

TITLE: Drooling

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RESIDENT PHYSICIAN: Sarah Rodriguez, MD

FACULTY PHYSICIAN: Byron J. Bailey, MD

SERIES EDITORS: Francis B. Quinn, Jr., MD and Matthew W. Ryan, MD

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Drooling, also known as ptyalism, can be defined as salivary incontinence or the spillage of saliva over the lower lip. It reflects a disturbance of the oral phase of deglutition which is associated with inefficient, uncoordinated swallowing and poorly synchronized lip closure. It is frequently associated with an abnormal increase in tone of the muscles that open the mouth. Drooling patients have difficulty managing normal salivary flow; sialorrhea, which some use interchangeably with drooling, indicates an increase in salivary flow which can lead to drooling. The functions of saliva include lubrication and moistening of food in order to facilitate mastication, protection of the mucosa from desiccation, prevention of dental caries, and a medium by which lysozyme, secretory IgA and salivary peroxidase are delivered to the oral cavity. There are three pairs of major salivary glands: the parotid, the submandibular, and the sublingual glands. Seventy percent of the 1.5 liters of saliva produced daily is from the submandibular gland; twenty-five percent is from the parotid glands and five percent is from the sublingual glands. Basal salivary flow is produced primarily by the submandibular gland; stimulated salivary flow is produced primarily by the parotid gland. Secretory innervation is via the autonomic nervous system, and is primarily parasympathetic. Origination of the preganglionic parasympathetic fibers to the submandibular and sublingual glands is the superior salivary nucleus in the medulla; these fibers travel with the nervus intermedius in the facial canal. They continue with the facial nerve to exit the mastoid segment at the chorda tympani. These fibers are then conducted through the petrotympanic fissure to the submandibular ganglion where preganglionic parasympathetic fibers synapse with postganglionic fibers. These fibers are then distributed to the submandibular and sublingual glands. Parasympathetic postganglionic fibers to the parotid gland have their origination in the inferior salivary nucleus in the medulla. These leave the brainstem with cranial nerve nine and travel to the petrosal ganglion. The tympanic branch of the glossopharyngeal nerve (Jacobson's nerve) passes through the middle ear on the promontory, where it may be submucosal or pass through a bony canal. After supplying the middle ear mucosa, this nerve then continues as the lesser superficial petrosal nerve. The lesser superficial petrosal nerve synapses with postganglionic parasympathetic fibers in the otic ganglion in the pterygopalatine fossa. The post ganglionic fibers then travel with the auriculotemporal nerve to supply the parotid gland.

Drooling is rarely seen in the normal child after the age of 18 months. Drooling can be acute or chronic. Acute causes of drooling include infections such as epiglottitis, peritonsillar abscess, or neoplasm. Chronic causes of drooling can be categorized into neurologic, medication effects, and indirect causes. Neurologic causes are most frequent and include cerebral palsy, amyotrophic lateral sclerosis, cerebrovascular accidents, Parkinson's disease, and congenital suprabulbar palsy. Medications associated with drooling include tranquilizers, anticonvulsants and anticholinesterases. Indirect causes include nasal obstruction, malocclusion, tongue size, head posture, sitting position, and emotional state. Consequences of drooling include maceration of skin around the mouth, chin and neck which may lead to secondary bacterial infections. Patients and caretakers must deal with the constant soiling of clothes, toys, and furniture. Psychosocially, the stigma and rejection associated with drooling takes a heavy toll on the patient. Workup includes a complete history and physical exam. It is difficult to objectively measure drooling. Historical features that should be taken into account include estimated quantity of saliva, use of bibs, and clothing changes required throughout the day. Factors that exacerbate drooling such as mouth breathing should be elicited. Nasal obstruction secondary to rhinitis, sinusitis or adenoid hypertrophy should be diagnosed and treated initially. History should also include the extent of any underlying neurologic disease. A history of hearing loss should be elicited as a unilateral hearing deficit may preclude trans-tympanic neurectomy. Physical exam must evaluate head posture, sores on the lips or chin, dental problems, tongue control, swallowing ability, and possible nasal airway obstruction. An audiogram should be performed if a trans-tympanic neurectomy is being considered. Some authors recommend barium swallow to rule out chronic aspiration or esophageal stricture as these can be contraindications to surgery.

Noninvasive modalities of treatment include speech therapy and behavioral therapy. Although speech therapy alone has yielded disappointing results, most physicians refer initially to a speech therapist for assessment prior to instituting more invasive therapy. The goal of speech therapy is to improve jaw stability and closure, to increase tongue mobility, strength, and positioning, and to improve lip closure (especially during swallowing), and to decrease nasal regurgitation during swallowing. In order to obtain good results, therapy must be started early in infancy. Treatment will be of limited value in severely retarded children or those who drool profusely. Behavioral therapy can incorporate cuing, positive reinforcement, and overcorrection or punishment. Behavioral therapy has been reported to be fairly successful in those patients with an adequate level of intelligence, but is labor intensive and regression is common. Dental appliances to control drooling have been tried by a small number of therapists. The appliance is used as a training device in concert with oral-motor therapy. It promotes mandibular stability, lip closure, better tongue position, and swallowing. Few individuals are experienced in their application and exactly which populations would benefit from such devices has not been elucidated. But these devices should be investigated as an alternative noninvasive therapeutic modality for drooling. Drugs used to treat drooling include glycopyrrolate and trihexyphenidyl. Studies have shown 70-90% response rates with glycopyrrolate but with consistently high side effect rates. Approximately 30-35% of families will choose to discontinue glycopyrrolate therapy due to unacceptable side effects. Side effects are anticholinergic in nature and include excessive dry mouth, constipation, urinary retention, decreased sweating and skin flushing, irritability and behavior changes. Efficacy and side effects of trihexyphenidyl are similar to glycopyrrolate. However, use of trihexyphenidyl may be of value in children with rigid or

dystonic cerebral palsy because of the potential benefit of tone reduction. Botox® (botulinum toxin type A) has recently been proposed as a potential medical therapy for drooling. Limited trials have been performed with most studies using adult subjects. However Jongerius, et al. report the use of botox injection into bilateral submandibular glands with good results and no side effects. Studies generally report good results with one study reporting recurrent jaw dislocation as a possible side effect of therapy. Radiation therapy has also been used to control drooling in patients with amyotrophic lateral sclerosis. These patients were generally too ill to undergo surgical treatment and had a short remaining lifespan thus eliminating the risk of secondary malignancy from radiation therapy. Xerostomia is a known side effect of radiation. In a recent study by Harriman, et al. a small dose of radiation to the submandibular and sublingual glands was shown to decrease drooling with few side effects and, more importantly, to have a positive effect on the quality of life in these terminally ill patients.

A variety of surgical techniques have been proposed as treatments for drooling. These range from duct re-routing procedures, removal of salivary glands, ligation of salivary ducts and various combinations of these procedures. Wilke, a Canadian plastic surgeon, was the first to propose a surgical approach to drooling. He performed parotid duct relocation to the tonsillar fossae to manage drooling in two patients with cerebral palsy. The results of these surgeries were not totally satisfactory and Wilke and Brodie concluded that it was necessary to remove the submandibular glands as these glands are responsible for the bulk of daily salivary output. The success rate of the combined procedure was 85%; however, the procedure was criticized for technical difficulty, external scars, mandatory tonsillectomy, prolonged hospitalization, and significant operative morbidity with swelling and dysphagia. Complications occurred in 35% of patients and included post operative cysts, parotid duct stenosis or fistulas, wound dehiscence, parotid swelling and suppurative parotitis, xerostomia, and increased dental and gingival infections. Parotid duct ligations have been used to treat drooling in many patients. Submandibular duct ligations have been avoided as the submandibular gland saliva is more viscous, alkaline, and contains a higher proportion of calcium and phosphate salts than parotid saliva, predisposing patients to calculi formation. The route of the submandibular gland is also “uphill” which also predisposes to calculi formation. Parotid duct ligation alone is technically easy with minimal morbidity and allows most patients to take liquids during the first postoperative day; however, complications such as low grade fever and parotid swelling during the early postoperative period are not uncommon. This procedure has also been associated with a 50% re-fistulization rate.

One of the best studied procedures with a large number of patients and long term follow-up data, is submandibular duct relocation. Addressing the submandibular gland makes sense as the duct opens in the anterior mouth and the gland produces the bulk of saliva. A recent review of cases by Crysedale, et al. includes 522 patients who underwent surgical management of drooling. Two hundred and twenty-six of these patients had submandibular duct relocation. This procedure includes identification of the submandibular duct orifice, removal of a mucosal flap around the duct orifice, skeletonization of the duct, creation of a submucosal tunnel posteriorly to the base of the anterior tonsillar pillar or just posterior to the base of the tonsillar pillar. The skeletonized duct is then routed through this tunnel and sutured in to place at the base of the anterior tonsillar pillar. The later group of patients involved in this review had submandibular duct relocation and sublingual gland excision. The sublingual gland excision was

added as the complications of ranula and lateral neck cyst formation were noted after submandibular duct relocation alone. Comparing the submandibular duct relocation (SDR) group to the submandibular duct relocation with sublingual gland excision group (SDRSGE), Crysdale found that 20 of the SDR patients developed ranula while none of the SDRSGE group developed this complication; also, 23 of the SDR group required additional surgery for the control of drooling while only 6 of the SDRSGE group required additional surgical intervention. Good outcomes were reported in the majority of 106 patients who completed follow up survey with a decrease in the mean “drooling score” from 8.1 to 4.9 on a scale of 2 to 9. O’dwyer, et al. have also reported their 15 year experience treating drooling in children with submandibular duct relocation. Of 53 patients studied, 94 percent of parents surveyed stated that their child had benefited from surgery and over half reported complete cessation of drooling within three months of surgery. Post-operative pneumonia from presumed aspiration occurred in three patients (all recovered) and ranula was seen in 4 patients. Fifteen of the patients studied had the procedure over 15 years ago and all patients maintained their initial improvement.

Trans-tympanic neurectomy is another surgical option for treating drooling. Goode and Smith and Frederick and Stewart have both reported 80% success rates with this procedure. Surgery consists of lifting a tympanomeatal flap, then locating and dividing the chorda tympani and the branches of the tympanic nerves. Failure to divide all the branches of the tympanic plexus will yield poor results. Fifty percent of patients have a hypotympanic branch of the tympanic plexus which is frequently covered by bone. Therefore, it is essential to drill down the inferior aspect of the promontory to complete this procedure. There are few complications associated with this procedure, although late recurrences of drooling may occur due to nerve regeneration. There has been concern that bilateral trans-tympanic neurectomies with resultant loss of taste would be tolerated poorly. However, a group of patients who had both chordae tympani divided had no loss of weight or appetite.

Intraductal laser photocoagulation of the bilateral parotid ducts has recently been introduced as another surgical alternative to the treatment of drooling. A recent study by Chang, et al. reported results of this therapy in 48 patients with cerebral palsy. Early complications included transient facial swelling in all patients. One hematoma, two infections and two cystic formations occurred in this series of patients. There was no obvious xerostomia or scar formation. Forty patients demonstrated remarkable improvement in drooling severity, seven patients showed significant improvement and one patient showed no improvement. This less invasive means of surgical therapy deserves more thorough investigation.

In conclusion, the successful management of drooling requires a multidisciplinary approach. Noninvasive modalities should be exhausted prior to medical or surgical management. A trial of medication is warranted for those patients who still have significant drooling after behavioral or speech therapy. If these modalities fail and surgery is considered as an option, expectations of the patient and/or parents should be elicited. A full understanding that the goal of surgery is reduction of drooling without xerostomia should be communicated to the patient and/or family. Each different surgical procedure has its own set of complications or morbidities, such as respiratory compromise, infection, cyst or hematoma formation, xerostomia, pain, swelling, or visible scarring. There is no agreement on the surgical approach that is most effective with the least complications or side effects. Areas that deserve more thorough investigation include the

use of intraoral appliances and the use of intraductal photocoagulation of the parotid ducts. Comparative studies of the various surgical techniques in common usage would also be helpful.

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