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Long-term impact of saliva control surgery in children with disability☆

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Abstract Sialorrhea is a common problem in children with disability, often negatively affecting socialization, self-esteem, and burden of care. Saliva control surgery is an available option to manage this problem, particularly when other conservative methods have failed. As little is known about the long-term impact of surgery, we followed up 62 patients who had combined bilateral submandibular duct translocation and bilateral sublingual gland excision at our pediatric hospital between 1994 and 2014. Eligible individuals were identified through a search of ICD procedure codes. When families of patients were contacted successfully, they were invited to complete a 14-item questionnaire designed specifically for this study. The results indicated that long-term outcomes of surgery were very good; 13/62 (21%) individuals no longer had a drooling problem and another 30 (48%) experienced only mild to moderate drooling. Although 84% families reported some or major improvement in drooling, 9 families reported that they would not go through the experience again because of a difficult recovery period, lack of effectiveness of the intervention, changes in saliva consistency that caused coughing and gagging, and dental decay. None of the collected variables were predictive of good or poor outcome.

☆ This work has not been presented, wholly or in part, at any meeting.

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The study indicated that surgical intervention is effective in the long term in the majority of cases and can be recommended to other families who attend our saliva control clinic.

Introduction

Drooling ("dribbling" or "poor saliva control" or "sialorrhea") impedes socialization, interpersonal relationships, and integration into school and community life. Secretions damage books, clothing, and computer equipment. Loss of self-esteem and confidence is a major issue. In addition to the social implications for both the child and their parents, excessive drooling can cause significant skin maceration and requirement for frequent changes of clothes and bibs, thereby necessitating an increased amount of personal care for both individuals and their carers.[1] Drooling is a common problem. It occurs in 40% of children with cerebral palsy[2] and is frequent in both children and adults with other neurological conditions.

Treatment options for excessive drooling include behavioral and oral motor programs, intraglandular botulinum toxin injections, oral medication, and surgical procedures. However, there is a lack of evidence as to which approaches are most effective, hence prompting the call for further research.[3] Surgery is usually undertaken when more conservative approaches have failed, when drooling is so severe that conservative measures are unlikely to achieve a satisfactory outcome, or when compliance with conservative measures is challenging because of severe intellectual and/or physical disability.

Extensive literature suggests that surgical management using a variety of surgical procedures provides significant subjective relief in approximately 80% of children with drooling.[4–6] However, in a meta-analysis of 42 studies with a median sample size of 18 and median follow-up duration of 8.1 months (range 0.1–50 months), it was concluded that evidence regarding surgical outcomes for drooling was of low quality and the results heterogeneous.[7]

Surgical procedures include duct ligation or rerouting, sublingual or submandibular gland excision, and varying combinations of these procedures. The preferred procedure at our institution is bilateral submandibular duct transposition (BSMDT) and bilateral sublingual gland excision (BSLGE) with occasional parotid duct ligation following failure of the other procedures. Sublingual gland excision was added because, in an early report of BSMDT surgery, 8% of children developed a ranula, a salivary cyst in the floor of the mouth due to damage to the sublingual ducts. To avoid this complication, we followed the authors' recommendation of simultaneous excision of the sublingual glands.[8]

A previous study undertaken by our group reported on 72 children who underwent BSMDT + BSLGE surgery between 1993 and 2003. Drooling was assessed using the frequency and severity scale[9] and the number of daily bib and clothing changes preoperatively and at 1 month, 6 months, 1 year, 2 years, and 5 years postoperatively.[10] At 2 years, the median score for frequency of drooling had decreased significantly, but most children remained drooling occasionally or frequently. The median score for severity of drooling had decreased from profuse to moderate at 2 years. The median number of daily bib and/or clothing changes decreased from 4 to 0. Seven children had major postoperative problems with hemorrhage or swelling, and five children who had a relatively poor response proceeded to have additional parotid duct ligation.[10]

The aim of our study was to determine the long-term impact of BSMDT and BSLGE surgery performed at Royal Children's Hospital, Melbourne, between 1993 and 2014 to determine whether good surgical outcomes, as reported previously, were maintained for a longer period.

Methods

The long-term follow-up study was conducted at the Murdoch Children's Research Institute in Melbourne, Australia, in collaboration with Saliva Control Clinic at The Royal Children's Hospital, Melbourne. This multidisciplinary monthly clinic involves a speech pathologist, dentist, pediatrician, plastic surgeon, and clinical nurse coordinator. Families are advised about treatments to improve their child's drooling, and surgery is offered when other more conservative management has not been successful.[11] The project was approved by Human Research Ethics Committee at The Royal Children's Hospital. The authors adhered to the STROBE guidelines (http://www.strobe-statement.org/) while undertaking the study.

Participants

We planned to recruit families of patients who underwent BSMDT and BSLGE surgery between the years 1993 and 2014. Families were not contacted if the person who underwent the surgery had died or if the family did not speak English. Approximately 8–10 children per year undergo BSMDT and BSLGE surgery; hence, we anticipated that there would be approximately 170 eligible young people. Given substantial time had lapsed, and to contact all of them would be challenging; therefore, we aimed for a sample of size between 60 and 80 participants for this study.

Recruitment

Potential participants were identified using ICD codes, and eligibility was confirmed from the medical records and from our saliva control clinic database. A participant information letter was sent to the latest known address, if available, and included a questionnaire. The letter explained that the questionnaire could be completed electronically, on paper, or by a telephone interview. Families were contacted by telephone two weeks after the mail was sent to check whether they had received the information and whether they were willing to participate to determine how

they would complete the questionnaire. If families agreed to complete the questionnaire electronically or on paper and then failed to return it, they were contacted two more times at two-weekly intervals. If they agreed to complete the questionnaire by telephone, they were asked to specify a convenient time. Completion of the questionnaire was deemed evidence of consent to participate.

Questionnaire

A 14-item questionnaire was designed to determine the frequency and severity of drooling at an extended period post-surgery (in some cases up to 24 years) and the presence or absence of any other significant negative or positive outcomes postsurgery. To assess drooling, the Thomas-Stonell and Greenberg scale provided a 5-point scale for severity and a 4-point scale for frequency of drooling.[9] The questionnaire also included the frequency of bib and clothing changes per day and overall rating of change in drooling since the surgery. Other items related to problems with dry mouth; changes in saliva consistency; changes in eating, drinking, and swallowing; occurrence of dental disease; and overall satisfaction with the surgical procedure. Demographic data collected included age at surgery, diagnosis, mobility (independently mobile, walks with aids, or wheelchair dependent), speech (normal speech, articulation problems, limited speech, or no speech), eating/drinking abilities (normal, some difficulties, or nonoral feeding), and the presence or absence of intellectual impairment. Respondents were also able to provide free text comments. The questionnaire was designed in REDCap[12] so that families could complete the questionnaire online if they wished, or if completed on paper or by interview, the data could be entered later.

Statistical analysis

Data of characteristics and long-term outcomes for the study group were presented as proportions for categorical data and as mean values, standard deviations, and ranges for continuous numerical data. Chi-square analyses were performed to ascertain whether the long-term outcome differed according to sex, main diagnosis, intellectual disability, open mouth, mobility, head control, speech, eating and drinking abilities, and oral habits. A good long-term outcome was defined as a subjective report of some or major improvement in drooling since the saliva control surgery.

Results

Of 158 operations performed between January 1994 and July 2014, 95 families were lost to follow-up. Excluding 1 incomplete response, surveys were completed for the remaining 62 individuals (35 males, 27 females) who had surgery at a mean age of 11.5 years (range 4.3–19.9 years). In all cases, the surgery involved bilateral translocation of the submandibular ducts (BSMDT) and bilateral excision of the sublingual glands (BSLGE). Surveys to assess the long-term outcome of surgery were completed between January and August 2018, with two being self-reported. The mean length of follow-up was 12.5 years (range 3.6–24.3 years). Additional characteristics of the study group are shown in Table 1.

Table 1 Demographic and clinical characteristics of the study group.

Characteristic	$n = 62$
Age (years) at follow-up, mean [SD]	23.8 [6.0]
Sex, n (%)	
Male	35 (56.4)
Female	27 (43.6)
Main diagnosis, n (%)	
Cerebral palsy	46 (74.2)
Other	16 (25.8)
Age (years) at surgery, mean [SD]	11.4 [4.4]
Duration of follow-up (years), mean [SD]	12.5 [6.0]
Mobility, n (%)	
Walks independently	23 (37.1)
Uses walking frame/sticks	5 (8.1)
Walks at home, uses wheelchair outside of home	7 (11.3)
Wheelchair dependent	27 (43.5)
Speech, n (%)	
Normal	2 (3.2)
Some articulation problems	12 (19.4)
Limited speech	14 (22.6)
No speech	34 (54.8)
Intellectual disability, n (%)	
Yes	49 (79.0)
No	13 (21.0)
Holds head up, n (%)	
Not at all, or little of the time	4 (6.5)
Some of the time	23 (37.1)
All of the time	35 (56.5)
Eating and drinking, n (%)	
Eats normally	11 (17.7)
Some eating and drinking difficulties	39 (62.9)
Partially tube fed	5 (8.1)
Totally tube fed	7 (11.3)
Projecting teeth, n (%)	
No	48 (77.4)
Yes	14 (22.6)
Teeth grinding, n (%)	
No	43 (69.4)
Yes	19 (30.6)
Tongue thrust, n (%)	
No	44 (71.0)
Yes	18 (29.0)
Mouth open, n (%)	
All the time	17 (27.4)
Most of the time	25 (40.3)
Seldom	13 (21.0)
Never	7 (11.3)

Long-term outcomes of surgery were very good (Table 2); 13/62 (21%) individuals no longer had a drooling problem, and another 30 (48%) were reported to experience only mild to moderate drooling. Drooling was never profuse. In terms of frequency, drooling was constant in 4, frequent in 18, and occasional in 27 individuals and, for the majority (38/62),

Table 2 Long-term outcomes of saliva control surgery.

Outcome	$n = 62$
Drooling severity, n (%)	
Dry, never drools	13 (21.0)
Mild: only lips wet	16 (25.8)
Moderate: wet on lips on chin	14 (22.6)
Severe: drools to extent that clothing becomes damp	19 (30.6)
Profuse: clothing, hands, objects become wet	0 (0.0)
Drooling frequency, n (%)	
Never drools	13 (21.0)
Occasionally	27 (43.6)
Frequently	18 (29.0)
Constantly	4 (6.4)
Number of bibs/clothing changes per day, n (%)	
Not at all	38 (61.3)
One	8 (12.9)
Two	5 (8.1)
Three	7 (11.3)
Four	2 (3.2)
Five or more	2 (3.2)
Perceived change in drooling since surgery, n (%)	
Major improvement	36 (58.1)
Some improvement	16 (25.8)
Little improvement	7 (11.3)
No improvement	3 (4.8)
Experience of recovery period, n (%)	
Very traumatic experience	11 (17.7)
Difficult experience	14 (22.6)
Mildly uncomfortable	20 (32.3)
Comfortable	17 (27.4)
Child's current concern about drooling, n (%)	
Not at all	52 (83.9)
Somewhat concerned	7 (11.3)
Concerned	3 (4.8)
Dry mouth, n (%)	
Yes	5 (8.1)
No	54 (87.1)
Sometimes	3 (4.8)
Thick or frothy saliva, n (%)	
Yes	9 (14.5)
No	53 (85.5)
Dental decay, n (%)	
Yes	8 (12.9)
No	52 (83.9)
Uncertain	2 (3.2)
Eating, n (%)	
Improved	7 (11.5)
Worsened	4 (6.6)
No change/Uncertain	50 (81.9)
Missing	*1*
Drinking, n (%)	
Improved	5 (8.2)
Worsened	1 (1.6)
No change/Uncertain	55 (90.2)
Missing	*1*
Swallowing, n (%)	
Improved	9 (14.8)
Worsened	1 (1.6)
No change/Uncertain	51 (83.6)
Missing	*1*
Do it again?, n (%)	
Yes	52 (83.9)
No	9 (14.5)
Uncertain	1 (1.6)

did not necessitate any bib or clothing changes. Only 10/62 (16%) families reported little or no improvement in drooling. In contrast, 36 (58%) families reported major improvement in drooling. Although the recovery period experience was recalled as difficult or traumatic for 25/62 individuals, 52/62 (84%) families thought that, in the same circumstances, they would proceed with the saliva control surgery, but one family expressed uncertainty. The reasons provided by the nine families who reported that they would definitely not go through the experience again were the combination of the trauma and pain during the recovery period, lack of effectiveness of the intervention in reducing drooling over an extended period, changes in saliva consistency that caused coughing and gagging, and perceived negative effect on the teeth. A few carers commented that drooling re-emerged as a problem many years after the surgery.

In terms of secondary outcomes and side effects, saliva control surgery did not appear to greatly change eating, drinking, and swallowing abilities. Known side effects of the surgery were only experienced by a small minority; a dry mouth was reported for 5 individuals, thick or frothy saliva for 9, and dental decay greater than what was perceived as expected for 8.

On univariable analysis, none of the collected variables including sex, main diagnosis, intellectual disability, open mouth, mobility, head control, speech, eating and drinking abilities, and oral habits were predictive of good outcome based on reported long-term major or some improvement in drooling.

Discussion

This study reports long-term outcomes of the surgical procedure for drooling, BSMDT + BSLGE. The results were very good for most of the participants, and 52/62 (84%) participants stated that they would undergo the surgery again under the same circumstances. Our results were compared favorably with those of other studies that have shown good outcomes with shorter follow-up periods, most of which have followed up patients for periods of months rather than years. In one study, caregivers of 16 children judged the efficacy (percent improvement) of the procedure to be 75% at six weeks postsurgery.[13] In a study of 28 children followed up for at least 3 months, a statistically significant difference ($p < 0.001$) was noted between preoperative and postoperative mean values for severity and frequency of drooling and also bib changes per day.[14] Comparing BSMDT + BSLGE surgery with BSMDT surgery alone with a

follow-up period of at least one year, the parents of 89% of children in the combined group were satisfied with the procedure and would recommend it to others.[15] In a larger study involving 72 children and adolescents with a mean age at the time of surgery of 15 years, 2 months, mean visual analog scale scores demonstrated a significant reduction in drooling when measured at baseline, 8, and 32 weeks after surgery. This was accompanied by a decrease in the amount of daily care required and improvement in social contact with other children.[16] Persons with adequate head stability and posture benefited most from the treatment.[17]

Participants in our study made positive comments about their satisfaction with the procedure, such as "When I look at any pre-operation photos I remember how wet she used to get – it was socially unacceptable. It was great to have something to cure one of her problems as many of them can't be cured." For 10 participants, however, there was little or no improvement, and unfortunately, none of the collected variables enabled us to predict which of those participants were likely to have little or no response to the surgery. As part of our group's research on the effectiveness of botulinum toxin injections into the salivary glands, we found no relationship between response to botulinum toxin injections and response to surgery.[18] Further research is required in this area.

The major limitations of this study were the lack of a face-to-face assessment, and the reliance on parental memory of the situation before surgery, which, in some cases, were many years. Yet families were interested to participate and provide comments, particularly, if they felt that this would assist both parents and clinicians in the future.

Many treatments are instituted in childhood and early adolescence, yet there is lack of knowledge as to whether these treatments are beneficial in the long-term. Long-term follow-up studies are essential to determine whether interventions in childhood have been effective so that scarce resources are not wasted on futile and cost-ineffective treatments. Our study has shown that this procedure is effective in the long term in the majority of cases and hence can be recommended to other families but with the understanding that approximately 1 in 5 children will not benefit.

Conflict of interest

None.

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