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HEAD AND NECK

Bilateral submandibular duct relocation by high-frequency radiosurgery

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Abstract The aim of this study was to investigate the efficacy and safety of radiosurgical bilateral submandibular duct relocation in neurologically impaired children and young adults suffering from excessive drooling. The enrolled patients were referred with excessive drooling after the failure of 6 months of oral-motor training and conservative methods. The exclusion criteria were dental caries and recurrent aspiration pneumonia. During 2000–2007, 14 children and young adults with persistent drooling underwent radiosurgical bilateral submandibular duct relocation and sublingual gland excision. A retrospective case note review was performed and a questionnaire study was conducted. The oral-motor function was assessed on a 4-degree scale preoperatively, 4 months postoperatively and after an average follow up time of 8–26 months. A majority of the patients (79%) achieved a satisfactory reduction in sialorrhoea. The average time of onset of an improvement in the drooling was 3 weeks (ranging from immediately to 5 months). The parents of 10 (71%) patients were happy with the outcome and would recommend the procedure to the parents of other children. The complications included three cases of transient sublingual swelling in the early, and two cases of ranula in the late postoperative period. The average duration of surgery was 48 min, i.e., about 30% less than for the previously favoured cold knife technique. Radiosurgery furnishes a new therapeutic approach for neurologically disabled children suffering from excessive drooling. It combines the advantages of the cold knife and laser techniques: it is easy, safe, precise and effective, with excellent tactile and haemostatic features.

Keywords Drooling · Radiosurgery · Submandibular duct relocation · Treatment outcome

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Introduction

Drooling in the normal infant is a psychological phenomenon that usually resolves after the age of 6 months. Sialorrhoea or excessive drooling is a major cause of a 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 is also a persistent feature in other 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 impacts of drooling are generally underestimated. The continuous sialorrhoea results in the soiling of clothes and constant facial irritation, and the unsightliness leads to the 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 or cuddled, and is thus deprived of affectionate social interaction. In older children, drooling is embarrassing and detrimental to peer bonding [4].

The management of drooling in childhood involves a multidisciplinary team approach, with the participation of a

paediatric neurologist, a family care physician, a dentist, a speech therapist and an ENT surgeon [4, 5].

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

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

In the group of patients reported in the present paper, the procedure undertaken was submandibular duct relocation with the use of radiosurgery. We hypothesized radiosurgery (combining the advantages of the cold knife and laser techniques) would be an effective and safe procedure with appropriate tactile and haemostatic features.

Materials and methods

Patients

Between January 2000 and July 2007, 14 children and young adults with persistent drooling underwent bilateral submandibular duct relocation through the use of high-frequency radiosurgery at the Heim Pál Hospital for Sick Children. A retrospective review of the case notes was performed, and various data were collected. The study group consisted of 8 males and 6 females. The range of age at surgery varied from 8–21 years (mean 15.5 years**).** All the patients were mentally normal, but demonstrated a neurological deficit: 12 of them suffered from cerebral palsy, while the aetiology of the remaining 2 patients was hypotonia.

All the patients were accompanied by a caretaker 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 had failed to respond to a 6-month course of conservative treatment (speech/physiotherapy and pharmacological treatment). The exclusion criteria were: a history of aspiration pneumonia and/or significant dental caries. All surgical procedures were performed under general anaesthesia by the first author (G.K.), using the same technique.

Radiofrequency surgery

A radiofrequency apparatus (Sugitron 4.0 Dual RF/120 IEC–Ellman International Inc.) was applied using 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 pass from the electrode tip to the patient and are returned to the machine via a neutral antenna plate. The impedance to the passage of the radio waves through the tissue generates heat within the cells, which boils the intracellular tissue water, creating steam (cellular volatilization), and the resulting vapourization results in either cutting or coagulation of the tissue. Since radiofrequency generates less heat than conventional cautery, less collateral thermal damage occurs, and the healing is therefore faster.

Questionnaire survey

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

Ethical consideration

The study was approved in advance by the local research and ethics committee of the Heim Pál Hospital for Sick Children.

Surgical technique

Following tonsillectomy, the tip of the tongue was elevated with a strong suture, exposing the floor of the mouth (2.0 Vicryl, Ethicon). The radiosurgical instrument (Surgitron 4.0 Dual RF/120 IEC–Ellman International Inc.) was switched on, the lingual frenulum was cut horizontally with the use of a wire electrode, and the incision was extended beneath both carunculas (CUT mode, fully filtered waveform, digital setting power 8–10, which corresponds to a power of 10–15 W) (Fig. 1). 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 (Fig. 2). With 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, fully rectified waveform, digital power setting 30–40, which corresponds to a power of 40–45 W). A submucosal tunnel was then created from the anterior incision to the base of the anterior faucial pillar adjacent to the base of the tongue (Fig. 3). The ducts were next pulled through the

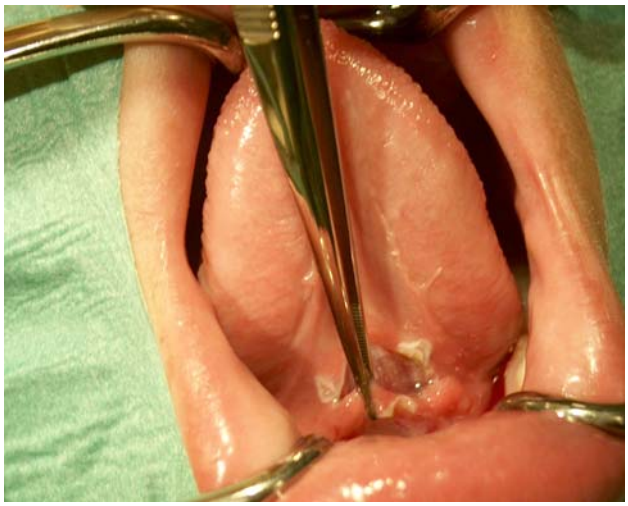

Fig. 1 The lingual frenulum was cut horizontally and the incision was extended beneath both carunculas

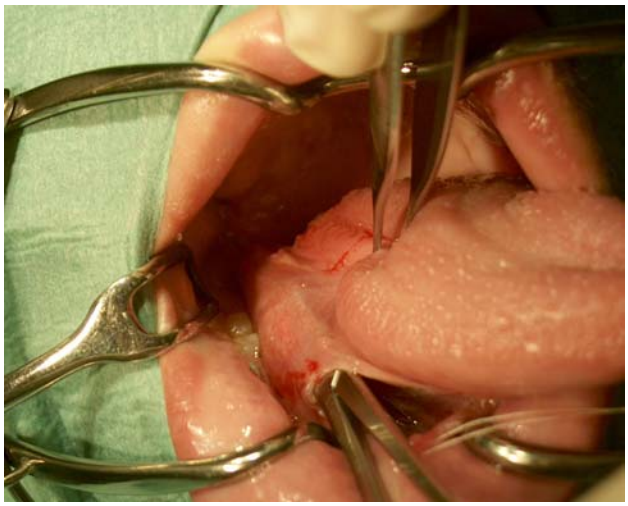

Fig. 3 Creation of a submucosal tunnel from the anterior incision to the base of the anterior faucial pillar adjacent to the base of the tongue

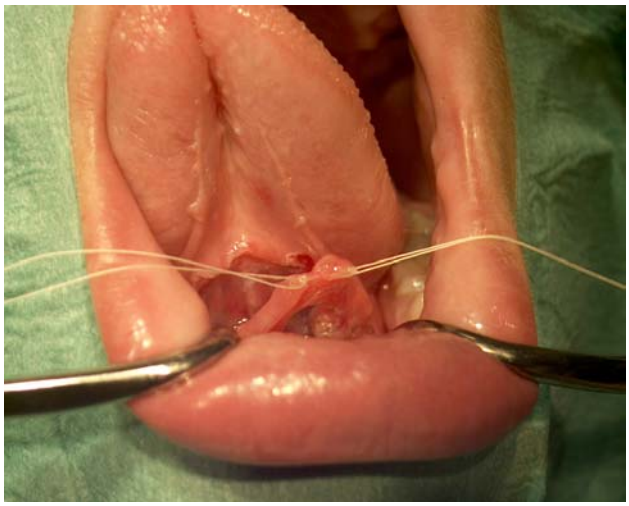

Fig. 2 The openings of the ducts were separated in the midline. 6.0 Vicryl sutures were inserted into the soft tissue cuff adjacent to the openings of the ducts

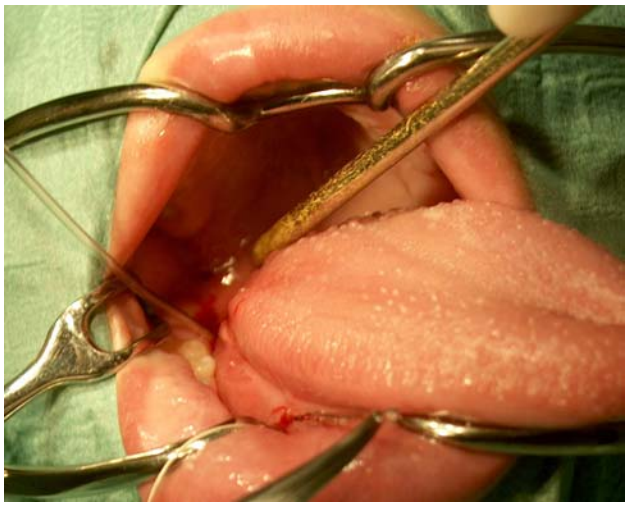

Fig. 4 The ducts were pulled through the tunnels and anchored to the faucial pillars. Note the bloodless surgical field

tunnels and anchored to the faucial pillars (Fig. 4). 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 on average on the third postoperative day.

Results

The questionnaire study resulted in 12 responses (84%). The carers of the remaining two patients were interviewed by phone. Table 1 shows the changes in the oral-motor function 4 months postoperatively and currently. The average time of onset of the improvement in drooling was approximately 3 weeks, ranging from immediately up to 5 months. Table 2 shows the satisfaction rate of the carers as concerns the surgical procedure based on the Crysdale criteria [16]. Only the parents of three patients (21%) considered that the surgical outcome was poor. Table 3 shows to what degree patients individually benefited from the surgical procedure.

The parents of ten patients (71%) were happy with the outcome and would recommend the procedure to the parents of other children, the parents of two children (14.5%) were not sure, while the parents of the remaining two patients (14.5%) would advocate against the procedure.

The follow-up time of 8–26 months, a majority of the patients (79%) achieved a satisfactory reduction in the level of sialorrhoea. The retrospective review of the case notes

Table 1 Changes in the oral-motor function 4 months postoperatively and currently

Grade	Description	Number of patients		
		Preop	Postop 4 months	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 Results of surgery based on the criteria of Crysdale

Only the parents of 3 patients (21%) considered that the surgical outcome was poor

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

showed that the average duration of surgery time was 48 min, which is some 30% less, than the average length of the cold knife technique that we previously favoured (75 min). The length of hospital stay ranged from 2 to 4 days, with a median stay of 2.4 days. No early postoperative complications (including haemorrhage, high temperature, healing difficulty or cracked lips) occurred. Three patients complained of mild swelling in the sublingual area, but it did not compromise their feeding. Late complications occurred in two patients, including ranula formation, which required surgical excision. No lingual palsy or transient submandibular swelling was noted in any patient. Neither an enhanced frequency of aspiration pneumonia nor an 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, a constant open mouth posture, poor lip and tongue control, a hypoactive gag reflex, diminished intraoral sensation and constant tongue thrusting activity [17]. The management includes both non-surgical and surgical steps. The non-surgical steps 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, first described by Laage-Hellman in 1969 [18], has subsequently become the most widely performed procedure in sialorrhoea. The advantages are a scarless procedure, few complications and a high success rate. The rationale behind the operation is that 70% of the saliva secreted in a resting state, i.e. between meals, is from the submandibular glands (500–700 ml over a 24-h period), and this thick and mucoid saliva gives the appearance of the "drooling child" [4, 7, 16, 19]. One of the reasons for the success of the relocation procedure is that the saliva coming into contact with the base of the tongue

Table 3 The degree of individual benefit of our patients following surgery

	Name (initials)	Age (years)	Diagnosis	Initial grade of drooling	Late grade of drooling
1.	B.H.	8	Cp	Severe	Severe
2.	K.SZ.	11	Cp	Severe	Severe
3.	K.L.	17	Hypotonia	Severe	Moderate
4.	K.N.	9	Cp	Moderate	Mild
5.	M.M.	19	Cp	Severe	Moderate
6.	L.P.	20	Cp	Moderate	Mild
7.	G.Z.	22	Cp	Severe	Severe
8.	B.O.	14	Cp	Moderate	Moderate
9.	SZ.P.	16	Cp	Moderate	Moderate
10.	L.R.	20	Cp	Moderate	Mild
11.	D.A.	14	Cp	Severe	Mild
12.	R.A	13	Hypotonia	Moderate	Moderate
13.	M.P.	17	Cp	Moderate	Moderate
14.	F.E.	19	Cp	Moderate	Mild

Cp Cerebral palsy

initiates a swallow reflex [5]. In a study in which glandular function was evaluated, by means of technetium scanning, Hotaling et al. [20] demonstrated a maintained long-term postoperative function in at least one gland.

There are two debatable points regarding the procedure. The first is the need or not for tonsillectomy. O'Dwyer et al. [17] stated that tonsillectomy should be performed prior to the procedure if there is a history of recurrent tonsillitis or if grossly enlarged tonsils are involved. Crysdale advocates a one-stage procedure and removes tonsils large enough to obstruct the relocated ducts [21]. The second debatable point is the synchronous excision of sublingual gland tissue. Crysdale et al. [22] reported that patients undergoing synchronous excision of the sublingual gland tissue had significantly fewer complications that required additional surgery (i.e. the excision of ranulas) and their view is widely accepted. However, in their recent publication, Glynn and O'Dwyer [19] observed, that the addition of sublingual gland excision increases the morbidity and consequently they no longer excise these glands.

In our case series, two ranulas formation occurred as a late complication, apart from the excision of the sublingual glands. This relatively high number is not related to the radiosurgical technique; given the small number of cases it must be a coincidence.

Today, radiofrequency surgery is widely used in otolaryngology. The most popular applications relate to the inferior turbinate, the soft palate, and the base of the tongue [23, 24]. We presumed that introduction of this method for bilateral submandibular gland relocation would have several advantages. It combines the advantages of the laser and cold knife techniques. Bridenstine [25] found that biopsies carried out with radiofrequency incision exhibited thermal damage zones of 75 microns, which is comparable to the situation with the CO_2 laser. Other studies have confirmed only minimal tissue damage and comparable biopsy margins following 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 minimum amount of thermal damage to the adjacent tissues. It induces no carbonization, less scarring and fast healing. It also allows an excellent tactile feedback, and less pain and swelling. It minimizes the risks of injury to the surrounding structures (the veins of the floor of the mouth, the hypoglossal nerve and the lingual nerve). It combines the potential of short operating time with a cost-effective machine. And last, but not least, the procedure does not require any particular safety precautions.

The details and the advantages of the submandibular duct relocation procedure have been widely discussed in the literature [4, 7, 16, 17, 22]. However, this is the first literature description of radiosurgical submandibular duct relocation. Our case series is rather small, but it clearly demonstrates the advantages of radiosurgery. We were especially pleased to detect no short-term complications compromising the feeding, and the only long-term complications were two cases of ranula. All the other aspects of the post-surgical result were comparable to those of other case series in the literature [4, 7, 16, 17, 22].

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

Conclusions

Radiosurgical bilateral submandibular gland excision comprises favourable surgical approach for neurologically impaired children suffering from drooling. Our case series, though rather small, clearly demonstrates the advantages of this technique: it combines the advantages of the cold knife and laser techniques in this difficult region; it is easy, safe, precise and effective, and displays excellent tactile and haemostatic features.

References

1. Crysdale WS (2002) The management of drooling. In: Bluestone CD, Stool SE, Alper CM et al (eds) Paediatric otolaryngology, 4th edn. Saunders, Philadelphia, pp 1138–1148
2. Tahmasebbi JF, Curzon ME (2003) Prevalence of drooling in children with cerebral palsy attending special schools. Dev Med Child Neurol 45:613–617
3. O'Dwyer TP, Conlon BJ (1997) The surgical management of drooling–a 15 year follow up. Clin Otolaryngol 22:284–287
4. Lal D, Hotaling AJ (2006) Drooling. Current Opin Otolaryngol Head Neck Surg 14:381–386
5. Crysdale WS, White A (1989) Submandibular duct relocation for drooling: a 10 year experience with 194 patients. Otolaryngol Head Neck Surg 101:87–92
6. Panarese A, Ghosh S, Hodgson D, McEwan J, Bull PD (2001) Outcomes of submandibular duct re-implantation for sialorrhea. Clin Otolaryngol 26:143–146
7. De M Adair R, Golchin K, Cinnamond MJ (2003) Outcomes of submandibular duct relocation: a 15-year experience. J Laryngol Otol 117:821–823
8. Jongerius PH, van Tiel P, van Limbeek J et al (2003) A systematic review for evidence of efficacy of anticholinergic drugs to treat drooling. Arch Dis Child 88:911–914

9. Jongerius PH, Rotteveel JJ, van Limbeek J et al (2004) Botulinum toxin effect on salivary flow rate in children with cerebral palsy. Neurology 63:1371–1375
10. Gerlinger I, Szalai G, Hollódy K, Németh A (2007) Ultrasound-guided intraglandular injection of botulinum toxin A in children suffering from drooling. J Laryngol Otol (in press)
11. Wilkie TF (1967) The problem of drooling in cerebral palsy: a surgical approach. Can J Surg 10:60–70
12. Dundas DF, Peterson RA (1979) Surgical treatment of drooling by bilateral parotid duct ligation and submandibular gland resection. Plast Reconstr Surg 64:47–51
13. Guerin RL (1979) Surgical management of drooling. Arch Otolaryngol 105:535–537
14. Kelm C, Mair EA (1999) Four-duct ligation: a simple and effective treatment for chronic aspiration from sialorrhea. Arch Otolaryngol Head Neck Surg 125:796–800
15. Arnold HG, Gross CW (1977) Transtympanic neurectomy: a solution to drooling problems. Dev Med Child Neurol 19:509–513
16. Crysdale WS (1980) The drooling patient: evaluation and current surgical options. Laryngoscope 90:775–783
17. O'Dwyer TP, Timon C, Walsc MA (1989) Surgical management of drooling in a neurologically damaged child. J Laryngol Otol 103:750–752
18. Laage-Hellman JE (1969) Retroposition augl submandibularis utforsgong som behandling vid drazling. Nord Med 82:1522
19. Glynn F, O'Dwyer TP (2007) Does the addition of submandibular gland excision to submandibular duct relocation give a better overall result in drooling control? Clin Otolaryngol 32:103–107
20. Hotaling AJ, Madgy DN, Kuhns LR, Filipek L, Beleenky WM (1992) Postoperative technetium scanning in patients with submandibular duct diversion. Arch Otolaryngol Head Neck Surg 118:1331–1333
21. Crysdale WS (1992) Drooling. Experience with team assessment and management. Int J Pediat Otorhinolaryngol 9:241–248
22. Crysdale WS, Raveh E, Mc Cann C, Roske L, Kotler A (2001) Management of drooling in individuals with neurodisability: a surgical experience. Dev Med Child Neurol 43:379–383
23. Sapci T, Usta C, Evcimik F, Bozkurt Z, Aygun E, Karavus A, Peker M (2007) Evaluation of radiofrequency termal ablation results in inferior turbinate hypertrophies by magnetic resonance imaging. Laryngoscope 117:623–627
24. Powell NB, Riley RW, Troell RJ et al (1998) Radiofrequency volumetric tissue reduction of the palate in subjects with sleep-disordered breathing. Chest 113:1163–1174
25. Bridenstine JB (1998) Use of ultra high frequency electrosurgery (radiosurgery) for cosmetic surgical procedures. Dermatol Surg 24:397–400
26. Pinczower EF, Gates GA (1998) Current therapy in otolaryngology-head and neck surgery. Mosby Year Book, Washington, pp 485–490
27. Arnrup K, Crossner CG (1990) Caries prevalence after submandibular duct repositioning in drooling children with neurological disorders. Pediatr Dent 12:94–97