Dermoid Cyst Occupying the Entire Floor of Mouth. Surgical Management and Review of the Literature

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Abstract

Dermoid cysts constitute almost 6.9% of all cysts in the head and neck region, representing less than 0.01% of all the cysts of the oral cavity. They occur more frequently in the lateral third of the eyebrow with 26% of them found in the floor of the mouth. Rarely they increase in size compromising speech, chewing, swallowing endangering also upper airway patency. Their stepwise structural approach and management consists in FNA biopsy, imaging studies mainly CT scan and finally surgical enucleation. In this report, we describe a young man who presented at the outpatient clinic of the Oral and Maxillofacial Surgery department at General State Hospital of Athens “Georgios Gennimatas” to evaluate a cystic sublingual lesion, occupying the entire floor of the mouth.

Keywords: Cyst; Dermoid; Epidermoid; Enucleation; Mouth

Introduction

Dermoid cysts constitute almost 6.9% of all cysts in the head and neck region, representing less than 0.01% of all the cysts of the oral cavity [1]. They occur more frequently in the lateral third of the eyebrow with 26% of them found in the floor of the mouth [2].

Histologically they present as cystic malformations lined with squamous epithelium. Further histological classification divides them into epidermoid (when they are lined with simple squamous epithelium), dermoid (when skin adnexa are found in the cyst wall) and teratoid (when other kind of tissue, such as muscle, cartilage and bone are present) [3,4]. Embryologically they are thought to originate from the overlying surface ectoderm that fails to separate from the underlying structures. An abnormal infolding of the ectodermal layer (hence the other often used term inclusion cyst) along normal embryonic lines of fusion of the facial processes or within the neural axis is thought to be the etiopathogenesis [5]. Their peak of incidence is in the second and third decades of life [6,7].

Clinically, they usually appear as a slow-growing asymptomatic mass, usually located in the midline, above or below the mylohyoid muscle. Depending on their relationship with the mylohyoid muscle, they may manifest either as a sublingual swelling (when above the muscle), or as a sub-mental swelling (when below the muscle). Frequent patient complaints are: tongue elevation, difficulty in swallowing, breathing or sleeping, as well as speech alteration or even the development of a double-chin [8,9].

As their initial course is often asymptomatic, dermoid cysts are usually diagnosed only after they have reached a considerable size. Their definite and recommended treatment is surgical excision via an intraoral or an extraoral access, depending on the lesion’s size and...
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As a general principle, according to Longo., et al. [11] those who are located above the muscle are excised via an intraoral approach and those who are below the muscle are excised via an extraoral approach. In this report, we describe a young man who presented at the outpatient clinic of the Oral and Maxillofacial Surgery department at General State Hospital of Athens “G. Gennimatas” to evaluate a cystic sublingual lesion, occupying the entire floor of the mouth.

Case Report

A 37-year-old man presented at our department with a bulging mass of the floor of his mouth. Physical examination showed a large, smooth nontender swelling that involved the submental, submandibular, and sublingual areas on both sides. The mass was not freely movable, did not move with swallowing and caused elevation of the floor of the mouth. The overlying mucosa was pink and healthy appearing. Salivary flow from Wharton’s ducts was abundant and clear; no purulence was noted as well.

The patient was afebrile and there was no gross facial asymmetry. The neck was supple and without lymphadenopathy. Jaw range of motion was within normal limits. His dental hygiene was good with no excessive caries and intraoral lesions and physical examination and blood test revealed no special findings. His medical history was free for any systemic and chronic disease or medications.

His main complaints were difficulty in speaking, in eating, in swallowing and in breathing during his sleep as also drooling. He reported that he had this lesion for almost 18 years as also four to five attempts of needle aspirations at the regional Oral and Maxillofacial Surgery department at the country of his origin.

The patient underwent an axial computed tomography (CT) scan that revealed a well-defined large cystic lesion with a diameter of 7 cm at the floor of the mouth, occupying the submental submandibular and sublingual spaces equal in both sides. Fine Needle Aspiration biopsy (FNA) of the cyst was also performed, but it was non-diagnostic. A tentative diagnosis of large midline dermoid cyst lying above the mylohyoid muscle was made.

Surgical excision of the lesion was performed via an intraoral transverse incision of the overlying mucosa, between the ducts of the submandibular glands. With a combination of sharp and blunt dissection the cystic wall was separated from adjacent structures and mobilized, taking special care to preserve the lingual artery and nerve on both sides. At one point during dissection, the cystic wall was perforated, releasing a cheese-like, purulent exudate the lesion was delivered through the intraoral incision with meticulous hemostasis of the resultant traumatic surface, followed by closure of the wound with sparse single stitches. The specimen was sent for pathology evaluation. The patient had an uneventful postoperative course with immediate improvement of speech and swallowing, and discharged home on the third postoperative day.

Histopathology revealed a cystic cavity lined by stratified epithelium with a distinct granular layer, containing lamellated keratin and showing no calcification. The presence of hair adnexa in the cystic wall was noted, and there were foci of chronic inflammation.

Discussion

An intraoral dermoid cyst grows slowly, and many of the patients appear when symptoms like aesthetic appearance issues (double chin), speech and voice alteration, and difficulties in swallowing and in breathing become apparent. Cysts may enlarge to a limit that can pose a critical risk to the airway, requiring immediate surgical intervention [10].

Differential diagnosis of cystic lesions in the floor of mouth and midline neck includes ranulas (simple or plunging), dermoid and epidermoid cysts, thyroglossal duct cyst, branchial cleft cysts, cystic hygroma, cystic lymphangioma, inflammatory processes infectious and noninfectious like cellulitis/abscess, Lundwig’s angina, submandibular duct obstruction (stenosis/calculi), vascular malformations like high flow arteriovenous malformations or low flow hemangiomas and lymphangiomas, heterotopic gastrointestinal cyst (HGIC) and benign neoplasms like lipomas or malignant neoplasms of the mucosa or salivary glands like squamous cell carcinomas, lymphomas and salivary gland tumors [1,13].
The diagnostic investigation for a suspected dermoid cyst includes aspiration (FNA) and imaging modalities like Ultrasonography and particularly CT scan and/or MRI [14]. Aspiration produces thick creamy cheese-like fluid, material, that may be subjected to bacteriological culture. Ultrasonography should be the examination of first choice in characterizing neck masses, as it is inexpensive, noninvasive, and accurate in experienced hands. It helps in differentiating solid from cystic lesions, enables aspiration biopsies, and helps in characterization of cyst contents and intraluminal excrescences as also may distinguish and evaluate blood vessels and vascularity of masses in head and neck region. CT and MRI allow precise localization of the lesion in relation to regional anatomic structures, thus allowing the surgeon to accurately plan appropriate surgical approaches [14,15].

Surgical enucleation is the only effective treatment and the recurrence is rare. Various surgical approaches have been described, dependent on the position of the lesion relative to the geniohyoid and mylohyoid muscle. There are two main approaches, the intraoral and the extra oral, for large dermoid cysts resection. Longo., et al. [11] recommended the intraoral approach with a transverse incision between the submandibular gland’s ducts orifices or a midline incision extending from the floor of the mouth to the tip of the tongue for the treatment of large lesions presenting above the mylohyoid muscle. They can be effectively utilized in large, deeply seated, non-infected lesions, leading to a very good cosmetic and functional result free of complications. Alternatively, an extraoral approach through a transcervical incision and blunt and sharp dissection through the muscles, geniohyoid and/or mylohyoid, is utilized for very large dermoid cysts involving simultaneously the floor of the mouth and the submental space, reaching occasionally the hyoid bone and in cases of severe infection that compromise patient’s airway [16]. Recently an intraoral endoscopic minimally invasive approach has been described to be safe and effective by Kim J.P., et al. [17] but there are limited series of case so far in order to extract safe conclusions and definitely that approach in not recommended for large lesions that can pose a risk for the patient’s airway.

In the case described above, the cyst was enucleated through an intraoral approach and a transverse incision to the overlying mucosa through the external orifice of the cystic wall. The decision was made upon cysts' size and location, patients' symptoms and imaging evaluation. Our experience from other cases as well as the relevant literature, support the notion of dissecting the entire cystic wall from adjacent structures without perforating it as this facilitates complete removal of the lesion and decreases the possibility of future recurrences.

**Figure 1a:** Dermoid cyst wall covered by keratinised stratified squamous cell epithelium. The underlying connective tissue shows inflammatory infiltration with the presence of multinucleated giant cells due to cyst wall disruption (H&Ex5).
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**Figure 1b:** Dermoid cyst wall with stratum corneum and the presence of a sebaceous gland (H&Ex5).

**Figure 2:** CT Scan of Head and Neck area with i.v contrast, indicating the extend of the cyst all over the floor of the mouth.

Figure 3: View of the enlarged and elevated overlying mucosa of the floor of the mouth.

Figure 4: Immediate exposure of the underlying cystic wall after a transverse incision between the ducts of submandibular glands.

Figure 5: Cyst is dissected separating from the adjacent tissues.

Figure 6: Cyst completely enucleated, wall ruptured during final steps of dissection, this allowed easy access to its posterior wall making dissection and separation from the tissues easier.
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