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Bilateral Submandibular Duct Relocation with High Frequency Radiosurgery

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Summary
The aim of this study was to describe and investigate the efficacy and safety of radiosurgery assisted bilateral submandibular duct relocation for neurologically impaired children suffering from excessive drooling.
Patients referred with excessive drooling after failure of 6 months of oral-motor training and conservative methods were included into the study. Exclusion criteria were: dental caries and patients with recurrent aspiration pneumonias. In the period of 2000 – 2007, fourteen children and young adults with persistent drooling underwent radiosurgery assisted bilateral submandibular duct relocation and sublingual gland excision. A retrospective case note review was performed and a questionnaire study was conducted.
Oral-motor function assessment was carried out with the use of a 4 degree scale preoperatively, 4 months postoperatively and currently. The average follow up time was 8-26 months; a majority of the patients achieved a satisfactory reduction in sialorrhea (79 %). The average time of onset of improvement in drooling was 3 weeks (from immediately to 5 months). 10 parents (71%) were happy with the outcome and would recommend the procedure to the parents of other children.
The complications included three transient sublingual swelling in the early and 2 ranulas in the late postoperative periods. Average of the surgical time was 48 minutes, about 30 % less, comparing with the previously favoured cold knife technique.
Radiosurgery opens a new therapeutic approach for neurologically disabled children suffering from excessive drooling. It combines the advantages of both cold knife and lasers being easy, safe, precise and effective, and having excellent tactile and haemostatic properties.
Key words: drooling, radiosurgery, submandibular duct relocation, treatment outcome

Introduction
Drooling in the normal infant is a psychological phenomenon that usually resolves after the age of 6 months. Sialorrhea or excessive drooling is a major cause in poor quality of life and a common problem in a neurologically impaired child. It can occur in 10 - 58 % of children with cerebral palsy and also a persistent feature in other cases of congenital and acquired neurological disorders [1, 2]. The cause is often multifactorial, but it appears to be primarily a defect in the oral phase

of swallowing. This is brought about by a combination of poor head control, an inability to close the mouth, poor lip control and disordered tongue mobility [3].

The medical, psychosocial and economic impact of drooling is underestimated. The continuous sialorrhea results in soiling of clothes, constant facial irritation and the unsightly nature results in segregation of these patients from other members of society. Ineffective swallowing may result in aspiration pneumonia. A drooling child is less likely to be held, and is thus deprived of affectionate social interaction. In older children, drooling is embarrassing and detrimental to peer bonding [4].

Management of drooling in childhood involves a multidisciplinary team approach with the participation of a pediatric neurologist, family care physician, dentist, speech therapist and an ENT surgeon [4,5].

The significance of drooling and the therapeutic recommendations depend on the clinical status of the individual affected and the degree of sialorrhea [5]. Physiotherapy (oral motor therapy) is aimed at correction of head and jaw posture. This is also used for increasing the mobility and strength of tongue and lip movement, however it has a long-term benefit only in a small number of cases [4,6,7]. Pharmacotherapy (anticholinergics, Botox) is of use in the short term but confers little long-term benefit [8-10].

There have been various surgical approaches described in the literature including parotid duct rerouting and submandibular gland excision [11-12] parotid duct ligation with submandibular gland excision [1] submandibular duct relocation or excision [13], four duct ligation [14] and transtympanic neurectomy [15].

In the group of patients reported here submandibular duct relocation with the use of radiosurgery was the procedure undertaken. We hypothesized that combining the advantages of cold knife and lasers, radiosurgery will be an effective, safe procedure with tactile and haemostatic properties.

Materials and methods

Patients

Between January 2000 and July 2007, 14 children and young adults with persistent drooling underwent bilateral submandibular duct relocation with high frequency radiosurgery (BSDRwHFR) at the Heim Pál Hospital for Sick Children. A retrospective review of case notes was performed, and the following data were collected. The study group consisted of 8 males and 6 females. The age range varied from eight years to 21 years (mean: 15,5 years**).** All patients were mentally normal and demonstrated a neurological deficit: 12 of them suffered from cerebral palsy, while the aetiology of the remaining two patients was hypotonia.

All patients were accompanied by a care taker and were assessed prior to surgery by an ENT surgeon and a speech therapist. A full ENT examination was carried out with specific emphasis on the assessment of head posture and control, the oral seal and lip and tongue control. The patients selected for surgery failed to respond to a 6 months course of conservative treatment (speech/physiotherapy and pharmacological treatment). Our exclusion criteria were: history of aspiration pneumonias and/or significant dental caries. All procedures were performed under general anaesthesia by the first author (G.K.) and using the same technique.

Radiofrequency surgery

A radiofrequency apparatus (Sugitron 4.0 Dual RF/120 IEC – Ellman International Inc.) was applied with a wire electrode to cut through the lingual frenulum and the mucous membrane (CUT mode), and to prepare the submandibular duct in the submucosal tissue adjacent to the lateral lingual sulcus (CUT/COAG mode). The optimum radiosurgery wave is a frequency of 4,0 MHz. The radio waves are transformed from the electrode tip to the patient and are returned to the machine by a neutral antenna plate. Impedance to the passage of the radio waves through the tissue generates heat within the cells, which boils intracellular tissue water, creating steam (cellular volatilization), and the resulting vaporization results in either cutting or coagulation of tissue. Since radiofrequency generates less heat than conventional cautery, less collateral damage is seen therefore healing is faster.

Questionnaire survey

A questionnaire survey was conducted to determine the symptomatic improvement, parent/carer satisfaction and the complication rate. Non-responders were followed up by telephone.

Ethical consideration

The study was approved by the local research and ethics committee of the Heim Pál Hospital for the Sick Children prior to commence.

Surgical technique

Having carried out tonsillectomy before**,** the tip of the tongue was elevated with a strong suture, exposing the floor of the mouth (2.0 Vicryl, Ethicon ®). The radiosurgical instrument (Sugitron 4.0 Dual RF/120 IEC – Ellman International Inc ®.) was switched on and with the use of a wire electrode the lingual frenulum was horizontally cut and the incision was extended beneath and below both carunculas (CUT mode, fully filtered waveform, digital setting power 8-10, which corresponds 10-15 Watt power) A 6.0 Vicryl suture was inserted into the soft tissue cuff adjacent to the openings of the ducts, which were separated in the midline. With the gentle elevation of these sutures the submandibular ducts were followed and prepared in the submucosal region until the lingual nerves were visualized (CUT/COAG mode, wire electrode, digital power setting 30-40, which corresponds to 40-45 Watt power). A submucosal tunnel was then created from the anterior incision to the base of the anterior faucial pillar adjacent to the tongue base. The ducts were then pulled through the tunnels and anchored to the faucial pillars. Once we were convinced that the ducts were not strangulated the sublingual glands were excised also radiosurgically and the wounds were closed with sutures. During the procedures the haemostasis was excellent, the surgical field was almost bloodless. The patients were discharged in average on the third postoperative days.

Results

In the questionnaire study there were 12 replies (84 %), the remaining two carers were interviewed by the phone. **Table 1** shows the changes of the severity of oral motor function preoperatively, four months postoperatively and currently. The average time of onset of improvement in drooling was i.e. three weeks and this ranged from immediately to five months.

Table 1. Change of the severity of oral motor function preoperatively, 4 months postoperatively and currently.

Grade	Description	Number of patients		
		Preop	Postop	Currently
Mild	Slight limitation of activity	0	7	5
Moderate	Moderate limitation of activity	8	5	6
Severe	Unable to carry out useful activity	6	2	3
Total		14	14	14

Table 2 shows the carers' satisfaction rate concerning the surgical procedure using the Crysdale criteria (16). Only three parents (21%) reported poor surgical outcome.

Parents were asked also whether or not they were happy with the results of the procedure. 10 parents (71%) were happy with the outcome and would recommend the procedure to the parents of other children. Parents of two children (14,5 %) were not sure, while two parents (14,5 %) would advocate against the procedure.

Table 2. Results of surgery with the us of Crysdales'criteria. Only 3 parents (21%) reported poor surgical outcome.

Crysdale's criteria	Description	Number of patients (n: 14)
Excellent	No saliva on chin, lower lip may be moist	4
Good	Saliva on chin only	4
Fair	Drooling reduced but still loss of saliva off chin	3
Poor	No change	3

The average follow up time was 8-26 months; a majority of the patients achieved a satisfactory reduction in sialorrhea (79 %). The retrospective review of the case notes showed that the average of the surgical time was 48 minutes, which is about 30 % less, comparing with the average length of the cold knife technique what we previously favoured (75 minutes). The length of hospital stay ranged from two days to four days with a median stay of 2,4 days. No early postoperative complications including haemorrhage, high temperature, healing difficulty, cracked lips occurred. Three patients complained of mild swelling in the sublingual area, without compromising their feeding. Late complications including ranula formation occurred in two patients which required surgical excision.

No lingual palsy, transient submandibular swelling occurred in any patients. Neither an increase in aspiration pneumonias nor increase in dental caries was reported during the follow up period.

Discussion

A number of factors have been identified that cause a predisposition to drooling in a neurologically impaired child, including a defect in the oral phase of swallowing, poor head control, constant open mouth posture, poor lip and tongue control, a hypoactive gag reflex, diminished intra-oral sensation and constant tongue thrusting activity [17]. The management includes both non-surgical and surgical means. The non-surgical means include physiotherapy [4, 6, 7] and pharmacotherapy [8-10].

Surgery is the treatment of choice in patients with severe drooling unresponsive to conservative management. Submandibular duct transposition was first described by Laage-Hellmann in 1969 [18] and since then is the most widely performed procedure in sialorrhea. The advantages are a scarless procedure, low complications and high success rate. The rationale behind the operation is that 70 % of saliva secretions in a resting state i.e. between meals are from the submandibular glands (500-700 ml over a 24 hour period), which being thick and mucoid gives the appearance of the "drooling child" [19]. One of the other reasons for success of the relocation procedure is that saliva coming in contact with the base of tongue initiates a swallow reflex [5]. In a study evaluating glandular function by technetium scanning, Hotaling [20] demonstrated maintained long- term postoperative function in at least one gland.

There are two debatable points regarding the procedure. The first is the need for tonsillectomy. O'Dwyer state that a tonsillectomy should be performed prior to the procedure if there is a history of recurrent tonsillitis or if grossly enlarged tonsils are encountered [17]. Crysdale advocates a one-stage procedure and removes the tonsils which are large enough to obstruct the relocated ducts [21]. The second debatable point is the synchronous excision of sublingual gland tissue. Crysdale state that patients undergoing synchronous excision of the sublingual gland tissue had significantly fewer complications that required additional surgery (i.e. excision of ranulas) [21] and his view is widely accepted. However, in their recent publication, Glynn and O'Dwyer state, that the addition of sublingual gland excision increases morbidity and they no longer excise these glands [16].

Today, radiofrequency surgery is widely used in otolaryngology. Inferior turbinate, soft palate, and tongue base applications are the most popular applications. [23, 24]. We felt, that introduction of this method into bilateral submandibular gland relocation has several advantages. It combines the advantages of both laser and cold knife. Bridenstine found biopsies done with radiofrequency incision to have thermal damage zones of 75 microns, which comparable to the CO_2 laser [25]. Other studies have confirmed minimal tissue damage and comparable biopsy margins with scalpel incision [26]. The micro sharp tip of the radiosurgical probe concentrates the radiofrequency energy, resulting in improved cutting and haemostasis precision and ensures the least amount of thermal damage to adjacent tissues. This induces no carbonisation, less scarring and fast healing. It also allows excellent tactile feedback, and less pain and swelling. It minimizes risks of injury to the sur-

rounding structures (veins of the floor of mouth, hypoglossal nerve, lingual nerve). It combines the potential of short operating time with a cost effective machine. At last but not at least the procedure does not require any safety precautions.

The details and the advantages of the submandibular duct relocation procedure has widely discussed in the literature [4, 7, 17, 19, 22]. However, our report is the first in the literature to describe the radiosurgery assisted submandibular duct relocation. Our case series is rather small, however clearly demonstrates the advantages of radiosurgery. We were especially pleased to experience, that no short term complications compromising the feeding occurred, and only 2 ranulas were diagnosed as long term complications. All the other aspects of the post surgical result were comparable to the other case series in the literature [4, 7, 17, 19, 22].

Independently of the surgical method used, it is difficult to predict which patient will have an unsuccessful outcome following surgery. O'Dwyer found in their series that the patients who failed to improve had the most severe oral-motor dysfunction [17]. We feel that especially in cases where the head control is unchanged following longstanding conservative treatment other surgical options need to be considered (i.e. excision of submandibular glands). We are aware of the fact, that dentists and maxillofacial surgeons have expressed concerns about dental caries[27]. We do ensure, however, that careful attention is paid to dental hygiene by a pediatric dentist in the years following the procedure.

Conclusion

Radiosurgery assisted bilateral submandibular gland excision opens up a more favourable surgical approach for neurologically impaired children suffering from drooling. Our case series is rather small but clearly demonstrates the advantages of this technique: it combines the advantages of both cold knife and lasers in this difficult region, being easy and safe, precise and effective with excellent tactile and haemostatic properties.

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