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Transoral approach to excision of massive dermoid cysts in pediatric patients: A case series

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ABSTRACT

Floor of mouth (FOM) dermoid cysts are a rare but clinically significant lesion of the head and neck. Classically, large cysts have been excised via a transcervical approach, although the limits of the transoral approach are expanding. The majority of FOM dermoid cysts present in the midline, but true lateral cysts have been reported. In this case series, we describe 3 pediatric patients who all underwent successful transoral excision of massive dermoid cysts, along with a literature review. Massive cystic lesions were identified on preoperative imaging. The dermoid cysts averaged 6 × 4.3 × 5 cm and were successfully removed via a transoral approach. Two were removed via a median ventral FOM incision and one via a lateral *trans*-sublingual approach to the FOM. When needed, the geniohyoid muscle was divided to gain access to the cyst. All cysts were removed intact, although partial, controlled decompression was performed to aid in dissection. There were no post-operative complications. FOM dermoid cysts are a rare but important cause of pediatric neck masses that present a challenging but satisfying surgical opportunity. The transoral approach is a viable option for massive dermoid cysts superior to the mylohyoid.

1. Introduction

A true dermoid cyst is a cavity lined with epithelium showing keratinization and presenting identifiable dermal appendices, as described by Meyer in 1955 [1]. These are usually congenital in nature, and present in the 2nd to 3rd decades of life [2]. Floor of mouth (FOM) dermoids are a small minority of the overall incidence, but are of particular clinical importance due to the risk of airway complications and should therefore be dealt with promptly [3–6]. The precise location of the lesion determines the presenting symptoms. Dermoids located above the mylohyoid protrude superiorly, displacing the tongue, and cause dysphonia and dysphagia [7]. Differentiating dermoid cysts from other pediatric FOM lesions such as ranulas is important due to differing surgical therapies.

Traditionally, transoral approaches are reserved for small cysts [2,8,9], those above the mylohyoid [5,10] or the genioglossus [11,12], although several reports have advocated for an expanded purview of the transoral approach [2,6,10,13–15]. In this article we review 3 cases of massive FOM dermoid cysts excised transorally, with removal of a sublingual gland and division of the genioglossus when necessary. This case series is exempt from IRB review per the UK Office of research integrity policy C1.0100.

2. Case reports

2.1. Patient 1

Patient 1 is a 17-year-old male who presented to the ED with odynophagia an enlarging "submental" mass for the past 12 days, 7 of which he had been on amoxicillin. CT scan (Fig. 1) demonstrated a 7 × 6.1 × 4.8 cm mass in the sublingual space extending from the attachment of the genioglossus near the mandible to the hyoid. In the or, he was intubated transnasally, prepped with peridex, and injected with lidocaine 1% with 1:100,000 epinephrine into the midline tongue along the ventral surface and the FOM down to the mandible. A sharp mucosal incision was made from the ventral surface of the tongue down to the alveolar ridge. The genioglossus and geniohyoid muscles were divided along the midline raphe to expose the dermoid in the sublingual space. The cyst was decompressed to improve visualization during dissection off the hyoid (Fig. 2). A penrose drain was left in place until discharge on

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Fig. 1. Axial CT with contrast of patient 1 revealing a large sublingual dermoid cyst.

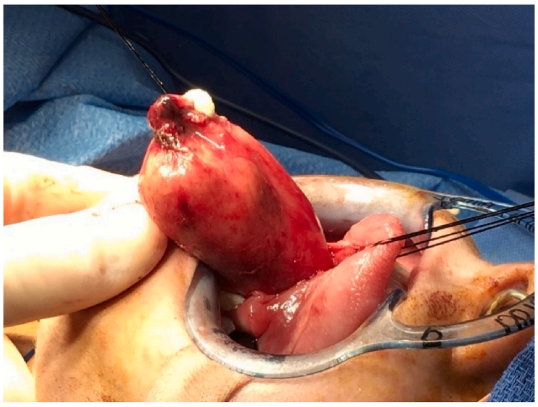

Fig. 2. A median ventral glossotomy incision is shown in patient 1. Controlled decompression was performed to allow better visualization during dissection.

postoperative day one. He was seen in clinic 1 week and 3 months postoperatively. His initial complaints of mild tongue tip numbness had resolved at the 3-month appointment.

2.2. Patient 2

Patient 2 is a 12-year-old girl who presented with complaints of increasing submental fullness over the past year. In the weeks prior to presentation, she contracted strep pharyngitis, and noticed rapid progression of the mass and difficulty with articulation. On physical exam, there was significant submental fluctuance and enlargement, which was more noticeable on the right. There were no overlying skin changes. The FOM was raised, with a translucent cystic appearance. The CT scan (Fig. 3) revealed a large homogenous FOM cyst, 6 × 4.5 × 6.5 cm in size, extending from tongue inferiorly to the level of the hyoid. The cyst was superior to the mylohyoid muscle and pierced the midline of the geniohyoid and genioglossus muscles. In the or, she underwent transnasal intubation, and her FOM was injected. A suture was placed through the tip of her tongue for retraction. An incision was made from the tip of the tongue on the ventral surface down to the alveolar ridge through the midline raphe. To aid in dissection, a small hole was made in the cyst and 20 mL of milky fluid was removed using a syringe and angiocath. Dissection was carried down to the mylohyoid. The cyst was removed intact. The genioglossus muscle was reapproximated, and the FOM was

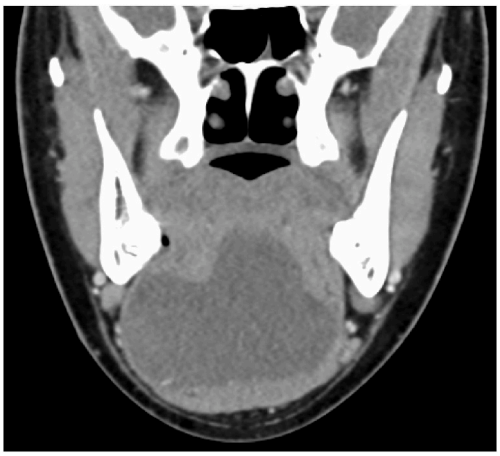

Fig. 3. Coronal CT with contrast of patient 2 revealing large FOM dermoid. Note the thinning of the mylohyoid as the dermoid expands laterally.

closed save a 1mm drainage port. Pathology revealed a benign cyst lined with keratinized squamous epithelium with skin appendages present in the cyst wall, consistent with a sublingual dermoid cyst. She did well postoperatively and was discharged home the next day on a weeklong course of postoperative antibiotics. At 1 week and 3-month follow-up, she denied any swallowing or breathing difficulties and displayed normal tongue range of motion on exam.

2.3. Patient 3

Patient 3 is a 14-year-old boy with a 1 year history of an asymptomatic stable right submandibular neck mass. He denied any pain, infection or drainage from the area, but was bothered cosmetically. On exam, there was fullness of the right FOM and the mass enlarged with tongue protrusion. The CT was consistent with a 5 × 2.5 × 4cm ranula (Fig. 4). After nasal intubation and local anesthetic injection into the FOM, the right Warthon's duct was cannulated with a lacrimal probe. Electrocautery was used to make a FOM incision lateral to the duct and dissection was carried out to identify the sublingual gland. After identifying the lingual nerve, the sublingual gland was excised to expose the large cyst. The cyst was dissected away from the submandibular gland, lingual nerve, and genioglossus muscle near the attachment to the genial tubercle. Chromic was used to close the incision, leaving a small area open superiorly for drainage. He did well postoperatively, without

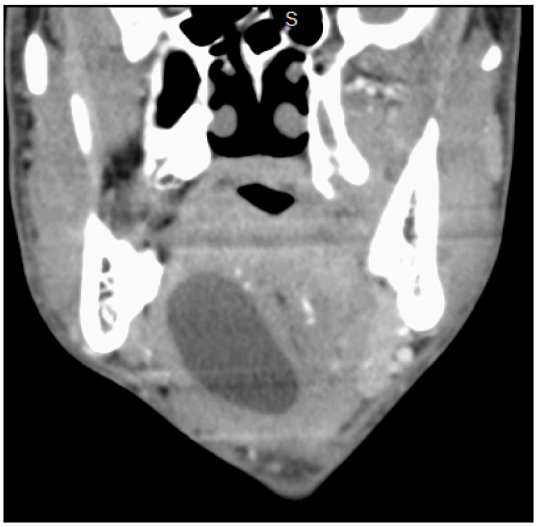

Fig. 4. Coronal CT of patient 3 revealing a large cystic mass in the right FOM.

dysphagia or significant pain.

3. Discussion

Dermoid cysts are one histological type of congenital germline fusion cysts [13], which are a rare cause of transoral cysts (0.01%), and rarely present in the FOM (1.6%) [16]. The term dermoid cyst is often ambiguously used to encompass all three histological subtypes of congenital germline fusion cyst, namely the epidermoid, dermoid, and teratoma subtypes [17]. These lesions are theorized to originate from midline ectodermal and mesodermal tissue trapped during the fusion of the first and second brachial arches and are therefore predominantly midline [1]. Only 13 cases of lateral dermoids have been reported in the medical literature [18–22]. There is some contention as to the origin of lesions off the midline, but many assume that they started in the midline and expanded laterally [1,3,11,22,23], while others propose that they arise from a first pharyngeal pouch [12,22]. Dermoid cysts can be further classified by their anatomic location, although inconsistent usage of the terms sublingual, submental, and submandibular in reference to lesions either superior or inferior to the mylohyoid or geniohyoid muscles obscures current recommendations [2,5,10,11,21,24].

FOM dermoid cysts are located in the sublingual space, which is superior to the mylohyoid and medial to the anterior bellies of the digastric muscles. The geniohyoid muscles, another commonly referenced anatomic landmark, are in the sublingual space. In contrast, the submental space is inferior to the mylohyoid and therefore not in the FOM. In 2007, Teszler et al. performed an exhaustive literature search in an attempt to standardize this nomenclature, but their precisely defined categories do not enjoy widespread usage [20,21]. At the very least, the term 'submental cyst' should be limited to cysts completely inferior to the mylohyoid muscle, and sublingual cysts with extension through the mylohyoid should be described as such. Regardless, it is critical to differentiate FOM dermoids from ranulas, due to the difference in surgical management; only the latter of which may be successfully treated with marsupialization and/or excision of the involved salivary gland [4,25]. Preoperative imaging is therefore helpful for surgical planning [3,8,11,26], although some authors suggest its role should be limited to large or recurrent lesions [25,27]. US, CT, and MRI are all adequate for diagnosis, although a T2 weighted MRI is the gold standard modality as it most accurately defines the relationship of the cyst to the mylohyoid [3,26].

Prior authors have advocated for transcutaneous approaches for large (>6cm) lesions located deep in the FOM [1,8,9,11,20,21]. While this approach is certainly safe, we present three cases of large FOM dermoids, safely excised transorally, without subsequent complication and with excellent cosmetic outcomes. No functional deficits were noted with division of the genioglossus or geniohyoid in the midline as the raphe allows for relatively atraumatic reapproximation. This technique has recently been the subject of a multicenter prospective observational study with 21 and 22 patients undergoing transoral and transcervical approaches, which found shorter operative times and superior cosmesis in the transoral group [28]. Interestingly, they employed an endoscope for the transoral cases, a technique they first described in 2010 [29]. This is the first reported case to our knowledge to describe removal of a sublingual gland to gain access to the cyst. The surgical technique was very similar to that described by Hughes et al. in their transoral approach to the submandibular gland [30]. This maneuver provided improved visualization of the lingual nerve and submandibular gland without resulting in any functional deficits. Given the obvious cosmetic benefit of the transoral approach, we support the trend to employ this route for excision of massive FOM dermoids.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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