Clinical prevalence of drooling in infant cerebral palsy

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Abstract

Objective: To determine the prevalence and severity of drooling in infant cerebral palsy (ICP) and analyze the possible surgical, pharmacological, myofunctional and novel alternative approaches to treatment of this disorder.
Methods: A clinical study is made of a group of patients with ICP (cohort) and aged between 4 and 34 years, visiting a dental clinic for disabled patients. The classification of Thomas-Stonell and Greenberg was used to assess the presence and severity of drooling.
Results: Of the total of 50 patients evaluated (52% males and 48% females), 58% presented drooling (mild in 44.4% and moderate to severe in 27.7%).
Conclusion: Over half of the patients with ICP presented drooling. Effective options are therefore needed for the treatment of this problem, which poses a series of negative effects for both patients and their care givers.

Key words: *Infant cerebral palsy, drooling, management of drooling.*

Introduction

Infant cerebral palsy (ICP) comprises a group of movement and postural development disorders that cause disability, and which originate during development of the fetal brain (1).
Despite progress in the prevention and treatment of certain causes of cerebral palsy, the number of affected children and adults has remained relatively constant or has even increased slightly in the last 30 years. This is because more critical premature and weak infants survive as a result of improved neonatal care (2,3).
Drooling is defined as an unintentional loss of saliva from the mouth, and is considered normal in infancy up until the age of 18 months when the oral motor muscles mature. Drooling beyond this age is not normal, and is often associated with neurological problems such as cerebral palsy or mental retardation (4-6).
Drooling manifests as an excessive output of saliva from the mouth secondary to alterations in voluntary oral motor activity, incorrect swallowing, or deficient lip sealing; in rare instances it is attributable to hypersialia - generally produced by the administration of antiseizure drugs or other psychoactive substances. Additional influencing factors are the emotional state of the patient, head posture, concentration capacity, dental malformations, the size control of the tongue, and deficient nasal respiration (7).
Patients with ICP generally present an intact swallowing reflex, though with a diminished capacity to control the initial voluntary preparatory phase, and a lack of coordination of the muscle activation sequence. Likewise, affected patients have problems coordinating swallowing and breathing, and this in turn may inhibit the swallowing reflex when the mouth is full of saliva - thus resulting in drooling (7-9).

Among the classifications of the severity of drooling, Thomas-Stonell and Greenberg in 1988 (4) defined five grades (Table 1).

Table 1. Drooling severity classification according to Thomas-Stonell and Greenberg.

Grade	Description
1	Dry, no drooling
2	Mild: humid lips only
3	Moderate: humid lips and chin
4	Severe: clothing begins to be affected
5	Profuse: clothes, hands and objects are wet

The management of drooling in children with neurological problems has received increased attention in recent years, and in fact an increased range of treatment options is presently available for such patients.

Surgery was one of the first options contemplated for the treatment of drooling. The principal surgical techniques comprised salivary duct relocation or ligation, or the removal of gland tissue. Chang et al. (4) relocated the submandibular gland duct and removed the sublingual glands, reporting excellent clinical results after the surgical procedure. However, side effects such as the loss of sphincter function after relocation of the orifice, and ranula formation, are problems that must be taken into account.

Bilateral relocation of the submandibular gland ducts is used, and tends to cause few adverse effects. The submandibular ducts are exteriorized to the oral cavity beneath the tongue, immediately below the lower teeth. Posteriorly, a tunnel is made on the floor of the mouth towards the tonsillar region, alongside the posterior portion of the tongue and towards the pharynx. Finally, the ducts are positioned through the tonsils and sutured; since they face towards the back of the mouth, swallowing of the saliva is facilitated (6).

More recently, intraductal laser photocoagulation of the parotid ducts has been proposed. This technique requires general anesthesia. The parotid duct is located and dilated with a catheter, after which an optic fiber measuring 600 μm in diameter is inserted from the intraoral orifice towards the terminal duct of the parotid gland. Finally, laser photocoagulation is carried out during 10 seconds at a power rating of 7 W in patients under 5 years of age (versus 10 W for older patients). In a clinical study carried out by Chang et al. (4), involving 48 patients, 83.3% showed significant reduction in drooling, and no patient presented signs of xerostomia (4).

Other treatment options involve drugs used to reduce saliva volume without altering swallowing function. In this context, cholinergic muscarinic receptor block reduces saliva output. However, a series of both local and central side effects may result, such as drowsiness, irritability, rapid mood changes, hostile behavior, fear, deficient coordination, nasal congestion, xerostomia, vomiting, diarrhea and bladder voiding difficulties.

Among the anticholinergic drugs used to control drooling, mention should be made of atropine sulfate, glycopyrrolate and scopolamine. The undesirable effect of these drugs is the pharmacological action sought in this case.

Mier et al. (5) conducted a double blind study involving 39 children administered either glycopyrrolate or placebo. The patients received body weight adjusted doses that in turn were subjected to four-fold increments. The results showed considerable improvement in drooling, particularly with the last treatment dose, while in contrast the greatest side effects were also noted with the third and fourth doses.

Regarding the use of transdermal scopolamine, the latter drug has been shown to offer a number of advantages over other alternatives, including easy administration, prolonged action (no patch replacement being needed in 72 hours), and a low incidence of side effects compared with other drugs exerting anticholinergic actions. While affording the same anticholinergic effects as any other anticholinergic agent, scopolamine offers the advantage of more potent action upon the salivary glands, with lesser action upon the heart and intestinal smooth muscle (5,10,11).

Botulin vaccine is presently used in fields such as neurology. It is employed as treatment for disorders such as cerebral palsy, tremor, bruxism, spasticity, rigidity and muscle dystonia. Its local administration allows precise selection of the muscle groups exhibiting hyperactivity requiring control via individualized doses, depending on the intensity of the disorder. The effect is local and reversible, yet sufficiently prolonged to allow repeated administration without the risk of side effects. The adequate use of this vaccine could improve patient quality of life by reducing the contractures and articular alterations that favor drooling (12).

Another management option is the so-called Baba Stop (13). This is a behavior modifying program developed for children with ICP that have good comprehension of verbal instructions and are amenable to social reinforcement efforts, in accordance to the existing cognitive level. Conditioning is based on the use of a temporizer that generates an acoustic, visual or tactile stimulus at fixed or variable time intervals. During the conditioning phase, the therapist gives instructions to the child to close the mouth and swallow when the conditioning stimulus is generated. This program was evaluated in a study of 12 patients, with positive results in all cases - though to variables degrees. Thus, drooling practically disappeared in some children,

while others showed a decrease and some proved able to control the problem; however, when the conditioning stimulus disappeared, drooling gradually returned. For this reason a portable temporizer was developed that is currently being evaluated. The most important consideration is that the patients can learn to swallow, after which the temporizer reminds them when to do so.

Finally, mention should be made of myofunctional therapy, which comprises a series of procedures and techniques used for the correction of orofacial muscle imbalance, the creation of new and normal muscular behavior, the reduction of negative habits, and improvement of patient esthetics (14).

Early myofunctional therapy in patients with ICP is very important in order to normalize respiration, swallowing and chewing as far as possible, and thus facilitate eating. Following thorough anamnesis and a good clinical examination, a program individualized to meet the requirements of each patient is established, involving a series of exercises targeted to concrete muscle groups. These exercises are gradually intensified in terms of number and difficulty as the patient improves. Castillo-Morales (15) proposed a combination of muscle exercises and specific functional orthopedic devices for each type of cerebral palsy.

Patients and Methods

The study group consisted of 50 patients with ICP and seen in a dental clinic for disabled patients, over a period of 8 months. There were 26 males (52%) and 24 females (48%). The inclusion criterion was a medical diagnosis of ICP. A case history was compiled, including patient data, associated diseases and the pharmacological treatment used. Information was also collected during the examination, related to the presence or absence of drooling and of other characteristics or anatomical alterations that contribute to the appearance of drooling. In this context, the 0-1-2 malocclusion index (MOI) of the World Health Organization was used, among other parameters:

Score 0: no anomalies or malocclusions.

Score 1: discrete anomalies, ≥ 1 rotated teeth, mild crowding or spacing.

Score 2: severe malocclusions.

In addition, sagittal anomalies were examined (molar and canine class according to the Angle classification), together with vertical anomalies including open bite and overjet, and transverse anomalies including crossbite.

Informed consent was obtained from the patient tutors for the evaluation, in compliance with the ethical norms of the institution. Likewise, the parents or care givers were questioned about the frequency of drooling, to contrast this information with that obtained in the clinic. The clinical evaluation was carried out by a single operator with the purpose of standardizing the results obtained.

After determining the presence or absence of drooling, the severity of the latter was assessed (Figures 1 and 2), based on the classification of Thomas-Stonell and Greenberg (Table 1).

Posteriorly, the data were tabulated and graphically represented to obtain the percentage results regarding the presence of drooling and its severity in the studied patient group. Statistical analysis (using the SPSS version 12 statistical package) yielded the absolute and percentage frequencies, and the association of qualitative variables was carried out by means of the chi-square test.

Fig. 1. Patient with ICP. Lack of lip sealing and severe drooling.

Fig. 2. Patient with ICP. Anterior open bite and mild drooling.

Results

Of the 50 patients with ICP included in the study, 26 were males (52%) and 24 females (48%), with an age range of 4-34 years (mean 15.5 years). A total of 29 cases suffered drooling (58%) to some degree within the classification of Thomas-Stonell and Greenberg, while 21 did not have this problem.

Mild drooling was recorded in 38.88% of the cases, while the remaining 61.11% drooling proved moderate or severe (grades 3 and 4). None of the patients presented grade 5 drooling according to the above classification.

Forty percent of the patients had a history of seizures, and in all these cases antiseizure medication was being prescribed. Of the 20 patients with such seizure antecedents, 17 (85%) suffered drooling while 3 (15%) did not. Comparison of these results by means of the chi-square test yielded significant differences (p=0.01).

On examining the presence of malocclusions related to the presence of drooling, we found that 15 patients had anterior open bite without lip sealing capacity; all but 3 cases (80%), suffered drooling to one degree or other (p=0.04). Three cases presented Angle class II (one case with overjet, another division 2 with overbite, and a third patient with edge-to-edge bite). One subject in the study also presented a class III molar relationship.

In the 30 patients without malocclusions, 14 suffered drooling. Likewise, on evaluating anomalous habits, 6 individuals showed some degree of bruxism, and in all of them clinical drooling was observed.

Discussion

Drooling is the unintentional loss of saliva from the mouth, and is normal in infancy up to the age of 15-30 months maximum. Thus, from the youngest age in our series (4 years), drooling is considered pathological (4,5). The presence of drooling in patients with ICP is not attributable to an excess production of saliva but mainly to a defect in swallowing (9).

Drooling poses important social limitations for many patients with neurological problems. In the present study, the disorder affected over half of all subjects (58%), in coincidence with the observations of Tahmassebi et al. (16), and exceeding the prevalences of 10% and 10-37% respectively reported by Dreyfuss et al. (10) and Mier et al. (5), respectively.

Other factors can be association to the etiology of drooling, such as antiseizure medication. In the present study, 40% of the patients had a history of seizures, in coincidence with the figures reported in the literature, where between 30% and 50% of all patients with ICP suffer seizures (16-18). This in turn requires the prescription of drugs which according to Sochaniwskyj (7) can contribute to excessive saliva production, and thus to a rise in the prevalence of drooling. In our case, most of the patients taking such medication (85%) suffered drooling.

The presence of malocclusions such as anterior open bite, lip hypotonicity and lingual incompetence complicate lip sealing and transport of the food bolus towards the posterior zone of the oral cavity. The initiation of swallowing is complicated as a result (7,19,20). Tahmassebi and Luther (20) have pointed to a possible association between lip sealing and the presence of drooling in children with ICP (20).

Vertical malocclusions such as open bite and overjet show an increased prevalence in the population with infant cerebral palsy. In this study, 30% of the sample had anterior open bite, in coincidence with the observations published by Giménez et al. (21) - with a prevalence of 62%. We observed a strong association between the presence of clinical drooling and anterior open bite with difficulties for lip sealing.

The high incidence of drooling in patients with ICP makes it necessary for the specialist to know and offer treatment alternatives based on the situation of each individual patient, with the aim of improving health (22) and quality of life.

Conclusions

The present study shows that the presence of drooling can be related to a number of factors, including antecedents of seizures and the use of antiseizure drugs, the presence of malocclusions such as anterior open bite and, and problems for effective lip sealing.

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