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Prevalence and distribution of developmental enamel defects in children with cerebral palsy in Beijing, China

XIAOBO LIN[1], WEIHONG WU[1], CHENGFEI ZHANG[2,3], EDWARD C.M. LO[2], CHUN HUNG CHU[2] & WARUNA L. DISSANAYAKA[2]

[1]Beijing Boai Hospital, China Rehabilitation Research Center, Beijing, [2]Faculty of Dentistry, The University of Hong Kong, Hong Kong SAR, and [3]School of Stomatology, Peking University, Beijing, China

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Aim. To investigate the prevalence and distribution of developmental enamel defects in children with cerebral palsy (CP) in Beijing, China.

Design. A total of 135 children aged 1.5–6 years with moderate or severe congenital CP diagnosed in Beijing Boai Hospital from year 2005 to 2009 were recruited. The children underwent dental examination at the hospital dental clinic.

Results. Enamel defects (opacity and/or hypoplasia) were found in 44 (32.6%) out of 135 CP children. Enamel hypoplasia was found in 35 (25.9%) of the CP children, opacity alone was found in 5 (3.7%) of the CP children, and mixed defects (opacity and hypoplasia) was found in 4 (3.0%) of the CP children. Most of the enamel defects were located symmetrically in the primary incisors and first molars. 42.4% of children with enamel defects were born prematurely (<37 weeks) where as only 23.2% of them were born at normal gestational age. No statistically significant difference in the prevalence of enamel defects was found in relation to birth weight ($P > 0.05$).

Conclusions. A high prevalence of developmental enamel defects was found among the children with CP. The prevalence of defects varied with the tooth type and was associated with gestational age of the children.

Introduction

The development of enamel in the primary teeth begins in the fourteenth week of intrauterine life and completes at the end of the first year of life[1]. The developing tooth germ is sensitive to a wide range of systemic disturbances and is unable to recover once it is damaged. Thus, the occurrence of enamel defects in primary teeth is likely to be related to injuries affecting ameloblasts or enamel maturation in the pre-, peri- and postnatal period of human development[2]. Systemic factors associated with developmental defects of enamel in primary teeth include: ingestion of chemicals such as fluorides, tetracycline, and thalidomides; prematurity/low birth weight; severe malnutrition, neonatal hypocalcaemia, vitamin D deficiency; deprivation of sunlight; hyperbilirubinemia; thyroid and parathyroid disturbances; maternal diabetes; neonatal asphyxia; certain viral infections; and genetic disorders such as amelogenesis imperfecta and tuberous sclerosis[3]. Local traumatic factors may also be responsible for some of the enamel defects. A study has found children who had undergone orotracheal intubation and mechanical ventilation during perinatal and postnatal period tended to suffer more defects on the maxillary teeth compared with non-intubated children[4]. Therefore, pregnancy and delivery are critical periods, imposing a number of changes that may modify the activities of the ameloblasts in the foetus.

Cerebral palsy (CP) is the most prevalent physical disability in childhood, and in the Western countries, the overall prevalence of CP is reported to be about 2 per 1000 live births[5]. In China, the overall prevalence of cerebral palsy is reported to be 1.6 per 1000 children aged below 7 years, with a higher prevalence of 9.7 per 1000 children who were

Correspondence to:
C. Zhang, 3A15, Prince Philip Dental Hospital, 34, Hospital Road, Hong Kong SAR, China.
E-mail: zhangcf@hku.hk

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born as multiples, which is six times higher than that of children born as singletons[6]. Such brain damage can develop during the delivery period because of disturbed circulation in the umbilical cord, premature birth, head injuries, or viral infection. One of the most common causes of brain damage leading to CP is hypoxia that may occur before, during or shortly after birth[7].

Cerebral palsy refers to a group of disorders of movement and posture that are attributed to nonprogressive disturbances that occurred in the developing foetal or infant brain[8,9]. Causes of CP are numerous, and the aetiology is multi-factorial. Risk factors are categorised by the timing of their proposed occurrence: prenatal, perinatal, and postnatal. The leading prenatal and perinatal risk factors for CP are low birth weight and premature birth. Other risk factors include neonatal encephalopathy, multiple pregnancy, infection and inflammation, and a variety of genetic factors[9,10]. The reported prevalence of CP was 0.2–0.4%; it increases with lower birth weight and higher immaturity. Increase in survival rate of infants after preterm birth has also led to an increase in CP rates. The outcome with respect to CP in the group of extremely low birth weight or immature infants remains a matter of specific concern, as prevalence seems to be rather stable at a high level[11].

Clinicians have long noted greater prevalence of disturbed enamel formation in young children with cerebral palsy or certain other congenital neurologic disorders[3]. The prevalence of enamel defects in primary dentition varies and is related to other factors, such as types of cerebral palsy, birth weight, gestational age, and the child's age at examination in the study[12]. Results of the few published studies have shown that the prevalence of enamel defects in the primary dentition of children with CP varies from 24% to 60%[3,12]. The prevalence of cerebral palsy has changed in the last 30 years; it has increased substantially among very preterm infants in association with and possibly as a consequence of large declines in infant mortality[5,8]. There is a lack of recent study on the prevalence of enamel defects among CP children. Information on Chinese population with CP is also not available currently.

The aim of this study was to evaluate the prevalence and distribution of enamel defects in primary teeth in Chinese preschool children with cerebral palsy and to explore its possible association with lower birth weight and preterm birth.

Materials and methods

The study population consisted of all the children aged 1.5–6 years with moderate or severe congenital CP diagnosed in the Beijing Boai Hospital (affiliated to the China Rehabilitation Research Center) from year 2005 to 2009. This study was approved by the Beijing Boai Hospital review board. Parental consent was obtained before the children were recruited in the study.

The medical history of each child, including the type of CP, birth weight, and gestational age, was retrieved from medical records in the Beijing Boai Hospital. All teeth were examined by a calibrated examiner. The teeth were cleaned with gauze, and examined in wet condition with a mirror and blunt probe. The types of enamel defects were recorded as demarcated opacities, diffuse opacities, and hypoplasia, using the developmental defects of enamel (DDE) index[13,14].

Statistics

The data were analysed using statistical software (SPSS 17.0, SPSS Inc., Chicago, IL, USA). The relationships between the dependent variable (enamel defects) and the independent variables were evaluated using chi square test (gender, gestational age, birth weight), fisher exact test (type of CP) and reconfirmed using logistic regression analysis. A probability value of less than 0.05 was considered statistically significant. A 95% confidence interval was used for the comparisons of different results within subgroups.

Results

Characterisation of subjects

No refusal was encountered and a total of 135 CP children were examined, 67.4% (91)

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were boys and 32.6% (44) were girls. Their age was from 1.5 to 6 years. The mean (±SD) age was 3.6 ± 1.2 years. Out of the 135 CP cases examined, 101 (74.8%) were spastic CP, 9 (6.7%) were athetoid CP, and 25 (18.5%) were mixed type CP.

Prevalence of hypoplasia, opacity alone and mixed defects (opacity and hypoplasia) in CP children

In the present study, the main defect found was enamel hypoplasia. It was found in 35 (25.9%) of the CP children. Enamel opacity alone was found in 5 (3.7%) of the CP children, and mixed enamel defects (opacity and hypoplasia) were found in 4 (3.0%) of the CP children.

Prevalence of enamel defects in CP patients

Enamel defects (opacity and hypoplasia) were found in 44 of the 135 CP children. They were found in 30 of the 91 male CP children, compared with 14 of the 44 female CP children. The prevalence in the two gender groups were almost similar (33.0% *vs* 31.8%, $P > 0.05$).

Prevalence of enamel defects in different types of CP

Enamel defects were found in 29 of the 101 spastic CP (28.7%) children, 4 of the 9 athetoid CP (44.4%) children, and 6 of the 25 mixed type CP (24.0%) children. The difference in the prevalence among the three CP subgroup children was not statistically significant ($P > 0.05$).

Effects of preterm birth and low birth weight on enamel defects of CP children

Out of the 135 CP children, 66 were preterm births (gestational age <37 weeks) and 69 were of normal gestational age (≥37 weeks). More enamel defects were found in children born prematurely (42.4%) than in children born at normal gestational age (23.2%) ($P < 0.05$). Table 1 shows that among the 53 cases with birth weight lower than 2.5 kg, 20 (37.7%) had defective teeth, while among the 79 cases weighing 2.5 kg or more at birth, 23 (29.1%) had defective teeth. There was no statistically significant association between DDEs and birth weight.

Table 1. Number of CP children with DDE according to selected factors.

	Normal Enamel	Defective Enamel	Total	*P*-value (Chi square)
Gender				
Male	61	30	91	0.894
Female	30	14	44	
Type of CP				
Spastic	67	34	101	0.480
Athetoid	5	4	9	
Mixed	19	6	25	
Gestational age				
<37 weeks	38	28	66	0.017
≥37 weeks	53	16	69	
Birth weight				
<2.5 kg	32	21	53	0.161
≥2.5 kg	59	23	82	

CP, cerebral palsy; DDE, developmental defects of enamel.

The distribution of enamel defects in the primary teeth of CP patients

Most of the enamel defects were located in the anterior teeth and first molars and were distributed symmetrically. The prevalence is slightly higher in the upper teeth than in the lower teeth. Overall, more than half of the enamel hypoplasia found was either in the maxillary incisors (37.7%), or in the mandibular incisors (26.8%) while around a quarter of the enamel hypoplasia were in the maxillary first molars (14.5%) or the mandibular first molars (10.4%). Opacity could be seen mostly located in upper and lower anterior teeth.

Logistic regression analysis

Logistic regression analysis was performed to overcome the confounding and nonsense factors. The final regression model revealed that only gestational age out of the four factors has a significant effect on enamel defects of children with CP (Table 2).

Discussion

This study used a cross-sectional design which involved CP children treated at the Beijing

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Table 2. Final model of logistic regression.

Factors	*β*(SE)	Odds ratio (95% CI)	*P*-value
Gestational age	−0.89 (0.38)	2.44 (1.16–5.13)	0.018
Constant	−0.30 (0.25)		0.220

Boai Hospital affiliated to the China Rehabilitation Research Center. The study enrolled a convenience sample of 135 children with CP. Beijing Boai Hospital being one of the major tertiary care centers in China; it recruits patients from all over the country. Patients with CP treated in a major hospital would likely to have better accessibility to health services. Therefore the results drawn in this study may not be representative of children with CP in China, but it at least provides insight into the status of the enamel defects of children with CP.

Defects in the structure of enamel can develop in the pre-, peri-, and postnatal periods. Severity of the enamel defects depends on the intensity, time and period of the aetiological factors influenced. More than 90 different factors have been found to be associated with developmental defects in the enamel of the primary and permanent dentition.[13] According to the previous studies, among the numerous factors associated with developmental defects in enamel of the primary teeth, prematurity and low birth weight have achieved particular prominence. The prevalence of enamel defects in primary dentition is significantly influenced by birth weight, gestational age and several systemic factors[15,16]. The reported prevalence of enamel defects in CP children varies from 24–68%[3,12]. In this study, enamel defects were found in 32.6% of the CP children. A similar prevalence in CP children was found in a previous study as well[12]. Defects in enamel can cause retentive areas which favour the accumulation of bacterial plaque, while hypocalcification can lead to a more rapid progression of carious lesions[13]. Furthermore, it is difficult for the children with CP to perform good plaque control practises by themselves. Thus parental assistance in tooth brushing and the use of fluoridated toothpaste are important measures in keeping good oral hygiene and preventing dental caries in children with CP.

In agreement with some previous studies[3,12], hypoplasia was the most frequent type of enamel defect found in this sample of Chinese CP children. In contrast, opacity was found to be the most frequent type of enamel defect in some other studies of non-CP children[2,13,17]. Enamel hypoplasia might be because of the disturbance of dental development, which consists of different phases, such as initiation of the dental lamina, histomorphogeneis, cytodifferentiation of ameloblasts and odontoblasts, and synthesis and secretion of ameloblast or odontoblast matrix proteins[18,19]. Formation of enamel opacity might be associated with injuries during calcification and maturation stages, which are the later stages of enamel formation resulting in qualitative changes[19].

In agreement with other studies[13,20], in this study it was found that enamel defects in the primary dentition were located mainly in the central and lateral incisors and first molars in the upper jaw. The tooth defect pattern suggests that there is a critical time period in which these teeth are most susceptible to develop enamel defects. Theoretically, in an 8-month foetus, enamel matrices of maxillary and mandibular anterior teeth are almost completely formed, and calcification is just beginning at the cusp tip of those teeth. At birth, the enamel matrices of an infant are fully formed, most of the enamel in anterior teeth and part of the enamel in primary first and second molars are calcified. Slower calcification has been observed in maxillary primary teeth than in mandibular primary teeth[20]. For example, if a disturbance occurs at 13th week in uterus, a hypoplastic lesion will appear on both maxillary and mandibular central incisors. If disturbance occurs at birth, all of the teeth will be affected to a certain degree[1]. The prevalence of enamel opacities was very low in this study (3.7%) and could be seen in nearly all teeth, though mostly in anterior teeth. In contrast the prevalence of hypoplasia was as high as 25.9%. This clinical evidence is strongly suggestive of disturbances in calcium homeostasis and/or direct injury to the ameoblasts associated with systemic disturbances. Selective involvement of only those ameoblasts that are active at the time of

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particular disturbance, some may die and stop secreting enamel, whereas others may recover and continue to secrete normal enamel over the defective spots, may account for the variability in development of the lesion[21].

Short duration of gestation and low birth weight, two highly but imperfectly correlated measures, are markers of neurological disorders. Premature birth is a major risk factor for cerebral palsy while the degree of the vulnerability to cerebral palsy may be related to the cause of the prematurity[3]. Prematurely born children show an increased prevalence of developmental defects in enamel in the primary dentition[22]. Our study showed that the CP children who were born prematurely had higher prevalence of enamel defects than that of the CP children born in full term. Logistic regression analysis confirmed the significance of this by identifying gestational age as the only factor that has a significant effect on enamel defects in children with CP out of all analysed factors. The exact mechanism and etiological factors underlining these enamel defects are not fully understood. The breast milk containing too little calcium and phosphorus for preterm infants, metabolic bone disease of prematurity with inadequate calcium and phosphorus supply, and deprivation of normal mineral stores can be important factors behind dental developmental defects in children born prematurely[16,23]. In this study, the prevalence of enamel defects was greater in the low birth weight group than in the children of normal group. The difference was not statistically significant, contrasting with some studies in which enamel defects were significantly more prevalent in low birth weight children[4,24], but agreeing with the result of one other study[13]. Very low-weight newborns normally present with a series of health problems such as pulmonary, cardiovascular, gastrointestinal, hepatic, immunological and renal immaturity as well as reduced metabolic reserves, which can affect the development of the dental tissues[25].

Conclusion

In conclusion, a high prevalence of developmental enamel defects was found among the children with CP. The main type of defect identified was enamel hypolasia. These defects were located mostly in incisors and first molars, and found to be associated with gestational age of the children.

What this paper adds

- This paper adds information on prevalence of enamel defects among Chinese children with CP, which was not available up to date.
- Prevalence of enamel defects in the primary teeth of children with CP in this study was 32.6%, and enamel hypoplasia was the most frequent type (25.9%) of defects identified.

Why this paper is important for paediatric dentists

- The high prevalence of enamel defects in children with CP demands special attention from the paediatric dentist in preventive and restorative dental care.

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