

Case of a Large Dermoid Cyst on the Floor of the Mouth Incidentally Diagnosed During Treatment of a Dentofacial Deformity

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Dermoid cyst and epidermoid cyst develop due to inflammation or fetal trauma, and dermoid cyst aberrantly include epithelial tissue. However, the relationship between the pathological tissue and lesion image in these diseases is unclear. Here, we report a rare case of a patient in whom a large dermoid cyst was incidentally detected on the floor of the mouth during treatment of a dentofacial deformity. The presence of the cyst was confirmed by computed tomography (CT) and histological examination. The 16-year-old female patient presented with a swelling extending from the floor of the mouth to the chin. On initial examination, a painless, elastic, soft swelling of the mouth floor was observed, but tongue movement, articulation, and eating were not impaired. CT revealed an oval, well-defined, cystic lesion approximately 60 × 40 × 32 mm in size. Intraoral cystectomy was performed under general anesthesia. At the 1-year follow-up, there was no sign of recurrence or occlusion. The patient's facial profile improved after postoperative orthodontic treatment.

Keywords: dermoid cyst, floor of the mouth, dentofacial deformity

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INTRODUCTION

Dermoid cysts are cystic lesions whose walls consist of epithelia and skin appendages. They develop via the invasion of ectodermal tissue that persists along the fusion line of the first and second branchial arches during fetal life or the invasion of epithelial tissue due to inflammation, external injury, or surgery [1]. Dermoid cysts can occur in different parts of the body. In a survey by New *et al.* [2], the head and neck region were involved in 6.9%, the area next to the anus in 44.5%, and the vicinity of the ovaries in 42.1% of the cases. Among the cysts in the head and neck region, ~50% occur in the orbit, 13% in the nose, and 23% on the floor of the mouth. The latter represent 1.6% of all dermoid cysts. Symptoms typically include painless swelling, but due to the cyst's slow growth, its progression largely goes unnoticed, although large cysts may result in pronunciation disorders, dysphagia, and respiratory disorders [3]. Here we report a case of a large dermoid cyst incidentally detected during treatment of a dentofacial deformity. Despite progressive enlargement of the lesion, the patient had not experienced any subjective symptoms.

CASE REPORT

A 16-year-old Japanese female came to our hospital to seek treatment for a dentofacial deformity. Despite the presence of a swelling extending from the floor of the mouth to the chin, she had no subjective symptoms (Fig. 1). Her general and systemic examinations were normal, and she had no history of trauma, surgery, or prolonged fever. An oral examination revealed a large swelling on the floor of

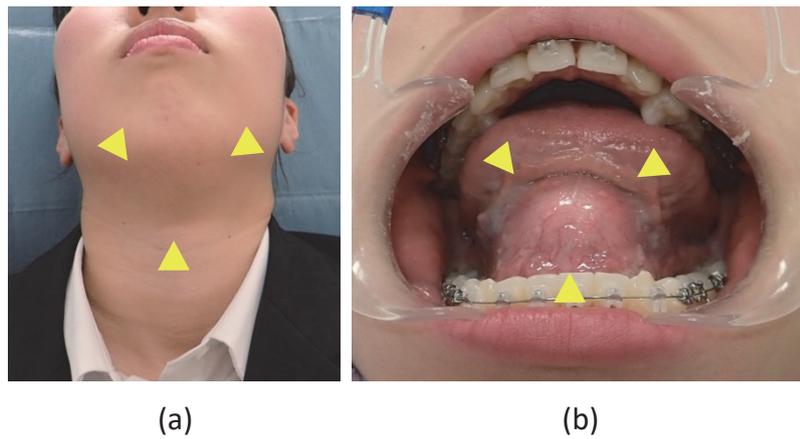


Fig.1. Preoperative neck and intraoral findings.
 (a) Submental swelling.
 (b) Sublingual swelling with a double-tongue appearance.

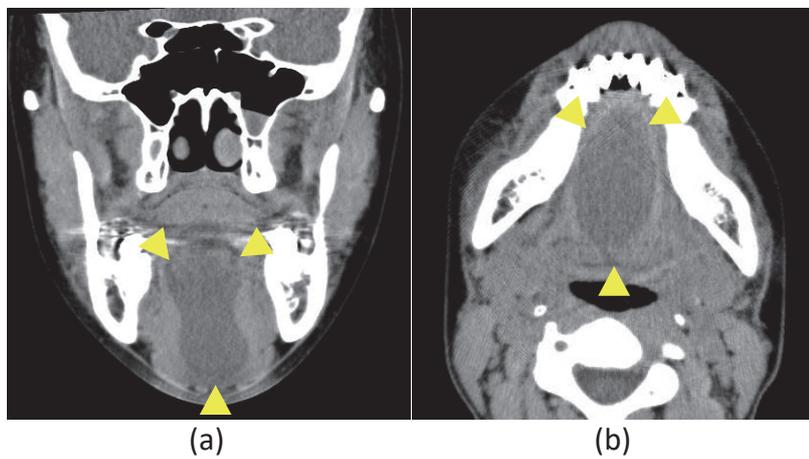


Fig. 2. Preoperative computed tomography reveals a large, oval, well-defined, cystic lesion.
 (a) Axial view.
 (b) Coronal view.

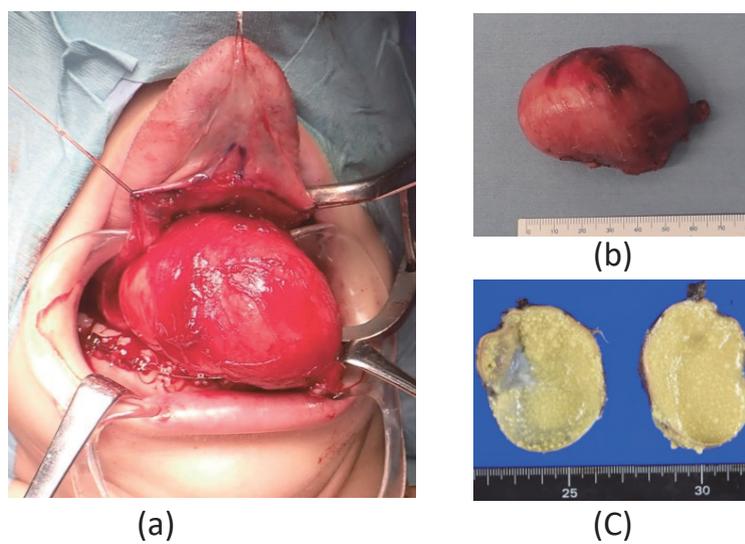


Fig. 3. Intraoperative findings.
 (a) The cyst was totally resected surgically using an intraoral approach.
 (b) Excised cyst.
 (c) Cut surface.

the mouth. The lesion was painless, soft, and elastic, but tongue movement, articulation, and eating were not impaired. Computed tomography (CT) imaging revealed the presence of a large (approximately 60 × 40 × 32 mm), oval, well-defined, cystic lesion (Fig. 2). A biopsy of the lesion revealed the extensive presence of keratin, suggestive of a dermoid cyst. The lesion was totally resected during a surgical procedure in which it was freed from the fibers of the mylohyoid muscle via sharp and blunt dissection using an intraoral approach with the patient under general anesthesia (Fig. 3). The resulting defect was closed in layers after ensuring hemostasis. Tracheal intubation and sedation were maintained to avoid the risk of postoperative airway stenosis due to swelling of the oral floor. Histopathological examination revealed a cystic lining by stratified squamous epithelium together with the presence of hair follicles, sebaceous glands, and sweat glands (Fig. 4). These features supported the diagnosis of a dermoid cyst. The postoperative course of the patient was uneventful. Eight months later, the maxilla was moved upward after Le-Fort I osteotomy to obtain a desirable amount of mandibular advancement after sagittal split ramus osteotomy. After postoperative orthodontic treatment, both the occlusion and facial profile were improved (Fig. 5). And at the 2-year follow-up, there was no sign of recurrence or occlusion (Fig. 6).

DISCUSSION

Most (> 90%) dermoid cysts that occur in the head and neck region have a maximum diameter of < 60 mm [4]. The differential diagnosis of a swelling on the floor of the mouth includes ranula, thyroglossal cyst, lipoma, hemangioma, and cystic lymphangioma [5, 6]. Magnetic resonance imaging (MRI) is useful for obtaining a diagnosis. A monocular cystic lesion typically presents as a lesion with low signal intensity in the T1-weighted images and high signal intensity in the T2-weighted images [1, 3, 4]. Most ranula and dermoid cysts are internally uniform and thus difficult to distinguish from each other, although dermoid cysts may be somewhat inhomogeneous due to the mixture of epithelial components [1, 3, 4]. Biopsy may therefore be useful to

clarify the preoperative diagnosis, although it may subsequently cause adhesion to surrounding tissues, scar formation, and recurrence [3]. In our patient, prior to dentofacial deformity surgery, she received a fixed orthodontic appliance as preoperative orthodontic treatment. As the metal artifacts precluded an MRI examination, the preoperative diagnosis of dermoid cyst was made by biopsy.

Although many cysts occur congenitally, the susceptible age is from the 10s to 20s. Cysts are often detected during puberty, when the growth rate, which was previously slow, increases, and the patients' sweat glands and sebaceous glands become functionally active. [7] In our patient, the slow growth of the lesion was such that she adapted to its presence and despite its large size, did not have subjective symptoms. In fact, the patient thought that weight gain had induced the swelling of the chin and floor of the mouth but because the changes did not interfere with daily life, she had ignored them.

Mayer's classification [7] is generally used to describe the histopathology of cysts. Dermoid cysts feature skin appendages such as hair, sebaceous glands, and sweat glands on the cyst wall, epidermoid cysts lack skin appendages, and teratoid cysts contain mesoblast and endoblast components in addition to ectodermal components, such as skin appendages, on the cyst wall.

Treatment consists of complete surgical removal via an intraoral or extraoral procedure or a combination of both depending on the preferred approach [4, 8] and the location of the cyst. According to the classifications of Bergmann [9] and Hagsaki [10], cysts located above the mylohyoid muscle are classified as sublingual, those below the mylohyoid muscle as submental, and those extending above and below the muscle as sublingual-submental. In general, the sublingual type is treated via an intraoral method, the submental type via an extraoral method, and the sublingual-submental type via a combined approach, although some surgeons advocate the use of an intraoral approach, at least initially, regardless of the location and size of the cyst. In addition, because damage to or persistence of the cyst wall can induce recurrence, perforation of the cyst wall should be avoided. A cyst with a relatively thick wall can usually be removed as a lump without per-

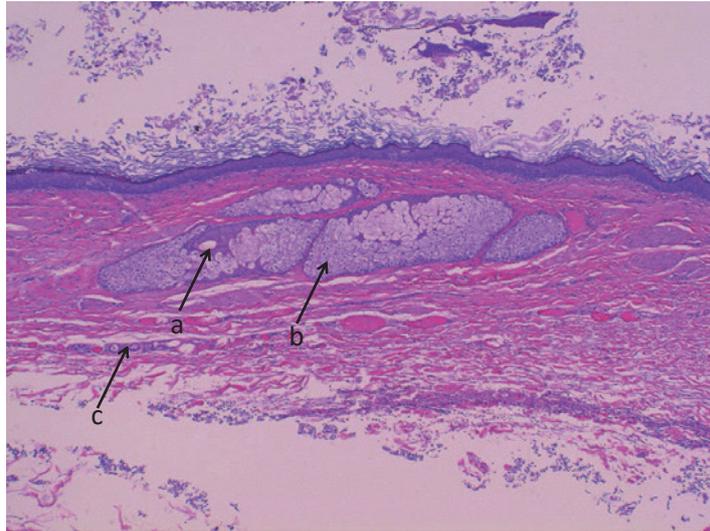


Fig. 4. Histopathological findings of the surgical specimens.

($\times 10$; hematoxylin and eosin staining)

Cystic lining with hair follicles, sebaceous glands, and sweat glands was observed during histopathological examination.

- (a) Hair follicles.
- (b) Sebaceous gland.
- (c) Sweat gland.

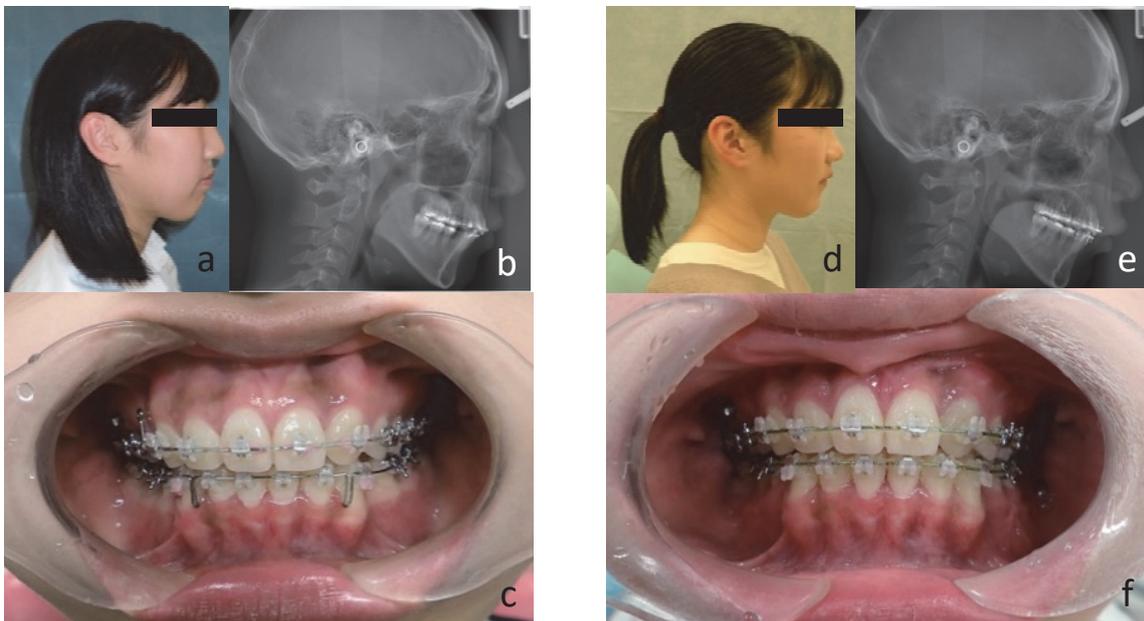


Fig. 5. Comparison of preoperative findings and postoperative results.

- (a) Preoperative facial photograph.
- (b) Preoperative cephalogram.
- (c) Preoperative intraoral findings.
- (d) Postoperative facial photograph.
- (e) Postoperative cephalogram.
- (f) Postoperative intraoral findings.

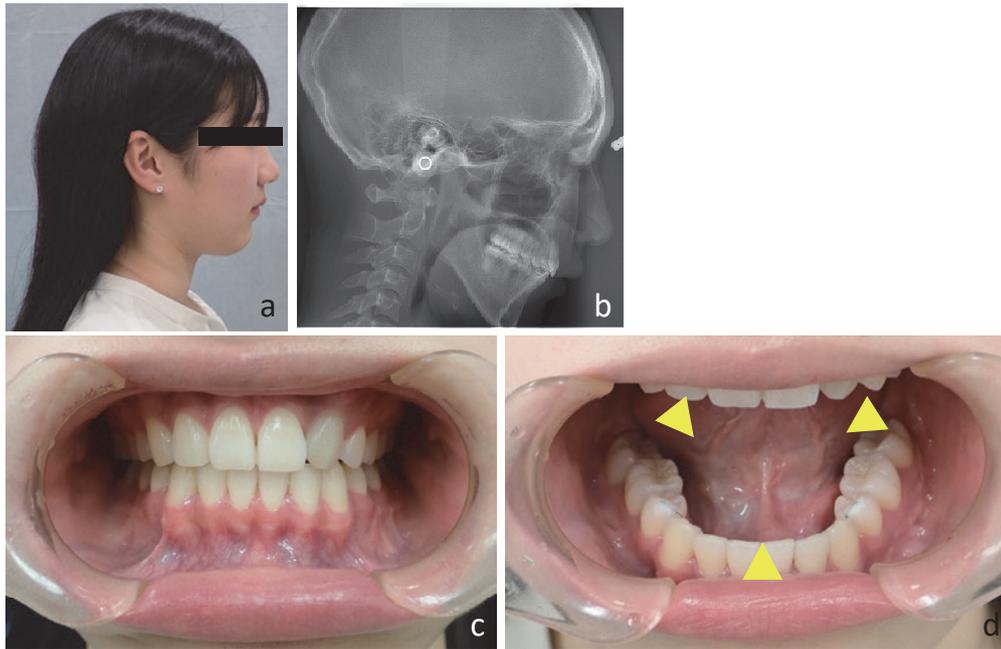


Fig. 6. Postoperative findings at the 1-year follow-up.
 (a) Postoperative facial photograph.
 (b) Postoperative cephalogram.
 (c) Postoperative intraoral findings.
 (d) Intraoral photograph taken at 2 years postoperatively.

foration, as was the case in our patient. However, Kondo *et al.* recommend partially opening the cyst intraoperatively prior to its removal to reduce its contents. This method should be considered depending on the location and size of the lesion [4].

Postoperative complications include the risk of upper airway obstruction due to swelling of the tongue at the floor of the mouth [3]. In our patient, this was managed with a steroid (Rinderon®) administered intraoperatively at 4 mg and then at 12 h after surgery at 2 mg, and with tracheal intubation to reliably maintain the airway. After surgery, no serious swelling developed, and the patient could be extubated on postoperative day 1. In patients with large lesions on the floor of the mouth, it is essential to consider postoperative airway obstruction and be prepared for a prompt response.

The prognosis of patients with dermoid cysts is generally good, as the thick cyst wall makes complete removal relatively easy and recurrence is rare. No recurrence was observed in our patient during 2 years of postoperative follow-up. However, although not reported in the head and neck area, the

malignant transformation of dermoid cysts has been reported, suggesting that careful long-term follow-up is necessary [4].

Dermoid cysts are often detected during a medical exam exploring the appearance of subjective symptoms. However, as far as we are aware, ours is the first case of a large (> 60 mm) cyst on the floor of the mouth detected incidentally during detailed examination of a dentofacial deformity. The causal relationship between dermoid cysts and dentofacial deformity is unknown, but patients with a dentofacial deformity should be examined for the presence of a lesion on the floor of the mouth.

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Conflict of Interests: None

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