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The impact of submandibular duct relocation on drooling and the well-being of children with neurodevelopmental disabilities



Saskia E. Kok ^{a,*}, Jan J.W. van der Burg ^{b,c}, Karen van Hulst ^d, Corrie E. Erasmus ^e, Frank J.A. van den Hoogen ^a

^a Department of Otorhinology and Head and Neck Surgery, Radboud University Nijmegen Medical Center, P.O. Box 9101, 6500HB, Nijmegen, The Netherlands

^b Department of Social Sciences/Pedagogical and Educational Sciences, Radboud University Nijmegen Medical Center, P.O. Box 9101, 6500HB, Nijmegen, The Netherlands

^c Department of Pediatric Rehabilitation, St. Maartenskliniek, Hengstdal 3, 6574 NA, Ubbergen, The Netherlands

^d Department of Rehabilitation, Radboud University Nijmegen Medical Center, P.O. Box 9101, 6500HB, Nijmegen, The Netherlands

^e Department of Pediatric Neurology, Radboud University Nijmegen Medical Center, P.O. Box 9101, 6500HB, Nijmegen, The Netherlands

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ABSTRACT

Objective: The aim of this study was to evaluate the impact of a reduction in drooling after bilateral submandibular duct relocation (SMDR) with sublingual gland excision on daily life and care, as well as social and emotional consequences in children and adolescents with neurodevelopmental disabilities.

Methods: This prospective cohort study included 72 children and adolescents (46 males, 26 females) with moderate to severe drooling, and their caregivers. Mean age at the time of surgery was 15 years 2 months (SD 4y 3mo). Fifty-two children were diagnosed with cerebral palsy and 20 had other non-progressive developmental disabilities. A caregiver questionnaire to document the impact of drooling on daily care and economic consequences, social interaction and emotional development and self-esteem was administered before, and 8 and 32 weeks after surgery.

Results: Following bilateral SMDR the mean Visual Analogue Scale (VAS, 0–100) scores demonstrated a significant ($p < 0.001$) reduction in the severity of drooling from 81 at baseline to 28 and 36 after 8 and 32 weeks, respectively. This was accompanied by a decrease in the amount of daily care required and reduced economic consequences. In addition, an increase in social contact with other children and adults was reported by caregivers after surgery.

Conclusion: Bilateral SMDR with sublingual gland excision provides a significant positive reduction in daily care of children suffering from drooling. Caregivers also report positive changes in their child's social interaction and sense of self-esteem.

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1. Introduction

Drooling is a chronic and serious problem in many children with cerebral palsy (CP) and other neurodevelopmental disabilities.

Abbreviations: CP, cerebral palsy; ENT, Ear, Nose, and Throat; GMFCS, Gross Motor Functional Classification System; QoL, quality of life; SMDR, submandibular duct relocation; VAS, visual analogue scale.

* Corresponding author. Radboudumc, Department of Otorhinology, 377, P.O. Box 9101, 6500HB, Nijmegen, The Netherlands.

E-mail addresses: saskia.kok@radboudumc.nl (S.E. Kok), j.vandenburg@maartenskliniek.nl (J.J.W. van der Burg), karen.vanhulst@radboudumc.nl (K. van Hulst), corrie.erasmus@radboudumc.nl (C.E. Erasmus), frank-kno.vandendoogen@radboudumc.nl (F.J.A. van den Hoogen).

About 10–37.5% of children with CP suffer from drooling due to dysfunctional oral motor control [1,2]. More recently, Reid et al. concluded from their population based study a 40% prevalence of drooling in children with CP, with 15% marked as severe drooling. In a subgroup of children with a mobility level on the Gross Motor Functional Classification System (GMFCS) of III or higher, they even found a prevalence of drooling up to 80% [3]. Drooling does not only affect the child, but has also a high impact on the well-being of their caregivers and the family. Besides practical implications, it has social and emotional consequences as well. Daily care requires frequently changing of clothes and bibs and the spilled saliva may also cause damage to toys, computers and communication devices [4]. In addition, drooling negatively influences the child's physical appearance, which may affect social interaction with peers, adults

or other children, elicit overt negative comments and result in lowered self-esteem. Drooling can negatively affect the estimate of the mental capacities of these children by others, even though cognitive functions are sometimes spared in this population [4]. Underestimation of the child's mental abilities may influence their opportunities for social interaction with peers and others.

Various therapeutic options have been described to reduce drooling in children. These options include oral motor therapies [5] and behavioural treatment [6], which may benefit children with sufficient cognitive and oral motor capabilities. Systemic anticholinergic drugs can reduce drooling, however they cause unwanted side effects in approximately 20% of the children [7]. Although Botulinum toxin injections have shown to be a safe, effective and only moderately invasive treatment, their effect is temporary and thus requires reintervention [8]. Nowadays, different surgical procedures are performed for a more definitive effect [9]. Submandibular duct relocation (SMDR), one of the proved surgical procedures to diminish drooling, was first described by Laage-Hellman in 1969 and since then it has been used extensively by other surgeons including Ekedahl [1] and Crysedale [10]. SMDR has formerly shown to be effective in about 80% of the children with the ability to swallow safely [11–14]. Although a reasonable number of mostly small retrospective studies on the effectiveness of SMDR in reducing drooling have been performed, evaluation of its effect on the impact on life quality is scarce. Most researchers only briefly report parent-, and patient satisfaction after surgery without comparing pre- and postoperative measurements [11,12,14,15]. Currently, prospectively collected data on changes in the impact of drooling on life before and after SMDR is lacking.

To evaluate change after intervention for drooling, objective as well as subjective measurements have been used. Some authors prefer objective measures such as the Drooling Quotient [16,17] or measuring salivary flow [18,19], whereas others find the counting of bibs more useful [15,20]. Blasco rightly pointed out that these objective measures should be enhanced with subjective measures on parent and patient satisfaction [20]. From this it can be stated that there is a need to evaluate the effect of treatment on the consequences of drooling. Despite the fact that there are several Quality of Life (QoL) questionnaires available, these are often generic or condition specific. Most do not address the consequences of drooling [21]. That is why our saliva control team developed a questionnaire in 2000 especially designed to evaluate changes in the impact of drooling on daily care and economic consequences, as well as the social and emotional consequences [4]. Previous research using this questionnaire, showed that reduced drooling after Botulinum toxin injections could ease daily care and have a positive influence on self-esteem and acceptance by peers and adults [22,23]. With this prospective cohort study we aim to measure changes in the impact of drooling on daily care and economic consequences, social interaction and emotional development and self-esteem after bilateral SMDR with sublingual gland excision in children with CP and other non-progressive neurodevelopmental disabilities and moderate to severe drooling.

2. Patients and methods

2.1. Study participants

Between March 2005 and December 2011, 72 children and adolescents with CP and non-progressive neurodevelopmental disabilities and moderate to severe drooling were treated with bilateral SMDR with sublingual gland excision, at the Radboud University Medical Center. Charts were reviewed for the present study and prospectively collected data of these patients were identified. After examination by the speech language therapist of

the team, all children appeared to have a safe pharyngeal swallow function. Before surgery, all included participants had at least moderate drooling, intermittent throughout the day (i.e. Teacher Drool Scale¹ score ≥ 3) [24]. Individuals older than 25 years were excluded from this study. None of the participants had undergone previous surgery to reduce drooling and no additional anticholinergic medication or Botulinum toxin injections were allowed during this study. Before this study, all children received oral motor therapy. Forty-four children (61.1%) were previously treated with Botulinum toxin injections and three children had extensive behavioural treatment prior to surgery, all without sufficient long lasting effect. This study was conducted in accordance with national and international ethical standards. Caregivers provided informed consent before surgery.

2.2. Procedure

Surgery was performed under general anaesthesia. The submandibular ducts were bilaterally relocated towards the base of the tongue and the sublingual glands were removed to prevent formation of a ranula. More details on the surgical procedure have been described previously [1,10,25].

2.3. Data collection

Participants and their caregivers visited our outpatient saliva control clinic for assessment and evaluation at baseline, and 8 and 32 weeks following surgery. At each visit, the children and adolescents were examined by one of two well-trained speech language therapists and one Ear, Nose, and Throat (ENT) surgeon, and at baseline also by a specialized child neurologist. During these visits, caregivers completed the written questionnaire on the impact of drooling.

2.4. The questionnaire on the impact of drooling

Our multidisciplinary team (speech language therapist, clinical paediatric psychologist, paediatric neurologist, rehabilitation physician and ENT surgeon) developed an extended questionnaire of over 60 items, which has been previously applied to evaluate both drooling severity in general and in daily conditions, and the impact of drooling on daily care and economic consequences, social interaction and emotional development and self esteem [4]. To enhance the applicability, we developed a short version by eliminating questions that were not distinctive in the evaluation of the effect of reduced drooling after Botulinum toxin injections in previously published research [22,23]. However, we kept questions related to emotional consequences of drooling, which only apply to a small subgroup of children who drool and are able to reflect on their social and emotional experiences and express them as well. Experts and four families gave their opinion about the short version of the questionnaire, which resulted in minor adjustments. Since 2005 this short version of the questionnaire has been applied at our center and it was used in the present study (see Appendix). The questionnaire consists of multiple-choice as well as open-ended questions and several Visual Analogue Scales (VAS).

2.5. Statistical analysis

To study the impact of bilateral SMDR, we compared the mean

¹ Teacher Drool Scale: 1. No drooling; 2. Infrequent drooling, small amount; 3. Occasional drooling, on and off all day; 4. Frequent drooling, but not profuse; 5. Constant drooling, always wet.

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