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Midline supramylohyoid mucocele arising from ectopic salivary gland tissue

KEYWORDS

Ectopic salivary gland tissue;
Geniohyoid muscle;
Midline;
Mylohyoid muscle;
Mucocele

In the oral and oropharyngeal region, most mucoceles arising from minor salivary glands are observed on the lower lip, and less frequently occur at various sites such as the ventral tongue, oral floor, buccal mucosa, lingual frenum, hard and soft palate, uvula, and retromolar and peritonsillar regions.¹ Mucoceles arising from ectopic salivary gland tissue are extremely rare in oral and maxillofacial regions.^{1,2} Here, we report a rare case of midline supramylohyoid mucocele arising from ectopic salivary gland tissue.

A 16-year-old woman with submental swelling was referred to our department. Computed tomography showed a well-circumscribed, lobulated homogeneous cystic lesion (32 × 31 × 24 mm) on the mylohyoid muscle, under genioglossus muscles, and between the geniohyoid muscles (Fig. 1A–C). The lesion was in contact with the hyoid bone. Magnetic resonance imaging revealed the lobulated heterogeneous cystic lesion had high signal intensity on fat-suppressed T2-weighted images (Fig. 1D and E). Ultrasonography showed a lobulated heterogeneous cystic lesion without blood flow (Fig. 1F). Fine needle aspiration (FNA) showed a yellowish gel (Fig. 1G). The preoperative diagnosis was midline supramylohyoid mucocele between the geniohyoid muscles. The patient underwent extraoral removal of the cystic lesion in a submental approach under general anesthesia. A 3-cm horizontal submental skin incision was made. After the incision of the platysma and mylohyoid muscles, the cystic lesion was exposed in the

surgical field (Fig. 1H). The supramylohyoid cystic lesion was dissected carefully from the geniohyoid muscles and was removed completely under the genioglossus muscles. There was no continuity between the cystic lesion and bilateral sublingual glands. The cystic lesion included yellowish mucus. The wound was closed, and the patient was discharged 2 days after surgery. The postoperative operative course was uneventful. The pathological diagnosis was mucocele (Fig. 1I).

Ectopic salivary gland tissue has been identified at various sites, including the skin, neck, hypophysis, mediastinum, prostate, rectum, vulva, thyroid gland, pituitary gland, external and middle ear, mastoid bone, maxilla, and mandible.^{1,3} To our knowledge, midline supramylohyoid geniohyoid mucoceles are extremely rare, and there were only two English reports of midline mucocele arising from ectopic salivary gland tissue between the bilateral geniohyoid muscles.^{1,2} In the present case, the midline mucocele was located on the mylohyoid muscle, under genioglossus muscles, and between the geniohyoid muscles, whereas the previously reported 2 cases had no extension to the mylohyoid muscles.

Differential diagnosis of midline deep sublingual cystic lesions includes dermoid/epidermoid cyst and thyroglossal duct cyst. In the present case, we suspected a radiologically dermoid/epidermoid cyst or thyroglossal duct cyst because of the midline supramylohyoid cystic lesion between the geniohyoid muscles. However, we diagnosed it

<https://doi.org/10.1016/j.jds.2023.09.012>

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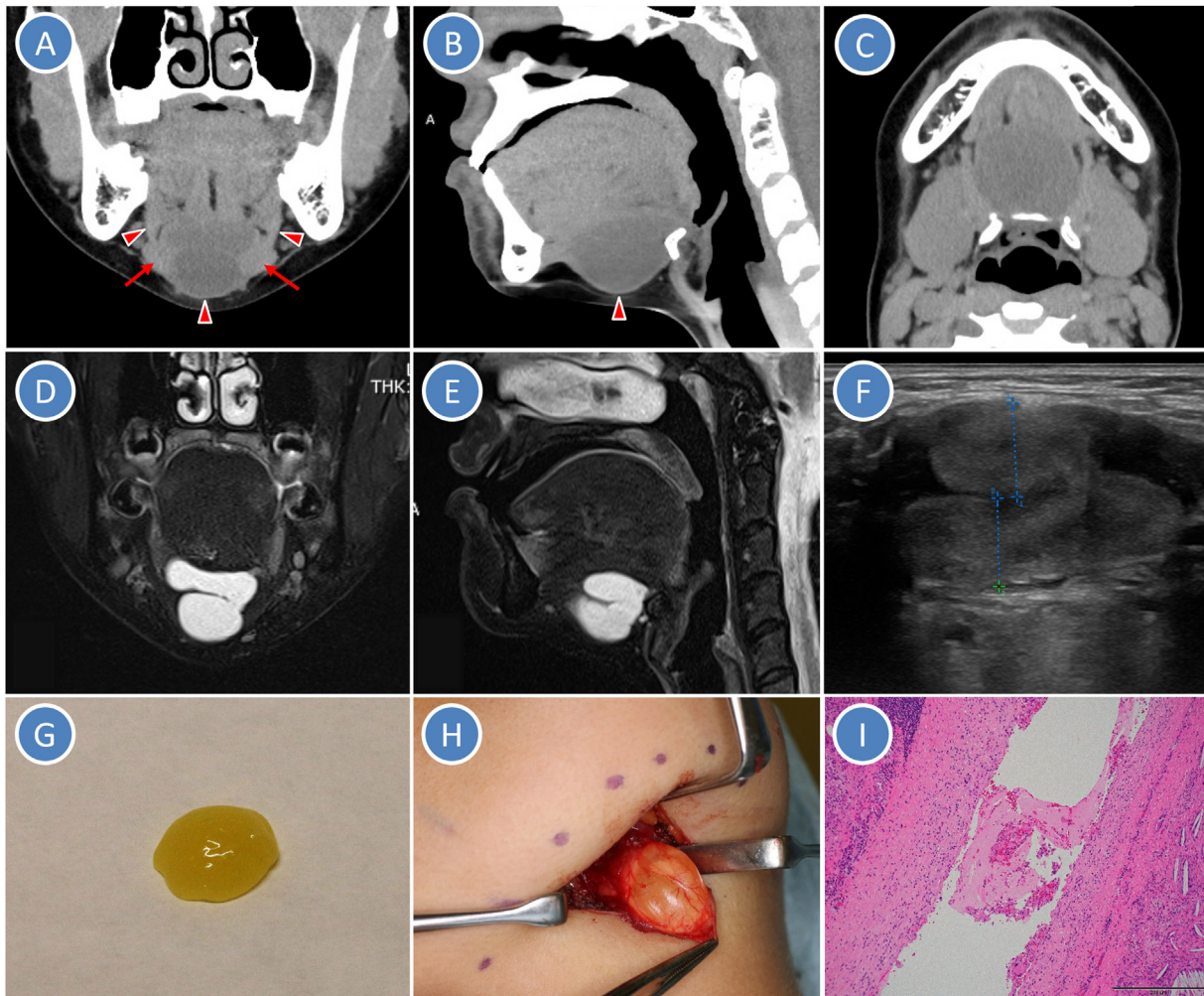


Figure 1 (A–C) Computed tomography (A: Coronal image, B: Sagittal image, C: Axial image.) showed a well-circumscribed, lobulated homogeneous cystic lesion on the mylohyoid muscle, under genioglossus muscles, and between the geniohyoid muscles. (D, E): Magnetic resonance imaging (D: Coronal image, E: Sagittal image) revealed the lobulated heterogeneous cystic lesion had high signal intensity on fat-suppressed T2-weighted images. (F) Ultrasonography showed a lobulated heterogeneous cystic lesion without blood flow. (G) Fine needle aspiration showed a yellowish gel. (H) The cystic lesion was exposed in the surgical field after incision of the platysma and mylohyoid muscles. (I) Pathological examination of the specimen revealed a mucocele composed of the fibrous cyst wall with a severe lymphoplasmacytic cell and histiocyte infiltrate and the presence of cholesterol slits (Hematoxylin and eosin staining). There are mucoid materials and red blood cells in the lumen of the mucocele. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

preoperatively as mucocele because FNA showed yellowish gel. We considered that the origin of the mucocele was ectopic salivary gland tissue, because the bilateral sublingual glands were not continuous with the mucocele on the mylohyoid muscle, under genioglossus muscles, and between the geniohyoid muscles. Because the common differential diagnosis of the midline deep sublingual cystic lesions does not include mucocele arising from ectopic salivary gland tissue for the rarity, oral surgeons and radiologists should consider mucocele in the differential diagnosis and broaden the possibility of correct diagnosis.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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Received 10 September 2023

Final revision received 11 September 2023

Available online 21 September 2023