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Is congenital malformation a risk factor for caries development in Swedish adolescents?

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Abstract

Objective. The aim of this study was to investigate whether various forms of congenital malformations are risk factors for approximal caries development in Swedish adolescents. ***Materials and methods.*** This longitudinal register-based cohort study included all adolescents ($n = 18\ 142$) of 13 years of age who resided in the county of Stockholm, Sweden, in 2000. The cohort was followed until individuals were 19 years of age. Dental caries (decayed, missing and filled teeth/surfaces (DMFT/S)) were collected from the Public Health Care Administration in Stockholm. Data concerning pre- and perinatal factors and parental socio-demographic determinants were collected from Swedish National Registers. ***Results.*** In a logistic regression analysis, neither congenital malformation nor any sub-group of congenital malformation registered at birth were significantly associated with an enhanced risk of approximal caries increment in adolescents between 13–19 years of age. The final multivariate logistic regression model, adjusted for possible maternal and family socio-demographic confounders, showed that congenital malformation of the 'circulatory system' was significantly associated with a decreased risk of approximal caries increment, between 13–19 years of age (OR = 0.33; 95% CI = 0.12–0.88). ***Conclusions.*** Congenital malformation should not be considered as a risk factor for approximal caries development in Swedish adolescents today. Noticeably, adolescents with congenital heart diseases exhibited less risk of developing approximal caries, which was probably related to prevention programs allocated to these children in Sweden.

Key Words: *children, congenital heart disease, dental caries, longitudinal study, register-based study*

Introduction

A congenital disease is a condition existing at birth or that develops during the first month of life, regardless of causation. In a congenital malformation, the development of a structure is arrested, delayed or misdirected early in embryonic life. The effects are permanent and can involve many different organs including the brain, heart, lungs, liver, bones and intestinal tract. These defects can occur for many reasons, including inherited (genetic) conditions, toxic exposure of the fetus (for example to alcohol), birth injury and, in many cases, for unknown reasons [1]. In Sweden, 20 children of 1000 are registered with some kind of congenital malformation every year. Of these, 15 children of 1000 were live births and ~ five children of 1000 constitute terminated pregnancies [2].

The most common congenital malformations in Sweden are congenital heart diseases (CHD), hypospadias, Down syndrome (DS) and cleft lip and palate (CLP) [3]. There are controversial results as to whether congenital malformation in general is associated with enhanced risk of dental caries. It has been reported that children with CHD exhibit more untreated caries lesions [4–9] and higher caries prevalence in primary teeth [4,5,10] compared to healthy controls, although there are case-control studies demonstrating controversial results [7,9,11]. In subjects with permanent teeth, however, most studies demonstrate similar caries prevalence in CHD subjects and in controls [6,7,9–11].

Regarding children with CLP, a systematic review was unable to find any evidence for the relationship between CLP and increased prevalence of dental caries [12], although some studies demonstrate

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enhanced risk of dental caries in children with CLP [13–16]. Furthermore, it is well reported in scientific literature that there is lower caries prevalence in Down syndrome individuals when compared with age-matched non-DS individuals [17,18].

In Sweden, there is a well-organized social health system, established for many years, including dental services free of charge to all children between 3–19 years of age. In addition, special preventive programs are allocated to different groups, for instance children with CHD. As far as we know, caries development over time has not yet been investigated in children with different forms of congenital malformations and, consequently, it is unclear whether congenital malformation is related to an enhanced risk for dental caries in children. The purpose of this study was, therefore, to investigate approximal caries increment during the teenage period in relation to various forms of congenital malformations.

Materials and methods

The present study was designed as a retrospective longitudinal register-based cohort study and is based on information collected from data sources at the Public Health Care Administration in the county of Stockholm, as well as from Swedish National Registers at Statistics Sweden (SCB) and National Board of Health and Welfare. The study was approved by the Regional Ethical Board in Stockholm, Sweden, and the study protocol was approved by the Swedish Data Inspection Board, a federal Swedish agency that serves as an institutional review board for linkages of databases.

All adolescents (n = 18 142) aged 13 years who resided in the county of Stockholm, Sweden in 2000 were included in the study. This cohort was followed until individuals were 19 years. During this period, the subjects received regular dental check-ups either at the Public Dental Health Service, at private practitioners or at the Division of Pediatric Dentistry, Department of Dental Medicine, at Karolinska Institutet. A total of 15 538 adolescents had clinical as well as radiographic dental examinations both at ages 13 and 19 years. The sample attrition rate was 14% and the most common reason for sample attrition was that the individual had moved out of the area. Of the examined subjects (n = 15 538), information about congenital malformation or not was collected in 13 826 which, thus, constituted the final study cohort.

Population-based registers

The usefulness of registers in epidemiological research is facilitated by the Personal Identification Number (PIN), which is a 10-digit number unique to all residents and recorded in all health and census registers [19]. The PIN permits linkage to each individual between different registries. Information from the Medical Birth Register (MBR), the Total Population Register (TPR), the Total Enumeration Income Register and the Education Register was used in the present study. To protect personal integrity, the use of registers and personal identification numbers in research is strictly regulated according to legislation and regulations from the National Data Inspection Authority and requires permission from regional ethical research boards at the universities.

The medical birth register. The MBR was established in 1973 and is administered by the National Board of Health and Welfare. Its purpose is to compile information on ante- and perinatal factors and their importance for the health of the infant. One advantage of the MBR is its large database content, containing maternal and neonatal data of nearly all infants born in Sweden (coverage 97–99%) [20]. Information about pregnancy, delivery and newborn babies was collected for the register. Mothers-to-be were followed from their first visit at the public maternity healthcare clinic (usually between week 10–12), during pregnancy, at delivery, to 8–12 weeks post-delivery.

The following variables were collected from the MBR: sex, weeks of pregnancy, birth weight, parity, maternal age, smoking habits, maternal weight gain, as well as mother's height and weight at the first visit to the public maternity healthcare clinic. The Body Mass Index (BMI) was calculated based on the mother's height and weight. In the statistical analysis, all the above variables collected from MBR were dichotomized (Table I).

Concerning the variable congenital malformations, information was collected from the MBR and based on WHO's International Classification of Diseases. The subjects were categorized according to their main diagnosis of congenital malformation. Since our subjects were born in 1987, the ninth version (ICD 9) and also its basic structure were used. Thus, all the subjects' diagnoses were put together according to appropriate sub-category in the chapter entitled 'congenital malformations, deformations and chromosomal abnormalities'. However, the variable 'congenital malformations' was categorized into 12 sub-groups: Children with no congenital malformation; children with congenital malformation in the circulatory system; in the respiratory system; in the musculoskeletal system; in the digestive system; in the nervous system; in the urinary system; in genital organs; in eye, ear and neck; cleft lip and palate; chromosomal abnormalities or other congenital malformations. Since there were few subjects with congenital malformations in the respiratory system (n = 3), nervous system (n = 4) and urinary system (n = 2), these subjects were added to the sub-group of 'other congenital malformations'. Finally, the variable

Table I. Bivariate logistic regression analysis with pre-, perinatal and sociodemographic factors as exposure and approximal caries increment (ΔDMFSa > 0) as outcome.

Variables	Subject with ΔDMFSa > 0 (n = 5184)/Total number of participants (n = 13 826)	Odds ratio[a]; 95% CI	p-value[b]
Child's characteristics			
Congenital malformations			
No	5050/13,449	1.00	
Yes	134/377	0.92; 0.74–1.14	0.428
Congenital malformation sub-groups			
Circulatory system	12/69	0.35; 0.19–0.65	0.001
Musculoskeletal system	63/160	1.07; 0.78–1.47	0.681
Digestive system	4/13	0.74; 0.23–2.40	0.615
Cleft lip and cleft palate	8/16	1.66; 0.62–4.43	0.309
Eye, ear, face and neck	10/23	1.28; 0.56–2.92	0.558
Genital organs	28/57	1.61; 0.95–2.70	0.074
Chromosomal abnormalities	6/22	0.62; 0.24–1.60	0.324
Other congenital malformations	3/16	0.38; 0.11–1.35	0.135
Sex			
Girl	2908/7670	1.00	
Boy	3038/7752	1.06; 0.99–1.13	0.104
Weeks of pregnancy			
> 37	4653/12,353	1.00	
≤ 37	515/1417	0.95;0.84–1.06	0.330
Birth weight			
0–2499 g	239/621	1.05;0.89–1.24	0.568
2500–4499 g	4790/12,824	1.00	
≥ 4500 g	148/363	1.16;0.93–1.43	0.185
Parity			
1–2	4042/11,165	1.00	
≥ 3	1142/2661	1.33;1.22–1.44	< 0.001
Caries experience at 13 years			
DFT = 0	2282/7930	1.00	
DFT ≥ 1	3723/7607	2.37;2.22–2.54	< 0.001
Maternal characteristics			
Age			
< 29 years	2647/6644	1.00	
≥ 29 years	2537/7182	0.83;0.77–0.88	< 0.001
Smoked in early pregnancy			
No	3185/9058	1.00	
Yes	1411/3261	1.41;1.30–1.53	< 0.001
BMI in early pregnancy			
0–24.99	2739/7747	1.00	
≥ 25.00	483/1125	1.23; 1.08–1.39	0.001
Weight gain during pregnancy			
< 20 kg	3123/8591	1.00	
≥ 20 kg	538/1419	1.07;0.95–1.20	0.258

Table I. (*Continued*).

Variables	Subject with ΔDMFSa > 0 (n = 5184)/Total number of participants (n = 13 826)	Odds ratio[a]; 95% CI	p-value[b]
Country of birth			
Born in Sweden	3974/11,219	1.00	
Born abroad	1915/4012	1.67; 1.55–1.79	< 0.001
Marital status in 1999			
Married	3857/9662	1.00	
Unmarried	2223/5412	1.18;1.10–1.26	< 0.001
Educational level 1999			
> 12 years	1918/5883	1.00	
10–12 years	2567/6499	1.35;1.25–1.45	< 0.001
≤ 9 years	1325/2692	2.00;1.83–2.20	< 0.001
Family characteristics			
Social welfare allowance 1999			
No	5203/14,064	1.00	
Yes	735/1350	2.04;1.82–2.28	< 0.001
Income level 1999			
High income range (≤ lower 25% percentile)	4206/11,759	1.00	
Low income range (> lower 25% percentile)	1732/3655	1.62;1.50–1.74	< 0.001

[a] Odds ratios of reference categories have been set to 1.00.
[b] p-values refer to binary logistic regression analysis.

'congenital malformations' was categorized in nine sub-groups that are presented in Table II.

The total population register. Since 1968, Statistics Sweden has kept the TPR. The following variables were collected from the TPR: Country of birth and marital status in 1999. The variables were dichotomized (Table I) in the statistical analyses.

The total enumeration income register. Data on individuals' annual income tax, founded on income tax returns and tax authority decisions, are collected by the National Swedish Tax Board. The board then sends summary statistics to SCB. The information is further processed to create variables such as income from employment and disposable income, which can then be assembled on the basis of information on certain relationships and presented on a family and

Table II. Approximal caries experience at 13 and 19 years and approximal caries increment (ΔDMFSa) between 13–19 years of age in relation to congenital malformations or not (n = 13 826).

Caries indices	DMFSa, 13 years Mean/SD	DMFSa, 19 years Mean/SD	ΔDMFSa increment (13–19 years) Mean/SD
Healthy (n = 13 449)	0.30/0.88	1.51/2.93	1.21/2.68
Congenital malformation (n = 377)	0.27/0.88	1.58/3.21	1.32/2.91
Congenital malformation sub-groups			
the circulatory system (n = 69)	0.23/0.73	0.42/1.14	0.19/0.65
the musculoskeletal system (n = 160)	0.36/1.14	1.81/3.48	1.45/3.07
the digestive system (n = 13)	0.46/0.97	1.69/3.38	1.23/2.80
cleft lip and cleft palate (n = 16)	0.00/0.00	2.00/2.78	2.00/2.78
eye, ear, face and neck (n = 23)	0.22/0.52	2.78/4.51	2.57/4.36
genital organs (n = 57)	0.26/0.64	2.25/3.93	1.98/3.70
other congenital malformations (n = 17)	0.06/0.25	0.56/1.50	0.50/1.51
chromosomal abnormalities (n = 22)	0.00/0.00	0.95/1.68	0.95/1.68

D, decayed; M, missing; F, filled; T, teeth; S, surfaces; a, approximal.

individual basis. Information is also available about the receipt of both social welfare allowances. From this register, information was collected regarding the family's receipt of social-welfare allowance, as well as disposable income on a family basis in 1999. Both variables were dichotomized and family income was expressed as low income (less or equal to the lower 25% income percentile, which corresponds to ~ 253 000 SEK in 2010) or high income (higher than the lower 25% income percentile) (Table I).

The education register. This register was established by SCB in 1985 and is annually updated with information about the highest formal education attained by each individual, from elementary to post-graduate level. From this register, data was collected about mothers' educational level in 1999. In the statistical analysis, educational level was categorized according to years of schooling; as: low (≤ 9 years), medium (10–12 years) and high (> 12 years) (Table I).

Data collection concerning dental caries

In Stockholm, data on manifest caries lesions (based on clinical and radiographic examination) in children and adolescents are sent to the Public Health Care Administration from the Public Dental Health Service, private practitioners and the Division of Pediatric Dentistry, Department of Dental Medicine, at Karolinska Institutet and analyzed at ages 3, 7, 13 and 19 years. Since 2000, all these data have been linked to the PIN. The registration sheets consist of the following caries counts: DT, FT, DSa (a = approximal) and FSa. Manifest caries was recorded on smooth surfaces as the minimal level that can be verified as a cavity and detectable by probing and, in fissures, by a catch of the probe under slight pressure. Approximal caries on the radiographs was recorded as manifest caries when the lesion clearly extended into the dentin. Caries development was calculated as the approximal caries increment between ages 13 and 19 years (i.e. the difference between the DMFSa values at age 13 and 19 years, respectively) and was subsequently dichotomized to distinguish between subjects (n = 5184) with approximal caries increment (ΔDMFSa > 0) and subjects (n = 8642) without approximal caries increment (ΔDMFSa = 0). The dichotomized approximal caries increment was further used as the outcome variable in logistic regression analyses.

The dental caries file of the study cohort was sent from the Public Health Care Administration to the SCB where the data file was linked with registers containing information on probands' relevant socio-demographic factors. The key to identifying individuals was kept within SCB and not disclosed to investigators.

Statistical analysis

Data analyses were carried out using the Statistical Package for the Social Sciences (SPSS, version 18.0). Statistical analyses included frequency tables, cross tables, non-parametric test, bivariate- and multivariate logistic regression analyses. The non-parametric test 'Kruskal-Wallis test' was used to study differences in distribution concerning approximal caries increment (ΔDMFSa) in sub-groups of congenital malformations compared to healthy subjects. The odds ratios (OR) with 95% CI were used to estimate the effects of 'congenital malformations' exposure. The outcome variable, approximal caries increment (ΔDMFSa) between 13–19 years of age, was dichotomized in subjects with approximal caries increment (ΔDMFSa > 0) and subjects without.

To analyze whether 'congenital malformations' exposure was a risk factor for approximal caries increment during the teenage period, the variable was adjusted for potential confounders in different models. All the variables were classified (as shown in Table I) and then entered into models in different groups and adjusted multiple logistic regression analyses were performed for each group of determination to evaluate whether congenital malformations were associated with the outcome. In Model I, no adjustments were made. In Model II, adjustment was made for the child's total caries experience at 13 years of age. In Model III, adjustments were made for child characteristics: sex, parity, birth weight and weeks of pregnancy. For Model IV, adjustments were made for maternal characteristics: Age, marital status, country of birth, educational level, smoking in early pregnancy, BMI in early pregnancy and weight gain during pregnancy. In Model V, adjustments were made for family characteristics: Income and receiving social welfare allowance. Finally, in Model VI, adjustments were made to include all sub-groups.

Results

In Table II, caries experience at ages 13 and 19 years and caries increment between these ages are described in relation to the occurrence of various forms of congenital malformations registered at birth. Of the cohort (n = 13 826), 37.5% (n = 5184) exhibited an approximal caries increment (ΔDMFSa > 0) during the follow-up. Of these, 5050 were born healthy and 134 with some kind of congenital malformation. Adolescents with congenital malformations in the sub-groups 'eye, ear, face and neck', 'cleft lip and cleft palate' and 'genital organs', with a mean value of 2.57, 2.00 and 1.98, respectively, exhibited the highest approximal caries increment. The mean approximal caries increment (ΔDMFSa) in children, born healthy, was 1.21, compared to 1.32 for children with some kind of congenital malformation. The lowest

Table III. Logistic regression analysis with caries increment (ΔDMFSa > 0) between 13–19 years of age as outcome and congenital malformations as exposure.

Variables	Model 1 OR (95% CI)	Model II OR (95% CI)	Model III OR (95% CI)	Model IV OR (95% CI)	Model V OR (95% CI)	Model VI OR (95% CI)
Congenital malformations						
No	1.00	1.00	1.00	1.00	1.00	1.00
Yes						
Circulatory system	0.35 (0.19–0.65)	0.35 (0.19–0.65)	0.34 (0.18–0.63)	0.36 (0.13–0.94)	0.36 (0.19–0.67)	0.33 (0.12–0.88)
Musculoskeletal system	1.07 (0.78–1.47)	1.06 (0.76–1.46)	1.08 (0.79–1.49)	0.91 (0.58–1.43)	1.08 (0.79–1.49)	0.94 (0.59–1.50)
Digestive system	0.74 (0.23–2.40)	0.69 (0.21–2.27)	0.74 (0.23–2.42)	1.29 (0.31–5.44)	0.79 (0.24–2.58)	0.98 (0.23–4.21)
Cleft lip and cleft palate	1.66 (0.62–4.43)	1.91 (0.72–5.10)	1.65 (0.62–4.42)	2.19 (0.72–6.63)	1.76 (0.66–4.70)	2.31 (0.74–7.20)
Eye, ear, face and neck	1.28 (0.56–2.92)	1.27 (0.55–2.93)	1.42 (0.61–3.29)	0.98 (0.28–3.40)	1.29 (0.56–2.94)	1.08 (0.31–3.78)
Genital organs	1.61 (0.95–2.70)	1.60 (0.94–2.71)	1.59 (0.94–2.68)	1.81 (0.94–3.47)	1.59 (0.94–2.69)	1.59 (0.81–3.11)
Chromosomal abnormalities	0.62 (0.24–1.60)	0.72 (0.28–1.83)	0.64 (0.25–1.63)	0.62 (0.17–2.28)	0.61 (0.24–1.56)	0.64 (0.17–2.37)
Other congenital malformations	0.38 (0.11–1.35)	0.41 (0.12–1.46)	0.39 (0.11–1.39)	0.22 (0.03–1.74)	0.40 (0.12–1.42)	0.23 (0.03–1.84)

Model I = Unadjusted; Model II = Adjusted for child's caries experience at 13 years; Model III = Adjusted for child's characteristics (sex, birth weight, weeks of pregnancy and parity); Model IV = Adjusted for maternal characteristics (age, country of birth, smoking in early pregnancy, BMI in early pregnancy, weight gain during pregnancy, educational level and marital status); Model V = Adjusted for family characteristics (income and social welfare allowance); Model VI = Adjusted for caries experience, child's characteristics, maternal characteristics and family characteristics.

mean value of approximal caries increment was found among subjects born with a congenital malformation in the circulatory system, 0.19 (Table II). The results of the non-parametric test demonstrated a significant difference in the distribution of approximal caries increment compared to healthy adolescents only for the sub-group 'congenital malformation in the circulatory system' ($p \leq 0.001$). No significant differences were found for other sub-groups of congenital malformations.

Bivariate analysis

The bivariate logistic regression analysis with approximal caries increment between ages 13–19 years as a dependent variable (ΔDMFSa > 0), did not demonstrate any significant association between congenital malformation (OR = 0.92; 95% CI = 0.74–1.14) and approximal caries increment during the teenage period (Table I). Concerning the sub-groups, only congenital malformations in the circulatory system exhibited a significant association with the outcome (OR = 0.35; 95% CI = 0.19–0.65). The analysis demonstrated that children with CHD exhibited less risk of developing approximal caries lesions during the teenage period compared to healthy children (Table I). In addition, the following potential confounders were also significantly associated with the outcome: Caries experience at 13 years, parity, maternal age, smoking in early pregnancy, BMI in early pregnancy, country of birth, marital status, educational level, income and receiving social welfare allowance (Table I).

Multivariate analysis

The logistic regression analysis concerning 'congenital malformations' exposure and the outcome 'approximal caries increment between 13–19 years of age' fitted a multilevel model, adjusting for potential confounders (Table III). In Model I with no adjustments, the results showed that the sub-group 'congenital malformations in the circulatory system' was significantly associated with ΔDMFSa > 0 between 13–19 years of age (OR = 0.35; 95% CI = 0.19–0.65). Although 'total caries experience at 13 years of age' was entered into Model II to control possible confounding, 'congenital malformation in the circulatory system' was still significant with the outcome (OR = 0.35; 95% CI = 0.19–0.65). In addition, no more sub-groups of congenital malformations were significantly associated with ΔDMFSa > 0. Model III was adjusted for possible confounders of the child-related variables; 'gender', 'parity', 'weeks of pregnancy' and 'birth weight'. Despite the adjustments, the sub-group 'congenital malformations in the circulatory system' was still significantly associated with ΔDMFSa > 0 (OR = 0.34; 95% CI = 0.18–0.63). In Model IV, adjustments were

performed related to maternal characteristics (age, marital status, country of birth, educational level, smoking in early pregnancy, BMI in early pregnancy and weight gain during pregnancy) and, in Model V, adjustments were performed related to family characteristics (income and receiving social welfare allowance). Although this study adjusted for possible maternal and family socio-demographic confounders, the significant association was unchanged. Finally, in Model V, all groups of potential confounders were entered and, despite this, the sub-group 'congenital malformations in the circulatory system' still exhibited a significant decreased risk of ΔDMFSa > 0 (OR = 0.33; 95% CI = 0.12–0.88).

Discussion

The novel finding in this register-based longitudinal cohort study reveals that children with congenital malformations registered at birth do not represent a group with enhanced risk of developing approximal caries during adolescence. Noticeably, children with congenital malformations of the circulatory system exhibited decreased risk of developing approximal caries lesions.

There is a great advantage of using population-based registers compared with common dental health surveys when identifying various risk factors for dental caries [21,22]. In this study we used the MBR register as our source of information. An advantage of using MBR is that it covers ~ 97–99% of deliveries in Sweden [21]. The quality of MBR has been evaluated three times, in 1976, 1998 and 2001, with the conclusion that it is a valuable source of information for reproduction epidemiology [23,24]. The validity of information on pre-natal factors used in the statistical analyses is probably accurate since the information was collected prospectively before birth. Furthermore, all information about the family was collected independently of the study outcome, which reduced problems with recall and interviewer bias. In addition, it was possible to adjust for these potential confounding factors in the statistical analyses. However, we were not able to include information on dietary or oral hygiene habits in the present study since this information is not available in any Swedish national registers. This is a limitation of our study and, consequently, we were unable to evaluate these variables describing lifestyle, oral attitudes and behaviors with respect to their influence and interactions on dental caries development.

Concerning the diagnosis of congenital malformations, information was collected based on WHO's international classification of diseases (ICD). We used the version of ICD 9 including the basic structure since our subjects were born in 1987. The basic structure of ICD is founded on the standardization of the nomenclature for the names of diseases and their basic systematization in the hierarchically structured category [25]. In the scope of ICD, all fields of medicine are represented and the nomenclature and classifications are essential parts of scientific methodology in healthcare [25].

In register-based studies, there is a risk of random errors because the diagnosis of manifest caries can sometimes be under- or over-reported due to several examiners. However, random errors are affected by increasing the size of the study, and will be reduced to zero if a study becomes infinitely large [26]. In this study, the final study cohort consisted of 13 826 children and the risk of random error is, therefore, of minor importance.

An analytic multi-level statistical model was used to investigate whether congenital malformation at birth should be considered as a risk factor for approximal caries development during adolescence. In this model, we adjusted for potential socio-demographic confounders since dental caries is multi-factorial and prone to socio-demographic inequalities [27]. The child's caries experience at 13 years of age was also adjusted for, since caries prevalence/experience is a valid predictor of future caries [28]. After such adjustments, the present study reveals that children with congenital malformation do not represent a group of children with enhanced risk for dental caries. One has to consider that children with congenital malformation represent a heterogenous group of subjects, which means that within the group it might be subgroups of patients with enhanced or less risk for caries development.

When sub-grouping the subjects, we demonstrated that children born with CHD exhibited a decreased risk of approximal caries increment between 13–19 years of age compared to healthy children. Unfortunately, we are not able to compare our results with other studies in literature since longitudinal cohort studies are lacking. However, previous case-control studies demonstrate no significant difference of caries prevalence in permanent teeth between CHD subjects and controls [6,7,9–11]. One has to consider, however, that there are studies demonstrating higher caries prevalence in primary teeth of CHD children compared to controls [4,5,10].

The reason for the contradictory result may be related to the fact that special preventive programs have been allocated to Swedish children with CHD since ~ 20 years ago. These children are recommended to be referred to a specialist in pediatric dentistry already when the child is 1 year of age for caries risk assessment including the preventive program [29]. The enhanced caries risk reported among pre-school children with CHD might be due to parents' lack of resources and adequate attention to their children's oral health (i.e. poor dietary and oral hygiene habits) during the first years of life due to long periods in hospital with surgery and illness. In

addition, being the parent of a child with CHD has been associated with problems such as sadness, anxiety and anger [30]. In light of the advanced prevention programs allocated to children with CHD in Sweden over a long period of time, the result in this study is well compatible with the view that children with CHD in Sweden demonstrate lower dental caries development during adolescence compared to controls. The lower caries development in children with CHD, during the teenage period found in our study, indicates that Swedish dental health services take care of this group of patients with congenital heart diseases efficiently with respect to caries development. However, one has to take into account that the results are representative for Sweden and cannot be generalized globally.

Additional explanations for the contradictory results regarding caries risk of CHD subjects in our study might be that most previous studies are dealing with severe heart diseases, whereas, in our study, all subjects with CHD diagnosed at birth are included. In addition, in most studies, the total number of cases with heart disease was included independently of when the diagnosis was settled. The relatively low number of subjects with CHD ($n = 69$) in our study compared to the incidence of CHD in Sweden (8–10/1000 live births) is explained by the fact that the co-morbidity children such as those with Down syndrome, Turner syndrome, 22q11 and Noonans syndrome, are categorized in their main diagnosis group and consequently not included as CHD subjects in the analysis.

The present study is the first longitudinal investigation and unfortunately we are not able to compare our results with other studies. Since our study is a longitudinal register-based cohort study with a high number of participants, we therefore assume that the novel finding has high validity.

A limitation in this study was that information about the use of medical products was lacking and, therefore, adjustments for medicines taken were not included in the statistical analytic model.

Regarding other forms of congenital malformation, no significant associations, neither negative nor positive, in relation to approximal caries development during the teenage period could be demonstrated. Also in this context, a systematic review concluded that there was no evidence for the assumption that CLP children have increased prevalence of dental caries [13], although significant positive association has previously been reported in children with cleft lip and cleft palate [14–16]. Children with Down syndrome are reported to exhibit lower risk of dental caries compared to controls [17,18]. However, in this study, the sub-group 'chromosomal abnormalities' did not demonstrate decreased risk of approximal caries development during the teenage period compared to subjects without congenital malformations.

In conclusion, children with congenital malformations do not represent a group with enhanced risk of caries development during adolescence in Sweden. Noticeably, children with congenital malformations of the circulatory system exhibited less risk of developing approximal caries lesions compared to healthy adolescents between 13–19 years of age, which is probably related to the fact that prevention programs have been allocated to these children over a long period of time.

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