Case Report
A Curious Case of a Pain in the Neck

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Received 11 November 2014; Revised 17 December 2014; Accepted 5 January 2015

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Abstract The article presents a case study of a 21-year-old Polish female with a short history of an enlarging, painful, midline neck swelling and odynophagia. She underwent a transcervical excision of an epidermoid cyst, which was abutting her airway posteriorly, with nasal intubation and close observation in ICU. She has enjoyed full relief of her symptoms and an excellent cosmetic outcome. The article discusses the challenges of diagnosis and management of midline neck swellings. In particular, avoiding needle aspiration and management of the airway are highlighted.

Keywords epidermoid cyst; neck swelling; surgery; transcervical; airway; fine needle aspiration

1. Background
Epidermoid cysts are benign lesions that can arise anywhere in the body, but tend to occur most often at sites of embryonic fusion [1]. They are characterized histologically by a cystic space lined by simple squamous epithelium. When they contain skin adnexae they are called “true” dermoid cysts, and when they contain tissues derived from all three germ layers they are called teratoid cysts. The term “dermoid cyst” has been used loosely in the literature to refer to any of these three cysts [2].

Eighty percent of epidermoid cysts arise in the ovaries and testes. Cysts arising in the head and neck are uncommon and account for only 7% of all epidermoid cysts [1]. Moreover, epidermoid cysts account for only 0.01% of all oral cavity cysts, with the majority of these occurring in the sublingual area [3].

Traditionally, the pillars of diagnosis have been imaging, in combination with fine needle aspiration and excision biopsy. The number of similar lesions that arise in the head and neck area complicates the clinical picture and poses diagnostic and management challenges.

We report a case of a sublingual epidermoid cyst, presenting as a painful anterior midline neck mass, abutting the airway and requiring urgent transcervical excision.

2. Case report
A 21-year-old Polish female presented to the Emergency Department with a sudden increase in size and pain from an anterior, midline neck swelling over 4 days. A small, stable, and painless swelling had been present for 3 months prior to her presentation. She had no past medical or surgical history and no family history of note. She was an active smoker with a five-pack-year history.

The swelling was visible extraorally as a large, rounded, midline neck lump (Figure 1). It was nontender to palpation with no overlying skin changes. She was afebrile with no lumps elsewhere. Intraoral examination revealed a solitary, midline, dome-shaped swelling in the floor of the mouth (Figure 2). It was firm, smooth, and fluctuant to palpation with no associated discharge. The overlying mucosa was
The swelling did not change with tongue movement, and milking of the sublingual and submandibular salivary glands produced thick, salivary secretions.

Flexible laryngoscopy revealed a patent airway and a normal nasopharynx, oropharynx, and larynx. Her blood results on admission were unremarkable.

A decision was made to avoid fine needle aspiration until the extent of the swelling was fully delineated by imaging. CT revealed a $4 \times 4 \times 4$ cm mass in the floor of the mouth between the genioglossus muscles. Of greatest concern, the sagittal view showed that the swelling, through its involvement of the base of the tongue, had expanded the pre-epiglottic space and had narrowed the airway to a near critical impasse of 0.37 cm posteriorly (Figure 3).

We decided to proceed with an urgent transcervical excision of the swelling under general anesthetic (Figure 4). The patient was nasally intubated and transferred to ICU for a close observation post-op.

Excision yielded a well-circumscribed, cystic mass of tissue (Figures 5 and 6). Sectioning revealed caseous material throughout the lesion. Microscopic examination revealed a cyst lined by squamous epithelium only, and containing laminated keratin. These appearances were consistent with a ranula.
diagnostic of a benign epidermoid cyst. At her last review in the Outpatient Department, the patient had made a full recovery with an excellent cosmetic outcome (Figure 7).

3. Discussion

Epidermoid cysts are benign lesions that can occur anywhere in the body. Cysts in the head and neck area are rare, accounting for just 7% of all epidermoid cysts. Only 0.01% of all oral cavity cysts are of epidermoid histology [3]. Cysts with the size and extent of airway involvement seen in our case are even more unusual.

Meyer classified these tissues histologically into epidermoid (lined by simple squamous epithelium), dermoid (containing skin adnexae), and teratoid (containing tissues derived from ecto-, endo- and mesoderm) [2]. Cysts can also be classified as congenital or acquired based on their evolution but there is no difference between the two either clinically or histopathologically [4].

The etiology of epidermoid cysts is unclear. It has been postulated that congenital cysts may arise from the entrapment of epithelial remnants during embryological development. Traumatic implantation of epidermal cells may be implicated in the pathogenesis of acquired cysts. Others have suggested that epidermoid cysts may be simply a variant of the more common thyroglossal cyst [5].

Regardless of etiology, these cysts are unlikely to become clinically apparent until they reach a large enough size to exert a mass effect on surrounding structures. This most commonly occurs between the ages of 15 and 35 with no gender predilection [5].

Upward displacement of the tongue by a sublingual swelling can result in dyspnoea and dysphagia. Inferior extension of the swelling gives rise to a characteristic “double chin” appearance.

Fine needle aspiration, US, CT, and MRI images provide important information on the location and extent of the lesion and facilitate optimal preoperative planning.

Fine needle aspiration has been widely advocated in the literature as a safe, reliable, and economical test for evaluating sublingual swellings [6]. One retrospective case-series study of histopathologically confirmed head and neck epidermoid cysts used FNAC as an essential component of their diagnostic workup [7]. Other studies have suggested that FNAC should be considered only as corroborative evidence after imaging or that FNAC may not be diagnostic at all [8].

One of the key learning points from our case is that FNAC is not appropriate in the initial workup of all neck lumps. Given the involvement of this lesion with the base of the tongue, even a small bleed from a FNA could have created a rapidly critical airway. While FNA is an important tool in the assessment of metastatic nodal disease, thyroid, and parotid lumps, it should be avoided when there is any risk that the mass is abutting the airway or when the mass is pulsatile. Coagulopathies and anticoagulant medications should also be considered contraindications to a FNA.

The most likely differentials for a cystic lesion in the floor of the mouth are a ranula, a thyroglossal duct cyst, and a cystic hygroma. The central location of our patient’s swelling favored a cyst over a ranula.

The treatment of an epidermoid cyst is a complete surgical excision for relief of symptoms and aesthetic disability. Malignant transformation of epidermoid cysts to basal or squamous cell carcinomas is rare but recognized. Recurrence is also uncommon after complete excision [1].

There have traditionally been two approaches to the surgical excision of sublingual swellings. Small sublingual swellings above the geniohyoid were thought to be amenable to the transoral approach, while large swellings below the geniohyoid were thought to be amenable to the external or transcervical approach. Huge swellings could be excised through a combined approach.
Our patient’s cyst was noted on CT to be located superior to the mylohyoid. Both transoral and transcervical excisions were considered. Given the size and location of the swelling, it was felt that patient safety was more important than the cosmetic outcome. It was decided to use the transcervical approach as this offered greater control of any potential bleeding at the base of the tongue.

Protection of our patient’s airway was crucial to the successful outcome of surgery. This patient was not suitable for return to the general wards as a post-op hemorrhage could have been rapidly fatal. For this reason, she was nasally intubated and transferred to ICU for observation for 24 h. Anecdotal evidence suggests that when post-op bleeds happen, they almost always occur within the first six hours after surgery. Another option for ventilaing this patient would have been to create a tracheostomy. However, given the patient’s age, her own preference, and the availability of an ICU bed, it was decided that nasal intubation was the most appropriate means of airway management.

4. Conclusion
Epidermoid cysts are very rare benign lesions in the oral cavity but should be considered in the differential diagnosis, particularly for midline masses. Fine needle aspiration is a useful diagnostic tool but should be avoided when there is a risk that the mass is abutting the airway. The transcervical approach is the most appropriate for large cysts based below the geniohyoid and airway protection is a key principle of surgery.

Consent The patient has given her express consent to be part of this case report.

References