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Bilateral submandibular duct rerouting: Assessment of results on drooling in cerebral palsy cases

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ABSTRACT

Objectives: Drooling is a physiological phenomenon in infants which becomes unusual and even pathologic after 18 months of age. Cerebral palsy (CP), the most common etiology for physical disability, mostly occurs in cases of normal-intelligence kids who are socially active and therefore their disorders require special attention. One of the major problems kids with CP face is excessive drooling and several therapeutic methods have been suggested to treat this problem. In this study described herein, bilateral submandibular duct rerouting (BSMDR) surgery was performed to investigate its effect on drooling in children with CP.

Methods: From March 2007 to April 2011, 16 children aged 6–16 years old with cerebral palsy who suffered from excessive drooling were recruited. A thorough physical examination was performed and a questionnaire was completed for each case. Those who met the inclusion criteria and provided an informed consent were selected for BSMDR surgery. They were followed-up twice, 10 days and 6 months after the operation to evaluate the degree of drooling or other possible side effects of the surgery.

Results: Sixteen patients entered the study and underwent surgery. On the first follow-up visit 87.50% presented overall improvement, of which 56.25% showed good to excellent improvement in contrast to 31.25% who exhibited fair improvement. On the second follow-up an overall improvement was observed in 81.25%, of which 43.75% showed good to excellent improvement compared to 37.50% with fair improvement.

Conclusion: Considering that during both the first and second follow-up visit only two cases (12.5%) did not respond to treatment, it could be concluded that BSMDR surgery is an effective treatment for reducing drooling in CP children.

1. Introduction

Drooling, defined as the excessive secretion of saliva from the mouth – sometimes referred to as sialorrhea is a common disorder considered as normal in infants less than 18 months of age. Drooling should gradually disappear with neuro-muscular development in the child. If it continues after the age of 4 it is regarded as pathologic phenomenon and requires intervention [1,2]. Drooling is common in cerebral palsy (CP) patients [1,3].

CP is the most common etiology for physical disability in children. CP occurs in 2–2.5% of 1000 live births. The statistics has remained unchanged over the last 40 years in spite of all the advances made in this field [4]. Although the exact etiology is still unknown, the main risk factors in CP's occurrence include low birth weight and premature pregnancy. Probable etiologies are categorized in three groups: before, during and after birth [4,5]. The most common disorder in children with CP is mobility problems. However, 10–45% of these children might suffer from visual, auditory and learning disorders, or even seizure.

Around 5–10% of CP patients complain of drooling which causes social isolation, especially in those who demonstrate good mental function and learning abilities. The various factors playing a role in the occurrence of drooling consist of difficulty in the oral phase of swallowing, macroglossia, inability to close the mouth, poor head

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control, and disordered tongue mobility [1,2,6]. The constant flow of saliva from the mouth causes cheilitis, perioral ulcers, secondary infections, recurrent soiling of clothing and bibs requiring frequent changing, and also psychosocial problems due to social isolation [7].

Studies have shown that the treatment of drooling results in less social isolation and so promotes greater participation in society. Although drooling is considered pathologic after the age of 4 and should be treated, in cases of normal development who only suffer from drooling or in CP patients with mild to moderate drooling, treatment could be postponed till the age of 6. In cases in which drooling is due to an acute neurologic deficit, there could be a wait of 6–12 months before the initiation of treatment [1].

Therapeutic modalities for drooling consist of physiotherapy, anticholinergic drugs, intraglandular botulinum toxin injections [8], and surgical techniques [9]. BSMDR is one of the best surgical options currently available. This is due to BSMDR's technical ease, minimal invasiveness and favorable outcomes in comparison to other surgical methods which have long been used for this purpose [9,10]. The study discussed herein was performed with the intent to investigate the results and compare both the short- and long-term outcomes of BSMDR surgery in 16 children with CP suffering from excessive drooling.

2. Materials and methods

Between March 2007 to April 2011, 16 CP children aged 6–16 years who had been suffering from drooling were hospitalized in the ear, nose and throat unit of Dr. Sheikh & Imam Reza Teaching Hospitals for undergoing BSMDR surgery. Cases with +4 tonsillar hypertrophy and occlusive problems were excluded from the study due to difficulties in rerouting surgery. Cases with athetoid CP or with mild drooling under the age of six were also excluded. A full medical history was taken, a thorough physical examination was performed and an informed consent was obtained from each patient's parents prior to hospitalization. Among them two patients had previously received medical therapy consisting of Botox injections, whereas the rest had no history of being treated for drooling.

Before the operation, the patient's drooling severity and frequency were recorded as shown in Table 1 [2]. The patients were divided into two groups, severe and very severe. After surgery, there were two follow-up visits: a short-term visit on days 7–10 and a long-term visit on months 3–6.

Response to treatment was recorded based on Crysdale's criteria [6] as shown in Table 2. The presence of aspiration and coughs both before and after surgery was also studied in every patient. SPSS software package, version 13 was used for all data analyses.

The study protocol was fully approved by the Research Council Ethics Committee of Mashhad University of Medical Sciences.

BSMDR surgery was performed under general anesthesia. After injecting Lidoadrenalin solution in the floor of the mouth, the Wharton's duct opening was dissected from the oral mucosa with a 3–5 mm margin, bilaterally. The oral floor was also incised at the level of the duct up to half of the route; the duct was dissected and separated from the sublingual gland. Afterwards, a submucosal tunnel was constructed in the floor of the mouth up to the anterior pillar of the ipsilateral tonsil. The freed duct was directed into this tunnel. In the final step, mucus along with the duct opening was sutured behind the anterior pillar. The mouth floor was then sutured separately by an absorbable thread (Fig. 1).

Table 1
Drooling classification.

Mild	Wet labial line
Moderate	Wet from inf. lip to chin
Severe	Dirty clothes with saliva
Very severe	Wet hands and clothes

Table 2
Response to treatment based on Crysdale's criteria.

Outcome	First follow-up		Second follow-up	
	Frequency (n = 16)	Percent	Frequency (n = 16)	Percent
Excellent	2	12.5	3	18.75
Good	7	43.75	4	25
Fair	5	31.25	6	37.5
Poor	2	12.5	2	12.5
No follow-up	0	0	1	6.25
Total	16	100	16	100

3. Results

In this before–after study, 11 (68%) patients were male and 5 (32%) female. Their age ranged from 6 to 16 years (mean = 12.00, median = 10.50). They were divided into severe (n = 7, 43.75%) and very severe (n = 9, 56.25%) groups based on the severity of the disease.

A treatment response was achieved in 14 (87.6%) of the 16 patients on the first follow-up. In 9 (56.3%) cases, the response was desirable (good to excellent improvement) while in the other 5 (31.3%) cases a fair response was observed. Two (12.5%) were unresponsive to therapy. On the second follow-up, 3–6 months after surgery, one patient was excluded due to having missed a follow-up visit. The desirable response decreased to 7 cases, one having been excluded and the response in another case having changed from good to fair. In total, a fair response was seen in six patients whereas the response to treatment was poor in two patients.

On the second follow up, response to treatment was 81.2% (13 cases). All non-responsive cases to therapy in both follow up visits were male. No statistically meaningful correlation was found between the level of treatment response and variables such as initial drooling severity, age, sex, tonsillar hypertrophy, protruded tongue, and saliva quality. This could be described by the small number of studied cases which itself was due to ethical considerations.

In this study three patients had complained of pulmonary symptoms and a recurrent cough before surgery. Full recovery in all was reported on the 6-month post-op follow-up. One patient complained of post-surgical coughs which had not been present

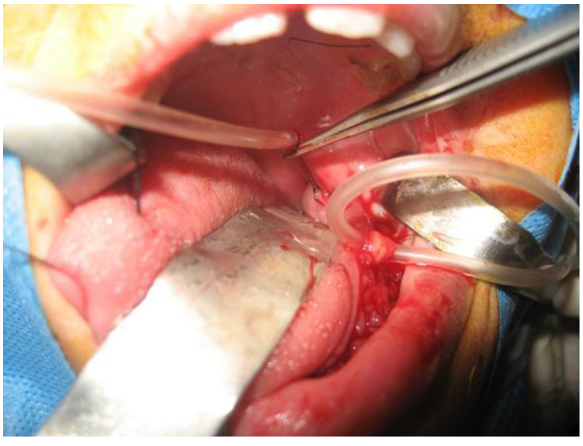

Fig. 1. Suturing of warton duct to anterior tonsillar pillar.