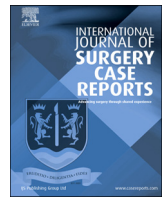




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A rare case of dermoid cyst arising in the upper lip

Yusuke Matsuzaki*, Hiroshi Kato, Shuji Yoshida, Akira Watanabe, Masayuki Takano

Department of Oral and Maxillofacial Surgery, Tokyo Dental College, 2-9-18 Kanda-Misakicho, Chiyoda-ku, Tokyo 101-0061, Japan

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ABSTRACT

INTRODUCTION: Generally, dermoid cysts commonly arise from the anus and ovaries. Oral and maxillofacial lesions are most commonly observed in the midline of the floor of the mouth, and lesions arising from the upper lip are relatively rare.

PRESENTATION OF CASE: A 50-year-old man was referred to our hospital due to swelling of the left upper lip. Clinical examination revealed an elevated alar base, Gerber protrusion and nasal deformity. Ultrasonography revealed a clearly defined 30-mm lesion with more hypoechoic bands. Meanwhile, magnetic resonance imaging revealed a 30-mm mass below the orbicularis oris of the left upper lip. The lesion had a homogeneous, low-signal intensity on T1-weighted imaging. Thus, based on these findings, a dermoid cyst was suspected. The lesion was then removed en bloc without the overlying skin while the patient was under general anaesthesia. Histopathological examination revealed a cystic cavity lined by an orthokeratinised stratified squamous epithelium. However, skin appendages were not found. Based on the clinical and histopathological features of the lesion, a diagnosis of epidermoid cyst was made. Swelling of the left upper lip, nasal deformity and Gerber protrusion significantly improved after surgery.

CONCLUSION: Although dermoid cysts arising from the upper lip are rare, the diagnostic accuracy for dermoid cyst can be improved with the combined use of ultrasonography and other imaging modalities even though these lesions are difficult to distinguish from differential diagnosis.

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1. Introduction

Dermoid cysts, which are usually congenital, are believed to arise from the entrapment of ectodermal tissue, and they may also be caused by the entrapment of epithelial debris due to trauma, inflammation or surgery [1,2]. Generally, these cysts commonly arise from the anus and ovaries. In the oral region, the floor of the mouth is the most common site, and dermoid cysts arising from the upper lip are relatively rare [3]. Herein, we present a case of an epidermoid cyst arising from the upper lip.

This work has been reported in line with the SCARE criteria (Agha) [4].

2. Case presentation

A 50-year-old man was referred to our hospital due to swelling of the left upper lip. The patient had been aware of the condition for quite some time. However, he did not mind until it gradually worsened about a year back. Clinical examination revealed an elevated alar base, nasal deformity and Gerber protrusion at the left side of the upper lip. An elastic hard, painless and mobile subcutaneous lesion was found under the upper lip. In the oral cavity, a smooth

surface and clear boundary were observed under the left upper lip mucosa (Fig. 1A–C). A panoramic radiograph revealed no abnormal findings (Fig. 2A). Ultrasonography showed a clearly defined 30-mm lesion with more hypoechoic bands (Fig. 2B), and posterior enhancement and a fine echogenic spot were observed within the lesion. Magnetic resonance imaging (MRI) revealed a 30-mm lesion located under the orbicularis oris of the left upper lip. Moreover, the lesion had a sharp margin, with a homogenous low-signal intensity on T1-weighted imaging. In addition, the lesion exhibited a homogeneous high-signal intensity on T2-weighted imaging and short TI inversion recovery (Fig. 2C). No obvious bone resorption was observed. Based on these findings, we believed that the lesion was a dermoid cyst, and surgery was then performed while the patient was under general anaesthesia. The mucous membrane was incised just above the lesion intraorally, and the lesion capsule was removed without the surround tissues (Fig. 3A). We excised a part of the orbicularis oris and the levator labii superioris because the lesion capsule had conglutinated to these muscles, and the detached levator labii superioris was sutured to the orbicularis oris. The resected specimen was yellowish-white in color, and its size was about 30 × 25 × 10 mm. The cystic lesion contained a white, cheesy material (Fig. 3B). Histopathological examination revealed a cystic cavity lined by an orthokeratinised stratified squamous epithelium. However, no skin appendages were observed. Based on the clinical and histopathological findings, a diagnosis of epidermoid cyst was made (Fig. 4A, B). Swelling of the left upper lip,

* Corresponding author.

E-mail address: matsuzakiyusuke@tdc.ac.jp (Y. Matsuzaki).

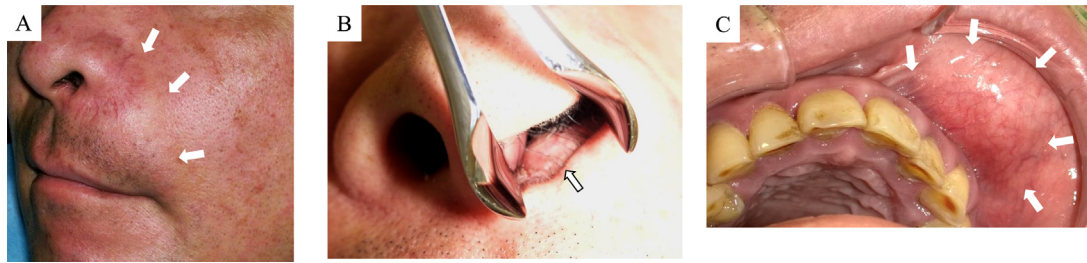


Fig. 1. At the first visit to our department, the imaging findings revealed an elevated alar base, Gerber protrusion and swelling below the mucosa of the left upper lip (white arrows).

- (A) Lateral view.
- (B) Intranasal view.
- (C) Intraoral view.

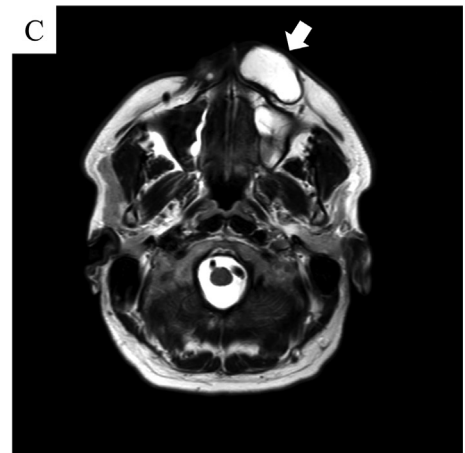
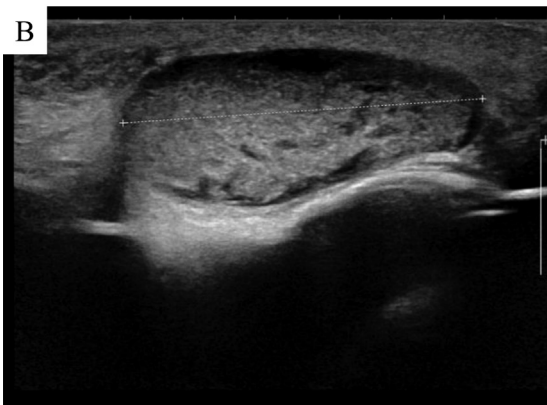
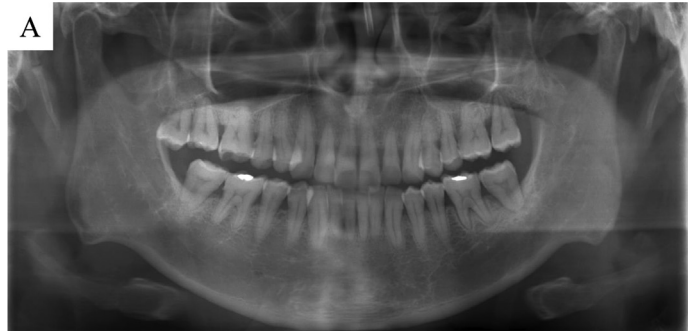


Fig. 2. (A) Panoramic radiography did not reveal abnormal findings.
 (B) Ultrasonography revealed a clearly defined 30-mm lesion with more hypoechoic bands.
 (C) Axial brain magnetic resonance imaging (T2-weighted image) revealed a 30-mm lesion located below the orbicularis oris of the left upper lip (white arrow).



Fig. 3. Intraoperative findings.
 (A) The mucous membrane was incised just above the lesion in the oral cavity.
 (B) The resected specimen was yellowish-white in color, and its size was about 30 × 25 × 10 mm. The cystic lesion contained a white, cheesy material.

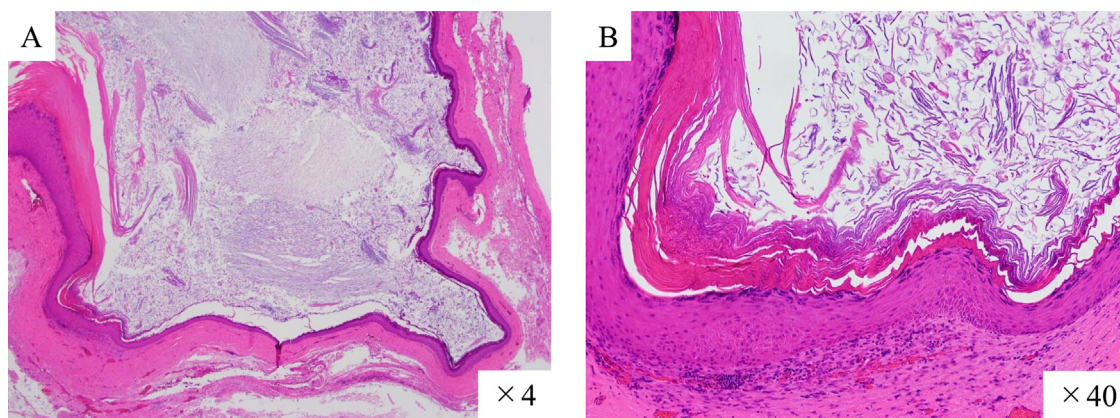


Fig. 4. Histopathological findings.

(A) The cystic cavity was lined by an orthokeratinised stratified squamous epithelium (A: haematoxylin and eosin staining, $\times 4$).

(B) The connective tissue wall had nerve and smooth muscle tissues. However, no skin appendages were observed (B: haematoxylin and eosin staining, $\times 40$).

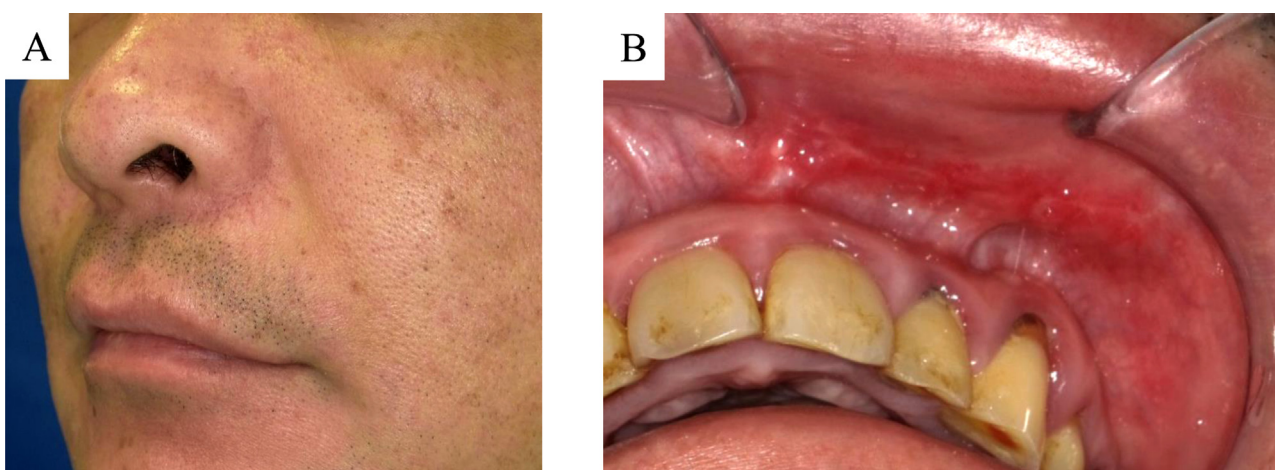


Fig. 5. Image of the facial appearance and oral cavity 1 year after surgery.

(A) Lateral view.

(B) Intraoral view.

nasal deformity and Gerber protrusion significantly improved after surgery, and no recurrence was observed after 1 year of follow-up (Fig. 5A, B).

3. Discussion

Dermoid cysts have been histopathologically classified as epidermoid, dermoid and teratoid cysts. The epidermoid cyst, as in the current case, is lined with a simple squamous epithelium and connective tissue, and the dermoid cyst is an epithelium-lined cyst containing skin appendages. The teratoid cyst is also an epithelium-lined cyst containing mesodermal or endodermal elements, such as the muscle, bone, teeth and mucous membranes [1,2]. Dermoid cysts, which are congenital, are caused by the entrapment of ectodermal tissues during midline closure of the first and second branchial arches [5] and the abnormal burial of a lateral cervical

cyst or the residual tuberculum impar [6]. Moreover, they can be attributed to the entrapment of epithelial debris due to trauma, inflammation or surgery and epidermal atheromas [7]. The lesions are commonly observed in individuals aged between 15 and 35 years [1]. However, there is no gender predilection.

Generally, dermoid cysts commonly arise from the anus and ovaries. Oral and maxillofacial lesions are most commonly observed in the midline of the floor of the mouth, and lesions arising from the upper lip are relatively rare. Nearly 34% of dermoid cysts are found in the head and neck, of which 6.5% are located in the floor of the mouth [8,9]. Seven cases of dermoid cysts arising from the lip have been reported in previous studies, thereby underscoring the rarity of arising from this site (Table 1) [3,10–15]. These cysts are observed not only in young but also in middle-aged individuals. In this case, the patient was aged 50 years. Due to the absence of any previous history of trauma or surgery, the cyst was likely con-

Table 1

Summary of cases of previously reported Dermoid cyst arising in lip.

Author	Year	Sex	Old	Location	Diagnosis	Size
Wang WC et al. [10]	2005	M	13	Left lower lip	Epidermoid	10 mm
Kormanik KE et al. [11]	2006	M	3-month	Mid upper lip	Dermoid	20 mm
Toriyama et al. [12]	2008	F	6-month	Mid upper lip	Dermoid	8 mm
Herlin et al. [13]	2011	M	47	Upper lip on both sides	Dermoid	18 mm, 7 mm
Phukan JP et al. [14]	2014	M	52	Right upper lip	Epidermoid	25 mm
Dogan F et al. [15]	2014	F	3-month	Mid upper lip	Epidermoid	20 mm
Mahalaskshmi et al. [3]	2016	M	38	Left lower lip	Epidermoid	20 mm

genital and caused by the gradual increase in the entrapment of ectodermal tissues during the closure of the maxillary prominence (MxPs), the medial nasal prominence (MNP) and the lateral nasal prominence (LNP). Union of the facial prominences occurs between the sixth and the eighth week of development. The lateral growth of the MxP pushes the nasal pits toward the middle face region. The ventral tip of the MNP extends further rostrally and ventrolaterally to begin fusion with both the LNP and the MxP at the medial end of their boundary junction. During fusion of these processes, apoptosis of epithelial cells occurs in the seam of the contacting MNP and the LNP, resulting in the connection of mesenchymal cell compartments. By the eleven weeks of development, the nostrils and lip are correctly formed by the completion of fusion of the MNP, LNP and MxP [16]. According to literature data, dermoids that arising in the midline of the upper lip are found in childhood. On the other hand, the dermoids that arising in the side of the lip are treated at a relatively middle age. As a result, poor apoptosis of epithelial cells occurs in the seam of the contacting MNP and LNP, and then fusion of the MxP. Therefore, it is thought that finding of the dermoid cysts was delayed because the epithelial remnant remained deeper than that of the midline of the upper lip. The mass had progressed just below the subcutaneous tissue in the external nose. However, there were no obvious surgical findings of adhesion of the capsule to the subcutaneous tissue. Therefore, combined resection of the skin was not performed.

Benign neoplastic conditions that may arise from this region include salivary adenomas, vascular lesions, abscesses, fibromas and lipomas [3]. MRI is considered the gold standard for diagnosis [8,17]. Dermoid cysts have different signal intensities on T1-weighted image and may be hyperintense (due to the presence of sebaceous lipids) or isointense relative to the muscle. They are usually hyperintense on T2-weighted image. Previous studies have shown that the lesion has a clear demarcated rim and commonly has a heterogeneous internal appearance [17]. In this case, ultrasonography revealed highly echogenic intracystic foci, which were attributed to the presence of keratin and sebaceous materials. This finding is consistent with the ultrasonography findings of dermoid cysts [18]. However, in some cases reports, the lesions were difficult to distinguish from lipomas [19]. Thus, the combined use of ultrasonography and other imaging modalities may improve diagnostic accuracy [8].

The treatment of dermoid cysts is surgical excision with the use of either the intraoral or extraoral approach based on the location of the dermoid cysts [20]. In all previous case reports, surgical excision was performed, and no recurrence was observed. However, about 5% of dermoid cysts progress to squamous cell carcinoma. Thus, continuous follow-up may be important [21]. In the current case, the levator labii superioris was sutured to the orbicularis oris, because the mass was just below the orbicularis oris, and removing it caused the orbicularis oris and the levator labii superioris to rupture. And, to prevent an increase in the alar width, pulled both nose inward using elastic surgical tape. Those treatment induced the alar base inward and the nasal deformation improved, without postoperative dysfunction. Based on a previous study, the prognosis after surgery is good, and the occurrence of postoperative complications is low [22].

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The authors have no sources of funding for research.

Ethical approval

A case report does not require approval from the Ethics Committee in our hospital.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

MT, HK and YM performed surgery. YM drafted the manuscript. HK and MT participated in the correction of the work. KO, SY and AW were responsible for perioperative management. YA and KM was responsible for pathological diagnosis.

Registration of research studies

No research study involved in this case report. Not applicable.

Guarantor

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Provenance and peer review

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Declaration of Competing Interest

The authors have no conflict of interest to declare.

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