The prevalence of malocclusion in children with cerebral palsy

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SUMMARY The prevalence of malocclusion in children with cerebral palsy was studied by comparison with a normal control group. The prevalence of drooling and prematurity was also assessed as well as the degree of mental handicap. Results showed an increased prevalence of malocclusion in children with cerebral palsy. Cerebral palsied children are likely to have a significantly increased overjet ($P<0.001$) when compared with normal children. The comparatively small sample sizes precluded firm conclusions being drawn regarding other group comparisons (such as comparing children with cerebral palsy with and without a mental handicap) but there maybe a tendency towards the more handicapped group having a Class II malocclusion.

Introduction

Cerebral palsy (CP) can be defined as a 'disorder of movement and posture due to a defect or lesion of the immature brain' (Bax, 1964). The severity of the brain lesion can vary greatly but the prevalence of cerebral palsy has remained reasonably constant at approximately 2 in 1000 live-births by school age in industrialized nations (Paneth and Kiely, 1984). It is therefore a comparatively common condition.

The dental status of children with CP is similar in many ways to that of unaffected children. They suffer from caries, periodontal disease, malocclusion, enamel hypolasia, bruxism and trauma (Lyons, 1960; Siegel, 1960; Wessels, 1960; Nunn and Murray, 1987; Pope and Curzon, 1991). The present study concentrates on malocclusion in these children.

There are conflicting reports regarding the prevalence of malocclusion in children with CP. Some have found an increased prevalence of malocclusion (Jackson, 1956; Lyons, 1956, 1960; Album *et al.*, 1964; Strodel, 1987), but others have found the prevalence of malocclusion to be within normal limits (Gum, 1962; Magnusson, 1964; Rosenbaum *et al.*, 1966; Miller and Taylor, 1970). It has been suggested that malocclusion may only be increased in the most severely brain damaged children (Brown and Schodel, 1976).

The muscles of the face and oral cavity play a role in facial growth and occlusal development (Houston *et al.*, 1992). As the tone and function of the orofacial muscles with CP can be abnormal, the facial growth and occlusion of these children may be outside normal limits. Drooling of saliva, sometimes a major problem in these children, could be a manifestation of particularly poor orofacial muscle function, and it is possible that children with CP who have a habit of drooling saliva may have increased malocclusion.

This study therefore set out to examine the prevalence of malocclusion (by using study models) in a group of children with CP when compared with a normal control group. Pharoah *et al.* (1987) have suggested that the presence of a mental handicap may be a marker of the severity of brain damage, with children with no mental handicap being the least severely damaged. Consequently, the children with CP and a mental handicap (CP + MH) were compared with those who had CP but no mental handicap (CP–MH). Those children with a habit of drooling saliva were also identified in order to assess whether they had an increased prevalence of malocclusion due to their apparently very poor orofacial function. Finally, the prevalence of prematurity was also investigated: it has been found that this is not uncommon in children with CP (Hagberg *et al.*, 1989).

Subjects and methods

The study population consisted of children who had CP, and a control group of children who had no known physical or mental handicap.

Special schools in the Leeds area which were likely to have pupils with CP were identified. A letter was sent to each school outlining the aim of the study and requesting a meeting to discuss it. Seven special schools were approached and the Senior Dental Officer from Bradford Health Authority. Where no reply was obtained from a school, a further letter was sent which was followed-up with a telephone call. Despite this, only two special schools agreed to participate. One contained children who had an IQ of 70 or more, the other contained children with an IQ of 70 or below. The Head Teachers of the schools who did not agree to take part in the study felt that the children in their care would find it very difficult to have impressions taken of their teeth, due to the severity of their mental or physical handicap.

Once a school had agreed to take part in the study, a visit was made to meet the school nurse involved in the day-to-day health care of the children, in order to discuss the study requirements. The children with CP were identified from the medical records held by the schools and a letter (detailing the reasons for and requirements of the study) were sent to the parents or guardians of the children, together with a consent form. The parents or guardians were asked to return a consent form in a pre-paid envelope: altogether, 41 consent forms were sent out and 31 parents or guardians consented to allow their children to take part in the study.

Children with CP who attended the Department of Paediatric Dentistry at the Leeds Dental Institute as out-patients, or who had dental treatment under general anaesthesia as in-patients were also asked to take part in the study. Information letters and consent forms were sent out to parents and guardians as indicated above in order to obtain impressions of the childrens' teeth (where these would not have been obtained as part of their routine treatment) or to seek permission to use the models when these had been obtained as part of their routine treatment.

The control group of children were selected from those attending the Department of Paediatric Dentistry at the Leeds Dental Institute. Once again, information letters and consent forms were sent out to parents or guardians in order to obtain impressions of the childrens' teeth (where these would not have been obtained as part of their routine treatment) or to seek permission to use the models when these had been obtained as part of their routine treatment.

Children were only included in the study if they had their first permanent molars, upper permanent central incisors and all permanent lower incisors erupted; had never knowingly had orthodontic treatment and they (or their carers) did not admit to their digit sucking. The children selected to be in the two groups were similar in age, sex and race.

Age, race and sex of children in the study and control groups

The children in both groups in the study ranged from 6–16 years of age. There were 34 children in each group. Race was determined from physical appearance and most of the children were White–Caucasian. In both groups, one child was a Negro and one was Indian/Pakistani. The remaining 32 children were White–Caucasian. In both the study population and control group there were 18 males and 16 females.

Mental handicap
Twelve of the children with CP (35 per cent) were mentally handicapped (CP + MH) with an IQ less than 70, and 22 (65 per cent) had an IQ greater than 70 and no obvious mental handicap (CP–MH). The term 'mental handicap' is used throughout this paper rather than the more up-to-date expression 'learning difficulty' in order to allow comparison with earlier work.

Classification of cerebral palsy
The motor and topographical classification was used to identify the type of CP of each child. The majority of the children were diagnosed as spastic with most of these having spastic quadriplegia. The types of CP found in the study population are shown in Table 1.

Alginate impressions of the upper and lower arches of the cerebral palsied and control children were taken and study models constructed with the aid of a wax bite. Two bites were recorded in order to increase accuracy when putting the study models into occlusion. When

Table 1 Distribution of cerebral palsy in a study population of children in Yorkshire.

Classification	n	Percentage
Spastic quadriplegia	16	47
Spastic hemiplegia	2	6
Spastic diplegia	9	26
Athetoid	2	6
Hypotonia	2	6
Ataxia	3	9

the two bites recorded were very different, the one in which the teeth most readily occluded was used. The plaster study models obtained from the impressions were placed in occlusion and various measurements made using a metal ruler and a vernier caliper.

Clinical assessment

Lip competence

This was assessed by direct observation of the lips of the children using the method of Ballard (1953). If the mandible was in the physiological resting posture and the lips were in apposition without contraction of orbicularis oris and mentalis muscles, the lips were competent. If the child had to contract the orbicularis oris and mentalis muscles vigorously in order to close the lips, the lips were recorded as being incompetent. Ideally, an assessment of lip competence would have been made on at least two occasions to assess reliability but it was felt that it might be unreasonable to further disrupt the school schedule in the case of the children with CP.

Drooling of saliva

Drooling of saliva was recorded as being present if the child was obviously seen to be drooling at the time of examination. However, this assessment was made prior to any physical examination, in order to avoid actually stimulating drooling to commence. Alternatively, it was recorded if the parents, guardians or school nurse noticed a considerable amount of drooling of saliva or if the child was wearing a bib. Other approaches such as the weighing of cotton wool rolls (Blackwell et al., 1978) were tried but were found to be impossible to use due to the lack of co-operation in the CP + MH group.

Premature birth

The parents were asked whether their child or children had been born prematurely and a record was made of the degree of prematurity based upon weeks of gestation. It could be argued that a measurement such as birthweight would be a more reliable way of assessing prematurity but such detailed information would have been virtually impossible to come by.

Measurements from study models (by DFL)

Overjet (mm)

The maximum horizontal distance between the maxillary and mandibular central incisors in occlusion was measured using a metal ruler.

Overbite (mm)

The maximum overlap of the mandibular central incisors by the maxillary central incisors was marked on the study models and measured with a metal ruler.

Anterior open bite (presence or absence)

An anterior open bite was recorded as being present if the lower incisors were not overlapped in the vertical plane by the upper incisors, and did not occlude with them.

Posterior crossbite

A posterior crossbite was recorded as being present if the buccal cusps of the lower first permanent molars occluded outside the buccal cusps of the upper first permanent molars. Lingual crossbite was not included in this study.

Incisor irregularity

The degree of mandibular incisor irregularity was measured according to the irregularity index of Little (1975). Spacing was ignored; spacing can be represented as a negative value, but it makes the incisor irregularity index meaningless (Little, 1975). The following groupings were used initially: 0 mm = perfect alignment; 1–3 mm = minimal irregularity; 4–6 mm = moderate irregularity; 7–9 mm = severe irregularity; 10 mm = very severe irregularity.

For purposes of comparison, these were later split into two categories (mild and severe irregularity) by combining the perfect and minimal irregularity groups and the moderate and severe irregularity groups.

Incisor relationship

The incisor relationship was classified in the antero-posterior plane (Houston et al., 1992):

Class I   The lower incisor edges occluded with

or immediately below the cingulum plateau of the upper central incisors.

Class II   The lower incisor edges lay posterior to the cingulum plateau of the upper incisors.

Division   1—The upper central incisors were proclined or of average inclination and there was an increase in overjet.

Division   2—The upper central incisors were retroclined. The overjet was average but could be increased.

Class III The lower incisor edges lay anterior to the cingulum plateau of the upper incisors. The overjet was reduced or reversed.

Arch width (mm)

The width of the maxillary and mandibular arches was measured at just one point (Proffit and Ackerman, 1986). The maxillary arch width was measured in millimetres as the distance between the mesial occlusal pits of the upper first permanent molars. The mandibular arch width was similarly measured as being the distance between the central midline fissures of the lower first permanent molars.

Arch circumference (mm)

Arch circumference was measured by addition of the measurements of four segments in each arch. These segments were: from the mesial surface of the first permanent molar to the distal surface of the canine on each side of the mouth, plus the distal surface of the canine to the midline between the central incisors and from the midline to the distal surface of the opposite canine.

Reproducibility of measurements from study models

Ten sets of study models were selected from the records store of the Department of Paediatric Dentistry at Leeds Dental Institute, and the above variables measured on two separate occasions by DLF. Each model was given a code number and the variables listed above were measured. The models were re-coded and all the measurements repeated. The results are shown in Table 2. The overall intra-examiner agreement for all the readings was 97.5 per cent.

Table 2 Reproducibility of measurements of 10 study models taken from the records store of the Department of Paediatric Dentistry at Leeds Dental Institute.

Measurement	Percentage agreement
Overjet	88
Overbite	93
Presence of anterior overbite	100
Presence of posterior crossbite	100
Lower incisor crowding	95
Incisor relationship	100
Upper arch width	99.2
Lower arch width	99.1
Upper arch circumference	99.5
Lower arch circumference	99
Dental age	100

Results

The data were entered into an IBM compatible computer for data analysis using the program 'Microsoft Works, Version 3.0' (Microsoft Corporation, Redmond, WA, USA). After data entry, the following comparisons were then made for each of the variables measured: (i) the control population was compared with children with CP; (ii) the children with CP with a mental handicap were compared with those with no mental handicap.

Results of the main group comparisons

Statistical analysis of data involved Student's *t*-test, Chi-squared and Fisher's exact test. Student's *t*-test was used to analyse overjet, overbite, arch width and arch circumference. Results are expressed as arithmetic means and the standard deviation. All other data were analysed using Chi-squared, or Fisher's exact test when numbers were small.

Premature births

None of the control children were born prematurely but 23 of those with CP were. ($\chi^2 = 34.7$ with one degree of freedom and $P < 0.001$).

Comparing children with CP with and without a mental handicap, six out of 12 with mental handicap were premature and 17 out of 22 with no mental handicap were premature. A higher proportion of those with no mental handicap were born prematurely, but using the Fisher's

exact test the difference was not significant ($P = 0.14$).

Drooling of saliva

None of the children from the control group drooled saliva. Of the children with CP, eight drooled saliva; three of these had a mental handicap and five did not. Using Fisher's exact test, this difference was not significant ($P = 1.0$).

Lip competence

Few children with CP had competent lips. In all, 20 of the control group of 34 children had competent lips, but only four of those with cerebral palsy did. ($\chi^2 = 16.5$ with one degree of freedom, $P < 0.001$; this result was significant at the 0.1 per cent level.)

Comparing the children with CP with and without a mental handicap, one out of the 12 children with a mental handicap had competent lips and three of the 22 with no mental handicap had competent lips. Using Fisher's exact test $P = 1.0$ which was not significant.

Overjet

The results of the measurement of overjet were analysed using Student's t-test and are shown in Table 3. When the control group was compared with the CP group the result for overjet was highly significant; $P < 0.001$. When the CP + MH group were compared with the CP – MH group, $0.01 < P < 0.02$ which was significant at the 5 per cent level.

Table 3 Results of overjet comparisons for the normal control children and those with cerebral palsy (CP) with and without a mental handicap.

| Group | n | Measurements in mm | | | |
		Mean	±SD	t	P
Control	34	4.0	2.1		
CP	34	6.4	3.3	3.54	<0.001
CP + MH	12	8.3	3.2		
CP – MH	22	5.5	2.8	2.70	<0.02

CP + MH = children with cerebral palsy and a mental handicap; CP - MH = children with cerebral palsy but no mental handicap.

Table 4 Results of overbite comparisons for the normal control children and those with cerebral palsy (CP) with and without mental handicap.

| Group | n | Measurements in mm | | | |
		Mean	±SD	t	P
Control	34	3.0	1.3		
CP	34	4.4	1.9	3.37	<0.005
CP + MH	12	3.7	1.1		
CP – MH	22	4.9	2.0	1.59	>0.05

CP + MH = children with cerebral palsy and a mental handicap; CP - MH = children with cerebral palsy but no mental handicap.

Overbite

The results of the measurement of overbite were analysed with Student's t-test and are shown in Table 4. Comparing the control group with the CP group gave the only statistically significant result $0.001 < P < 0.005$ which was significant at the 1 per cent level. The mean overbite of the CP + MH group was slightly smaller than the mean for those in the CP – MH group but this was not statistically significant.

Incisor relationship

The incisor relationship was classified as Class I (Cl.I), Class II division 1 (Cl.II/1), Class II division 2 (Cl.II/2) and Class III (Cl.III). The results are shown in Table 5 and were analysed by comparing those with a Class I incisor relationship with those with a Class II or III incisor relationship. The children with CP were compared with the control group of children ($\chi^2 = 0.53$ with one degree of freedom. This was not significant).

When the CP + MH children were compared

Table 5 Results of incisor relationship comparisons for the normal control children and those with cerebral palsy (CP) with and without a mental handicap.

Group	n	Class I	Class II/1	Class II/2	Class III
Control	34	18	13	1	2
CP	34	15	17	2	0
CP + MH	12	2	9	1	0
CP – MH	22	13	8	1	0

CP + MH = children with cerebral palsy and a mental handicap; CP – MH = children with cerebral palsy but no mental handicap.

with those with no mental handicap using Fisher's exact test, $P = 0.03$, which was significant at the 5 per cent level. This was the only significant result and none of the children with a mental handicap had a Class III incisor relationship and only one had a Class II division 2 incisor relationship. The test therefore showed that a statistically significant number of children with a mental handicap had a Class II division 1 incisor relationship.

Other variables

There were no significant differences between any of the comparisons in the following variables: anterior open bite, posterior crossbite, incisor irregularity, arch width or arch circumference.

Discussion

The present study adds to the evidence that there is an increased prevalence of malocclusion in children with CP. Previous studies have not compared children with CP and a mental handicap with those with no mental handicap. This has been attempted in this study but owing to the small size of the sub-groups, some caution is needed in interpretation of the statistical results. Nevertheless, it appears that lip competence may have some relevance in the aetiology of increased overjet in children with cerebral palsy and may also be of relevance with respect to a drooling habit in the CP + MH group.

Lip position, however, may be of greater significance although this was not investigated in this study. Furthermore, as already acknowledged, assessment of lip competence would have benefited from further study of the reliability of such assessment. In addition, 20 out of 34 (nearly 60 per cent) of children in the control group used in this study were assessed as having competent lips. However, Walther (1960) found that only 20 per cent of 11–13-year-old children had competent lips, whilst 46 per cent of children over 11 years were found to have competent lips in a study by Tulley (1964). There is clearly a discrepancy between the studies which may be due to a number of factors such as differences in age groups and ranges, whether or not orthodontic treatment had been undertaken and the difficulty of the assessment itself.

Our finding of an increased overbite in children with CP contrasts with work by other authors such as Magnusson (1964) and Rosenbaum et al. (1966) who found a reduced overbite in children with CP. However, such a finding might be expected where this is associated with an increased overjet (Houston et al., 1992).

Prematurity also requires further analysis. Many of the children with CP were born prematurely according to the criteria used, but none of the control children were born prematurely. It is therefore difficult to rule out the possibility that prematurity may have a role to play in malocclusion prevalence. The current study cannot answer this question satisfactorily.

It is clear that a number of areas exist which require more detailed study and we cannot say that having an increased overjet 'causes' lip incompetence and drooling or vice versa. Despite this, workers have treated children with CP using a variety of functional appliances and forms of physiotherapy to control malocclusion and drooling, apparently with some success (Castillo-Morales et al., 1982; Fischer-Brandies et al., 1987; Limbrock et al., 1990). However, studies with larger sample sizes are needed to establish whether treatment should be directed towards the more severely mentally handicapped children (possibly those with the greatest overjet). In addition, little information is available regarding what if any changes occur in lip competence with maturation in children with CP. This would be of great relevance to any operator undertaking orthodontic treatment as stability of overjet reduction for example, could be questionable. At the same time, drooling may also be affected by lip position or competence. It is interesting to note that spontaneous improvements in drooling do appear to occur: two children in this study had apparently drooled when younger but had subsequently ceased.

Conclusions

1. Children with cerebral palsy have a significantly increased overjet and overbite and are likely to have incompetent lips.
2. The only significant difference between cerebral palsied children who have a drooling habit and those that do not, is that more of those with a drooling habit have incompetent lips.

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