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Risk of divorce and likelihood of having additional children among families with children with spina bifida: A Swedish population-based longitudinal register study

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ABSTRACT

Background: To raise a child with disability might present challenges and affect the functioning of the family unit. In this study, the risk of divorce for parents of children with spina bifida and the probability of having additional children were analysed.
Methods: Longitudinal, matched case-control, data between 2004 and 2014 from multiple linked Swedish Population Registers were analysed using Cox proportional hazard models with interval censoring.
Results: The results showed a reduced risk of divorce among parents of children with spina bifida compared to parents of children who did not have spina bifida. Some indications of heterogeneous effects were noted; a stronger protective association was noted among parents who are married compared to cohabiting, have higher education, and where the mother is older at the birth of the child with spina bifida (34 + years). No association was found on having additional children after the birth of a child with spina bifida.
Conclusion: The results should be understood in the Swedish context, which is known for its comprehensive welfare system. Future research should investigate the mechanisms behind these results.
What this paper adds?: This study contributes to the field by utilizing population-based register data, which is rare for spina bifida research. Comparison to prior studies indicates that there is substantial heterogeneity across disabilities indicating that while some research can be conducted at the broad disability level, in certain contexts it might be inappropriate to study disability as a group or generalising the results from one disability to the next.

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1. Introduction

Children with disabilities or special healthcare needs often require additional care and attention compared to typically developing children, a responsibility that normally falls on the parents. The added burden can lead to negative consequences for the parents, for example in terms of emotional and financial stress (Bourke-Taylor et al., 2014; Gallagher & Hannigan, 2014). Parents of children with spina bifida have been reported to have increased levels of psychological distress and family dysfunction as well as lower quality of life (Holmbeck & Devine, 2010; Valença et al., 2012; Vermaes et al., 2005). However, families have also been noted to manifest increased family resilience (Holmbeck & Devine, 2010; Ridosh et al., 2016). Still, there is a concern that family dysfunction can lead to an increased risk of divorce and, among those who do not divorce, a reduced likelihood of having additional children.

Divorce is associated with a number of adverse effects, especially for women who tend to be the primary caregiver and their children (Bourke-Taylor et al., 2014; Huff & Hartenstein, 2020), in that it increases financial insecurity and reduces socioeconomic status. This, in turn, is associated with negative long-term outcomes for the children in terms of health, education, and labour market achievements (Demir-Dagdas et al., 2018; Holmbeck & Devine, 2010; Jacobs et al., 2001). One concern is therefore that having a child with disability could lead to a dual challenge where consequences are associated with both the disability itself and adverse family outcomes during childhood (Loft, 2011).

Spina bifida is a congenital neural tube defect that often results in complex health issues that include neurological (e.g., hydrocephalus), urological (e.g., neurogenic bladder), and musculoskeletal (e.g., hip dislocations) health issues (Dicianno et al., 2008). Secondary conditions such as pressure injuries and obesity are common and can influence the quality of life of the children and their parents (Lindquist et al., 2022). Additionally, at a group level, individuals with the most severe and the most frequent type of spina bifida, myelomeningocele, often display a cognitive phenotype that includes executive dysfunction. This is not the same as intellectual disability and is signified by difficulties related to planning, initiating, and executing tasks (Zabel et al., 2011).

The purpose of this study was to investigate how having a child with spina bifida affects family formation in terms of the risk of divorce and the likelihood of having additional children, and if the observed relationships varied across parental characteristics. We hypothesised that parents of children with spina bifida have an increased risk of divorce and a reduced likelihood of having additional children following the birth of the child with spina bifida compared to parents of children without spina bifida.

1.1. Previous research

A number of studies have found that having a child with disability is associated with an increased risk of divorce compared to parents with a child without disability, for example in Denmark for disabilities and chronic conditions (Loft, 2011), in the United States (US) for autism spectrum disorder (Hartley et al., 2010), and in the United Kingdom (UK) for early cognitive delay (Hatton et al., 2010). However, other studies have found no effect on divorce, for example after having a child with cerebral palsy in Sweden (Müller et al., 2022) and Denmark (Michelsen et al., 2015), as well as disabilities more generally in the US (Mailick Seltzer et al., 2001). Likewise, several studies have found a protective effect on the likelihood of divorce, for example for disabilities in Norway (Lundeby & Tøssebro, 2008; Tøssebro & Wendelborg, 2017) and the US (Namkung et al., 2015), and for Down's syndrome in the US (Urbano & Hodapp, 2007), comparing to parents with children without these diabilities. Few prior studies have explored the role of spina bifida in family formation, and existing studies generally consist of older descriptive studies or include spina bifida as one of many diagnoses in a more generic disability group (Mauldon, 1992; Singh, 2003).

Prior research suggests a reduced probability of having additional children among parents of children with disabilities. Studies from Denmark and Sweden have noted that having a child with cerebral palsy, compared to having a child without cerebral palsy, is associated with a reduced likelihood of having additional children (Michelsen et al., 2015; Müller et al., 2022). A Norwegian study found that families with children with disabilities had fewer siblings than the comparison group (Lundeby & Tøssebro, 2008). Similarly, a Turkish study found that parents of children with disabilities did not have additional children as a result of care-related time constraints (Şimşek et al., 2015). However, a US study found no effects on the likelihood of having additional children (Wehby & Hockenberry, 2017).

2. Methods

2.1. Study design and participants

We identified all persons with a diagnosis of spina bifida in Sweden born between 1991 and 2014. Eligible participants were identified using ICD10 code Q05 from the National Patient Register and the National Birth Register. A comparison group consisting of individuals without spina bifida or cerebral palsy was drawn from the Register of the Total Population at a ratio of five persons without spina bifida for each person with spina bifida, matched for birth year, sex, and municipality of residence. This means that we compare to a general population group of parents of children without spina bifida or cerebral palsy. Cerebral palsy was excluded from the comparison group due to practical reasons in the data ordering process as the overarching project studies both spina bifida and cerebral palsy (for details see: Alriksson-Schmidt et al. (2019). The parents of both cases and controls were identified through the Multigenerational Register. Yearly information on parental education, civil status, and family constellations was linked from the Longitudinal Integrated Database for Health Insurance and Labour Market Studies and the Register of the Total Population. Parental health information (diagnoses and health care utilisation) was extracted from the National Patient Register.

We only have information on parental health from 2001. Thus, the main analysis includes only parents of children with spina bifida

born 2004–2014, (2261 children of which 287 have spina bifida). We include the full sample born 1991–2014 in a sensitivity analysis without parental health data (6610 children of which 926 have spina bifida). Ethical approval has been obtained (dnr: 2018/1000 and 2021–00164). All data can be accessed, after normal application procedures, from respective register holders.

2.2. Outcomes

We examined the effect of having a child with spina bifida on two outcomes: the likelihood of divorce/separation and the probability of having an additional child. The sample is conditioned on parents living together at the time of the birth of the child. Divorce is defined as the parents no longer residing at the same address, irrespective of if they were married or co-habiting, even if they reconcile and move back together at a later date during the study period. Cohabiting is common in Sweden, and we thus included the separation of both married and cohabiting parents in our definition of divorce. Potential differences based on marital status are explored in the heterogeneity analyses. Whether the parents had additional children was deduced based on information on the date of birth, the number of children in the household, and the birth order.

2.3. Control variables

The covariates to control for potential confounding included the birth year of the child, the sex of the child, marital status, age of the mother at birth (categorized as 17–29, 30–33, 34–48 years), and presence of older siblings (yes/no). Because place of residence has previously been linked to the risk of divorce (Lyngstad, 2011), we included a variable indicating the region of residence at the time of birth of the child. Parents were considered to have a foreign background if either the mother or the father was born outside of Sweden. The education level of the household was measured as the highest level of completed educational attainment of the parents (mandatory

Table 1
Descriptive statistics of parents with and without a child diagnosed with spina bifida born between 2004 and 2014.

	No Spina bifida (n = 1974)	Spina bifida (n = 287)
Girl	1060 (53.7)	150 (52.3)
Multiple births	**6 (0.3)**	**7 (2.4)**
Older siblings	1153 (58.4)	157 (54.7)
Married	944 (47.8)	140 (48.8)
Mother's age at birth of child	784 (39.)7	120 (41.8)
Young (17–29 years)	572 (29.0)	86 (30.0)
Middle (30–33 years)	618 (31.3)	81 (28.2)
Older (34–48 years)		
Mother immigrated to Sweden	387 (19.6)	62 (21.6)
Father immigrated to Sweden	**369 (18.7)**	**72 (25.1)**
Highest parental education	**87 (4.4)**	**21 (7.3)**
Mandatory education	758 (38.4)	125 (43.6)
Secondary education	**1129 (57.2)**	**141 (49.1)**
Higher education		
Mother's mental health diagnosis	1769 (89.6)	251 (87.5)
None	95 (4.8)	18 (6.3)
At least 1	111 (5.6)	18 (6.3)
Unknown		
Father's mental health diagnosis	**1863 (94.4)**	**258 (89.9)**
None	**43 (2.2)**	**13 (4.5)**
At least 1	65 (3.3)	17 (5.8)
Unknown		
Mother's CCI	1796 (91.0)	260 (90.6)
0	55 (2.8)	8 (2.8)
> 0	122 (6.2)	19 (6.6)
Unknown		
Father's CCI	**1846 (93.5)**	**255 (88.9)**
0	**47 (2.4)**	**14 (4.9)**
> 0	81 (4.1)	18 (6.3)
Unknown		
Mother smoked before pregnancy	**1579 (80.0)**	**194 (67.6)**
No	257 (13.0)	48 (16.7)
Yes	**138 (7.0)**	**45 (15.6)**
Unknown		
Mother smoked during pregnancy	**1727 (87.5)**	**218 (76.0)**
No	122 (6.2)	26 (9.1)
Yes	**124 (6.3)**	**43 (15.0)**
Unknown		

Note: Significant differences at the 5 % level are presented in bold based on the chi-square test for categorical variables and the *t*-test for continuous variables. Abbreviation: CCI Charlson Comorbidity Index.

(≤ 9 years), secondary (≤ 12 years), or higher education (>12 years)). Parental health prior to birth of the child was measured in terms of mental and physical health for the mother and father respectively. For the purpose of this study, mental health problems were based on having a diagnosis of anxiety (ICD10 codes: F41, F42, F93.0–93.3, F06.4), other neurotic, stress-related and somatoform disorders (ICD10 codes: F40.1, F43, F44, F45, F48), depression (ICD10 codes: F30-F39), burnout (ICD10 codes: F43, Z73), sleep disorder (ICD10 codes: G47, F51.0, F51.8, F51.9), puerperium disorder (F53), and/or substance use disorders (ICD10 codes: F10-F16, F18-F19) in the National Patient Register in the three calendar years prior to the birth to the child. Parental physical health was measured using the Charlson Comorbidity Index (CCI) (Ludvigsson et al., 2021). The CCI was coded as (0) having no comorbidities vs. any comorbidities (>0). Finally, we also included whether the mother smoked three months before pregnancy or smoked during the last trimester (Idstad et al., 2015). Sample characteristics are presented in Table 1, where differences between groups are analysed using t- and Chi^2 tests.

2.4. Statistical analysis

Prior studies in the field have predominately used logistic regressions and we followed this tradition by presenting the probability of divorce at 1–3, 4–6 and 7–9 years of age of the child in the supplementary material (A1 & A5). For the main analysis, we applied the Cox proportional hazard (PH) model for interval-censored survival-time data. The Cox PH model takes timing into account instead of only focusing on whether the event occurred within the study period. Our data are interval-censored, as we only observe the year and not the exact date of a divorce or the birth of an additional child. Complete spells were defined as ending in either divorce/separation or the birth of an additional child, whereas a partial spell occurs due to right censoring, i.e., when the child or one parent dies, emigrates, or is lost to follow-up, and if the spell reached the end of the study period.

We first ran the model without any control variables, after which we included basic controls such as age and marital status, followed by a set of full controls except for parental health variables. The final model also included parental health variables.

The proportional hazards assumption is crucial for the Cox model (Basu et al., 2004). We checked if this assumption was fulfilled by plotting the estimated log-log survival curves for both groups. Parallel plots indicated that the proportional hazards assumption had not been violated. To test the robustness of our results in different models, we ran parametric survival analyses with interval-censoring. The choice of a parametric model was based on the Akaike Information Criterion (AIC) (Akaike, 1974). We also conducted a sensitivity analysis using a Cox model without interval censoring.

Lastly, we performed heterogeneity analyses to investigate if there were differences in effect based on parental demographics and socioeconomic characteristics (mother's age, parental education, marital status, and presence of older siblings). By interacting having a child with spina bifida with the characteristic of interest, the analysis showed the hazard ratios within subgroups. The likelihood ratio test was used to indicate if the interaction improved the model, thus indicating a heterogeneous effect. All results were considered significant at p-values < 0.05 and all analyses were performed using Stata 17.

3. Results

Table 2 shows the risk of divorce based on the Cox proportional hazard model while the logistic regression results are presented in the supplementary material (Table A1). Both models show a protective effect of having a child with spina bifida on divorce, an effect that increases the more potential confounders are controlled for. The full specification reports a hazard ratio (HR) of 0.54 (full regression output is available in the supplementary material A2). Fig. 1 depicts the hazard rates for the groups in terms of the risk of divorce where the parallel characteristics of the curves indicate that the proportional hazard assumption is not violated.

Fig. 2 shows the smoothed hazard curve and cumulative hazard estimates of divorce using the full model specification in Table 2. Estimating the hazard ratio of divorce using the full sample of children born between 1991 and 2014 shows no association between having a child with spina bifida and the risk of divorce (Model 2 in Table 3). This should be compared to a significant hazard ratio of 0.59 (Model 3 in Table 2) in the base case estimates on the 2004–2014 study period. This indicates that the effect of having a child with spina bifida on divorce rates have changed over time. Alternative model specifications resulted in estimates similar to the base case

Table 2
Hazard ratios of time to divorce for parents with a child with spina bifida compared to parents with a child without spina bifida.

	Model 1 No controls HR [95 % CI]	Model 2 Basic controls HR [95 % CI]	Model 3 Controls without health HR [95 % CI]	Model 4 All controls HR [95 % CI]
No spina bifida	1.00 (ref)	1.00 (ref)	1.00 (ref)	1.00 (ref)
Spina bifida	**0.64**	**0.62**	**0.59**	**0.54**
	[0.45, 0.89]	**[0.44, 0.87]**	**[0.41, 0.83]**	**[0.38, 0.76]**
Observations	2261	2261	2261	2261

Note: Main analysis using the Cox proportional hazard model with interval censoring for divorce on the sample of children born 2004–2014 in a stepwise regression. In Model 1, no control variables were included, Model 2 controlled for the sex and year of birth of the child, the marital status of the parents and the mother's age at birth of the child. In Model 3, the region of residence, the presence of older siblings, immigration background of parents, and parental education were added. Finally, in Model 4 the mother's and father's mental health and comorbidity status and whether the mother smoked before and during pregnancy were added. Results significant at the 5 % level are presented in bold with confidence intervals in brackets. Full model output can be found in the supplementary material (A2). Abbreviations: HR Hazard ratio, CI Confidence interval,

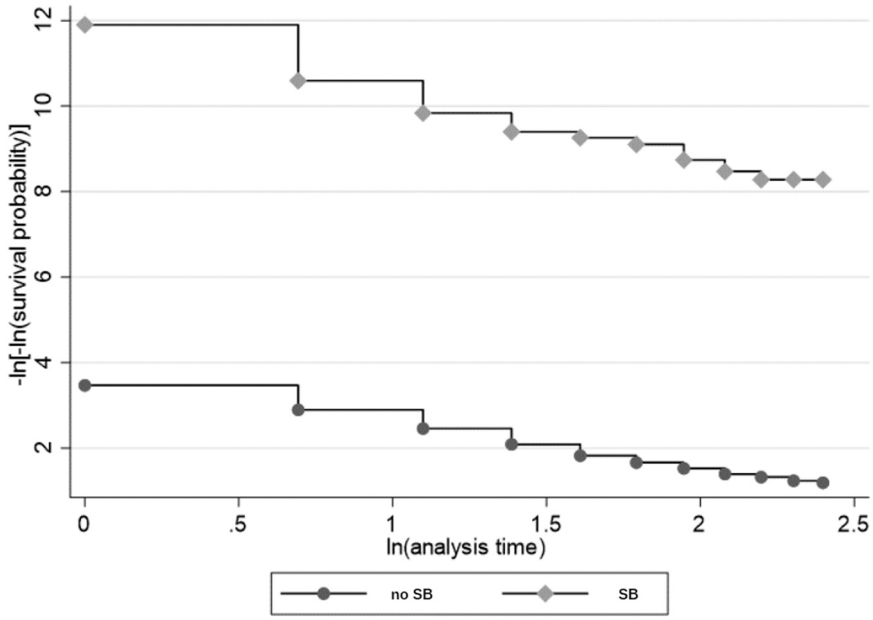

Fig. 1. Proportional Hazard assumption test for having a child with spina bifida on the risk of divorce**.** Note: The figure shows the log-log survival curves for the treatment and comparison groups (i.e., having a child with SB or without SB) regarding the risk of divorce. The hazard curves are adjusted for all covariates from the full specification (see Table 2). Parallel development of the plots indicates that the proportional hazards assumption is not violated. Abbreviation: SB Spina Bifida.

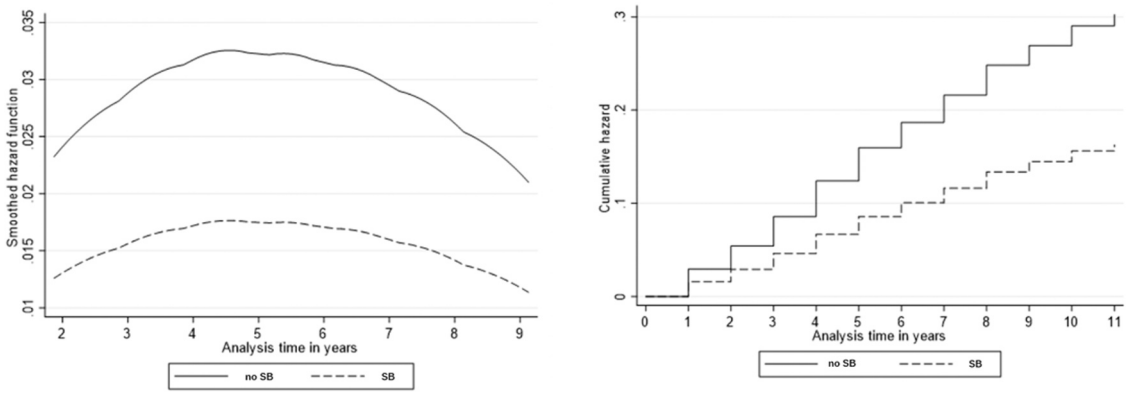

Fig. 2. Smoothed hazard curve and cumulative hazard estimates of divorce**.** Note: The figure shows the smoothed hazard rates and cumulated failure estimates of divorce for parents of children with and without SB, based on the full model specification in Table 2. The smoothed hazard rates on the left show the likelihood of divorce across groups in each period, given that divorce did not happen in any previous period. The cumulated failure rates on the right display the proportion of divorced parents across time. Abbreviation: SB Spina Bifida.

results. Note that the time ratio (TR) used in the log-logistic parametric model has the opposite interpretation to the HR used in the Cox PH model. A time ratio above 1.00 indicates that the time to divorce is longer for parents of children with spina bifida compared to the comparison group and is therefore in line with a hazard ratio below 1.00.

We find no indication of a heterogeneity effect over parental characteristics, based on the likelihood ratio test. However, some variations in the effect size were noted across subgroups (Table 4), where having a child with spina bifida was associated with a stronger protective effect for mothers in the youngest and oldest age categories compared to the middle category, as well as for married compared to cohabitating parents. No protective effect was found for parents with low education while the strongest reduction in risk of divorce was noted for parents with higher education. All heterogeneity results should be interpreted with caution given the non-significant likelihood ratio tests.

Table 3
Sensitivity analysis of divorce risk for parents with a child with spina bifida.

	Base case HR [95 % CI]	Full Sample HR [95 % CI]	Parametric: Loglogistic survival model TR [95 % CI]	Cox proportional hazard model without interval-censoring HR [95 % CI]
No spina bifida	1.00 (ref)	1.00 (ref)	1.00 (ref)	1.00 (ref)
Spina bifida	**0.54**	0.91	**1.90**	**0.55**
	[0.38, 0.76]	[0.80, 1.03]	**[1.35, 2.66]**	**[0.39, 0.77]**
Observations	2261	6610	2261	2261

Note: The sensitivity analysis for the likelihood of divorce. The full sample model included children born 1991–2014 and is specified as Model 3 in Table 2. The parametric model and Cox PH model control to full model specifications as Model 4 in Table 2. The log-logistic parametric model was chosen based on the AIC score. Significant results at the 5 % level are presented in bold. The full model outputs are presented in the supplementary material (A3). Abbreviations: HR Hazard ratio, CI Confidence interval, TR Time ratio.

Table 4
Heterogeneity analysis of divorce risk for parents with a child with spina bifida.

	Mother's age HR [95 % CI]	Parental education HR [95 % CI]	Marriage status HR [95 % CI]	Older siblings in household HR [95 % CI]
Spina bifida x Mother's age category	**0.51**			
Spina bifida x 17–29 years	**[0.32, 0.81]**			
Spina bifida x 30–33 years	0.72			
	[0.38, 1.38]			
Spina bifida x 34–48 years	**0.41**			
	[0.17, 0.98]			
Spina bifida x Parental education		0.98		
Spina bifida x Mandatory		[0.34,2.83]		
Spina bifida x Secondary		**0.61**		
		[0.40, 0.93]		
Spina bifida x Higher		**0.30**		
		[0.14, 0.67]		
Spina bifida x Civil status				
Spina bifida x Cohabiting			**0.63**	
			[0.43, 0.93]	
Spina bifida x Married			**0.36**	
			[0.17, 0.76]	
Spina bifida x Child has older siblings in the family				
Spina bifida x No				**0.59**
				[0.37, 0.96]
Spina bifida x Yes				**0.49**
				[0.29, 0.81]
p-value of LR test	0.5132	0.1128	0.1576	0.5656
Observations	2261	2261	2261	2261

Note: The heterogeneity analysis tested the interaction of having a child with spina bifida with the mother's age, parental education, marital status, and the presence of older siblings in the family on the likelihood of divorce. Significant results are presented in bold. Abbreviations: HR Hazard ratio, CI Confidence interval, LR Likelihood ratio test.

Table 5
Hazard ratios of time to additional children for parents with a child with spina bifida compared to parents with a child without spina bifida.

	Model 1 No Controls HR [95 % CI]	Model 2 Basic controls HR [95 % CI]	Model 3 Controls without health HR [95 % CI]	Model 4 All controls HR [95 % CI]
No spina bifida	1.00 (ref)	1.00 (ref)	1.00 (ref)	1.00 (ref)
Spina bifida	1.14	1.08	1.00	0.99
	[0.94, 1.39]	[0.88, 1.32]	[0.82, 1.23]	[0.80, 1.22]
Observations	2261	2261	2261	2261

Note: The Cox PH model with interval censoring for additional children on the sample 2004–2014 in a stepwise regression. Model 1 has no control variables included, Model 2 controlled the sex of the child and year of birth of the child, the marital status of the parents and the mother's age at birth. In Model 3, the region of residence, presence of older siblings, immigration background of parents and parental education were added. Finally in Model 4, the mother's and father's mental health and comorbidity, whether the mother smoked before and during pregnancy were added. Full model output can be found in the supplementary material (A6). Abbreviations: HR Hazard ratio, CI Confidence interval, CCI, Charlson comorbidity index.

3.1. Additional children analysis

In the second part of the analysis, we found no effect of having a child with spina bifida on the likelihood of having additional children, neither in the logistic regression analysis (see supplementary material A5) nor in the Cox PH model with interval censoring (Table 5).

The test of the proportional hazard assumption indicated that the proportional hazard assumption might be violated. We therefore proceeded to estimate a parametric Gompertz model with interval-censoring (Table 6). However, the results of the parametric estimation were very similar to the base case estimate, indicating that the results were not driven by uncertainties around the proportional hazard assumption. The additional sensitivity analyses support the base case estimate of no effect of having a child with spina bifida on the likelihood of having additional children. The heterogeneity analysis reveals no variations in effect across parental characteristics (supplementary material A8).

4. Discussion

This study investigated the risk of divorce and the probability of having additional children for parents with a child with spina bifida in Sweden. We used a large population-based, nationally representative, dataset that included several potentially important confounders generally not included in previous studies. We found that parents of children with spina bifida have a 46 percent reduced probability of divorce compared to parents with a child without spina bifida, a finding that was robust across model specifications.

Our results indicated that having a child with spina bifida is associated with relationship stability, which has also been noted in a Scandinavian study on intellectual and developmental disabilities (Tøssebro & Wendelborg, 2017) and a US study on Down syndrome (Urbano & Hodapp, 2007). The stability of the relationship could be attributed to stronger mutual obligations among parents, as suggested by Lundeby and Tøssebro (2008), potentially created by the early life challenges and adjusting to parenting a child with disability. It has been suggested that families with children with spina bifida exhibit a "resilience-disruptive" characteristic to family functioning. This includes disruption of certain family functions and substantial resilience in other functions (Holmbeck & Devine, 2010), making the overall outcome difficult to predict. The reduced risk of divorce for parents with a child with spina bifida compared to parents with a child without spina bifida could also be understood in the context of the perceived high cost of separation, both in terms of an uncertain financial situation in the future and the reduced likelihood of re-partnering (Lundeby & Tøssebro, 2008). Unfortunately, we cannot establish the mechanism behind the lower divorce rates in the current study, which is left for future research.

The comparison group used in this study was created based on a random draw from the general population (although matched in terms of sex, birth year and municipality of residence of the child) and can therefore include other disabilities (except cerebral palsy) and chronic diseases according to their prevalence in the population. Thus, we are not comparing parents of children with spina bifida to parents of healthy children only. Our results should therefore be interpreted as the average effect of having a child with spina bifida.

Many parents who receive a diagnosis of spina bifida during pregnancy choose to terminate the pregnancy (Bodin et al., 2018). Fifty percent of known prenatal cases of spina bifida resulted in a terminated pregnancy in Sweden during the study period (Socialstyrelsen, 2015). There is therefore a risk of selection bias in our study that hinders a causal interpretation of the results. If parents who chose to have the child with spina bifida and parents who chose to terminate the pregnancy are different in characteristics that are associated with the risk of divorce, such as religiousness or initial stability of partnership, our estimates are likely overestimated. However, the pregnancies with known prenatal spina bifida that are not terminated appear to be predominantly late diagnosis, i.e., after gestational week 23 (Nikkilä et al., 2006), which is the latest point where termination can be approved in Sweden. This would argue against selection bias, although further studies on termination decisions of pregnancies with known spina bifida are needed to establish this. Nevertheless, the results from the current study have external validity given that regardless of who and why women choose to terminate a pregnancy, those who decided to carry to term are included in the study and for all intents and purposes, it is the divorce risk for them that is of interest. Overall, the results of the current study should be interpreted with some caution in terms of causality. A descriptive interpretation would, at the very least, show that this particular group of parents are not worse off compared to the general population in terms of divorce and family formation.

As mentioned earlier, previous studies have found mixed results in associations between having a child with disability and the risk

Table 6
Sensitivity analysis for the likelihood of having additional children for parents with a child with spina bifida.

	Base case HR [95 % CI]	Full Sample HR [95 % CI]	Parametric: Gompertz survival model HR [95 % CI]	Cox proportional hazard model without interval-censoring HR [95 % CI]
No spina bifida	1.00 (ref)	1.00 (ref)	1.00 (ref)	1.00 (ref)
Spina bifida	0.99	0.92	1.01	1.00
	[0.80, 1.22]	[0.82, 1.03]	[0.83, 1.23]	[0.82, 1.22]
Observations	2261	6610	2261	2261

Note: Sensitivity analysis for the likelihood of having additional children. The base case corresponds to column 4 in Table 5. The full sample model includes all children with CB born 1991–2014 and is specified as Model 3 in Table 5. The parametric model and Cox PH model control to the full model specifications (Model 4) in Table 5. Full model outputs can be found in the supplementary material (A7). Abbreviations: HR Hazard ratio, CI Confidence interval.

of divorce. Some studies have found no association e.g. (Baeza-Velasco et al., 2013; Mailick Seltzer et al., 2001; Michelsen et al., 2015; Namkung et al., 2015), while other studies have found an increased risk of divorce (Hartley et al., 2010; Hatton et al., 2010; Loft, 2011), comparing to parents with a child without disability.

Sweden, like other Scandinavian countries, is known for its comprehensive welfare system and universal healthcare with free child health services. In this type of setting, the financial burden, and thereby the stress on the family unit, of having a child with a disability could be expected to be lower compared to countries with less generous welfare systems. It is then, perhaps, more interesting to note the difference in results compared to Müller et al. (2022) who used the same statistical approach and identical data material and found no effect on divorce rates after having a child with cerebral palsy in Sweden. This indicates that there is substantial heterogeneity across disabilities and that it might be inappropriate to study disability as a group. Thus, we should be cautious in generalising the results from one disability to the next and likewise, applying results for the general disability area to specific conditions in certain areas of research.

The estimated association varied across parental characteristics such that higher parental education and being married were associated with a stronger protective effect on divorce, which is in line with previous research (DeRose & Wilcox, 2017; Holmbeck & Devine, 2010; Loft, 2011). In a Scandinavian setting, Loft (2011) noted that being married was positively associated with the length of the relationship, suggesting that the effect of marriage could be interpreted as a proxy for the stability of the relationship. The results further indicated a stronger protective effect of having a child with spina bifida on the likelihood of divorce if there was an older sibling in the family. The presence of siblings can provide a supportive role alleviating some of the stress on the parental relationship. This additional support may ease the stress in taking care of a child with disability, potentially reducing the risk of divorce (Namkung et al., 2015; Wolfe et al., 2014). It is also possible that this effect is due to the longer experience of being a parent, which is expected to reduce the overall burden of having an additional child with disability.

No effect was found of having additional children after the birth of a child with spina bifida. The available literature on this is inconclusive with some studies reporting similar findings to ours (Wehby & Hockenberry, 2017) and others reporting a reduced likelihood of having additional children after the birth of a child with a disability in the Scandinavian context (Lundeby & Tøssebro, 2008; Michelsen et al., 2015). In the broader European context, reduced intentions and desires to have additional children have been reported (Di Giulio et al., 2014; Şimşek et al., 2015). The Swedish study on cerebral palsy using the same methodological approach and data material as the current study found a reduced likelihood of having additional children for parents of children with cerebral palsy (Müller et al., 2022). Taken together, this highlights the different consequences of having a child with disability across disability-specific factors, again cautioning against studying disability as a homogeneous group and generalising the results between disabilities.

Following the Becker-Lewis quantity-quality model (Becker & Lewis, 1973), a reduction in the number of additional children would be expected given the need to consider how the additional children would affect the resources (financial and time) available for existing child/children. However, there are a number of possible explanations as to why this is not the case in the current study. Namkung et al. (2015) argue that the quantity-quality trade-off could be more complex in families with children with disabilities with an increased (compensating) valuation of having additional children also as support for the child with disabilities. It is also possible that the characteristics of the contextual situation with a welfare system that provides certain services and financial support coupled with an overall high standard of living in Sweden might weaken the necessity of the quantity-quality trade-off. An important factor in the Becker-Lewis model is the budget constraint and although all families are constrained, families in affluent contexts have more room to re-allocate resources from consumption of other goods. The interesting question deriving from these results is why families with a child with cerebral palsy are reducing the number of additional children while families with a child with spina bifida do not. This could potentially be a consequence of characteristics of the disabilities as well as care and financial burden or differences in the response of the Swedish welfare system across disabilities. The potential selection bias stemming from termination of pregnancies should also be considered here, and future studies need to consider if there are unobserved confounders that could explain the observed results. Cerebral palsy is most often not identified in utero as opposed to spina bifida.

Overall, the results of the current study indicate that families with children with spina bifida have parents who are less likely to divorce compared to the control group of families with children without spina bifida and are equally likely to have additional children as families with a child without spina bifida. However, the results should be understood in the context of the Swedish welfare state that provides support. The consequences of having a child with disabilities might be different in other countries with different support structures. However, the effect of welfare support is unknown, it could serve to mitigate the stress on the family unit, or it could enable parents to separate, knowing that the (financial) consequences will not be that great. Future studies should try to discern the role of welfare support on the consequences for the family unit of having a child with disabilities.

4.1. Strengths and limitations

Some limitations to this study should be considered. Although hazard rates help explain how the past impacts the future, there is implicit conditioning as the risk of divorce is conditioned on that it did not happen in the preceding years. This makes it difficult to draw causal conclusions as it may cause an imbalance of confounders between the two groups (Aalen et al., 2015). Furthermore, although we have access to comparatively rich data, we cannot rule out the possibility of unobserved factors biasing the results, especially regarding termination of pregnancy as discussed above. In addition, spina bifida is a rare condition and even though we have the full national population sample, we have a fairly low number of births included in the analysis. The strengths of this study are the longitudinal population-based sample including all children with spina bifida born during the study period, together with a representative general population comparison group. In addition, we have employed advanced statistical models and control for

confounders that are rarely, if ever, controlled for in prior research on the topic.

5. Conclusion

Our study indicates that parents of children with spina bifida are less likely to divorce and as likely to have additional children as parents in the general population. The results highlight differences with other types of congenital/early onset disability on family formation, stressing the need to consider the characteristics of specific disabilities rather than treating disability as a homogeneous group in this context of research. It is left for future studies to try to disentangle the mechanisms behind the differences in the impact of childhood disabilities on family outcomes, which is necessary in order to construct effective policies and support structures where needed.

CRediT authorship contribution statement

Mtutu R. Samu: Writing – original draft, Formal analysis. **Johan Jarl:** Writing – review & editing, Methodology, Formal analysis, Data curation. **Vibeke Müller:** Methodology, Formal analysis, Data curation, Conceptualization. **Alriksson-Schmidt Ann:** Writing – review & editing, Funding acquisition, Formal analysis.

Ethics approval

Ethical approval has been obtained (dnr: 2018/1000 and 2021–00164)

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Conflict of Interest

No competing interests to declare.
A Poster Presentation of this paper has been presented at European Academy of Childhood Disability, Barcelona in 2022.

Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.ridd.2025.105043.

Data Availability

The data that support the findings of this study are available from the register holders after standard application procedures.

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