

Epidermoid Cyst of the Buccal Mucosa Diagnosed by Magnetic Resonance Imaging and Ultrasonography: A Case Report and Review of the Literature

Dai Ichikawa¹, Seigo Ohba^{1,2}, Hitoshi Yoshimura¹, Shinpei Matsuda¹, Yoshiaki Imamura³, Kazuo Sano¹

¹Division of Dentistry and Oral Surgery, Department of Sensory and Locomotor Medicine, Faculty of Medical Sciences, University of Fukui. ²Department of Regenerative Oral Surgery, Nagasaki University Graduate School of Biomedical Science. ³Division of Surgical Pathology, University of Fukui Hospital

Abstract

Objective: Epidermoid cysts are rare lesions in the oral cavity. Intraoral epidermoid cysts are most commonly located in the floor of the mouth; they are less common in the lips and buccal mucosa. Cysts arising from the buccal mucosa are often difficult to distinguish from other lesions. We herein report a case of epidermoid cyst in the buccal mucosa diagnosed with imaging.

Methods: A 65-year-old Japanese man presented with a painless soft mass in the left buccal mucosa. MRI and ultrasonography were used to diagnose the mass correctly.

Results: The well-circumscribed mass was covered with healthy buccal mucosa. The lesion showed low intensity in T1- and high intensity in T2-weighted magnetic resonance imaging. The intensity did not decrease in fat-suppressed T2-weighted images. The interior of the lesion was homogeneous. Ultrasonography revealed a well circumscribed lesion with a heterogeneous interior. Surgical excision via an intraoral approach was performed under general anesthesia. Histopathology revealed that the cyst wall was lined with stratified squamous epithelium. The cystic cavity was filled with laminated keratin. Skin adnexa were not observed in the cyst wall. Based on histopathological findings, the lesion was diagnosed as an epidermoid cyst. No recurrence or other adverse events were observed in 6-month follow-up.

Conclusion: The combination of MRI and US can be useful to diagnose dermoid cysts and epidermoid cysts.

Key Words: Epidermoid cyst, Magnetic resonance imaging, Ultrasonography

Introduction

Epidermoid and dermoid cysts are derived from ectodermal tissue. Cysts containing skin adnexa in the cyst wall are termed dermoid cysts and those not containing adnexa are termed epidermoid cysts [1]. Both congenital and acquired etiologies have been proposed for the development of (epi)dermoid cysts. According to the theory of congenital development, ectodermal elements migrate into the facial midline where the first and second branchial arches fuse [2]. According to the theory of acquired development, the epidermis migrates into the deep tissue as a result of a physical trigger such as trauma and develops into an (epi)dermoid cyst [2].

Epidermoid cysts can occur anywhere in the body, and are most common in the ovary and testicle [3]. Only 1.6% of epidermoid cysts occur in the oral cavity [4]. Intraoral epidermoid cysts are most commonly observed in the floor of the mouth and are seldom found in the lips or buccal mucosa [5].

Various imaging methods may be used to diagnose epidermoid cysts. Magnetic resonance imaging (MRI) is the most useful method for diagnosis. The lesion appears as a well-circumscribed mass on MRI. The signal intensity of epidermoid cysts is low in T1-weighted images and high in T2-weighted images [6]. The intensity of the cyst interior is usually uniform. Computed tomography also shows a uniform cyst interior [7]. Ultrasonography (US) reveals a well-circumscribed, smooth mass with a heterogeneous interior [8].

Surgical removal is a common treatment for epidermoid cysts [9]. Cysts recur in less than 3% of cases [10,11]. Complete removal of the cyst wall is essential to avoid recurrence [6].

It is often difficult to differentiate lesions arising from the buccal mucosa; therefore, correct diagnosis is essential to plan adequate treatment. We herein report a case of epidermoid cyst in the buccal mucosa correctly diagnosed with a combination of MRI and US.

Case Presentation

A 65-year-old Japanese man was referred to the Department of Dentistry and Oral Surgery at the University of Hospital. He had noticed a painless swelling in the left buccal mucosa for 10 years. The patient had a prior history of anal fistula surgery 2 years previously and bladder diverticulum surgery 1 year previously. He had been receiving chemotherapy for gastric

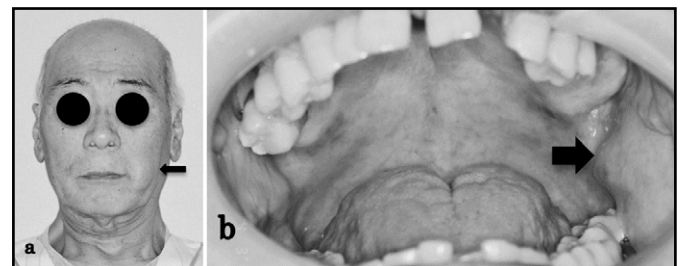


Figure 1. Preoperative facial and intraoral appearance. a) A well-circumscribed painless soft mass is visible in the left cheek (arrow). b) The soft, movable mass in the left cheek was covered with healthy mucosa (arrow).

Corresponding author: Seigo Ohba, DDS, PhD, Department of Regenerative Oral Surgery, Nagasaki University Graduate School of Biomedical Science, 1-7-1 Sakamoto, Nagasaki 852-8588, Japan, Tel: +81 95 819 7704; Fax: +81 95 819 7705; e-mail: sohba@nagasaki-u.ac.jp/seigoohba@gmail.com

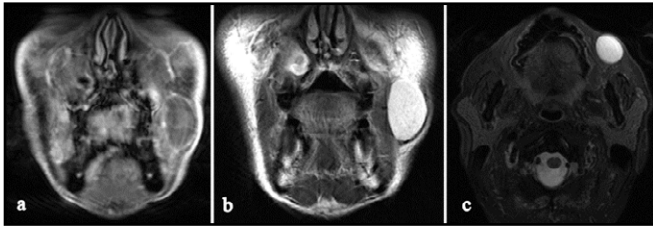


Figure 2. MRI findings. Coronal sections are shown in a and b; axial sections are shown in c. The intensity of the lesion was slightly lower than that of muscle in T1-weighted image (a) and the same level as that of adipose tissue in T2-weighted image (b). The density of the mass did not decrease in fat-suppressed T2-weighted images (c). The interior of the lesion was homogeneous.

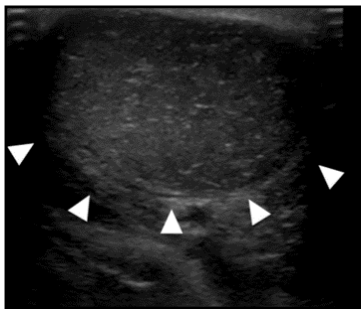


Figure 3. Ultrasound findings. A well-circumscribed mass with a heterogeneous interior is seen (arrowheads).

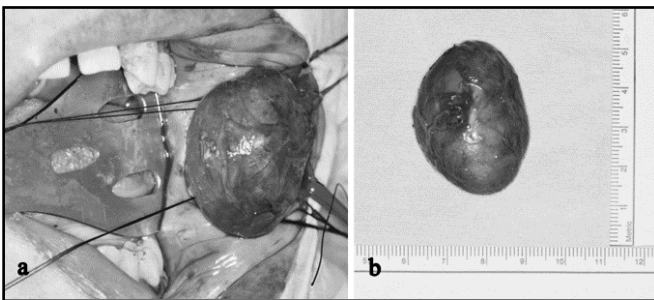


Figure 4. Intraoperative findings. The ovoid lesion was enucleated en bloc without damage. The 40 × 30 × 20-mm mass was soft and had a smooth surface.

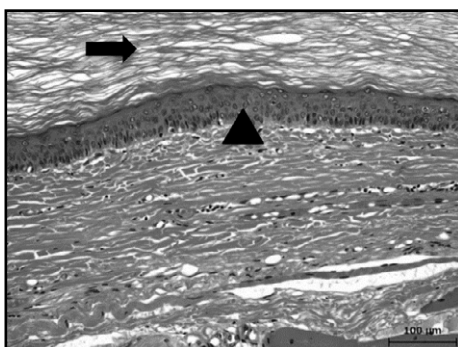


Figure 5. Histopathology. The cyst wall consists of stratified squamous epithelium (arrowheads). The cystic cavity is filled with laminated keratin (arrows).

cancer over the previous year. The patient's grandfather had also suffered from gastric cancer.

A 35×30 mm painless, soft, well-circumscribed mass was observed in the patient's left buccal mucosa (Figure 1). There was no submandibular lymph node swelling or tenderness. The mass was covered with healthy mucosa and was not attached to mucosa or skin.

Imaging Examinations

MRI: Axial and coronal sections revealed an ovoid subcutaneous cystic lesion in the left buccal mucosa, approximately 35×25 mm in size (Figure 2). The mass showed low intensity in T1- and high intensity in T2-weighted images. The intensity of the mass did not decrease in fat-suppressed T2-weighted images. The interior of the lesion was homogeneous. There were no indicators of infection around the lesion. The thickness of the cyst wall was approximately 2-3 mm. There was no evidence that the lesion connected with the parotid gland. According to the findings, the mass had not been diagnosed correctly yet and there were still possible diseases including epidermoid/dermoid cyst, lymphangioma and mucocele. Therefore we performed additional imaging test, US to assess the border and inside of the mass.

US: A well-circumscribed ovoid lesion with a heterogeneous interior was seen on US (Figure 3). A connection between the lesion and the skin was suspected. Based on imaging and clinical findings, we diagnosed epidermoid cyst of the left buccal mucosa.

Progress

Surgical excision via an intraoral approach was performed under general anesthesia. A 30-mm horizontal incision was made in the left buccal mucosa parallel with the occlusal plane. The lesion was enucleated en bloc without damage (Figure 4). No recurrence was observed in 6-month follow-up.

Histopathology

Histopathology revealed that the cyst wall was lined with stratified squamous epithelium similar to epidermis (Figure 5). The cystic cavity was filled with laminated keratin. Skin adnexa were not observed in the cyst wall. Based on these findings, the lesion was histopathologically diagnosed as an epidermoid cyst.

Discussion

Epidermoid and dermoid cysts are derived from ectodermal tissue. The floor of the mouth is the most common intraoral location of these cysts. Occurrence in the buccal mucosa is rare. Only five cases of epidermoid cysts arising from the buccal mucosa have been reported in the English-language literature. Schneider et al. [12] first reported a case of epidermoid cyst of the buccal mucosa in 1978. Subsequently, Rajayogeswaran et al. [13], Ozan et al. [3] and Kini et al. [14] reported similar cases. These cases are summarized in Table 1. Lesions arising in the buccal mucosa can change facial appearance in their early stages. These lesions may also be observed easily in the oral cavity. However, small lesions in the buccal mucosa do not inhibit the oral functions of chewing, swallowing, or talking. Therefore, lesions in the buccal mucosa tend to be observed without any treatment over a long term, based on previous reports (Table 1). The patient in the present case noticed the mass in his buccal mucosa 10 years before presentation.

Several lesions present as well-circumscribed, non-attached masses in the buccal mucosa, including epidermal cyst, lipoma, lymphangioma, hemangioma, pleomorphic

Table 1. The cyst wall was lined stratified squamous epithelium in all cases. Surgeries were performed in all cases. In any case, there were not abnormal findings around the mass and no submandibular lymph nodes showed significant changes. There is no recurrence.

References	Age	Sex	Site	Duration of disease	Episode	Size (mm)	Clinical findings	Imaging findings	Histopathology
Schneider et al. (1978)	36	F	Right	unknown	none	40 × 5 × 2	mobile, soft	none	The cyst wall contained melanin.
Schneider et al. (1978)	30	F	Left	3 years ago	none	22	mobile, soft	none	The cystic cavity was filled with keratin.
Rajayogeswaran et al. (1989)	25	M	Left	1 years ago	bite	20 × 15	mobile, covered by healthy mucosa	none	The cystic cavity was filled with keratin. There were partial sebaceous glands in the cystic cavity.
Ozan et al. (2007)	38	F	Left	6 months ago	none	20 × 30 × 40	mobile, covered by healthy mucosa	none	The cystic cavity was filled with keratin.
Kini et al. (2013)	25	M	Left	2 years ago	none	15 × 15 × 15	mobile, covered by healthy mucosa	US: compressible and non vascularised	The cystic cavity was filled with keratin.
Present Case	65	M	Left	10 years ago	none	40 × 30 × 20	mobile, covered by healthy mucosa	MRI: low-density in T1- and , high-density in T2-weighted images. US: A well-circumscribed lesion was detected with heterogeneous inside.	The cystic cavity was filled with keratin.

adenoma, inflammation, and mucocele. Correct diagnosis of such lesions is important for appropriate treatment. Therefore, imaging plays an important role as a non-invasive method of diagnosis.

Epidermal cysts originate from the epidermis or from the epithelial component of infundibular hair follicles. These tissues migrate into the subcutaneous layer and fill with keratinous material, resulting in an epidermal cyst [15]. These cysts are clinically characterized as spherical masses without spontaneous pain or tenderness. While the clinical findings of the present case were similar to an epidermal cyst, epidermal cysts are commonly hard, whereas the present lesion was soft. Epidermal cysts are attached to the skin. In contrast, epidermoid cysts do not generally attach to skin [15]. The lesion in the present case was suspected to be connected with skin based on US images. However, MRI findings did not show this connection, and the mass was freely moving on palpation. Therefore, the lesion was considered not to be an epidermal cyst. No adhesion between the lesion and skin was observed at surgery.

Based on MRI findings, lipoma, lymphangioma, hemangioma, pleomorphic adenoma, inflammation, and mucocele were considered as differential diagnoses. All of these lesions have high intensity in T2-weighted images, as do epidermoid cysts.

Lipomas are soft masses with clinical behavior similar to that of epidermoid cysts. Lipoma was easily excluded in the present case because the intensity of the lesion did not decrease in fat-suppressed T2-weighted images (*Figure 2*).

Lymphangioma also presents as a well-circumscribed soft mass. On US, these lesions appear as multilocular masses with regular margins [16]. The US findings of lymphangioma are completely different from those of the present case.

Hemangioma is enhanced in contrast-enhanced MRI [17]. Moreover, feeder vessels are often found entering the lesion. In the present case, the lesion was not enhanced and no feeders were observed in contrast-enhanced MRI. Confirmation of blood flow around the lesion with Doppler US would allow more definitive exclusion.

Pleomorphic adenoma occurs commonly in the parotid gland as a well-circumscribed, spherical, soft or hard mass. Pleomorphic adenomas and epidermoid cysts can be difficult to distinguish clinically. Pleomorphic adenomas are often enhanced in contrast-enhanced MRI [18]. The lesion in the present case was not enhanced.

Inflammatory abscess is another differential diagnosis for epidermoid cyst. Signs of inflammation such as fever, redness, or tenderness around the lesion were not found when the patient initially visited our department, and he had not experienced any episodes of inflammation prior to visiting the hospital. There was no obvious source of infection around the lesion. Furthermore, blood test results were within the normal range (white blood cells: 5400 cells/ μ L, C-reactive protein: 0.12 mg/dl). Tissue surrounding an abscess is enhanced while the interior is not enhanced in contrast-enhanced MRI [19]. These MRI findings did not match those of the present case.

It was most difficult to distinguish the present case from mucocele. The buccal mucosa is a common site for mucocele

occurrence [20,21]. Mucoceles commonly present clinically as well-circumscribed, soft masses [22]. On MRI, they appear as well-circumscribed homogeneous lesions with high intensity in T2-weighted images, as do epidermoid cysts [20]. On US, the present case appeared as a well-circumscribed lesion with a heterogeneous interior. Fluid-filled lesions show anechoic findings on US [23]. Therefore, it was determined that the present lesion was not a mucocele.

It can be difficult to distinguish dermoid cysts from epidermoid cysts based on imaging findings. However, dermoid cysts usually have heterogeneous interiors on MRI [24] because of the presence of skin adnexa. In the present case, the interior of the lesion was homogeneous. Therefore, dermoid cyst was excluded from the differential list.

In general, epidermoid cysts do not transform into malignancies. However, malignant transformation has been reported [25]. Fleming and Botterell [26] reported that epidermoid cysts recur within 2 years. Based on these reports, further follow-up of the present case will be necessary.

Multiple imaging examinations often contribute to the correct diagnosis of epidermoid cysts. In the present case, the combination of MRI and US was useful in diagnosing a soft mass arising in the buccal mucosa. Of course, incisional or aspiration biopsies are necessary to determine the definitive

diagnosis of such lesions. However, biopsies damage the cyst wall, resulting in adhesion of the cyst wall to surrounding tissues. Adhesions make complete resection of lesions difficult, increasing the chance of recurrence. In the present case, clinical and imaging findings indicated a low probability of malignant tumor; epidermoid cyst and dermoid cyst were the primary differentials. Thus, biopsy was not performed, to reduce the risk of recurrence.

Conclusions

Soft masses arising in soft tissues are often difficult to differentiate by clinical findings and a single imaging modality alone. Therefore, multiple imaging examinations should be performed to correctly diagnose soft masses arising in the buccal mucosa. In particular, the combination of MRI and US can be useful to diagnose dermoid cysts and epidermoid cysts, as shown in the present case.

Consent

Authors got an informed consent form from a patient.

Competing Interests

There is no conflict of interest to declare

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