

Original Research Article

Bilateral submandibular duct relocation for chronic sialorrhea: a 15 years of experience in children with a neurological disorder

Imen Mehri Turki^{*} 

Stomatology Maxillofacial Aesthetic and Reconstructive Surgery Department. Mohamed Tahar Maamouri University Hospital, Nabeul, Tunisia

(Received: 21 February 2023, accepted: 12 July 2023)

Keywords:
Drooling / sialorrhea / Wharton duct / relocation / oral surgery / neurological disorders

Abstract – Background: Chronic sialorrhea is a troublesome condition for children with neurological disorders that affect swallowing. Various surgical procedures for reflecting drooling cases are helpful. Some of them could be followed by complications or lead to over-dryness of the mouth. This article aims to highlight the safety and effectiveness of bilateral submandibular duct relocation (BSDR) on fifteen years of experience. **Material and methods:** A retrospective chart review identified 25 patients with chronic sialorrhea who underwent BSDR. Clinical and demographic data were reviewed. Surgical outcomes were assessed with both Wilkie and Brody's criteria and the Teacher drooling (TDS) scale. **Results:** The mean age of children was 11.7 years. They had neurological disorders and dribbled saliva with an average of 4.28 according to the TDS which falls to an average of 1.12 after surgery. By referring to Wilkie and Brody's criteria, 88% of the results have been labelled as excellent and good for the others. **Conclusion:** Our 15 years of experience in managing neurologically impaired children with profuse drooling illuminates and supports our trend for BSDR without sublingual gland excision. This surgical procedure is safe, and effective against drooling, protects oral health, and provides a high rate of success.

Introduction

Drooling or chronic sialorrhea is a troublesome condition of extraoral leakage of saliva. It is not due to salivary hyperproduction but is rather a swallowing dysfunction secondary to a deficiency of neuromuscular or muscular coordination of the oral sphere including dentomaxillar and labial components. Drooling is considered physiologic for up to 18 months. Surgical treatment is delayed awaiting for child's development, or when non-invasive therapeutic modalities including oral motor therapy, anticholinergic drugs, and intraglandular botulinum toxin injections are not efficient [1,2]. The management of drooling is highly challenging for caregivers, distressing for the parents, and repugnant to patients which are typically cast out of society due to their neuromuscular diseases. So a radical solution with valuable and safe surgical management helps drool children.

This article aims to emphasize the safety and efficiency of the double submandibular duct relocation without sublingual gland excision through our 15 years of experience and

long-term follow-up of the postoperative outcome, using the teacher drool scale and Wilkie and Brody's criteria in 25 neurologically impaired children. Otherwise, outcomes of other surgical options have been also described.

Material and Methods

The study was exempted from approval by the local ethical committee because of its retrospective nature. The World Medical Association Declaration of Helsinki Ethical Principles for Medical Research Involving Human Subjects was followed. Written informed consent was provided by the child's legal guardian.

Patient's data collection, and assessment tools

A retrospective medical review was performed on 25 neurologically impaired children with a diagnosis of chronic sialorrhea. Pharmacological therapy for drooling was ineffective in these patients over a period of more than 6 months. The clinical data were reviewed regarding demographic data, drooling severity using the preoperative teacher drool scale [3] (TDS), aetiology of neurological deficit, postoperative

* Correspondence: turkiymen@yahoo.fr

Table I. Teacher drool scale.

Teacher Drool scale (TDS)	Score
No drooling	1
Infrequent drooling, a small amount	2
Occasional drooling, on and off all-day	3
Frequent drooling, but not profuse	4
Constant drooling, always wet	5

Table II. Wilkie and Brody's criteria.

Wilkie and Brody's criteria	
Excellent	Apparently normal salivary control
Good	Slight loss of saliva with or without dried froth on the lip
Fair	Improved but with significant saliva loss
Poor	Failure of significant control or too dry

complications, hospital stay, and recurrence. The assessment of the surgical result was classified on Wilkie and Brody's criteria [4] and the post-operative TDS which was calculated at a minimum of one month after the surgical procedure. The follow-up was completed in one up to two years. The data were analysed with Excell Logiciel.

Teacher Drool scale (TDS): It is a metric for assessing the level and frequency of saliva drooling in patients with chronic sialorrhoea that ranges from 1 if there is no drooling to 5 if profuse and constant drooling exists (Table I).

Wilkie and Brody's criteria were used to classify postoperative results. This assessment was based on the observation of the parents, caretakers, or caregivers (Tab. II).

Surgical procedure

All these children underwent the same surgical approach which was detailed in our previous report and first described in 1974 [1,5] which is the double submandibular duct relocation. It was as follows:

The patient was operated on under general anaesthesia with a nasotracheal tube. Lidocaine infiltration of the floor of the mouth has preceded Wharton's duct catheterism. A spindle-shaped incision centred by the orifice of this duct was made followed by a submucosal dissection around it. The proximal portion of the duct has been identified allowing the skeletonization of 2 cm up to 3 cm length of this salivary channel which was freely movable. Care should be taken for the lingual nerve. A tunnel was made in the floor of the mouth backwards to the palatal arches where the duct was rerouting and the mucous collar containing the orifice of this duct emerged through an incision made in the anterior pillar of the tonsillar fossa and was fixed. The mucosal defect of the floor of

the mouth was sutured, after meticulous haemostasis, without any wound tension. The whole procedure was then repeated on the other side. The perioperative antibiotic was given systematically and conducted for five days. An elbow fixing bracket was positioned bilaterally to avoid traumatic lesions by intraoral hand intrusion.

Results

The age of children and adolescents varied from 6 years and three months to 16 years and two months with an average age of 11 years and seven months (Fig. 1). The sexual predilection was male with a sex ratio of 2/1. All patients demonstrated neurological disorders, the aetiologies of which are shown in (Table III).

The excessive dribbling of saliva was observed, before BSDR, in 22 patients, and only three patients had occasional but continuous daily drool. Results have been labelled as excellent for 22 patients, and good for the others according to Wilkie and Brody's criteria (Fig. 2). Preoperative assessment of drooling revealed a TDS scale above 4 for 88% of patients, with an average of 4.28. This average dropped to 1.12 after the surgical procedure (Fig. 3). Hospital stays ranged from 2 and 4 days during which all patients were under corticosteroid and paracetamol for analgesia except one patient with congenital insensitivity to pain. The need for respiratory support following extubation was not observed. Only two patients had developed postoperative swelling which resolved spontaneously within 48 hours. Furthermore, there were not any haemorrhagic or infectious complications. Additionally, there was no lithiasis or salivary cyst formation at the long-term follow-up. Besides, over-dryness of the mouth was not reported as a postoperative complaint. As well as for the recurrence of drooling, did not occur.

Discussion

Drooling is physiological in infants and usually resolves after 18 months of age as a result of the maturation of orofacial motor function for obtaining swallowing system coordination. But intermittent drooling can persist until four years in the awake status. After that, it is mainly seen in neurologically impaired children [1,6]. When this condition is perpetuated with a constant and continuous pattern, it becomes a serious problem for these patients and their families. Naturally, It leads to psychological repercussions, significant negative economic impact, and social rejection. The inability to swallow adequately increases the risks of aspiration pneumonia, and the leakage of saliva favours also perioral infection, particularly by *Candida albicans* [7].

Swallowing is a physiological process that can be initiated voluntarily but is thereafter under a complex reflex control. Wilkie showed a disturbance in the saliva transport from the frontal parts of the mouth to the pharynx in children with cerebral palsy and drooling. He was the first surgeon in 1967,

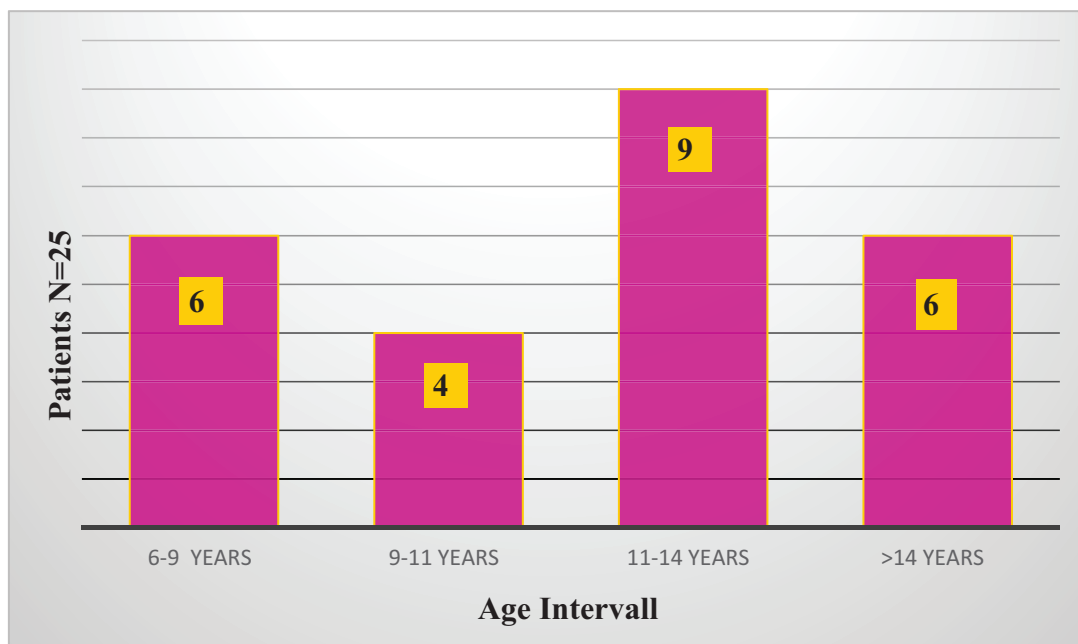


Fig. 1. Age distribution of drooling children.

Table III. The aetiology of neurological disorders and symptoms associated to drooling.

Neurological disorders	Drooling and associated symptoms	Number of patients
Down syndrome	Mental impairment, hypotonia, epilepsy	6
Cerebral palsy	Intellectual disability, dyskinesia, spastic hemiplegia	8
Partington syndrome	Intellectual disability, hand dystonia, dysarthria	3
Hypoxic encephalopathy	Hypotonia, cognitive impairment, epilepsy	3
Autism	Restricted behaviour, anxiety, and emotional reaction	3
Congenital insensitivity to pain	Lack of pain awareness, fingers amputation, anhydrosis	1
Wolf-Hirschhorn syndrome	Intellectual disability, microcephaly, hypotonia	1

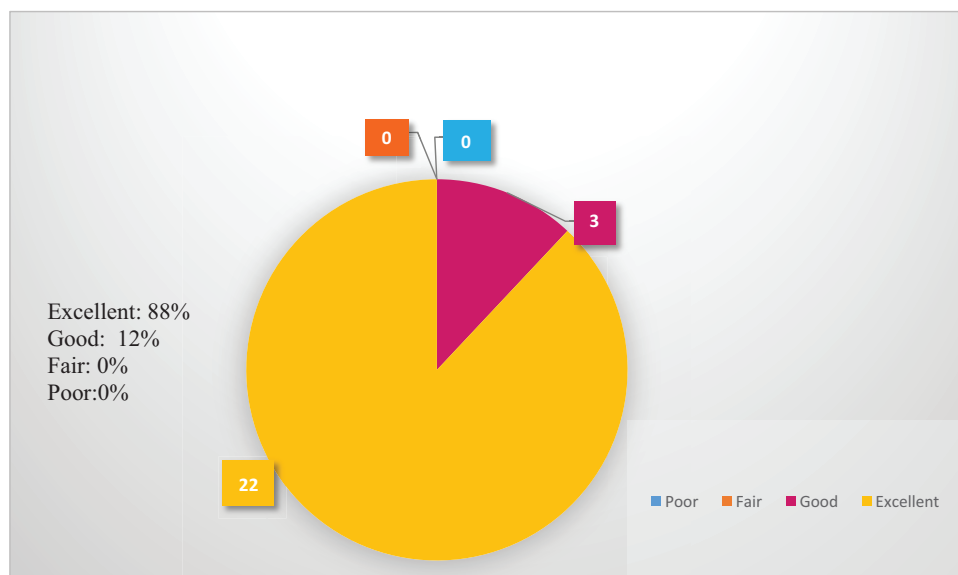


Fig. 2. Outcome assessment according to Wilkie and Brody's criteria.



Fig. 3. Distribution of children according to TDS before and after BSDR.

who described a surgical treatment for drooling consisting of bilateral parotid duct relocation and bilateral submandibular gland excision. The successful result was obtained at a range of 86% [4]. However dental caries and periodontal complications were observed due to xerostomia which also burdens food mastication and speech fluidity. The moisturizing of the oral cavity is then poorly assured by minor mucous glands which contribute just less than 10% of the volume of the whole saliva at rest [8]. Adding to these significant disadvantages of xerostomia, and external scar following bilateral submandibular sialoadenectomy, there is also a potential risk of multiple nerve injuries such as of the lingual nerve, the hypoglossal nerve, and the facial marginal nerve. Postoperative hematoma could easily occur and wound infection happen to lead to the worsening of the cervical scar [1]. Looking for fewer complications and fewer morbidity, bilateral parotid duct ligation combined with bilateral submandibular duct relocation was described [9]. The purpose of duct ligation is to obtain glandular atrophy. For this purpose, four duct ligation was also reported particularly to control aspiration pneumonitis [10]. However, complications inherent to xerostomia such as gingivitis, burning sensation of the mucous membrane, rampant caries, cheilitis, and tongue and palate crusting are frequently seen. Thus, four duct ligation seems to be an easy and safe procedure but 68.8% of drooling recurs within 6 months. This is attributed to an eventual erosion of the duct by tight ligature at the weak point of ligation leading to new opening and fistula formation [11].

Chorda tympani neurectomy is known to reduce the salivary flow rate of both the submandibular and sublingual glands, but it is not an efficient method for drooling management. Besides, it produces a loss of taste in the anterior two third of the tongue taking away the gustative pleasure from these children who have so little pleasure in their life. Furthermore, hearing loss is a possible complication of this neurectomy [12], which is in our regard more dramatic than sialorrhea.

The problematic saliva in the drooling child is produced by the submandibular gland which represents 70% of salivary secretion in a "resting state" [13]. Hence, rerouting saliva leakage from the Wharton duct directly in the pharynx is a good option against saliva stasis in the anterior portion of the mouth and then dribbling saliva. It also initiates the swallowing reflex while at the same time maintaining a moist healthy oral cavity due to parotid and sublingual saliva secretion. The most physiologic surgical procedure is the double submandibular duct relocation, as performed for our patients. It has been reported for the first time in 1974 by Ekedahl who associated a simultaneous sublingual duct ligation [5]. Instead of sublingual duct ligation, the excision of the sublingual gland was performed by other surgeons to prevent the occurrence of postoperative ranula [14]. This complication did not happen to our patients knowing that submandibular duct relocation was used without any additional surgical or medical treatment. Sublingual gland excision increases the surgical time and leads to more dissection in the floor of the mouth, adding morbidity and supplementary risk of bleeding and oedema which causes postoperative difficulties in respiration leading to respiratory

assistance [1]. Furthermore, other surgical teams revealed a higher rate of postoperative haemorrhage and pain with additional sublingual gland excision respectively in 13.7% and 36% of patients rather than 3% and 12% of patients when bilateral submandibular duct relocation was just performed [15]. So, they no longer use additional sublingual gland excision while they reported efficient results for drooling as shown in our surgical trend, where TDS falls from 4.23 to 1.12 after Bilateral submandibular duct relocation. Also, according to Wilkie and Brody's criteria, excellent results were labelled in 88% of our patients and good results for the others. Hence, we can judge that BSDR gives successful results. Similar studies with submandibular duct relocation performed alone or in combination with other procedures such as parotid duct ligation or sublingual gland excision reported a success rate ranging from 75% to 89% [7]. It has been reported that drooling recurrence was not successfully managed with additional surgical procedures. That means that the recurrence of drooling is not caused by surgical failure of the primary intervention, but other additional factors such as dental malocclusion, head position, and further medication like anticonvulsant drugs should be revised and well managed [1,6]. In the whole follow-up of our children, recurrence of drooling was not detected. Otherwise, some authors advocate that submandibular duct relocation cannot be performed in patients with a history of recurrent aspiration pneumonia, so procedures that decrease salivary output, are more advisable. However, aspiration pneumonia did not occur following the increase of the salivary flow in the pharynx for our neurologically impaired patients. As well as for the occurrence of duct lithiasis, salivary cysts, and sialadenitis which are reported in other experiences [1,16].

Conclusion

Our 15 years of experience in managing neurologically impaired children with profuse drooling illuminates and supports our trend for bilateral submandibular duct relocation without sublingual gland excision. This surgical minimally invasive procedure is the most conservative and physiological approach providing a scarless technique with low complications and a high success rate. Drool treatment effectiveness while maintaining the hydration of the mouth is crucial for oral health and oral function. The outcomes of this safe and effective surgical procedure improve significantly the quality of life of drooling children and their families.

Conflict of Interests

None

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

The principles stated in the Declaration of Helsinki "Ethical Principles for Medical Research Involving 'Human Subjects'" are respected. The study was exempted from approval by the local ethical committee because of its retrospective nature.

Informed consent

The surgical procedure was explained verbally, and written informed consent was obtained from the children's legal guardians.

Author's contribution

Conceptualisation, Methodology, Investigation, Writing original draft preparation, Reviewing and Editing: Imen Mehri Turki. The author has read and agreed to the published version of the manuscript.

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