A Case of a Dermoid Cyst Compressing the Airway

Laura R. Garcia-Rodriguez, MD; Sachin Pawar, MD; Michelle A. Michel, MD; Bruce H. Campbell, MD

ABSTRACT

Introduction: In this report we discuss the etiology, common locations, diagnostic approach, and treatment of a dermoid cyst.

Case Presentation: A 22-year-old man arrived at the emergency department complaining of submental fullness, an increase in snoring, choking, gagging, and difficulty breathing. The patient was taken to the operating room for a complete resection of a large dermoid cyst that was compressing his airway.

Discussion: Dermoid cysts are uncommon head and neck tumors mainly presenting in patients aged 15 to 35. The origin of dermoid cysts is thought to be congenital in most cases, but they can also develop from acquired factors such as trauma or surgical implantation that forces epithelial cells into deep tissues.

Conclusion: Although benign and often asymptomatic, dermoid cysts may cause other associated symptoms due to compression of structures in the head and neck.

INTRODUCTION

Dermoid cysts are uncommon masses that can arise in the head and neck area, most commonly in people aged 15 to 35 years,[1] during a period of maximal epithelial activity.[2] They are thought to be congenital in origin, but the literature also indicates they can arise due to implantation.[1] Diagnostic modalities include imaging such as ultrasound, computed tomography (CT) scan, and magnetic resonance imaging (MRI). Dermoid cysts must be considered in the differential diagnosis of head and neck masses. In this report, we present a case of an acute growth of a neck mass in a 22-year-old man that resulted in airway compression and caused him to go to the emergency department (ED). As a case report, this project was exempt from Institutional Review Board approval.

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Author Affiliations: Department of Otolaryngology and Communication Sciences, Medical College of Wisconsin, Milwaukee, Wis (Garcia-Rodriguez, Pawar, Campbell); Department of Radiology, Medical College of Wisconsin, Milwaukee, Wis (Michel).

Corresponding Author: Bruce H. Campbell, MD, Department of Otolaryngology, Medical College of Wisconsin, 9200 W Wisconsin Ave, Milwaukee, WI 53226; phone 414.805.5583; fax 414.805.7890; e-mail bcampbell@mcw.edu.

CASE PRESENTATION

A 22-year-old male presented to the emergency department with a 2-day history of increasing submental fullness and an associated increase in snoring, choking, gagging, and difficulty breathing. He reported a 2- to 3-year history of significant weight gain, up to 148.5 kg, which had caused him to develop a "double chin." However, after losing 15-20 kg over the past year, he became concerned because of persistent submental fullness. He had been sleeping propped up on pillows for the past year due to difficulty breathing at night, which he attributed to asthma. However, use of his albuterol inhaler did not alleviate his symptoms. Two days prior to his arrival at the ED, his primary care physician noted a submental mass and made an outpatient referral to our clinic for further evaluation. Instead, because of worsening respiratory symptoms in the interval, he presented to the ED for acute management.

Clinical examination of the patient's oral cavity and oropharynx was unremarkable. The floor of the mouth was soft and the tongue appeared normal with good mobility. Fiberoptic examination of the pharynx and larynx showed medial displacement of the left lateral pharyngeal wall and rightward deviation and rotation of the larynx. The airway was patent. Examination of the neck revealed a 10 x 15 cm soft neck mass extending from the submental region to the left mandibular angle and inferiorly just beyond the level of the hyoid. A contrast-enhanced CT scan of the neck was obtained (Figure 1), which was followed up by an ultrasound as recommended by radiology (Figure 2). An MRI was obtained to further define the location of the lesion with respect to adjacent musculature and other soft tissues (Figure 3).

Excision of the mass began with a skin crease incision followed by a subplatysmal flap elevated to the level of the mandible. The anatomy of the neck musculature was distorted due to stretching by the underlying mass. Dissection focused on establishing a plane between the mass and the surrounding

Figure 1. Post-contrast Axial CT Scan

Image demonstrates a large, well-defined floor of mouth mass that is hypodense compared to adjacent musculature. Multiple low-attenuation, rounded foci are noted scattered throughout the lesion. Note slight mass effect upon the oropharyngeal airway (arrow).

Figure 2. Sagittal Ultrasound Image

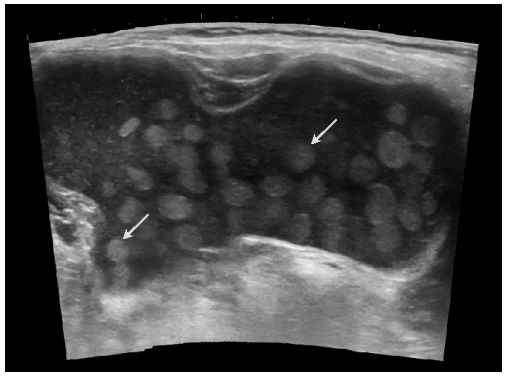


Image shows that the lesion is cystic, measures 13 x 8 x 5 cm, and contains multiple mobile, hyperechoic nodules (arrows) suspended within the fluid.

soft tissue. Dense musculature was dissected off the mass and retracted away. Once the plane around the mass was established, dissection was completed from lateral to medial and inferior to superior. The right marginal branch of the facial nerve and submandibular gland were identified and preserved. Once the mass was circumferentially exposed, it was delivered out of the neck, freeing it from its depths on the left side. The tissue tethering the mass to the neck was cut and the mass was delivered intact. Final inspection, irrigation, hemostasis, and drain placement were completed prior to closure.

Pathology revealed a cyst wall lined by focally keratinizing stratified squamous epithelium with underlying areas focally showing skin adnexae. The globules identified on the imaging studies were composed of keratinaceous debris with acute inflammation.

The postoperative course was rather eventful as the patient had submental swelling and drain output that exceeded the criteria for safe discharge. Drains remained in place for 3 days. Because of increasing swelling, his neck incision was reopened on postoperative day 5. Edema of muscle and soft tissue was present. The remainder of his recovery was uneventful. At follow-up 4 months later, he reported no problems with oral intake, articulation, or breathing. He no longer needed to sleep in a semi-recumbent position. He did not have asthma, and no longer required albuterol.

DISCUSSION

A dermoid cyst is an uncommon head and neck tumor that mainly presents in patients ages 15 to 35, with an equal distribution between males and females.[1] Depending on the source, the ratio of males to females can be 3:1.[3] It is thought that most are seen in this age range because of increased activity of epithelial tissues. With accelerated activity of hair follicles, sweat glands, dermis/epidermis, etc, there is increased filling of the cystic lumen, which is why these previously inconspicuous lesions are noticed with maturation of the patient.[2] One study analyzed 2063 neck masses, of which 252 (12%) turned out to be congenital.[4] Of these, dermoid cysts comprised 11% and thyroglossal duct cyst/fistulas represented 53%. Another study similarly reported that 9% of pediatric neck masses were dermoid cysts.[5]

The origin of dermoid cysts is thought to be congenital in most cases, but they also can be acquired. The congenital theory suggests that entrapped midline ectodermal tissue during fusion of the first (mandibular) and second (hyoid) branchial arches during the third and fourth week of fetal development result in a cystic structure.[1] Alternatively, the cysts may arise from the tuberculum impar of His, which forms the body of the tongue and floor of the mouth with each mandibular arch.[3] These cysts also can develop from acquired factors such as trauma or surgical implantation that forces epithelial cells into deep tissues.[1]

A review of 195 case reports indicated the most common location to be intraoral (58.3%), followed by sublingual (52%), and submental (26%).[1] From those case reports, it was also apparent that the vast majority of cysts were dermoid (72.9%), epidermoid (22.2%), and teratoma (4.9%). The simplest is an epidermoid cyst which is lined by simple stratified squamous epithelium. A dermoid cyst has the features of an epidermoid cyst, plus skin appendages such as hair, sweat glands, and seba-

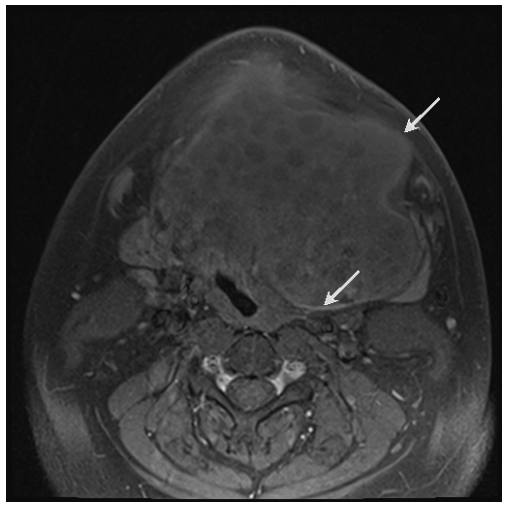
ceous glands. A teratoma is characterized by epidermoid and dermoid features, plus gastrointestinal or respiratory tissue and connective tissue derivatives.[1]

In the case described here, an axial CT scan with contrast (Figure 1) demonstrated a large hypodense floor of mouth mass centered to the left of the midline with numerous low-attenuation rounded foci within the lesion, which were thought to represent fat globules. The pathognomonic finding on CT scan and MRI is a "sack of marbles," the marbles indicating coalescence of keratinous material with lipid, which was seen on this image.[6] The exact location of the mass was difficult to determine on CT due to its large size, 10 x 12.6 x 6 cm, but was most likely located within the sublingual space as it crossed the midline in the anterior floor of mouth. There was mass effect on the oropharynx, hypopharynx, and supraglottic laryngeal airway, resulting in the patient's dyspnea, which was not alleviated by frequent albuterol use. A sagittal ultrasound image (Figure 2) showed a large ovoid cyst, 13 x 8 x 5 cm, with complex internal features. The lesion was filled with hypoechoic fluid with multiple well-defined hyperechoic nodules suspended within the fluid. The findings of the CT scan and ultrasound were consistent with a dermoid cyst. An axial-T1 weighted gadolinium-enhanced MRI with fat suppression, localized the mass to the sublingual space (Figure 3). The mylohyoid muscle sling was thinned and markedly peripherally displaced. The MRI demonstrated that the cyst was larger to the left of the midline, but crossed extensively into the right floor of mouth in the plane between the mylohyoid sling and the (superiorly displaced) geniohyoid muscles. The signal intensity within the fluid suggested that it was proteinaceous; however, the globules did not follow the signal intensity of mature fat. Figure 4 is an enlarged view of the cyst contents.

Similar cases have previously been reported in the literature with typical presentations such as a painless mass resulting in a double chin, dyspnea, dysphagia, or dysphonia. Recommended preoperative diagnosing techniques include ultrasound, CT scan, MRI, and fine needle aspiration biopsy.[3] MRI is usually necessary to determine the exact location within the surrounding soft tissues as the cyst wall may tightly adhere with its surroundings. Ultrasound is a reasonable initial imaging study as it is readily available, cost effective, and does not expose the patient to ionizing radiation. This is particularly helpful in the pediatric patient.[7]

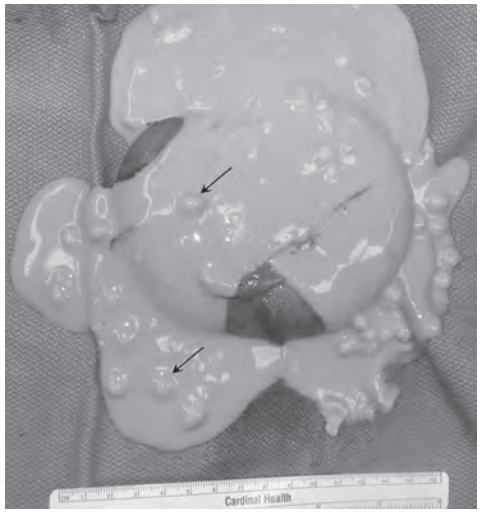
Most clinicians agree that treatment is complete surgical resection, and recurrence is rare if complete excision is achieved. To relieve symptoms of airway compromise prior to definitive treatment, aspiration may be attempted, although it may not be possible if the contents are too viscous. Prior to surgical intervention, if the mass is constricting the larynx and intubation is impossible, tracheotomy or decompression via aspiration may be necessary.[1] Complications can include infection, bleeding, anesthesia risk and incomplete resection, although some cases report no complications except moderate post-surgical edema, which also occurred in our patient.[3]

Figure 3. Axial T1 Weighted, Gadolinium-enhanced MRI with Fat Suppression



Shows no appreciable enhancement within the cyst, localization of the mass to the sublingual space. Minimal enhancement is noted at the periphery (arrows).

Figure 4. Cystic Contents Exposed after Resection of Mass



Globules are noted with arrows.

CONCLUSION

Dermoid cysts have characteristic clinical and radiographic fea-

tures that can aid in preoperative diagnosis. Although benign and often asymptomatic, dermoid cysts may cause other associated symptoms due to extrinsic compression of structures in the head and neck, including airway compromise. Currently, surgery is the mainstay of treatment and complete excision typically results in a very low recurrence rate.[2,3]

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REFERENCES

1. King RC, Smith BR, Burk JL. Dermoid cyst in the floor of the mouth. Review of the literature and case reports. *Oral Surg Oral Med Oral Pathol.* 1994;78(5):567–576.

2. Meyer I. Dermoid cysts (dermoids) of the floor of the mouth. *Oral Surg Oral Med Oral Pathol.* 1955;8(11):1149–1164.

3. Longo F, Maremonti P, Mangone GM, De Maria G, Califano L. Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques. *Plast Reconstr Surg.* 2003;112(6):1560–1565.

4. Al-Khabteeb TH, Al Aoubi F. Congenital neck masses: a descriptive retrospective study of 252 cases. *J Oral and Maxillofac Surg.* 2007;65(11): 2242-2247.

5. Connolly AA, MacKenzie K. Paediatric neck masses—a diagnostic dilemma. *J Laryngol Otol.* 1997;111(6):541-545.

6. Lin HW, Silver AL, Cunnane ME, Sadow PM, Kieff DA. Lateral dermoid cyst of the floor of mouth: unusual radiologic and pathologic findings. *Auris Nasus Larynx.* 2011;38(5):650-653.

7. Ayugi JW, Ogeng'o JA, Macharia IM. Pattern of congenital neck masses in a Kenyan paediatric population. *Int J Pediatr Otorhinolaryngol.* 2010;74(1):64-66.

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