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Lateral dermoid cyst of the floor of mouth: Unusual radiologic and pathologic findings

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Abstract

A lateral dermoid cyst is a rare lesion of the floor of mouth, with only 12 cases reported in the literature. We describe the case of a 60-year-old man with a slowly enlarging mass in the submandibular region. Magnetic resonance imaging demonstrated a lesion containing multiple uniformly rounded foci, creating a ''sack-of-marbles'' appearance. Needle aspirations showed atypical findings, and the mass was excised. Histopathology revealed a cyst containing a keratinizing stratified squamous epithelial lining with apocrine and eccrine glands. These findings were diagnostic of a dermoid cyst, which should be considered in the differential diagnosis of any midline or lateral cervical lesion.

Keywords: Dermoid; Neck mass; Neoplasm; Head and neck imaging; Head and neck pathology

1. Introduction

Dermoid cysts are uncommon, benign congenital tumors of ectodermal origin that can occur in any region of the body. Believed to arise during fetal development when ectodermal differentiation and entrapment occur along lines of embryonic fusion, these lesions can be divided into three distinct histopathologic subtypes, including epidermoid, dermoid and teratoid varieties. While the ovaries and sacral region are the most common location for these masses, nearly 7% occur in the head and neck region, and more specifically, dermoid cysts of the floor of mouth account for 1.6% of all cases [1]. Furthermore, of the dermoid lesions occurring the floor or mouth, only about 6% are found laterally and present in the submandibular space, with a total of only 12 such cases reported in the literature to date [2,3]. We present an additional case and provide uniquely detailed radiologic, surgical and histopathologic findings. This case is highly unusual in three respects: (1) that the lesion became clinically significant in the seventh decade of the patient's life; (2) that the lesion was located in the tongue base and submandibular regions where it exerted considerable mass effect; and (3) that the radiographic and gross histopathologic findings were exceptionally xenotypic in appearance.

2. Case report

A 60-year-old gentleman presented to our institution with a two-year history of a painless and slowly enlarging mass of the right anterior neck. On physical examination, a moderate-sized mass protruding from the right submandibular region and mild fullness of the right floor of mouth were

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noted. With bimanual intraoral palpation, a large, round, firm and non-tender mass could be appreciated. Fiberoptic nasolaryngoscopy, moreover, revealed base of tongue protrusion posteriorly, resulting in moderate vallecular effacement. Multiple fine needle aspirations (FNA) provided variable findings, including a biopsy that revealed atypical cells concerning for squamous cell carcinoma. A computed tomography (CT) scan demonstrated a 4.4 cm × 3.0 cm × 3.5 cm lesion with mixed attenuation and scattered heterogeneous calcifications (Fig. 1). Magnetic resonance imaging (MRI) further revealed a strikingly xenotypic mass containing multiple uniformly-rounded foci measuring up to 3 mm in diameter, creating a "sack of marbles" appearance on cross-sectional imaging (Fig. 2).

The patient was consented for a transcervical excision of the lesion and possible tracheotomy or postoperative intubation given the large size and the lateral submandibular presentation of the mass, as well as concern for post-operative local edema triggering airway compression at the level of the tongue base. Intraoperatively and following submandibular gland removal, the mass was noted to be deep to the mylohyoid muscle and surrounded by extensive fibrosis that encased the right lingual nerve. The main trunk of the nerve was preserved. The hypoglossal nerve was also adherent to the capsule of the mass, but was carefully dissected free from it and preserved. Following *en bloc* removal of the mass, incision through the capsule of the lesion demonstrated a large cyst containing numerous regularly shaped yellow, 3 mm spheroid fragments of equal size that had a paste-like consistency (Fig. 3). Histopathology revealed that the cyst contained an attenuated and keratinizing stratified squamous epithelial lining with rare underlying skin appendages, including apocrine and eccrine glands, within the cyst wall (Fig. 4). These findings were diagnostic of a dermoid cyst.

Due to the extensive dissection required to access the posterio-superior aspect of the cyst and concern for resulting oropharyngeal/tongue base edema, the patient remained intubated overnight while on parenteral corticosteroid therapy and was successfully extubated on the first postoperative day. At last follow-up the patient is doing well and without signs of recurrence. He initially had a praxia of the mild lingual and hypoglossal nerve praxia ipsilaterally that has subsequently resolved by six weeks post-operatively.

3. Discussion

Dermoid cysts of the floor of mouth are uncommon tumors found in the midline in the vast majority of cases [2,4] and typically present in the second or third decades of life as a painless, slow-growing mass in the floor of mouth, submentum or anterior neck [5]. True lateral cervical presentations are exceedingly rare, with only 12 cases of purely lateral cervical dermoid cysts reported in the literature. Of note, Teszler and colleagues developed an anatomico-surgical classification system of dermoid cysts of the floor of mouth to assist surgeons with the decision-making process of the surgical approach. In their algorithm, a dermoid cyst can be grouped into one of seven classes based on its median versus lateral location and relationships to the mylohyoid and geniohyoid muscles [3]. Here we have described a highly unusual case of a supramylohyoid dermoid cyst presenting the seventh decade of life as a lateral cervical mass.

Histopathologically, the terms "dermoid" or "dermoid cysts" have been used as umbrella titles to describe the three subtypes of these congenital cysts containing keratinous squamous material. Epidermoid cysts or epidermal inclusion cysts are lesions lined with a simple squamous epithelium with no adnexal structures. True dermoids are stratified squamous epithelial-lined cysts that contain skin adnexal structures, including hair, hair follicles, sebaceous and sweat glands. Finally, teratoid cysts are masses lined with a variety of epithelia, including stratified squamous and ciliated respiratory epithelia, and contain elements of ectodermal, endodermal and/or mesodermal origin [4]. The differential

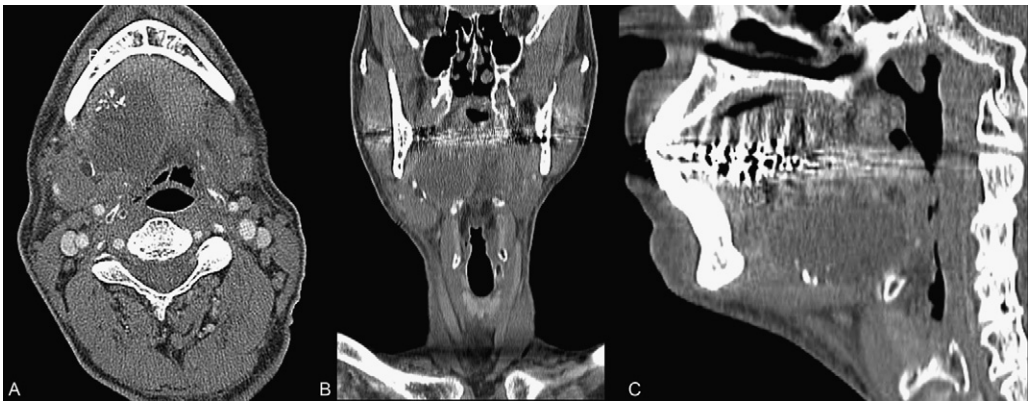

Fig. 1. Contrast-enhanced CT imaging in axial, (A) coronal, (B) sagittal, (C) soft tissue windows demonstrated a 4.4 cm × 3.0 cm × 3.5 cm mass containing heterogeneous calcifications and rounded fat attenuation.