MEGA JOURNAL OF CASE REPORTS

OPEN ACCESS

Case Report

Compiled Date: May 10, 2024

A Case of Sublingual Dermoid Cyst Causing a Diagnostic Challenge

Eiji Mitate[1*], Yasuhisa Sawai[1], Taichi Demura[1], Youta Yamauchi[1,2], Miho Hasumoto[1], Satoshi Wada[1], Hiroyuki Nakano[1], Noboru Demura[1]

[1]Department of Oral & Maxillofacial Surgery, Faculty of Medicine, Kanazawa Medical University, Japan

[2]Department of Oral & Maxillofacial Surgery, JA Toyama Kouseiren Takaoka Hospital, Japan

[*]**Corresponding author:** Eiji Mitate, Department of Oral & Maxillofacial Surgery, Faculty of Medicine, Kanazawa Medical University, 1-1, Daigaku, Uchinada, Kahoku, Ishikawa 920-0293, Japan, Tel: +81-76-281-3511; Fax: +81-76-281-3511

Abstract

This case report discusses the presentation and diagnosis of a sublingual dermoid cyst, which initially posed a diagnostic challenge due to its resemblance to a ranula. Dermoid and epidermoid cysts typically result from ectodermal tissue entrapment or epithelial tissue infiltration during embryonic development or trauma. Ranulae, on the other hand, are mucous cysts originating from minor salivary glands. The patient, a teenage girl, presented with a midline floor of the mouth swelling, initially diagnosed as a mucous cyst. Despite failed treatments, the patient opted for intraoral extraction to prevent scarring. Physical examination revealed a smooth, elastic swelling without tenderness, and imaging indicated a midline lesion with muscle displacement. Further investigation, including fine needle aspiration, identified solid keratinous material, leading to a revised diagnosis of a dermoid cyst. Surgical excision successfully removed the lesion, restoring normal tongue position without complications. The histopathological examination confirmed the dermoid cyst diagnosis. The case highlights the importance of careful evaluation and aspiration of cystic contents for accurate diagnosis, especially in distinguishing between dermoid cysts and ranulae, where confirmation of aspirated material (keratinous or liquid) and amylase quantification are crucial.

Keywords: Dermoid cyst; Diagnosis; Differential; Floor of mouth; Sublingual; Fine needle aspiration cytology

Introduction

Dermoid cysts, including epidermoid cysts, are believed to originate from ectodermal tissue entrapment during the embryonic period or from infiltration of epithelial tissue due to trauma or inflammation. However, ranulae are considered to be mucous cysts derived from the minor salivary glands of the sublingual or floor of the mouth. Despite the commonality of painless swelling, dermoid and epidermoid cysts tend to occur in the midline of the floor of the mouth, with yellowish cheesy or cottage cheese-like contents. In contrast, the ranula predominantly appears unilaterally with liquid contents. In this report, we present a case of a sublingual dermoid cyst that posed a diagnostic challenge in differentiating it from a ranula.

Case Presentation

The patient, a female in her late teens, first noticed swelling in the midline of the floor of her mouth and the sublingual region approximately one year before her initial visit to my department. The patient sought consultation with a nearby otolaryngologist, and a transparent viscous fluid was aspirated through a puncture from the sublingual region. The clinical diagnosis was a mucous cyst, which led to referral to the otolaryngology department of a nearby general hospital. Despite percutaneous aspiration and six sessions of local injection of OK-432, swelling did not reduce. Surgical excision through an extraoral approach was recommended; however, the patient preferred intraoral extraction to avoid skin scarring. Thus, she referred to my department.

The patient's height was 161 cm, and weight was about 70 kg. Her nutritional status was good. There were no notable families or medical histories.

A palpable swelling, approximately the size of a chicken egg, was observed in the midline of the lower jaw **(Figure 1A)**. The swelling on the floor of the mouth had a smooth surface with no redness or warmth **(Figure 1B)**. It was elastic and slightly firm but not tender. No fluctuation was observed. Because of swelling, the tongue was posteriorly displaced, although there were no complaints of difficulty breathing, articulation disorders, or swallowing difficulties. At this point, I thought that the slightly firm consistency was attributed to the previous injections of OK-432.

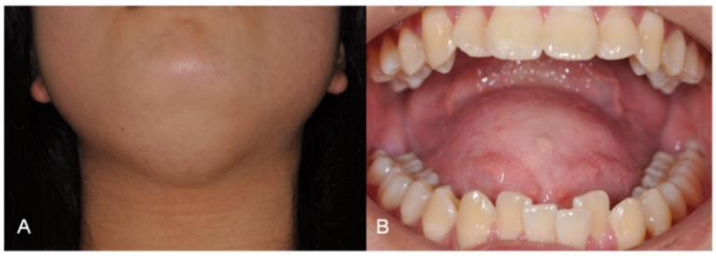

Figure 1: Initial medical examination.

(A) – The submental area is swollen.

(B) – The tongue is compressed backwards.

A computed tomography was performed for internal evaluation, which revealed a lesion in the middle line of the floor of the mouth with a uniform and slightly thickened membrane **(Figure 2)**. The genioglossus muscle was displaced downward.

Figure 2: Computed tomography. A well-defined circular area is present in the midline of the tongue.

(A) –Horizontal section

(B) – Frontal section

(C) – Sagittal section

Subsequent magnetic resonance imaging revealed a homogeneous lesion with low signal intensity on T1-weighted images **(Figure 3A-C)** and high signal intensity on T2-weighted images on the floor of the mouth **(Figure 3D and 3E)**. There was no clear extension below the genioglossus muscle, and the presentation was somewhat asymmetrical. The base of the tongue was posteriorly displaced. Coronal sections revealed separation of the tongue muscles, a finding less characteristic of the ranula. Having doubts about the diagnosis of ranula on imaging basis, fine needle aspiration was performed to reassess the diagnosis. However, no liquid component was aspirated; instead, a solid white keratinous material was collected. The aspiration cytology results confirmed the presence of keratinous material.

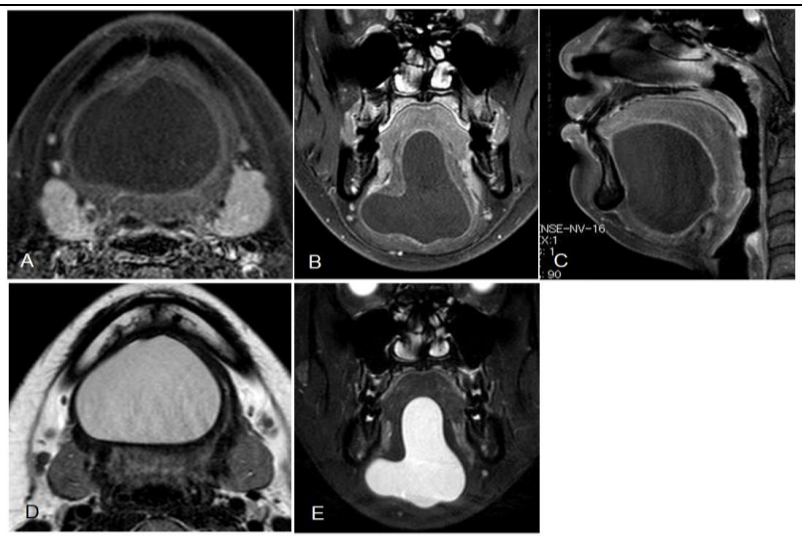

Figure 3: Magnetic resonance imaging

An internal homogeneous lesion is seen at the base of the mouth. It is low-signal on T1-weighted images (A, B, C) and high-signal on T2-weighted images (D, E). There is no extension below the hyoid muscle. The left-right morphology is slightly asymmetrical. The tongue is compressed posteriorly

(A, D) – Horizontal section

(B, E) – Frontal section

(C) – Sagittal section

Considering the location of the lesion in the midline separating the tongue muscles and the keratinous content, the clinical diagnosis was revised from ranula to dermoid or epidermoid cyst. A surgical excision was performed, and the lesion, located more toward the side of the oral cavity than the genioglossus muscle, was removed through an incision in the midline of the floor of the mouth **(Figure 4A)**. The excised specimen measured 70×45×40 mm weighed 61 g **(Figure 4B)**, and contained white keratinous material with no liquid component **(Figure 4C)**. Histopathological examination revealed a cyst wall composed of a stratified squamous epithelium with sebaceous glands **(Figure 4D)**. The final diagnosis was dermoid cyst.

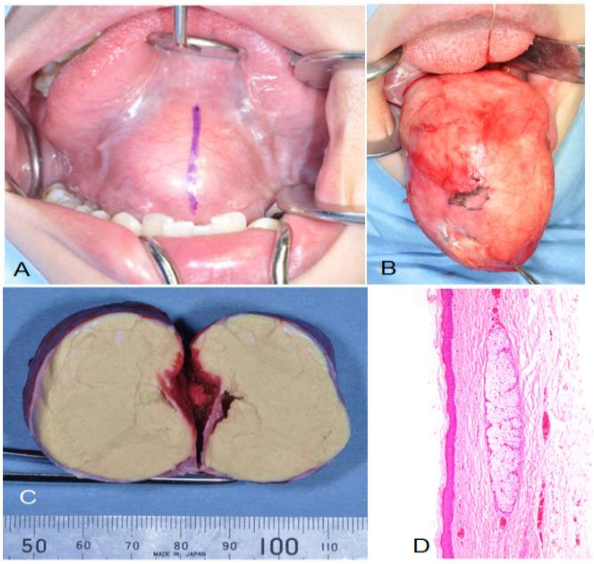

Figure 4: Intra-operative and pathological findings

(A) – The incision was made in the midline of the sublingual surface.

(B) – At the time of removal of the lesion.

(C) – Cross-section of the excised specimen. There was no moisture, only a white-grey muddy substance.

(D) – Histopathology image (Hematoxylin-Eosin Stain). The cyst wall consists of stratified squamous epithelium. Sebaceous glands are also present in the connective tissue.

Postoperatively, the tongue returned to its normal position within the lower dental arch, and there were no complications such as scarring or restriction of tongue movement.

Discussion

Dermoid cysts in oral floor lesion are very rare. About 7% of all dermoid cysts found in the body are formed in the head and neck region, with only a slight possibility of occurring within the oral cavity at approximately 1.6% [1]. Dermoid and epidermoid cysts occurred less than 0.01% of all oral cavity cysts [2]. King et al. [3], reported that out of 1,007 cases of pediatric head and neck tumors, 9.4% were reported to be dermoid cysts, with only 0.3% occurring within the oral cavity. Cramer et al. [4], reported a case that was diagnosed the swelling at the floor of the mouth as a lymphatic malformation initially; however, through preoperative Fine-Needle Aspiration (FNA), they obtained a diagnosis of a dermoid cyst, leading to subsequent surgical intervention. Our case was considered a ranula until the patient sought consultation at our department. Delayed FNA due to the previous aspiration and administration of OK-432 led to continued consideration of ranula even in our department. Regarding the site of occurrence, 82.8% of the floor of the mouth dermoid cysts is reported to be in the midline [5]. Although cases of unilateral dermoid cysts have been reported, leading to suspicion of ranula, in the literature, there are no cases in which ranula was observed in the midline. In terms of contents, dermoid cyst contents are typically described as cheesy or sludge-like. However, it was crucial to confirm whether the aspirated material was keratinous or liquid, and if liquid, amylase quantification could help differentiate between dermoid cysts and ranula.

Conclusion

In conclusion, when examining a patient with a history of previous treatments, it is essential to collect information from the referring physician. However, blind acceptance of these details without considering the possibility of discrepancies between the previous diagnosis and current clinical findings may require additional examinations. A comprehensive approach, combining all available information, is vital for an accurate diagnosis.

References

1. 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):1560-5.
2. Etarbi MS, Aishaish H, Khalifa O. Large Sublingual cyst in the floor of the mouth and submental space. Otolaryngol. 2017;7:287-91.
3. 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):567-76.
4. H Cramer, H Lampe, P Downing. Dermoid cyst of the floor of the mouth diagnosed by fine needle aspiration cytology: a case report. Acta Cytol. 1996;40(2):319-26.
5. Yutaka Konayashi, Koji Kino, Hirokazu Manita, Riri Ssatou, Kiyoshi Kikuchi, Hidemi Yoshimatsu, et al. Clinical Observation of Dermoid Cyst and Epidermoid Cyst in the Oral and Maxillofacial Region. J Jpn Stomatol Soc. 1998;47(1):101-107.

Citation of this Article

Mitate E, Sawai Y, Demura T, Yamauchi Y, Hasumoto M, Wada S, Nakano H, Demura N. A Case of Sublingual Dermoid Cyst Causing a Diagnostic Challenge. Mega J Case Rep. 2024;7(5):2001-2006.

Copyright