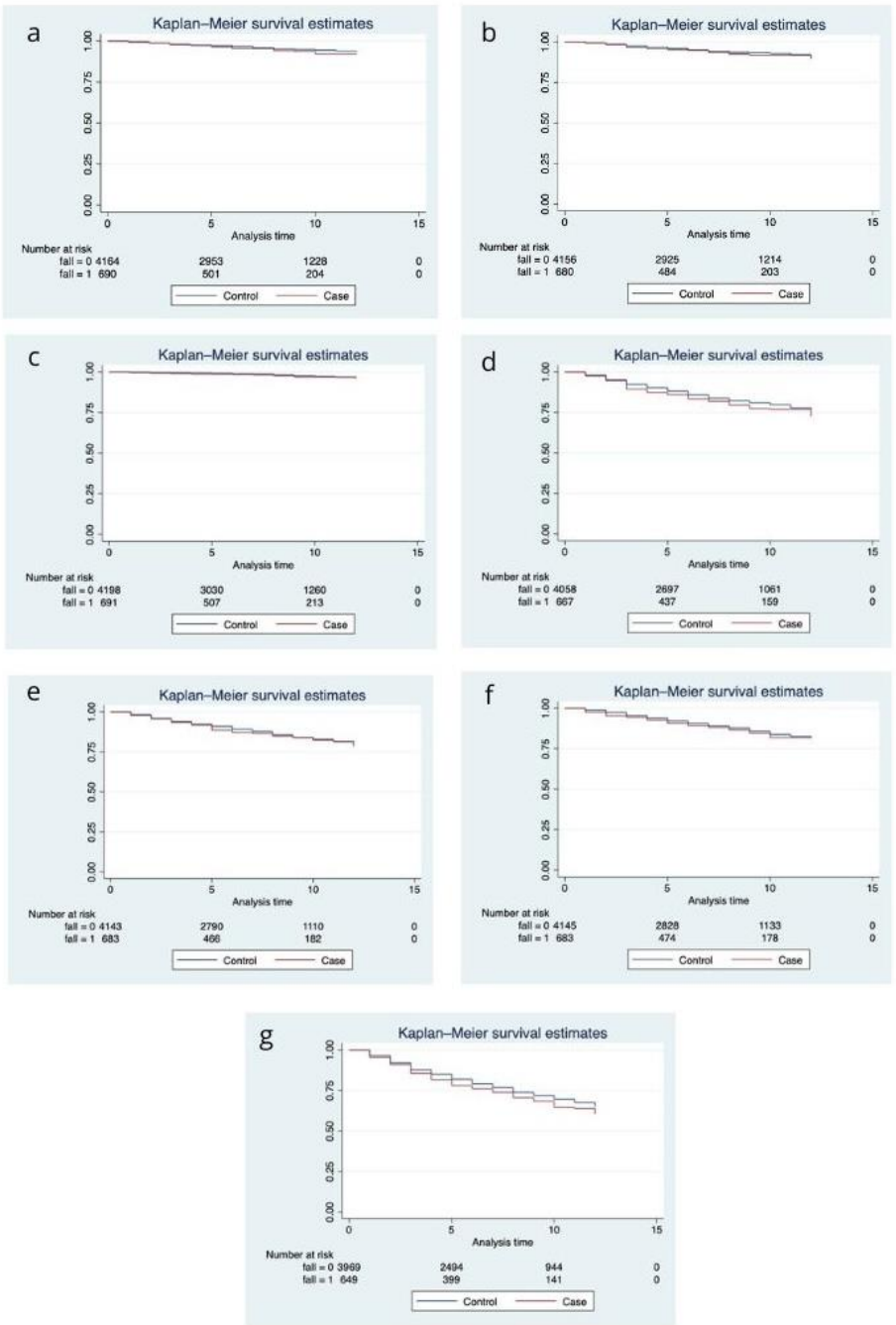
Statistical significance is shown in bold ($p < 0.05$).

	OR (95 % C.I.)		
	Model 1	**Model 2**	**Model 3**
Child with SB - Yes - No	**1.20 (1.01 – 1.43)** 1.00	1.02 (0.80 – 1.30) 1.00	0.98 (0.77 – 1.26) 1.00
Age (continuous)		1.02 (1.00 - 1.03)	1.02 (1.00 – 1.03)
Sex of the parent - Female - Male		**1.92 (1.62 – 2.28)** **1.00**	**1.87 (1.57 – 2.22)** **1.00**
Sex of the child - Female - Male		0.98 (0.84 – 1.15) 1.00	0.97 (0.82 – 1.14) 1.00
Foreign background - Non-Swedish - Swedish		0.84 (0.65 – 1.07) 1.00	0.85 (0.66 – 1.09) 1.00
Parental education - Mandatory or less - Secondary or higher		**1.63 (1.36 – 1.94)** **1.00**	**1.58 (1.32 – 1.89)** **1.00**
Parity - One child - More than one child		1.08 (1.00 – 1.17) 1.00	1.07 (0.99 – 1.16) 1.00

At least one condition at baseline - Yes - No			 **9.33 (5.60 – 15.53)** **1.00**

Figure 1

Kaplan-Meier survival models to time of event, i.e. instance of mental health condition, medication dispensed or death/end of follow-up in parents of children with SB and parents of children without SB or CP.



The table shows the following: a. mood disorders, b. anxiety, c. sleep disorders, d. antidepressants, e. anxiolytics, f. sedatives, g. any mental health outcome.

Appendix

Appendix 1

In the unadjusted models, several associations were found. Regarding mental health diagnoses, having a child with SB was associated with mood disorders during middle childhood (OR = 1.25, p = 0.030), and anxiety in infancy (OR = 1.28, p = 0.021), early childhood (OR = 1.30, p = 0.022), and middle childhood (OR = 1.19, p = 0.047). For medications, having a child with SB was associated with prescription of antidepressants in infancy (OR = 1.19, p = 0.018), toddlers (OR = 1.20, p = 0.014), early childhood (OR = 1.24, p = 0.007), middle childhood (OR = 1.31, p < 0.001), and early adolescence (OR = 1.95, p = 0.006). Prescription of sedatives was associated with having a child with SB in early adolescence (OR = 1.68, p = 0.048). An association between having at least one of the outcomes under study and having a child with SB was found in infancy (OR = 1.21, p = 0.003), toddlers (OR = 1.21, p = 0.003), early childhood (OR = 1.26, p = 0.001), middle childhood (OR = 1.29, p < 0.001), and early adolescence (OR = 1.77, p = 0.011).

Popular science summary

This study investigated whether having a child with Spina bifida (SB), a birth defect that can lead to various levels of disability, is associated with a higher presence of mood, anxiety, and sleep problems in parents. Using national data, we identified parents of children with SB and a group of parents of children without the condition. Our results show that, in general, parents of children with SB didn't have a higher risk. However, they had higher odds of developing mental health problems during certain developmental stages of their children, like middle childhood. Additionally, the severity of SB and its symptoms played a role in mental health outcomes. Parental mental health is critical as it can affect the well-being of both the parents and children and can decrease their quality of life. These findings highlight the need for a strong support system that helps parents of children with SB cope with the difficulties of having a child with disabilities, especially in certain stages of their child's life. By understanding the challenges these parents face and the specific factors involved in them, we can aim to develop better prevention strategies and support networks to improve the quality of life for them and their children.

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