**Relationship between lip position and drooling in children with cerebral palsy**

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**ABSTRACT. Aim** This was to determine whether there is any relationship between lip position and drooling in children with cerebral palsy (CP).

**Methods** One hundred and sixty individuals with CP (aged 4-18 years) agreed to take part in the study. The following data were collected in two ways. Firstly the presence or absence of drooling, the dental age, the incisal relationship and lip position were obtained by direct observation of the children. Secondly other data for age, sex, learning disability and type of CP were collected using a questionnaire/form.

**Statistics** This was by simple Chi squared analysis.

**Results** A significantly greater number of CP children who drooled had incompetent lips ($p<0.002$). When comparing CP children with drooling (mild + moderate + severe) and those without drooling there was a clear difference in distribution of it between those with and those without competent lips. This difference was found to be highly significant ($p<0.005$).

**Conclusion** Lip position and oral seal share a potentially important association with drooling.

**KEYWORDS**: Cerebral palsy, Lip position, Drooling.

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**Introduction**

Drooling is the unintentional loss of saliva and other oral contents from the mouth and is a normal phenomenon of infancy that subsides in early childhood, usually by 15-18 months. Cessation of drooling occurs as a consequence of physiological maturation of oral-motor function [Blasco and Allaire, 1992] but it can be both a clinical sign and symptom, being most often associated with neurological disorders. As it affects the general well being of the individual and may present problems for carers, specific treatment is essential in the overall intervention plan of the multidisciplinary team.

Problem drooling affects a small number of individuals, mostly those with neurological dysfunction in the form of motor deficits such as cerebral palsy (CP). However, as CP is diagnosed in 2-5 per 1,000 live births [Paneth and Kiely, 1984], it is estimated that 10%-37.5% of children with CP have severe enough drooling problems to interfere with daily social and practical functions [Budhraja et al., 1973; Ekedahl, 1974; Van De Heyning et al., 1980; Tahmassebi and Curzon, 2003]. This represents a significant population. The cause of drooling is not sufficiently addressed in the literature, although a number of factors are considered to be potential influences. In addition, head position, jaw stability, lip closure and spontaneous swallowing are considered prerequisites for normal development of speech, feeding and drool control [Crysdale et al., 1984].

Drooling occurs normally in children under 1 year of age or in those who are teething. Persistent drooling in children over 4 years of age usually signals oral motor dysfunction [Ogg, 1975]. Lip closure and jaw stability develop early in infancy, as by 36 months of age a child’s jaw stability is nearly fully developed [Iammatteo et al., 1990]. The effect of mouth closure on drooling has been reported by Ray et al., [1983] and also by Iammatteo et al. [1990], who suggested that the facilitation of mouth closure can be effective in decreasing drooling. However, the success of this technique cannot be generalized because the studies were carried out on only two and one subjects respectively.

Closure of the jaws is also required in order to achieve normal coordination of the tongue movements needed for salivary transport. Harris and Purdy [1987] reported that children with CP who drooled swallowed inefficiently and uncoordinatedly, with poor synchrony of lip closure.
A recent work by Franklin et al. [1996] has shown that children with CP, who have a drooling habit, are more likely to have incompetent lips and a greater prevalence of maxillary anterior tooth protrusion. However, only 8 out of 34 CP children in that study had a drooling problem. It was therefore felt appropriate to confirm the Franklin et al. study using a much larger sample size to assess any relationship between drooling and lip position.

Materials and methods

The study population. This consisted of children with CP who attended special schools. Out of the total of 17 special schools in the county of Yorkshire, 13 were identified as having pupils with CP and they all agreed to participate in the study. Six of the 13 schools were registered to contain children with severe learning disabilities, four with moderate to severe/profound learning disability, two with mild to moderate learning disability and one children with physical disabilities. Once ethical approval was confirmed, the head teachers of all special schools in the Leeds, Dewsbury and Airedale area (Yorkshire) were contacted by letter outlining the aim of the study and inviting participation. If no reply was received two weeks after sending the initial letter, a second letter was sent which was followed up with a telephone call. Once a school had agreed to take part in the study, a visit to meet the head teacher and school nurse was arranged. The aim of this visit was to discuss what the study entailed and the possible examination arrangements.

Children with CP were identified from the medical records held by each school. A list of the names and addresses of those children with CP in each school was obtained. Information letters were then sent to the parents/guardians of the children informing them of the reasons for the study and the need to carry out oral examinations, noting absence and presence of drooling. This study was originally based on positive consent. However, due to the very low response rate (25%), the ethics committees were again approached and approval to complete the study using negative consent was granted.

Data collection. The data was collected in two ways. Data concerning the presence or absence of drooling, the dental age, the incisor relationship and the presence or absence of lip seal were obtained by direct observation of the children. This was for periods of approximately 5 minutes for each child in their classroom. The observational sessions were undertaken by one investigator (JT) and were carried out in the mornings, and at least one hour post-prandial. Other data such as the age, sex, learning disability and type of CP were collected using a questionnaire/form that was completed by each parent/guardian of the children. These records were later confirmed by the medical records held at the schools. Learning disability was categorized into mild, moderate or severe; this information was obtained from the school medical records. Each child was identified for their CP condition using the motor and topographical classification of Minear [1956].

Problem drooling spans from mild to severe. Drooling of saliva was, therefore, recorded using the Blasco et al. [1990] categorization:
- mild drooling (saliva spills onto the lips but not beyond the vermilion border);
- moderate drooling (saliva reaches chin);
- severe drooling (characterised by dripping of saliva onto clothing).

Lip position was recorded as being competent or incompetent [Ballard, 1963]. If the lips met together at rest without contraction of orbicularis oris and mentalis muscles, then the lips were described as competent (Fig. 1). If the child had to contract the orbicularis and mentalis muscles vigorously in order to close the lips, then the lips were recorded as incompetent (Fig. 2).
Incisor relationship was recorded using the Houston et al. [1992] method which classifies it in the antero-posterior plane when the teeth are in occlusion as follows:

- Class I - the lower incisor edges occlude with or lie directly below the cingulum plateau of the upper central incisors.
- Class II - the lower incisor edges lie posterior to the cingulum plateau of the maxillary central incisors. There are two divisions:
  - Division 1 - the upper central incisors are proclined or are of average inclination. The overjet is thus increased.
  - Division 2 - the upper central incisors are retroclined.
- Class III - the lower incisor edges lie anterior to the cingulum plateau of the upper central incisors.
- Anterior open bite - when the teeth are viewed in occlusion and from the anterior, horizontal perspective, the incisal edges of the upper central incisors do not meet or overlap the lower central incisors.

Statistical analysis. Relationships between drooling and anterior oral seal and between incisor relationship and drooling were assessed using $\chi^2$ analysis. The $\chi^2$ test was used to assess quantitatively whether a set of frequencies followed a particular distribution, i.e. whether the observed frequencies differed significantly from those expected.

Results

Altogether 171 children (aged 4-18 years) participated and twenty-one parents/guardians did not allow their children to take part. Children who were known to have had previous surgery or were on medication to reduce their drooling were not approached. Two children were excluded from the study due to absence of any teeth and 9 children because of poor cooperation. Therefore, 160 out of the potential 171 children with CP were included.

The majority of the children were diagnosed as spastic (68%) with most of these having spastic quadriplegia (45%). Thirteen children were diagnosed as having CP but the type of CP could not be identified. Ninety-two of the children (58%) had a severe learning disability; 38 (24%) and 16 (10%) had moderate and mild learning disabilities respectively. In one subject with no drooling, the level of learning disability was not known and therefore this individual was excluded from the analysis. A further thirteen of the children (8%) in the study did not have any learning disability.

Table 1 shows the distribution of CP children with different degrees of drooling who had competent lips as compared with those with incompetent lips. The results of $\chi^2$ analysis showed that there was a highly significant difference in the distribution of drooling between those with and those without competent lips. In particular children with competent lips were more likely to be non-droolers and less likely to exhibit severe drooling than would have been expected by chance. The reverse was true of those with incompetent lips. From Figure 3 it can be seen that when comparing CP children with drooling (mild + moderate + severe) and those without there was a clear difference in the distribution of drooling between those with and those without competent lips. This difference was found to be highly statistically significant ($\chi^2=7.71$, 1 degree of freedom, p<0.005).

The majority of the CP children in this study population had a Class II Division 1 incisor relationship (39%). There were 35% of the children with Class I, 19% with anterior open bite and 7% with Class II Division 2 or Class III incisor relationship. The number of children with Class II Division 2 and Class III incisor relationships was very small and a meaningful statistical analysis was not possible, and their data were therefore excluded. A significant difference was found in the distribution between individuals with a Class II Division 1 incisor relationship and those with a Class I incisor relationship, those with mild, moderate and severe drooling and with none. As can be seen from Figure 4, individuals with a Class I relationship were more likely to display no drooling and less likely to have severe drooling. Those with a Class II Division I malocclusion were less likely to be non-droolers ($\chi^2=13.20$ with 4 degrees of freedom, p<0.010). Subjects with an anterior open bite appeared to mirror the overall drooling distribution that had been found for Class I individuals.
This study was unusual as, firstly it included a much larger sample size than most other similar studies on CP [Franklin et al., 1996; Zamzam, 1998], secondly there was an almost equal number of CP children with competent lips (82) and of CP children with incompetent lips (78). Previous authors such as Franklin et al. [1996] have reported that most of the CP children in their sample population had no anterior oral seal. This may represent a bias: families with drooling difficulties children are maybe more likely to volunteer to take part in a study which they see as potentially useful to their problem whereas unaffected individuals may see no need to take part in it. Zamzam [1998] has previously reported this problem and related this to the requirement for positive consent for that study.

Lip position as assessed using a standardised and reproducible method previously described by Zamzam and Luther [2001] does not appear to change with age in children with CP [Zamzam, 1998]. Furthermore, whilst a lack of oral seal at rest is common in the general population, it is evident that this alone does not cause drooling. For example, Ballard [1963] reported that fewer than 80% of the English population have an ideal anterior oral seal. However, in the present study, when CP children with competent lips were compared with those with incompetent lips, there was a highly significant difference in distribution of drooling. Those with competent lips were more likely to be non-droolers and less likely to exhibit severe drooling. Franklin et al. [1996] also found significantly more CP children with incompetent lips who drooled as compared with those who had competent lips.

In contrast to the present study, Zamzam [1998] did not find any difference in lip seal in CP children who drooled. However, as mentioned previously, that study was based on positive consent and the study population consisted mainly of children who drooled, i.e. 53 children out of 65 drooled. Therefore, it was not possible to eliminate the possibility of bias in those results.

Class II Division 1 incisor relationships have been reported to have a high prevalence in CP [Koster, 1956; Lyons, 1956]. The study by Franklin et al. [1996] on 34 CP children also showed significantly increased levels of Class II Division 1 incisor relationships. A high prevalence of anterior open bite in children with CP has been also reported [Koster, 1956; Lyons, 1956; Album et al., 1964], but others have found a similar prevalence to that of normal children [Rosenbaum et al., 1966; Strodel, 1987; Franklin et al., 1996]. In this present study the number of children with Class II Division 2 and Class III incisal relationships was very small. This is in agreement with other studies [Koster, 1956; Lyons, 1956; Franklin et al., 1996].

In our study significantly more individuals with a Class II Division 1 malocclusion suffered from drooling than those children with Class I incisor relationship. These results would therefore indicate that treatment of a Class II Division 1 malocclusion (in individuals with drooling) might encourage an anterior oral seal and thus, indirectly, the degree of drooling. However, as discussed below, this requires careful consideration. Furthermore, this study has not been able to show what is cause or effect.
The effect of mouth closure on drooling has been reported by Ray et al. [1983] and Iammatteo et al. [1990], who suggested that facilitation of mouth closure can be effective in decreasing drooling. The results of the current study therefore suggest that if lip seal can be achieved in CP children with drooling, then it may decrease the degree of drooling. This conclusion is in agreement with those few studies where the success of oral appliance therapy in the management of drooling has been reported. Nevertheless, this effectively means that orthodontic treatment is required (to reduce upper front tooth protrusion, which would help allow the lips to meet). Such treatment is itself difficult, frequently involving complex fixed appliances, extractions and high levels of cooperation among the patient, guardians and operator for lengthy periods (often 2 years or more). Furthermore, orthodontic treatment is not without risk, the most common problems are decalcification and caries [Mitchell, 1992; Kukleva et al., 2002] due to poor oral hygiene and/or poor dietary control, but other problems can also arise. For children with CP, orthodontic treatment is not common and may require significant parental/guardian and professional support [Becker and Shapira, 1996], but might still prove impossible, particularly in those individuals with the most severe disabilities.

Other aspects also require clarification. For example, amongst routine patients, at least 10% will commonly fail to complete treatment [Trenouth, 2003] and the stability of treatment outcome can also be problematic even in such ‘routine’ cases [Melrose and Millett, 1998; Blake and Bibby, 1998]. Therefore, if it was possible to assess more specifically whether a definite link exists among lip seal, incisor relationship and drooling, it would be extremely important not to raise false hopes and to establish that the necessary level of cooperation and support could be achieved before proceeding with any orthodontic treatment. It would also be necessary to consider whether changes could occur by maturation alone as this would influence the age at which any treatment should be started. For example, Haberfellner and Rossiwall [1977] presented a case report of a four and a half year-old CP boy with constant drooling. A modified orthodontic “monobloc” was provided. After 25 months of therapy, lip closure was found to be adequate and no further drooling occurred during this period. However, this improvement in drooling could have been due to maturation of the orofacial musculature or combination of both therapy and natural development; in a recent study by Tahmassebi and Curzon [2003], it was found that the degree of drooling decreased as the child’s dental age increased and the orofacial musculatures matured.

Untreated controls would also need to be available should any prospective, randomised, controlled clinical trials be undertaken. This could be difficult for the reasons discussed above; indeed previous studies have neglected to include controls. Fischer-Brandies et al. [1987] treated 71 CP children aged 4 to 14 years with orofacial regulation therapy for an average of 15 months. Forty children in the study had severe drooling, and they reported that appliance therapy resulted in a reduction of drooling in 28 of these children, though the therapy could not render the drooling to a ‘normal’ condition. However, Fischer-Brandies et al. [1987] stated that the orofacial therapy did not improve the open mouth habit or the spontaneous position of lips. Likewise Limbrook et al. [1990] used removable acrylic appliances with ‘acrylic bumpers’ labially to stimulate the lips and complete the lip seal. They reported that 67% of the cases treated by appliance therapy showed a reduction in drooling. Unfortunately, a major limitation of both of these studies was the lack of any control group.

Conclusions

Competent lips may be an important factor in the reduction of drooling, clearly not the only one as drooling was still found in individuals who had a Class I incisor relationship or competent lips. More research into the management of drooling, in affected individuals, by improving lip position and anterior oral seal is warranted. However, it would be essential not to underestimate the difficulties associated with such work and to adopt a cautious approach as success could clearly not be guaranteed despite lengthy, complex treatment, which is of itself not risk free.

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