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Surgical management of chronic salivary aspiration

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ABSTRACT

Aim of the study: Sialorrhoea and chronic salivary aspiration are a major problem in many neurologically impaired children causing embarrassment, skin issues and recurrent lower respiratory tract infections (LRTI). The aim of this study was to assess the efficacy of salivary gland surgery in the treatment of chronic salivary aspiration in such children.
Objectives: To compare admission rates for LRTI per annum before and after surgical intervention.
Methods: Retrospective review of all patients who underwent salivary management surgery for chronic aspiration under Princess Margaret Hospital's (PMH) Otolaryngology department from 2006 until 2013.
Results: Twelve patients were included in this review. Their ages ranged from 3 to 21 years (mean = 11.4). Their genders were equally distributed. Two patients had underlying congenital disorders; one had an acquired brain injury, while the majority (n = 9, 75%) had cerebral palsy secondary to a sustained perinatal injury.

Most patients (n = 11, 91.7%) had bilateral submandibular gland excision and parotid duct ligation as a primary procedure. One patient had a laryngotracheal separation. Two patients went on to have a second procedure. The mean follow up time was five years. Using Wilcoxon Signed-Rank test we showed that the median rate of admission per annum for LRTI pre-operatively was 1.0. This was reduced to 0.5 post-operatively, which was statistically significant ($p \leq 0.05$).
Conclusions: We hypothesize that the combination of bilateral submandibular gland excision and bilateral parotid duct ligation is effective in reducing admissions with aspiration pneumonia in neurologically impaired children, and therefore improves the quality of life in these patients.

1. Introduction

Sialorrhoea, or excessive drooling, is a significant health issue encountered by neurologically impaired children. The associated increased risk of aspiration in these patients leads to significant morbidity and reduced quality of life. The normal swallowing mechanism is in itself a highly complex process, which requires the coordination of a number of muscle groups. Consequently, any congenital or acquired condition which disrupts this mechanism can lead to failure of the swallowing process [1]. Such conditions include neuromuscular dysfunction, as in cerebral palsy; anatomical malformations, or sensory dysfunction [2].

Sialorrhoea is generally conceptualised as anterior and posterior drooling [3]. Anterior drooling refers to the unintentional loss of saliva from the mouth due to malfunction of the oral phase of swallowing [4]. The major challenges posed are cosmetic and psychological, with skin excoriation, frequent changing of soiled clothes, increased care requirements, as well as embarrassment and social isolation [4]. Posterior drooling occurs when saliva spills over the tongue into the faucial isthmus [3]. In normal circumstances this would activate the pharyngeal phase of swallowing, but in individuals with an impaired swallow, the normal physiological closure of the larynx followed by relaxation of the upper oesophageal sphincter is disrupted [3]. Pooled saliva then leads to congestion, gagging, choking, vomiting and aspiration [3].

Other factors that contribute to sialorrhoea and salivary aspiration include dysfunctional oral motor activity, tongue size, and dental malformations [5]. In children with neuromuscular dysfunction, the risk of aspiration may be further exacerbated by their positioning, which is typically supine [3].

In addition to sialorrhoea, many patients with neurological impairment experience gastro-oesophageal reflux disease (GORD), which significantly contributes to the risk of aspiration [2]. The

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majority of patients with longstanding aspiration will be fed via nasoenteric tube initially, and subsequently via gastrostomy or jejunostomy to avoid aspiration of feeds and medication [1]. In order to minimise aspiration of gastric contents, medical and/or surgical management of GORD is essential, with many of these patients undergoing fundoplication [2]. Despite these measures, these children often continue to aspirate and develop recurrent LRTIs, in which case salivary aspiration is addressed [2].

Chronic salivary aspiration is a major cause of morbidity in neurologically impaired children in particular and may cause them to suffer from recurrent lower respiratory tract infections (LRTI). Sequelae depend on frequency and amount of aspiration, immune status and baseline bronchopulmonary status [1]. Given that the aetiology of aspiration is multifactorial, management is best facilitated using a multi-disciplinary approach [1]. The aims of treatment are to decrease the incidence of LRTI, reduce hospitalisation rates and care requirements, and improve quality of life [2].

Strategies employed in the treatment of sialorrhoea and chronic aspiration can be broadly categorised into conservative measures, pharmacological therapy, and surgery. Behavioural modifications and feeding programmes are generally the initial management strategies considered [6]. Structural defects such as laryngotracheal clefts and tracheo-oesophageal fistulae are ideally corrected with surgery when possible [2].

Pharmacological interventions for sialorrhoea aim to reduce the amount of saliva produced in oral cavity and digestive tract. In the oral cavity, anti-cholingergic agents block muscarinic receptors, inhibiting stimulation of salivary glands [7]. The most commonly used anticholinergic medications are glycopyrrolate bromide, atropine, benztropine, benzhexol hydrochloride and scopolamine [7]. Given that anti-cholinergic medications block systemic muscarinic receptors, including in the central nervous system, they have a significant side effect profile [2] including xerostomia, increased viscosity of secretions, urinary retention, constipation, drowsiness, dizziness, headache, blurred vision, and seizures [7]. The consideration of side effects is essential in individuals who have severe neurological dysfunction and have an array of pre-existing co-morbidities, such as neurogenic bowel and bladder.

More recently, intra-glandular botulinum toxin type A (BoNT-A) and botulinum toxin type B (BoNT-B) injection have emerged as a treatment option to reduce oral secretions [2]. Botulinum toxins act by inhibiting the release of acetylcholine at the neuromuscular junction and reducing the amount of saliva produced by the salivary glands [7]. A 2012 Cochrane review [7] of treatment options for sialorrhoea in patients with cerebral palsy evaluated four trials involving BoNT-A. Although all studies showed some statistically significant change for treatment groups up to 1-month post intervention, there were methodological flaws associated with all of the trials [7]. As a result the existing evidence is considered to be inconclusive regarding the efficacy and safety of BoNT-A as an intervention [7]. Furthermore, BoNT-A and BoNT-B injections have been found to lead to deterioration in chewing and swallowing in some patients [8]. Other side effects include facial weakness, recurrent mandibular dislocation, and xerostomia [8]. A systematic review by Rodwell et al. [8] found that BoNT was an effective temporary treatment for sialorrhoea in children with cerebral palsy. However, serious adverse events, such as dysphagia, pooling of saliva and aspiration pneumonia were reported in a significant number of studies [8]. It was hence concluded that the potential for such serious complications must be taken into account when considering salivary gland BoNT injection in this population [8].

For patients with refractory drooling, there are a number of surgical options available. Surgical management includes submandibular gland excision (SMGE); submandibular duct ligation, parotid duct ligation (PDL), and tracheostomy to assist with bronchopulmonary lavage [2]. SMGE eliminates the major source of saliva production in resting state, while PDL decreases salivary secretion in response to food ingestion. In the most intractable cases, laryngotracheal separation (LTS) or diversion, as well as laryngectomy, may be indicated. The overriding drawback of these procedures is that, despite being highly effective in preventing aspiration, they concomitantly prohibit phonation.

The aim of this study was to appraise the efficacy of surgical management of posterior drooling using SMGE and PDL in reducing the incidence of aspiration pneumonia in children.

2. Method

A retrospective chart review was performed of all patients undergoing surgical management of sialorrhoea for chronic aspiration at Princess Margaret Hospital (PMH) from 2006 to 2013. At PMH, SMGE and PDL are considered for severe cases that have not responded adequately to medical treatment. LTS is reserved for intractable cases.

PMH is Western Australia's only tertiary paediatric facility, treating children and adolescents from around the state. There are approximately 250,000 inpatient and outpatient visits annually [9].

The patient list was obtained using theatre and clinic records. Information regarding patient demographics and diagnoses was collected using inpatient and outpatient notes. Feeding status, pre- and post-operative incidence of pneumonia, and oxygen saturation were also recorded. We assessed the number and type of procedures undergone by each of the patients, as well as surgical outcomes.

The data insertion and descriptive statistical analysis was performed using the Statistical Programme for Social Sciences (SPSS). The Wilcoxon Signed-Rank test was used to assess the pre and post-operative rates of admission for LRTI. Ethics approval was obtained from the Internal Review Board at Princess Margaret Hospital via the hospital's GEKO website.

The primary outcome measure used was reduction in hospital admission due to LRTI. The secondary outcome measures were length of hospital stay, and a reduction in secondary procedures. We hypothesised that the combination of bilateral submandibular gland excision and bilateral parotid duct ligation is effective in reducing re-admissions with aspiration pneumonia, and therefore the quality of life of these children.

3. Results

3.1. Patient demographics

Twelve patients were identified for this study. Their ages ranged from 3 to 21 years (mean 11.4 years). The ages of the patients at the time of surgery ranged from 1 to 18 years (mean 7.2 years). Genders were equally distributed, with 6 males and 6 females included in the review.

Of the twelve patients in this review, two had an underlying congenital disorder (Angelman syndrome and lissencephaly). One patient had sustained a traumatic brain injury in childhood, while the remaining nine patients (75%) had a diagnosis of cerebral palsy (Table 1)

Seven of our patients (58%) were born preterm, defined as birth before completion of 37 weeks gestation [10]. Two of these seven were very premature, defined as between 28 and 32 weeks gestation, while four (33.3% of our study population) were extremely premature at less than 28 weeks gestation [10].

There was a wide range of co-morbidities in this group, reflecting the complexity of the patient population. Seven of the