

A Case of Submandibular Gland Mucocele

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Submandibular Gland Mucocele: The mucocele occurring in the submandibular region is rare; most cases originate in the sublingual gland. Here, we report a rare case of mucocele originating in the submandibular gland. In this report, we present such a case in a 7-year-old boy, who was treated by an extirpation of cyst with submandibular and sublingual gland.

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INTRODUCTION

Mucoceles known as mucous extravasation phenomena are common lesions of the minor salivary glands. Mucoceles rarely occur in the submandibular region. Usually, their lesions derived from sublingual glands extend to the submandibular region through mylohyoid muscle and are called plunging ranula.¹ In this report, a rare case of a mucocele originating in the submandibular gland is presented.

CASE REPORT

A 7-year-old-boy was referred to our department complaining of a swelling in the right submandibular region. Two years earlier, he had visited a clinic because of a swelling of the right side of the floor of the mouth. He was diagnosed with a ranula and treated with absorptive puncture therapy four times.

On clinical examination, his face was asymmetric. Furthermore, elastic soft swelling, the boundary of which was unclear, was found from his right submandibular area to the submental region. The swelling was painless and was covered with normal skin. Salivary flow from the right sublingual gland decreased somewhat compared to the corresponding site. No swelling in the regional lymph node was noted.

A computed tomography (CT) scan showed a lesion with a clear

boundary outside submandibular gland in the upper right neck (Figure 1a).



FIGURE 1: a: Axial CT scan indicates a low-density lesion in the upper right neck (arrow heads). No tail sign was observed. b: Coronal T2-weighted MRI shows a high-intensity area with clear boundary circumscribed the submandibular gland (arrow heads). Sg: submandibular gland.

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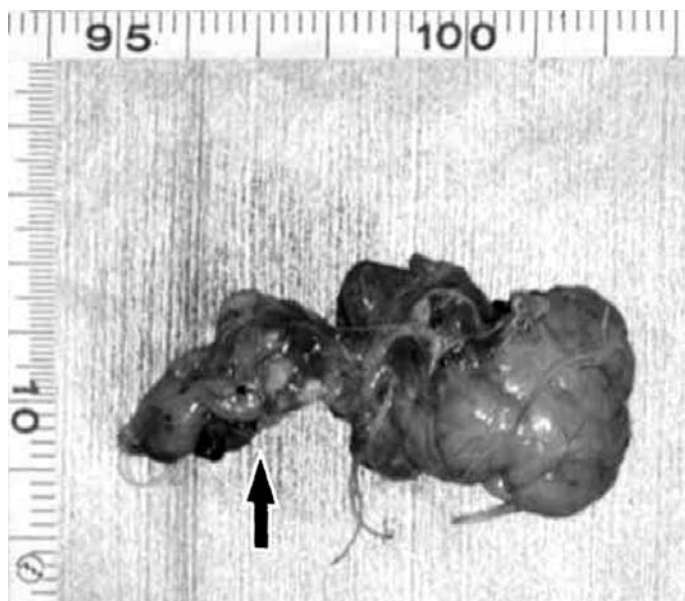


FIGURE 2: Gross examination of the excised specimen showed the cyst (arrow) linked to the anterior part of the submandibular gland.

T2-weighted magnetic resonance imaging (MRI) demonstrated a

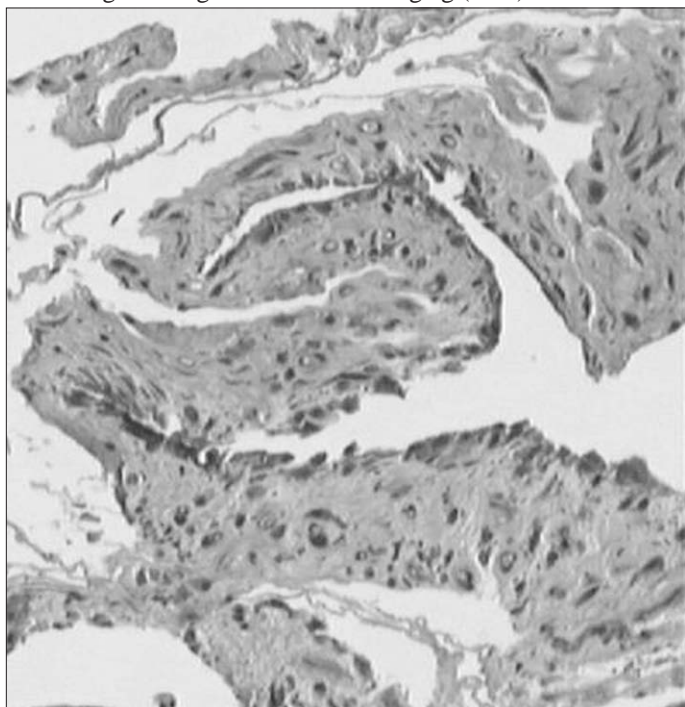


FIGURE 3: Histology shows fibroid cyst wall lacking an epithelial lining (H-E stain, ×200)

cyst-like lesion, irregular in shape from the anterior of the right medial pterygoid muscle to the lateral mandible and a high-intensity area with a clear boundary. The inside was homogeneous and filled with fluid-like material. The cyst circumscribed the right submandibular gland (Figure 1b).

Since no tail sign was noted, plunging ranula originating from sublingual gland was negative. In addition, there was no lymph node enlargement.

Cyst extirpation was performed under general anesthesia. The right submandibular site was incised, from which was separated the

platysma, and the whole cyst was exposed. The cyst wall linked to the submandibular gland and the anterior part lay adjacent to the sublingual gland. The cyst wall was quite thin and the surface was smooth. Pale yellow and viscous fluid was collected from the inside. The cyst was extirpated en bloc with the submandibular gland and the sublingual gland was also extirpated at the same time. The cyst was located next to the submandibular gland (Figure 2). Neither atrophy nor degeneration was noted in the submandibular gland.

Pathology report showed that there was no epithelial lining on the inside of the cystic wall. The cyst wall with abundant fibers was thinly folded and the arrangement of fibroblasts was irregular (Figure 3).

DISCUSSION

Submandibular gland mucocele is rare and to our knowledge, only 9 cases including ours have been reported in English literature.²⁻⁷ In these cases, patients ranged from 7 to 39 years of age. An 8:1 male gender distribution was seen from these studies. The cysts were differentiated from ranulas that showed a slight preponderance of females.⁸ However, a male slight predilection was reported for the plunging ranula.⁹

The discrimination of the salivary gland causing the mucocele in the submandibular region is clinically difficult but CT¹⁰ and MRI¹¹ findings are quite useful to make a differential diagnosis. Mucocele arising from the sublingual gland contains sublingual gland and develops to the inferior part of mylohyoid muscle. Therefore, a characteristic image known as tail sign can be observed.^{10,11} However, CT and MRI findings revealed the cyst circumscribed the right submandibular gland, and no tail sign was noted in this case. Those findings were very useful because we were able to evaluate that it was submandibular gland origin before the surgery.

The treatment of mucocele originating in submandibular gland was proposed as the following procedures; marsupialization, incision and drainage, extirpation of cyst alone, extirpation of the submandibular gland and/or the sublingual gland with the cyst. Radical treatment in cases of submandibular gland mucocele is necessary to remove the lesion with the submandibular gland. It has been reported that when the mucocele originating in the submandibular gland also involves or is close proximity to the sublingual gland, the prognosis after surgical removal of the lesion with submandibular and sublingual glands is excellent.³ In this case, the patient had a history of sublingual gland mucocele so that the sublingual gland was also removed at the same time. We once considered adopting a conservative therapy for the patient because he was young, but decided to remove the lesion with both salivary glands because the patient had suffered from symptoms for a long time and the patient and his family requested this treatment. Now, the prognosis is good and the patient is followed as an outpatient.

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