Bilateral submandibular gland excision for drooling: Our experience in twenty-six children and adolescents

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Accepted for publication 6 January 2015

Dear Editor,

Approximately 40% of children with cerebral palsy (CP) suffer from drooling, and it is considered severe in 15%.[1] Drooling is caused by a combination of several factors such as diminished awareness to swallow, poor posture and dysfunctional oral motor functions.[2]

We distinguish between anterior and posterior drooling. Anterior drooling is characterised by saliva spilled from the mouth that is clearly visible. Posterior drooling is defined as the spill of saliva over the tongue through the oropharyngeal isthmus, causing aspiration and associated pneumonias.[3]

Morbidity due to drooling has been widely described. Different therapies have been reviewed,[4,5] but there is no consensus regarding the optimal treatment strategy.

Surgical interventions are indicated when conservative measurements have failed, when a more long-term solution is desirable or when conservative measurements are not expected to improve drooling, for example in older patients or patients suffering from a progressive disease.

In individuals with combined anterior and posterior drooling, submandibular duct relocation is contraindicated. Bilateral submandibular gland excision may be an effective procedure instead. Previous studies regarding the efficacy of this procedure were based on small and heterogeneous populations.[4–6] In particular, no validated objective measurements were used. A previous meta-analysis compared different surgical intervention methods, but did not include studies on submandibular gland excision without parotid duct rerouting or ligation.[5]

We aim to be the first to provide both objective and subjective results of bilateral submandibular gland excision in young people with neurological disabilities who drool due to severe dysphagia.

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Materials and methods

Ethical considerations

The research was conducted in accordance with national and international ethical standards. Informed consent was provided before each intervention.

Study design

We analysed a historic cohort of children and adolescents who were examined at the Multidisciplinary Saliva Control Centre of the Radboud University Medical Centre Nijmegen, the Netherlands, between January 2001 and January 2014. Demographic data were collected preoperatively.

Surgical procedure

For submandibular gland excision, a skin incision of ≈5 cm in length was made under general anaesthesia, 4 cm below the border of the mandible. The platysma muscle was separated and the lower border of the gland exposed. If necessary, the facial artery was identified and spared if possible. The lingual nerve and hypoglossal nerve were identified and spared. After gland excision, a suction drainage was routinely placed for 1 day. Intracutaneous resorbable sutures were used.

Participants

Forty-five children and adolescents have undergone bilateral submandibular gland excision. This decision was made on expert opinion by our multidisciplinary team. Subjects were categorised by CP type, having epilepsy, severity of motor disturbance assessed by the Gross Motor Function Classification System (GMFCS), posture, developmental age, ability to eat and type of drooling (Table 1).

We excluded three cases with posterior drooling only, as drooling intensity was assessed by measuring visual saliva loss. In addition, we excluded twelve cases who had

Table 1. Patient characteristics

	Included for analyses $n = 26$
Sex, *n* (%)	
Male	13 (50)
Female	13 (50)
Mean age at intervention, year : month (SD)	
Submandibular glands excision	16 : 5 (6.69)
Botulinum toxin-A injections	12 : 0 (6.06)
Main diagnosis, *n* (%)	
Spastic CP	11 (45.8)
Spastic/dyskinetic CP	2 (8.3)
Ataxic CP	2 (8.3)
Dyskinetic CP	2 (8.3)
Other developmental disability	7 (29.2)
Missing	2
Developmental age, *n* (%)	
<4 years	20 (87)
4–6, IQ <70	1 (4.3)
>6 year	2 (8.7)
Missing	3
GMFCS level, *n* (%)	
I	1 (4)
II	3 (12)
III	2 (8)
IV	2 (8)
V	17 (68)
Drooling kind, *n* (%)	
Anterior	10 (38.5)
Antero-posterior	16 (61.5)
Epilepsy, *n* (%)	
Controlled	18 (72)
Intractable	2 (8)
No	5 (20)
Missing	1
Head position, *n* (%)	
Anteflexion	6 (24)
Retroflexion	1 (4)
Asymmetrical	7 (28)
Normal	9 (36)
Not registered	3 (8)
History of pneumonia, *n* (%)	
Yes	12 (46.2)
No	14 (53.8)
Use of benzodiazepine, *n* (%)	
No/unknown	24 (92.3)
Yes	2 (7.7)
Gastrostomy feeding required, *n* (%)	
Oral + feeding tube	5 (20.8)
Oral	12 (50)
Feeding tube	7 (29.2)
Missing	2

GMFCS: Gross Motor Function Classification System level descriptions; I: reduced speed, balance and coordination; II: limitations walking on uneven surfaces and inclines, and in crowds or confined spaced; III: walking indoors or outdoors on a level surface with assistance, wheelchair as needed; IV: reliance on wheelchair; V: no means of independent mobility; CP, cerebral palsy.

undergone surgery for drooling prior to our surgery. Three children were excluded because of incomplete or missing medical records. In one case, the period between botulinum toxin-A injection and surgery was <24 weeks. Ultimately, 26 cases were included for analysis.

Variables

Drooling was assessed at baseline and prospectively during follow-up visits in the outpatient clinic (8 and 32 weeks after treatment). Drooling intensity was evaluated using the drooling quotient (DQ), which is a validated, semi-quantitative direct observational method. The DQ is expressed as a percentage estimated from the ratio of observed drooling episodes and the total number of observations (DQ [%] = 100 × number of drooling episodes/20).[7,8] Successful therapy effect was defined as a higher than 50% reduction compared to baseline.

Severity of drooling during the prior 2-week period was scored by a visual analogue scale (VAS) score. Caretakers assign a drooling score by marking on a line from 0 (= no drooling) to 100 (= excessive drooling). A reduction of VAS score of >2 SD from baseline is considered clinically significant. The Thomas–Stonell and Greenberg classification, which consists of a 5-point scale for severity and a 4-point scale for frequency, was used as a second subjective score.

Statistical analysis

Descriptive statistics were employed to summarise patient characteristics. For DQ and VAS, univariate ANOVA with repeated measures analysis was used. The patient was set as random factor and time as fixed to evaluate whether treatment responses differed significantly over time. When significant, a *post hoc* pairwise comparison (Fisher's LSD) was performed to evaluate differences between means at different time points. Chi-squared (χ^2) test was used to confirm the association between antero-posterior drooling and recurrent pneumonias based on history. Data were

analysed using SPSS version 20.0 for Windows (SPSS Inc., Chicago, IL, USA).

Results

Characteristics of the 26 patients included for analyses are shown in Table 1. Diagnoses comprised of 17 patients with CP, two patients with a yet unknown disease and seven patients with a non-progressive developmental disability.

Observation and scoring by speech therapists revealed that a minority of 39% had anterior drooling only, compared to 61% with antero-posterior drooling. Correlation analyses revealed that 19 of 25 subjects with a combination of anterior and posterior drooling had suffered from recurrent pneumonias in the past, while none of the subjects with anterior drooling alone had suffered from pneumonia (Chi-squared-test; d.f. 1: value 17.917, $P < 0.001$). This result was in accordance with the observations by the speech therapists.

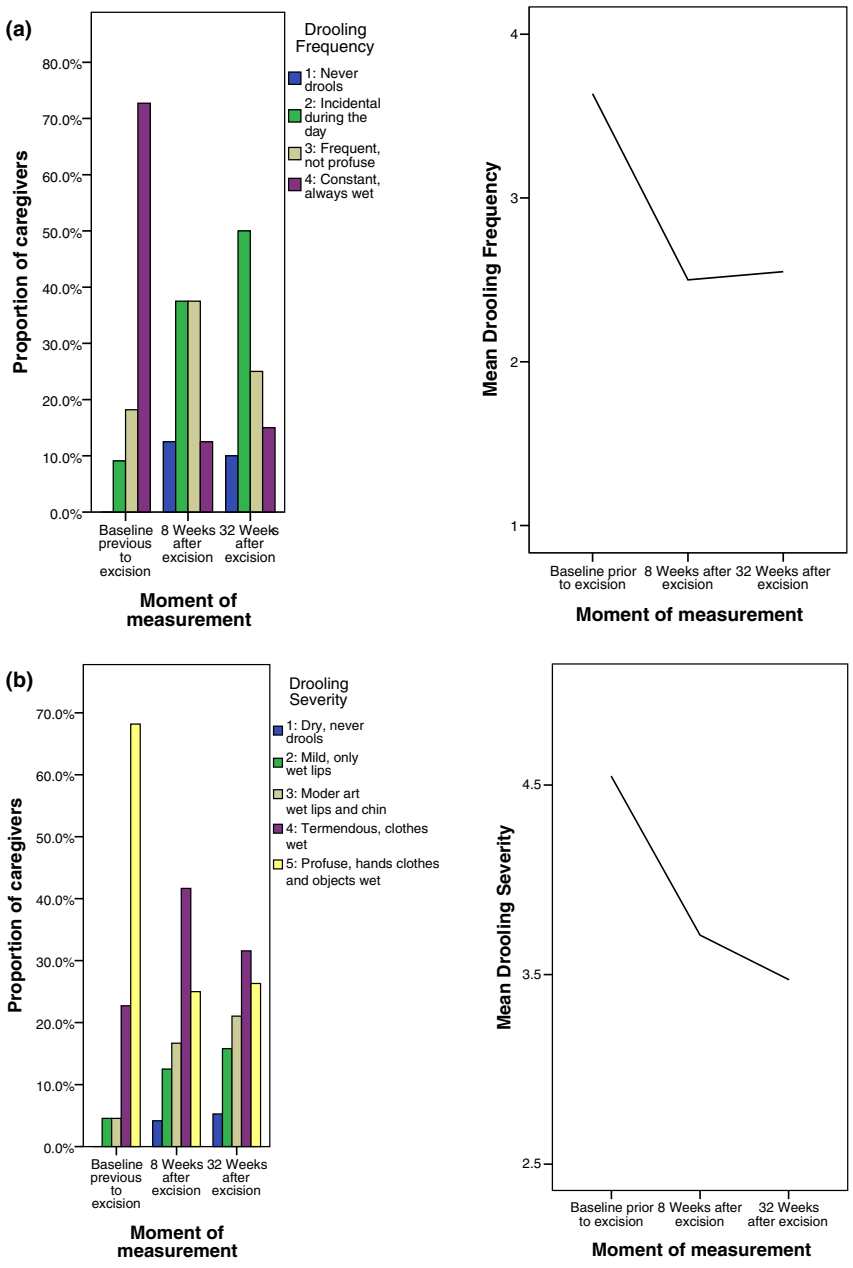

Fig. 1. (a) Drooling frequency score, marked by caregivers before and after surgery. (b) Drooling severity score, marked by caregivers before and after surgery

The average age at the time of surgery was 15.6 years (SD 6.72, range 2–38 years). Two subjects were transferred to the intensive care unit, due to bleeding requiring reoperation. One case of xerostomia was reported. No procedures resulted in damage of the marginal branch of the facial nerve, lingual nerve or hypoglossal nerve.

Subjective outcomes based on the Thomas–Stonell and Greenberg classification are shown in Fig. 1. At baseline, caregivers had assigned the highest score of 4 for drooling frequency (defined as 'constant, always wet') in 72.7% of cases. In contrast, at 8 and 32 weeks follow-up, scores lower than 4 were assigned in 87.5% and 85% of cases, respectively. Drooling severity was assigned the highest score of 5 (defined as 'profuse, hands, clothes and objects wet') by 68.2% of caregivers at baseline. At 8 and 32 weeks, this was reduced to 25% and 26.3%, respectively.

Univariate ANOVA with repeated measures analysis revealed a significant effect of time for both subjective VAS score ($P \leq 0.001$, d.f. 2, F 16,589) and objective DQ ($P = 0.002$, d.f. 2, F 7, 498) (Fig. 2). The marginal mean DQ was reduced from 33.5 at baseline to 17.1 at 8 weeks ($P = 0.008$) and to 9.9 at 32 weeks ($P = 0.001$) following surgical intervention. The estimated marginal mean VAS score improved from 75 at baseline to 34.7 after 8 weeks. Although the mean score was slightly higher (40.5) after 32 weeks, this was still significantly lower than at baseline ($P \leq 0.001$) (Table 2). Based on the treatment success criteria, 64.7% and 61.5% of subjects had at least a 50% reduction in DQ at 8 weeks and 32 weeks, respectively. Success rates based on a VAS score reduction by at least 2 SD were 55% at 8 weeks and 44% at 32 weeks.

Sample sizes for the analysis of treatment responses varied from 18 to 23, as we did not impute for missing data.

Discussion

Synopsis of key findings

We demonstrate that bilateral submandibular gland excision is an effective treatment for drooling, with an overall response rate of 63%. Subjective outcome measurements also showed significant improvements following surgery. We noticed the subjective improvement was slightly less after 32 weeks, in contrast to the objective results, where the effect progressed. These differences underline the importance of the use of standardised objective outcome measurements.

Comparison with other studies

Prior to surgery, patients have frequently undergone conservative treatments, with or without success.[3–5]

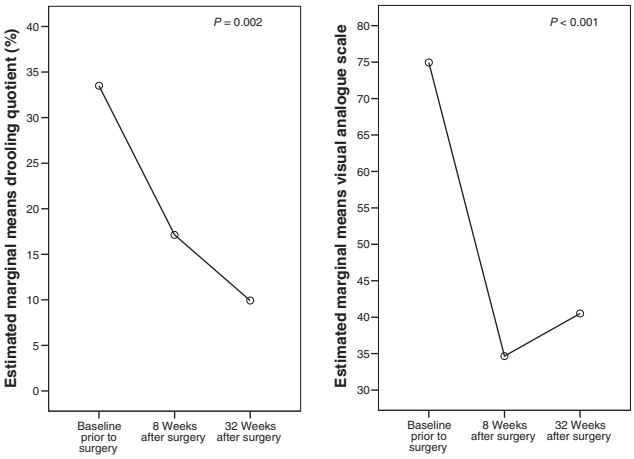

Fig. 2. Estimated marginal mean drooling quotient and visual analogue scale (VAS) scores during follow-up moments.

For the last decade, botulinum toxin-A injections in one or more major glands have offered a promising and well-tolerated treatment option.[3,6,9] A recent study by Scheffer *et al.*[9] using the same objective measurements methods reported a response rate of ≈50% after 8 weeks when injecting the submandibular glands. This is only slightly lower than our response rate of 64.7% after 8 weeks. However, the effect of botulinum toxin-A injections fades after 32 weeks; it remains effective in only 11.3% of cases. In contrast, bilateral submandibular gland excision resulted in a response rate of 61.5% at 32 weeks after surgery. Nevertheless, botulinum toxin-A injections come with the benefit of limited procedure-based morbidity and the fact that the temporary nature is expected. This makes it an attractive procedure in children between 4 and 8–10 years of age, when ongoing development still might solve the problem.

Bilateral submandibular gland excision is slightly less effective compared to submandibular duct relocation (response rate 81% in our clinic).[5,9] Differences in success rates can be explained by the multivariate causes of drooling and heterogeneity of the population. Submandibular duct relocation is only performed in those patients with only anterior drooling and a safe pharyngeal phase of swallowing. Excision of the submandibular glands is also performed in those with combined or posterior drooling, and thus patients with a more pronounced severe dysphagia. In addition, submandibular duct relocation may trigger a more frequent swallowing reflex due to pharyngeal saliva release after surgery. This could explain differences in success.

Ligation of the salivary ducts has recently gained popularity.[10] As for submandibular glands excision, ligation of the salivary ducts aims to reduce the amount of

Table 2. Mean differences between baseline and follow-up of drooling quotient and visual analogue scale (VAS)

	No. of observations	Missing observations	Absolute mean difference (95% CI)	Relative mean difference (%)	Significance (*P*)
Drooling quotient					
Baseline	21	5			
8 weeks	21	5	16.37 (4.5–28.2)	49	0.008
32 weeks	18	8	23.58 (10.6–36.5)	70	0.001
Visual analogue scale					
Baseline	23	3			
8 weeks	21	5	40.23 (25–55.6)	53.70	≤0.001
32 weeks	19	7	34.45 (18.5–50.4)	46	≤0.001

The mean difference is significant at the 0.05 level.
CI, confidence interval.

saliva produced. This procedure is attractive because of its surgical simplicity and because it carries a much lower risk for unsightly scars or nerve damage. Varying results have been reported, with response rates up to 73%. Unfortunately, further procedures are frequently required due to recurrence of drooling.[10]

Strengths and weaknesses of the study

We are aware of the fact that our group is very heterogeneous, especially in term of age, nevertheless we think that this does not influence our results. In addition, the mental age showed less heterogeneity. Although we used a small number of subjects and a short period of follow-up, we did use objective measurements during follow-up instead of only subjective measurements. Also long-lasting results are to be expected due to the nature of the intervention.

In our study, we used stringent exclusion criteria. Although we collected a historic cohort, all data were collected prospectively. Specifically, we created a cohort with an indication for treatment of either anterior or mixed antero-posterior drooling. This allows our findings to be more easily compared with future studies and to be clinically correlated to the follow-up measurements (DQ and VAS scores).

Conclusion

Our study shows that bilateral submandibular gland excision significantly reduces drooling in more than half of the children with a neurological disease. This procedure is especially attractive for those where submandibular duct relocation is contraindicated. In our opinion, subsequent studies should focus on larger sample sizes and posterior drooling.

Keypoints

- Drooling is a major problem in children and adolescents with neurological disorders. It has been suggested that excision of the submandibular glands may be an effective method for reduction of saliva.
- We analysed a historic cohort of 45 patients who have undergone submandibular gland excision for moderate-to-severe drooling in our clinic between January 2001 and January 2014. Twenty-six children were eligible for analysis. They were evaluated preoperatively (baseline) and at 8 and 32 weeks following surgery.
- Drooling intensity was significantly reduced following surgery compared to baseline. Drooling quotient was reduced from a baseline score of 33.5 to 17.1 after 8 weeks and 9.9 after 32 weeks ($P = 0.002$). On the basis of our success criterion, 63% of surgeries were successful. Similarly, subjective visual analogue scale score and drooling severity and frequency scores showed significant improvement following surgery.
- Bilateral submandibular gland excision is an effective therapy for drooling in young people with neurological disabilities, especially when submandibular duct relocation is contraindicated.

Acknowledgements

The authors are grateful to Mw. Dr. Petronella Peer for her statistical contribution. We would like to thank the participating patients for their cooperation.

Financial support

None.

Conflict of interest

None to declare.

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