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Associated health care cost in parents of children with Myelomeningocele in Sweden: A repeated cross-sectional analysis of region Skåne's health care utilisation data base.

Author
Abimbola Ajomale

Supervisor
Johan Jarl
Associate Professor in Health Economics
Lund University, Sweden

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Abstract

***Background*:** Studies have put spinal bifida which Myelomeningocoel (MMC) is a form at 6.1 per 10,000 births in Sweden. Other evaluations have pointed that disabilities could pose a burden on families despite the social net available within the Swedish society. It has been reported that depression, musculoskeletal disorders, and frequent health care visits are often likely to be reported in parents of children with developmental disability which might constitute a major public health problem.

Aim: The aim of this study is to explore the health care cost in parents of children with MMC compared to parents of typically developing children across the region Skåne, Sweden.

Methods: A repeated cross-sectional study design was adopted in the course of this work. The data were collected yearly from all persons with MMC born between the year 2001 and 2015 and their parents. Health care data from region Skåne was used. Health care cost was analysed adopting independent t test to compare the mean cost and its significant levels while adopting a linear regression model to evaluate pattern of cost by controlling for confounders in region Skåne. Parents of children with Myelomeningocoel health care cost was evaluated using descriptive statistics.

Results: Parents of children with MMC have significantly higher healthcare cost at one year before birth (SEK 6,054), at birth (SEK 7,644), and two years after birth (SEK 6,356) in region Skåne with female parents utilising public health care about seven times higher than their male counterparts.

Conclusion: Findings from the study showed significant higher health care cost in parents of children with Myelomeningocoel. Female parents were the only ones who utilise care for depression and musculoskeletal disorder. The study has an important implication in public health in looking at other factors that can cause disparity of care cost in gender within this study population.

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1. Introduction

About a billion people worldwide has been said to live with some sort of physical, sensory, mental or intellectual incapacitation which impact their lives on a daily basis[1]. Developmental disabilities are said to be a cluster of disorders emanating from injuries that affect a child's physical, learning, or behavioural capability[1]. Children with developmental disabilities stand a greater risk of having poor health, scholastic fulfillment, and wellbeing compared to children without disabilities[2]. Globally, disability has been noticed to put strains on families. These strains are said to have a far-reaching effect on them. The out-of-pocket costs of medical care and other services may be enormous. All of these potential effects could have repercussions for the quality of life in the parents such that if the parents are negatively affected, this in turn will have a negative effect on the child. Their living pattern, relationships and the society at large will be impacted too[3].

Studies have shown that there is often the burden of finance when caring for children with disability by their parents[4]. This financial burden could be associated with the health of the child, their education, and sometimes their social activities. Other activities that can put strain on the parent financially are from renting of equipment, devices, modified accommodation, bedding at home, specialized mode of transportation, medications and special diets[3,4]. Though, some of these financial expenses may be catered for where there are good health systems in place. Sometimes the parents may be eligible for reimbursement through the insurance outfit or through publicly funded health programs targeted at families with children with disabilities[4]. Some families with a child or children with one form of disability or the other may be predisposed to increased stress or have a negative effect on their mental and physical health[5]. This may be because parents may be at a risk of depression and anxiety, due to stressors, like increased caregiver demands and financial constraints. However, if such conditions are not mitigated promptly, parental depression may have an enormous impact on the family structure, and on the parent-child relationship dynamics[5].

Having to live with children with disability has been found to have profound effects on the family structure. Adversely, their time, financial cost, emotional demands, complex logistics all have far reaching effects that will percolate to the fabrics of the larger society[6]. Often times, care taking roles from the parents may lead one or both parents to jettison their career plans. Studies have shown cases where female family members are affected more because they are likely to take up the role of the primary care giver and may end up giving up their career or work. Bringing the burden of financial implication of taking care of a child with

disability in a society where male earn marginally higher than women may also open up the family to divide role and care responsibilities[7]. It could also become challenging or problematic when birth defects have a prognosis of being a life-long chronic situation that may necessitate continuing parental care past the age other children attain independence. In addition, studies have shown that depression or financial burden caused from having a child with deformity for example can interfere with correct implementation of care strategies for their child[4,8].

1.1 Myelomeningocoel

Myelomeningocele (MMC) is a form of spina bifida which is a developmental impairment which is said to be the most intricate congenital defect compatible with life. An incidence of about 1 in 4,000 neonates may be affected globally[6]. MMC occurs around the early weeks of pregnancy[6]. It has been described as multi-system affecting condition which may lead to musculoskeletal, urogenital or neurological anomalies. The quality of life and social adaptability in the affected individuals are often impaired. Hydrocephalus is often seen in children with MMC which might later affect their fine motor function and ability to plan[7]. 10% of the newly born children with MMC are with clinically obvious hydrocephalus while over the first week, the progression of hydrocephalous rises abruptly and are seen in about 85% of the cases[7,8].

1.2 Health care cost and parental health challenges

Parents with children having special needs often access more specialised care, and other educational services, which will often lead to complex economic burden as a result of their children's condition[9]. Studies have shown that health care cost incurred by these families can be substantial[10]. Other works have reported parent of children with severe disability sliding into poverty while taking care of the special need child and their own failing health[11]. A United States study put the percentage of families having financial burden for having a child with special health care need at 40%, and majority of these cost goes to parental physical and mental health challenges associated with their child's care[11].

Care giving as a parent might be a daunting task where it is possible to ignore one's health and wellbeing[12]. This is seen when parents are engrossed with their special need child. Studies have shown that Musculoskeletal Disorder (MSD) often becomes an issue to the parents when they are the primary care givers. They are commonly affected by an extensive class of musculoskeletal problems like back pain, neck pain, shoulder pain and leg pain which make

them incur extra health care cost[13]. Previous works globally and in Europe have established that caregivers tend to do more physical work (PW) and are prone to be affected with musculoskeletal disorders and poor mental health[11,13]. Also, the behavioral problems seen in children with these disabilities repeatedly strengthen the stress and trigger in parents tilting them into a poor mental health[5,12]. Parental depressive symptoms (PDS) has been found in some studies to occur in parents of children with Myelomeningocoel (MMC) as form of spina bifida up to about 70% of cases which brings extra health care cost to the parents[9].

Arguably, children with special needs often show externalised behaviour, like difficulty in communicating, poorly formed social interactions, and oppositional defiant disorder when compared to apparently developing children. Hence, studies have established that parental health challenges are tied with negative outcomes in children[9] which also elaborate stressors on the caregiver in terms of financial strains and care demand[9,10,14].

1.3 Aim

The aim of this study is to explore the health care cost in parents of children with MMC compared to parents of typically developing children across the region Skåne, Sweden.

To further add, this project will be the first of its kind in Sweden to the author's findings that will look at the parents of children with MMC considering health care cost with associated health challenges. The peculiarity of studying parent's health care utilisation in term of cost in this project is novel. Hence, this research is set out to complement large body of knowledge in understanding health care cost pattern in parents of children with MMC in Sweden as this might be the first register-based study.

1.4 Research Questions

1. What is the extra burden of health care cost (in-patient and out-patient care) for parents of children with MMC compared to parents without children with MMC in Sweden?
2. Is the incidence of clinical depression (ICD-10:F32) and musculoskeletal disorder (ICD-10:M20-M25) increase in parents of MMC compared to parents of children without MMC in Sweden?

2 Method

2.1 Study design

This study is a register based repeated cross-sectional research. New samples are collected at different time points which represent different health care cost estimation for each successive calendar year. This implies that respondents differ at earlier and subsequent years or times of survey. Hence, a combined analysis over specific period of time is considered in this study. These are health care cost five years before the birth of the child with MMC, at the year of birth, and five years after birth. The cases are parents of children with Myelomeningocoel. The cases are pulled from the national register comprising the population register by identifying children with MMC, then the parents were identified by the generational data. If MMC cases are found from 2001 to 2015, then they are included. Controls from the general population living in Skåne region and their parent are pulled from the total population register and the generational register. Hence, data were used from the population register, patient register (PAR), birth register (MFR) and the MMCUP register in Sweden from the year 2000 to 2015[15]. The acronym MMCUP denotes "Myelomeningocoel follow-up programme" in Sweden[15].

Parents of MMC children are predetermined to be residing within the region Skåne, and the child with MMC must be living with both parents within the same region. So, it means invariably that a parent must have a child with a confirmed MMC as diagnosis. The period of review for this study was put at 2001 to 2015. A total of ninety-nine cases (parents with children with MMC) and seven hundred and five controls were identified. A ratio of five controls was chosen to each case as determined by the research group. Though, the initial cases (parent of children with MMC) gave a total one hundred and ten before the condition of both parents living together with the child was introduced. However, the control population was narrowed down to the current number either because one or both parents might have moved out of region Skåne to other regions or their children may have other forms of disabilities and not suitable to be included in the control group.

The first step in establishing the control group was matching the children with MMC as living with both parents. Then, an exclusion of children with other congenital disability similar to MMC such as cerebral palsy was made to prevent a crossover. . The study will involve analyses for five years before the birth of the child, the year of birth and five years after all considered during successive calendar year.

2.2 Study setting

Region Skåne health care data base has health care cost imputed from the diagnosis related group (DRG) and this is put in cognizance as a region of interest for this study. Skåne region is a part of the country located in the southern part with a population of approximately 1.4million inhabitants[16]. Sweden being as a country is located in northern Europe with a parliamentary form of government with a flavor of monarchy[16]. The population in Sweden stands at 10.38 million from the Statistics Sweden, data from January 2021 with more than 80% of the population living within the urbanised regions[17]. Sweden runs a decentralized health care system being managed by either the regions, municipality, or the local authority. Hence, every county council, local authority or municipality are responsible for running and managing its own healthcare assets. Though the Swedish government has operated a socially responsible health care system for a very long period and was consolidated with the Health and Medical Services Act of 1982 which has a solid principle of equal access to health for all[16]. However, it is of note that healthcare services offered can be different, but the study setting can be representative of the whole country[18]. This can be attributed to the fact that the social and the welfare system that handles disability work the same throughout the country.

2.3 Independent Variables

The study has put into consideration some key independent variable on which the study is contingent. They are being a parent with a child diagnosed as having myelomeningocele, parent's place of birth, sex of the children, and the age of parent. The parent's sex also is considered to see any influence or pattern that might arise from their health care cost. The sex of the child might also be important to see if there is a pattern seen in care cost with respect to their sex.

2.3.1 Sex of parent

The study is also interested in seeing the pattern of health care cost within the sex of the parent of children with MMC. Cost of care might be higher around delivery for females. Hence, need to disaggregate parent's sex and see if there was a difference in their health care cost.

2.3.2 Parent's place of birth

Parent's place of birth is chosen to see if there will be influence in care cost for parents born in Sweden or abroad.

2.3.3 Sex of children

The sex of the children too is also considered to be able to see if there is a pattern that might have influence or drive the health care cost in either of the sex. If more cost is accrued caring for a male child of female child or vice versa. Or on the other hand if there is sex specific syndrome that is related to changes in care cost.

2.3.4 Age of the parent

The age of the parent is also considered to see if there are trends that may influence health cost pattern with respect to the health care cost in parents of children with MMC. There have been evidences of increased disability in children with advanced parental age[18].

2.4. Dependent Variables

Health care utilisation in terms of cost is the dependent factor that is been looked into in this study. Health care utilisation is a well-known attribute that is been studied in many research. Other dependent variables been looked at are depressive illness and musculoskeletal disorders in the parents of myelomeningocele children. Depressive episodes are said to be present in some parents taking care of children with some form of physical disability or the other[18]. Other studies have suggested that there is a consistent increase in physical workload as seen while taking care of children particularly those having some forms of neurological disorder or defomoities[19]. Hence, depressive illness is being evaluated by studying the health care cost from depressive illness care. This is such that the patterns of care cost seen are evaluated in parents of children with MMC in region Skåne in Sweden.

2.5 Health care cost

The health care cost in these parents is considered under various aggregates. The first of the aggregates is, outpatient cost, then inpatient cost, and primary care cost which will culminate to a total health care cost in the parents of the child with MMC. Diagnosis Related Groups (DRGs) was adopted to categorise hospitalisation costs and determines the amount of money

each service is worth[17]. These sets of cost are also looked into at the point of birth of the child, before birth, and after the birth of the child all of which are extracted from the data base.

2.6 Statistical analysis (Chi-Square, T test and linear regression)

The study has used descriptive statistics in presenting the relative health cost while having a child with MMC. Variables like sex of the parents, sex of the children with MMC, place of birth of the parent and the age of the parents were also considered. Chi-Square were used to present categorical variables. Independent t-test was employed with a p-value of <0.05 considered to be significant for the work. The independent sample t-test was used to compare the means of the health care utilisation in cost of the parents with MMC children and parents without MMC children as controls. The independent sample t test was adopted as a parametric test to assess for any statistical evidence that might be present in the mean cost to demonstrate any significant difference[20]. Multiple linear regressions were also employed to see the pattern in the health care cost by controlling for confounders.

The statistical analysis was done with SPSS version 27.0.1.0, a product of IBM in the United States of America. The software is one of the foremost packages employed in solving predictive analysis and hypothesis evaluation globally[21,22].

2.7 Ethical considerations

Ethical approval was given by the ethical review board at Lund with approval number Dnr: 2018/1000. Results from the study will be presented on an aggregated group level that will make it impossible to identify individuals. The data from the MMCUP were all collected voluntarily with the consent of all the participants in the registry[23]. All patients registered within the register were given their right of autonomy by adopting only ascribed number in keeping with the principle of autonomy in research[18]. The confidentiality of the subjects within this study was also well guided by not using any assigned number from the registry. The data was also not shared with anyone outside the research team. The principles of non-malevolence and beneficence [30,43] will be worth mentioning because the use of the subjects' data in this work is aimed at improving health outcome in the subjects. It is of note too that women may want to terminate their pregnancies if they have an inclination that the cost of care might be increased while taking care of a child with MMC.

3. Results

3.1 Descriptive statistics

Table 1 and 2 shows the distribution of sample characteristics and categorical picture of both the test and the control populations. It also reflects the distribution of musculoskeletal disorder and depression within the study population. Females constitute more than half of the parents of the MMC child at 52% compared to males. Two individual each had issues with musculoskeletal disorder (MSD) and depression among the parents of the children with MMC which constitute a prevalence of 2% respectively. The differences seen in gender in parent and children were not statistically significant so, the case and control groups are comparable. Parent's place of birth, age at birth, depression and musculoskeletal disorders were also not significantly different. Fig II shows the gender distribution of children with MMC based on their place of birth where about 76% were born in Sweden.

3.2 Independent t-test

Table 3 shows the mean health care cost with independent t- test showing levels of significance in mean between parents of children with MMC and parents of control group at the year of child's birth, a period of five (5) years before their birth and five years after. A significant difference was seen in the mean health care cost at the year of birth for the parent with children having MMC. One year before birth of children with MMC also showed a significant difference with (M= 16,355, SD=53554). A significant difference was seen in the mean health care cost at two years after birth for test group with (M= 14610, SD=36389). The health care cost difference were SEK 6,055, SEK 7,645, and SEK 6,357 higher in parents of children with MMC at one year before, at birth and two years after respectively.

3.3 Linear regression analysis

Table 4 presents the adjusted effect of MMC status on health care cost, controlling for sex of parent, sex of child, and the age of parent. The estimated extra cost when a parent has a child with MMC at one year before birth, at birth, one year after birth and two years after birth were estimated at 6053kr, 7559kr, 6118kr, and 3620kr respectively when other variables were controlled for. However, the unadjusted analysis using the t-test showed a health care cost of 6055kr, 7645kr, 5030kr, and 6357kr at one year before birth, at birth, one year after birth and two years after birth respectively.

MMC status and sex of the parent showed the most potent predictors at the year of birth with B: 7559; 95% CI: (1820-13298) with $p<0.05$ for a child born with MMC. Parent's gender at the year of birth too had B: 42402; 95% CI: (38630-46174). These show the degree of change that is observed in MMC and the sex of the parent. This association account for about 40% of the health care cost variance seen in the subject. One year and two years after birth also have 37% and 33% of the cost difference explained by the independent variable. On disaggregating parent's sex into male and female, estimate extra cost was higher in females. Table V shows that the total health care cost at four years before the birth of the children with MMC and at birth were accounted for by the female parent up to 80% and about 50% respectively.

Fig I show a line graph in semblance to the ACO (modeling framework of the accountable care organisation) model which emphasis health care cost relatively spiraling to a peak with debilitating illness.

4. Discussion

This study has demonstrated what the extra burden of health care cost (in-patient and out-patient care) for parents of children with Myelomeningocele in Sweden looks like. Also the incidence of common health challenges seen in them as depression and musculoskeletal disorder was elucidated. The result reflected an increase parental health care cost around the period pre delivery and immediate post delivery period in parents of children with MMC compared to the control population. 2% of the cases incurred cost for musculoskeletal disorder and depression. These findings show that having a child with MMC will influence increased health cost a year before and up to two years after delivery of such child. However, the sex of the parents too played a role in the cost pattern, though in a very small way. Being able to conceptualise a pattern in health care cost in this group may go a long way in predicting the trend which may ultimately inform policy and planning for imminent health care spending.

4.1 The burden of health cost in the parents

To create a succinct boundary and blend within the bigger domain of public health and econometrics[23], this study on health care cost is an important aspect of the health care system to be evaluated to mitigate the latent factors and economic facts that may bedevil health budgetary planning and health policy implementation. Sweden has been known for a steady health and medical care cost within the league of countries within the European Union[24] for a

couple of years. The country has maintained a consistent portion of its GDP which is about 11% over the years[25] on health allocation. The regional government and the municipality fund most of these bills through taxes. However, the central government also contributes to this spending leaving the patient to cover a small portion of the health care cost[26]. This study has shown a pattern of health care cost with a unique pattern in the parents of children with MMC which were marginally higher than the control group before the birth of MMC child. Previous studies have shown that having such a child can be a major stressor on the parent in terms of financial strains in period surrounding the child's birth and for years after[27]. Health care needs has been said to go a long way to inform health care cost. Then the quality of service and its availability too will influence its utilisation which will cumulate to the total cost of health care[28].

Apart from the one year prior to delivery, at birth, one and two years after delivery, the other period under review did not show a significant difference in health care cost. This might be due to an effective preventive health system in Sweden. Other explanation may be due to the social and welfare support system that encourage paid leave for parents in Sweden[16]. It is however possible that the sample size might account for the level of significance seen in the study considering a 6.1 per 10,000 birth of MMC in the Swedish general population. Since small sample size may also limit the level of significance[21]. The pattern of significance level seen in the study may also be attributed to the five years period before birth and after. There might be room for the possibility of having more significant levels if more years were evaluated.

The peak in health care cost seen in this thesis is also in keeping with the modeling framework of the accountable care organisation (ACO) which has developed a model of health care cost with regards to targeted diseases within particular population[29]. However, it will be of great importance to be able to adopt this kind of model or pattern seen in parents of children with MMC for other chronic diseases with regards to smart health budgetary allocation at the different regions.

4.2 The Swedish Disability framework and MMC

The study revealed a progressively increased health care cost from birth up till the fourth year compared to the control population. The LSS, known as the Swedish Personal Assistance System, which was backed by law or the Swedish Disability Act has made it possible to have children with disabilities receive subsidised customised help[30]. The disability care act in

section 20 made it clear that 'If anyone under the age of 18 is cared for in a home other than her or his own by virtue of this Act, her or his parents are liable to contribute to a reasonable degree to the municipality's costs for this care[30].The implication of this act shows that as the provision of the law, the municipality is compelled to bill such parents the maintenance allowance for the care of the child in question. It is apparent with this legal framework that the health care cost by the parent of children with MMC will definitely be more at these periods. Hence, this is in keeping with the findings in this study where there was a significant increase of SEK 7645 at the year of birth and SEK 6357 after two years of birth. Though, there are no records from the data if the children were institutionalised.

4.3 Parental health challenges of children with MMC

The results from this study indicate that about 2% of the parents of children with MMC had accrued health care cost on depression and musculoskeletal disorder management. These findings are consistent with previous works where parents with children having physical disabilities are said to be reporting frequent cases of stress, anxiety and depression[31]. There seem to be several possible explanations for these findings. Of such is the fact that there is a model that suggests that the child's chronic disability is a potential stressor[32]. In another instance, the chronic nature of this deformity like MMC with accompanying delayed developmental milestones in the child is also alluded as a functional care strain (fcs) on the parents most importantly[33]. Attending life's challenges and every day's care for the child has also been seen to compound depressive episodes in parents[33,34]. The study also showed a prevalence of 2% in parents incurring health care cost in managing musculoskeletal disorder. This finding is consistent with previous findings that suggest some musculoskeletal disorder (MSD) in caregiver taking care of children with cerebral palsy[35]. However, the previous study suggested that the care giver train index seem not to show a significant connection between MSD and the care giver[36]. It seems possible that this might be due to the improved and the mechanised way of moving children with MMC and the body mass index of the parent could also be suggestive[36].

There are still enough room to evaluate a closer relationship between musculoskeletal disorders and depression in parents of children with MMC if there are no limitations to the data collected or if such variables are ab initio designed to look out for them in a primary data.

4.4 Paradigm in health care cost

The study does find that female parents significantly have a higher health care cost than their male counterparts. This difference was found to be about eight times more than what the male parent consumes. This finding was actually consistent with previous findings that suggested that health care cost in women is twenty (20%) higher in terms of per capital cost[37]. Most of these health care cost findings are also said to be attributed to mental, behavioral, and musculoskeletal disorders[38]. There is a possibility that females are more particular about their health and may be due to early life's contact in health care utilisation regarding reproductive care[39].

There are still areas to be explored regarding shifts and pattern in health care cost in an environment where most health cost are subsidised by the state[24]. It also bring to mind if the concept of equality in shared child care commonly advocated for in Sweden are really meeting the target[40]. On the other hand, it could suggest that the females are still straddled with most burden of childcare within the union especially in this study that showed a significant care cost from the female parents. This will however be worth evaluating to be able to see if there are intrinsic factors that may be looked into in future research to deeply understand the dynamics of health care cost in Sweden for equitable planning.

4.5 Methodological considerations

The study adopted a repeated cross-sectional design which utilise data from the region Skåne health data base at different points in time[41]. Unlike where a single cross-sectional data provides a snapshot at an instance, repeated cross-sectional design gives room for evaluation of change with time both at the population scale and micro-scale[41]. This design format can be useful in regression models where patterns overtime are being evaluated[42].

The data used were accessed from region Skåne. Other agency responsible for data within the region is Socialstyrelsen; the National Board of Health and Welfare in Sweden[43]. The agency is an arm of the government functioning through the ministry of health and social affairs. Their functions are to provide social services, medical services, patient safety and issues of epidemiology[44]. One of their major functions is to also collect, analyse and process data for epidemiological use[44]. The choice of evaluating the health care cost was contingent on the fact that health care costs were only available for region Skåne[43,45]. It is often the case when combining several years of huge data collections to have increase numbers of variables and cases with the selection of paramount data of interest regarding each data period [44]. It has

however been encouraged that "unique survey year-specific" identifiers of interest are used to evaluate variables on the dataset[44]. Potentially, the role of parent in cost utilisation in the management of children with MMC will be seen across a period in time, and how the pattern is being influenced within the region. This will also give health planners the advantage of seeing MMC management from the angle of the parents compared to many studies using cost utilisation in the children[27,37].

4.6 Implications and directions for future research

This project might have a far-reaching implication in policy and understanding of health care cost in parents of children with disabilities within the region Skåne.

The study shows an important health care pattern in terms of cost in parents of children with myelomeningocele within the region. This is actually in keeping with some other global study where parents incurred more cost when they have a child with one deformity or the other. However, the model developed in the study implied that female parent had more health care cost than their male counterpart.

The outcome from the study is suggestive of a deeper understanding of the dynamics and mechanism of health care cost in the parent of children with MMC. Further evaluation in terms of causality might be added to unravel if there are direct links or marked disparity and lack of equitable cost for parents of children with MMC and other deformities. All of these properly put into perspective during the study design will also be able to inform smart policy and health care programmes in terms of equitable health care in region Skåne.

4.7 Strength and limitations

This is the first attempt at looking at health care cost using a regional data base in estimating parent's health care utilisation when having a child with MMC. It will be relevant and inform health policy within the region and beyond. The burden of having a child with disability could be far reaching, and the result could shed some light in understanding the dynamics of mitigating it for the greater good of the society.

There may be some possible limitations in this study, nonetheless while bearing this in mind; the result should be interpreted with caution. One of the major limitations that are striking is the absence of socioeconomic status of the population which could have been controlled for. Others are events in the parents surrounding antenatal period. The data did not show if the women had issues relating to their health during pregnancy. Or intra-partum or any period

post-partum or other complications due to having a child with MMC which may affect the health care cost in the parent.

5 Conclusion

The findings of this study have shown a distinction in health cost in parents of children with MMC. Health care cost will remain one of the most important markers in accessing ill health and its antecedent effects in this population. The disparity in health care cost in female parents which was significant suggests a notable pattern in this population.

Future work that will look more specifically into gender sensitive health care utilisation will go a long way to inform a sound health policy reform.

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Appendix

Tables

Table I: Sample Characteristics at the year of birth

Variable	Parents of children with MMC (n=99), n (%)	Parents of children without MMC (n=705), n (%)	*p*
Gender of Parent			
Female	52 (52.2%)	367 (52.1%)	0.93
Male	47 (47.5%)	338 (47.9%)	
Gender of Children			
Female	49 (49.5%)	393 (55.7%)	0.24
Male	50 (50.5%)	312 (44.3%)	
Parent's place of birth			
Abroad	24 (24.2%)	148 (21.0%)	0.47
Sweden	75 (75.8%)	557 (79.0%)	
Age of parent at the time of child's birth	Mean in years (Range)	Mean in years (Range)	
Female	31.85 (19-49)	32.55 (18-59)	0.55
Male	33.04 (22-46)	32.45 (19-52)	

Table II: Chi Square table showing health challenges as seen in parents of children with MMC and the control group

Variable	Parents of children with MMC (n=99) (%)	Parents of children without MMC (n=705)	*p*
Musculoskeletal Disorder			
Female	2 (2%)	8 (1.1%)	0.12
Male	0 (0%)	0(0%)	
Depression			
Female	2 (2%)	8 (1.1%)	0.12
Male	0 (0%)	0 (0%)	

Table III: Comparison of health care cost between parents of children with MMC child and control group at birth, five years before and five years after birth using independent t- test.

	Mean (SD) In Swedish Kronor (SEK)		Mean difference (95 % CI)	P-value
Year of birth	**MMC child**	**Control child**		
Five years before birth	9,217 (18992)	9,805 (30765)	-588 (-7020, 8197)	0.879
Four years before birth	12,591 (24197)	10,259 (21179)	2332 (-7628, 2963)	0.387
Three years before birth	15,099 (30019)	11,805 (27459)	3293 (-9858, 3271)	0.325
Two years before birth	14,495 (25951)	10,984 (20928)	3511 (-8414, 1392)	0.160
One year before birth	16,355 (53554)	10,300 (19053)	6055 (-11557, -552)	0.031
At the year of birth	35,092 (38606)	27,447 (33842)	7645 (-14905, -385)	0.039
One year after birth	13835 (34502)	8805 (24381)	5030 (-10222, 161)	0.058
Two years after birth	14610 (36389)	8253 (19048)	6357 (-11022, -1691)	0.008
Three years after birth	14203 (26056)	10516 (21728)	3687 (-8506, 1132)	0.133
Four years after birth	11441 (20386)	10963 (21571)	478 (-5060, 4104)	0.838
Five years after birth	8084 (14401)	11273 (26994)	-3189 (-2403, 8780)	0.263

Table IV: Linear regression analysis model for health care cost (SEK) in parents of children with Myelomeningocele compared to the control group at birth, five years before and five years after birth.

Variable	5 Years before birth **Adjusted B (CI at 95%)**	4 Years before birth **Adjusted B (CI at 95%)**	3 Years before birth **Adjusted B (CI at 95%)**	2 Years before birth **Adjusted B (CI at 95%)**	1Year before birth **Adjusted B (CI at 95%)**	Year of birth **Adjusted B (CI at 95%)**	1 Year after birth **Adjusted B (CI at 95%)**	2 Years after birth **Adjusted B (CI at 95%)**	3 Years after birth **Adjusted B (CI at 95%)**	4 Years after birth **Adjusted B (CI at 95%)**	5 Years after birth **Adjusted B (CI at 95%)**
MMC Status											
Child with MMC	-660 (-8268-6949)	2142 (-3060-7342)	3524 (-2938-9986)	3694 (-1093-8481)	6053* (598-11507)	7559* (1820 - 13298)	6118* (1511-10725)	3620 (-1138-8377)	498 (-3952-4947)	-3027 (-8579-2526)	2004 (-4115-8123)
Sex of Children											
Male	-841 (-6079-4396)	3160 (-396-6716)	3398 (-1033-7828)	874 (-2336-4085)	-479* (-4154-3196)	2082 (-1709-5874)	2327 (-682-5337)	93 (-3021-3206)	3439* (428-6451)	2119 (-1719-5956)	902 (-3437-5242)
Sex of Parent											
Female	5180* (-51-10410)	8439* (4923-11955)	10290* (5871-14708)	9449* (6257-12641)	8157** (4497-11817)	42402** (38630-46174)	6899* (3888-9911)	7768** (4655-10882)	9998* (6984-13011)	6347* (2507-10188)	6790* (2437-11142)
Parent's place of birth											
Abroad	-2992 (-6160-176)	2261* (58-4465)	-93 (-2706-2519)	731 (-1184-2646)	623 (-1510-2756)	-134 (-2433-2165)	1925** (-3663-187)	144 (-1970-1682)	882 (-908-2672)	441 (-1859-2741)	588 (-1936-3112)
Age of the parent	35.37 (-504-1265)	-157 (-456.-141)	-170 (-551-212)	281* (12.80-548)	185 (-121- 492)	-92 (-410-226)	86 (-172-343)	-291* (-544- 39)	-192 (-443- 59)	286 (-48- 620)	194 (-196-584)
R^2	0.004	0.047	0.033	0.052	0.025	0.40	0.37	0.33	0.058	0.015	0.01
n	67	74	80	86	100	99	96	94	97	94	88

* < 0.05 **< 0.01

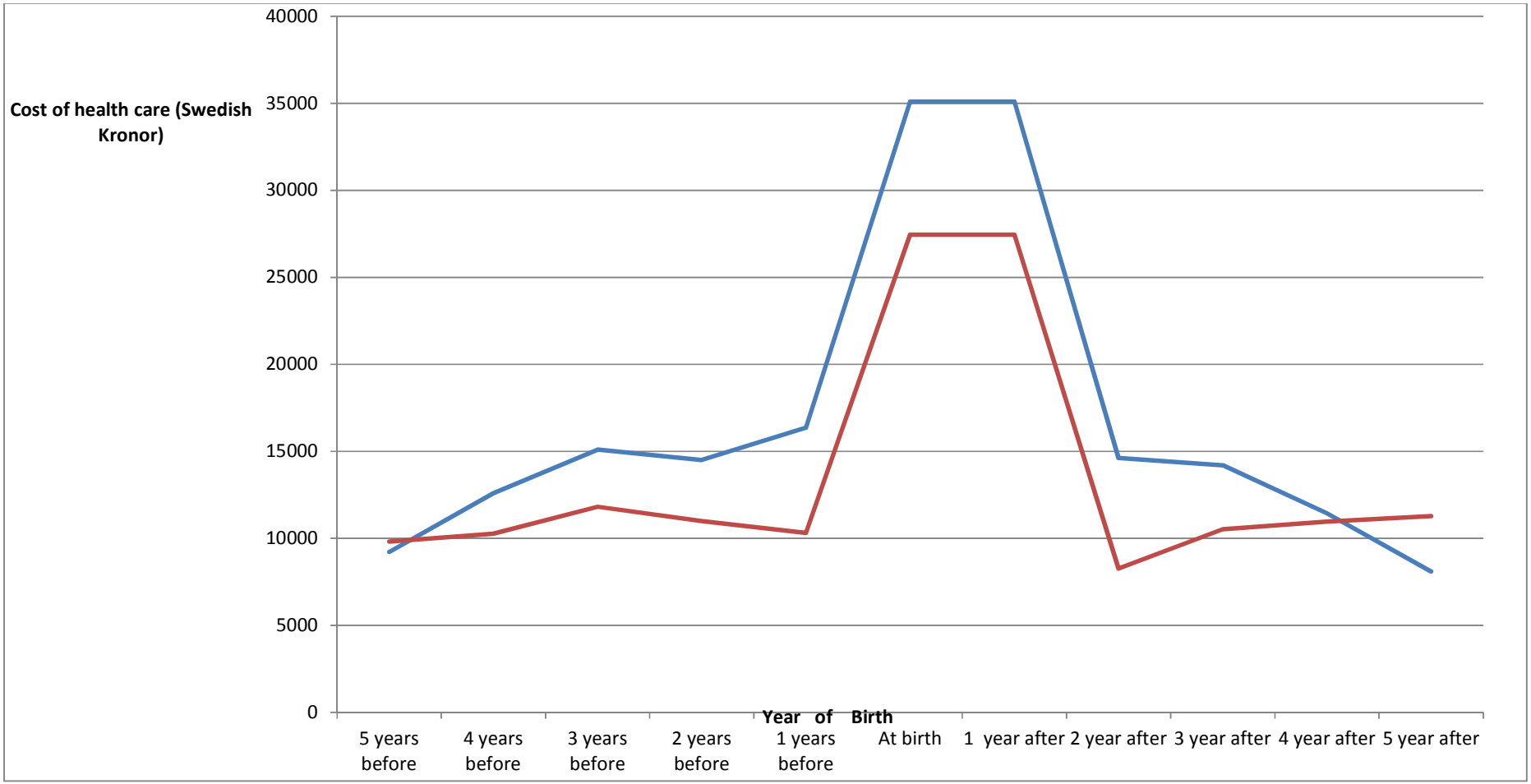
Table V: Linear regression analysis model for health care cost (SEK) showing disaggregation of the sex of parents of children with Myelomeningocele compared to the control group at birth, five years before and five years after birth.

Variable	5 Years before birth Adjusted B (CI at 95%)	4 Years before birth Adjusted B (CI at 95%)	3 Years before birth Adjusted B (CI at 95%)	2 Years before birth Adjusted B (CI at 95%)	1Year before birth Adjusted B (CI at 95%)	Year of birth Adjusted B (CI at 95%)	1 Year after birth Adjusted B (CI at 95%)	2 Years after birth Adjusted B (CI at 95%)	3 Years after birth Adjusted B (CI at 95%)	4 Years after birth Adjusted B (CI at 95%)	5 Years after birth Adjusted B (CI at 95%)
Sex of Parent											
Female	2329 (-157-4815)	3686** (984-6389)	3141* (-83-6366)	3337* (634-6040)	-1259 (-6682-4164)	12686** (9739-15633)	2317 (-1458-6091)	2812* (152-5471)	3185* (1208-5162)	2029** (-606-3451)	-716 (-3508-2077)
Sex of Children											
Male	4210 (-5132-13550)	6879 (-4070-17829)	11931 (-1018-24880)	6576 (-4277-17428)	-8293 (-30271-13685)	-3234 (-14961-8494)	12516 (-2773-27805)	5634 (-5194-16461)	-2100 (9947-5748)	60.3 (-5750-5629)	6790* (2437-11142)
Parent's place of birth											
Abroad	-1715 (-8076-4647)	1191 (-6206-8588)	-1946 (-9680-5789)	3777 (-2267-9821)	4901 (-8258-18059)	-783 (-7646-6080)	-6134 (-14354-2085)	-3642 (-11068-3785)	3158 (-1437-7753)	1587 (-1822-4995)	588 (-1936-3112)
Age of the parent	-163 (-935-608)	-475 (-1500-550)	-1151* (-2276--27)	798 (-242-1839)	834 (-966-2634)	-297 (-1286-692)	-13.3 (-1416-1389)	-280 (-1257-696)	-302 (-928-324)	399 (-111-909)	194 (-196-584)
R2	0.01	0.80	0.10	0.10	-0.02	0.45	0.08	0.08	0.14	0.12	-0.00
n	35	37	39	43	51	51	51	51	53	50	47
Constant	0.02	0.80	0.20	0.20	0.50	0.70	0.50	0.30	0.40	0.70	0.40

<0.005*
<0.001**

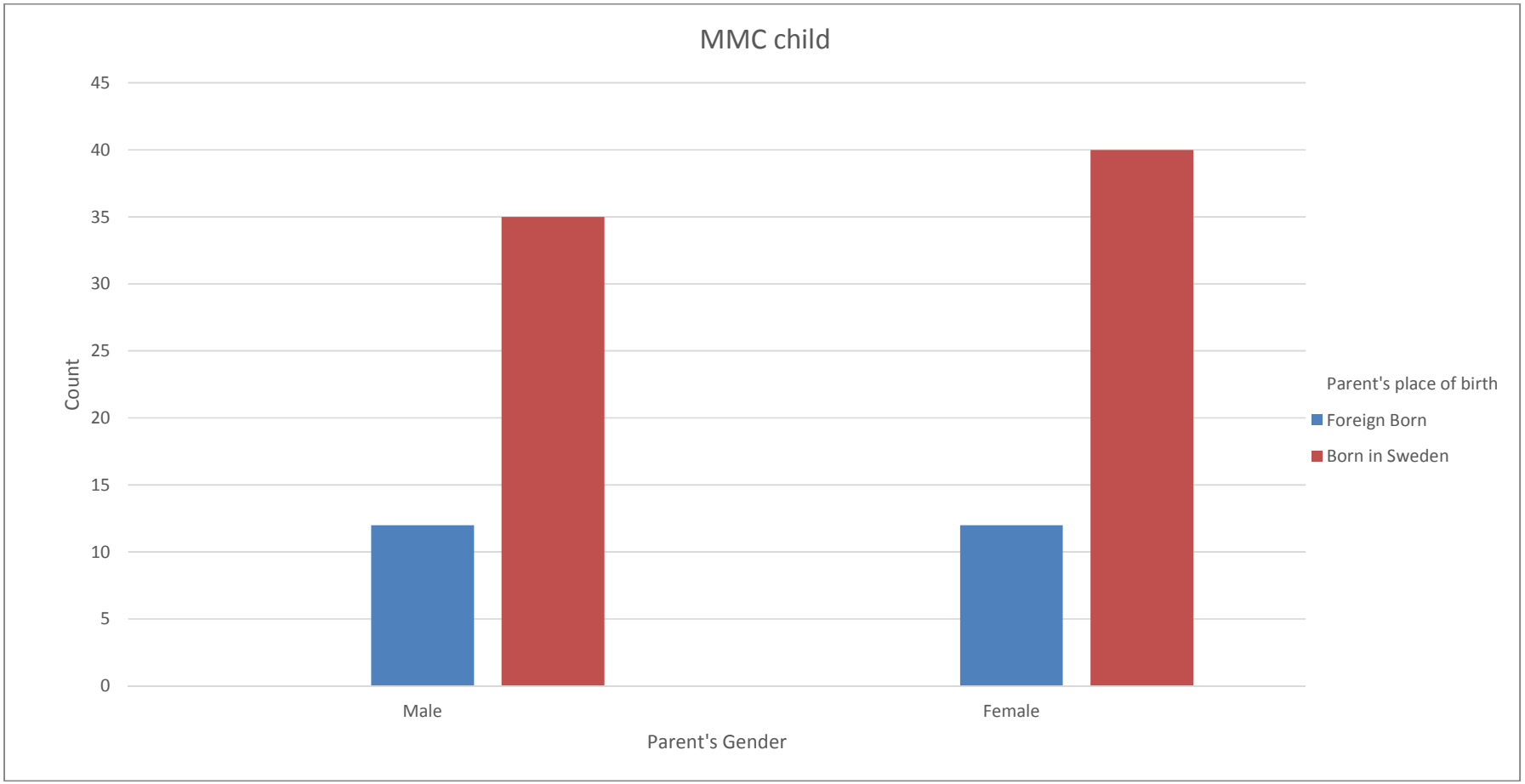
Figures

Fig I. Unadjusted Mean total health care utilisation cost (SEK) for parents of MMC (99) children and parents of control group (705).



Blue-MMC Red-Non MMC

Fig. II. Gender distribution of parents of children with MMC and place of birth.



Popular Science Summary

Children with backbone deformities (myelomeningocele) and their parents often face difficulties in accessing healthcare which are often expensive. However, in Sweden the government has put facilities in place for them. The robust data from the National board of health and Welfare in Sweden has helped this study to see a pattern in how parents of these wonderful children utilise health care cost on their health own health while taking care of their children. These spending patterns were looked into over about 11 years to be able to see how our policy makers can make the healthcare system better. The more we look, the clearer we should see.

This study revealed that parents of these children with MMC spend more than their counterparts. The health care cost difference were SEK 6,055, SEK 7,645, and SEK 6,357 higher in parents of children with MMC at one year before, at birth and two years after birth respectively. Some of these parents also have some levels of depressive episodes with body and bone problems while caring for their challenged children though not significantly different from the control group. Interestingly, the study found that female parent spends averagely 8 times more than their male peers when they have a child with MMC on their health. It is essential that this pattern seen in parents of children with back bone deformities from birth be well taken care of within the context of healthcare planning.

Finally, to create value for the money that drives healthcare, understanding of the pattern of health care cost must be known. Also, careful implementation of scientifically sound evidence will help our policy makers to create value for tax payer's money.

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