



Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology

CLINICOPATHOLOGIC CONFERENCE

Yellow-white tumor on the floor of the mouth

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CLINICAL PRESENTATION

A 48-year-old white woman was referred by her dentist for diagnosis and management of a painless lump beneath her tongue that was discovered during a routine examination prior to the construction of full dentures. The patient had been aware of the lesion for several months, but had ignored it due to the absence of pain and lack of interference with speech or mastication. Recently, she felt "tenderness" in the ipsilateral submandibular area. Her medical history was unremarkable and she denied irritation or trauma to the involved area.

Intraoral examination revealed an oval-shaped tumor on the left side of the floor of her mouth, along the course of the submandibular duct (Fig. 1). The surface of the tumor was smooth with a yellow-white color and mildly erythematous periphery. The mass measured 1.3 × 0.7 cm and was hard, well-circumscribed, movable, and painless on palpation. Decayed teeth roots covered by hyperplastic gingiva were present in the anterior mandible. No other lesions were observed. With external palpation, reduced saliva flow was noted from the

left submandibular duct compared to the right, and the left gland was slightly enlarged and tender. No regional lymphadenopathy was evident. A panoramic radiograph was non-contributory.

DIFFERENTIAL DIAGNOSIS

The floor of mouth is the most common intraoral location for two developmental epithelial cysts, the dermoid cyst and the oral lymphoepithelial cyst. Both lesions typically present as an asymptomatic, freely movable submucosal nodule that can vary in color from yellow-white to pink.^{1,2} The oral dermoid cyst typically arises in the midline of the floor of the mouth of children and young adults, but may present more laterally.² More than half of all cases of oral lymphoepithelial cyst occur in the floor of the mouth of adults.¹ While keratin and sebum are responsible for the yellow-white color of many dermoid cysts, abundant lymphoid tissue and keratin contribute to the appearance of lymphoepithelial cysts.³ The dermoid cyst has a soft to dough-like consistency with characteristic pitting on pressure,² and the lymphoepithelial cyst may be either soft or firm on palpation.¹ Despite the similarities in clinical appearance to the present case, neither of these cysts are commonly associated with signs or symptoms of salivary gland obstruction.

Sialolithiasis or calculus formation represents roughly half of salivary gland disease affecting the major glands of adult patients. In addition, it is among the most common causes of acute and chronic infections in the head and neck region.⁴ Eighty percent of all sialoliths occur in the submandibular gland, possibly due to the increased alkalinity of its saliva, higher concentration of calcium and phosphate, higher mucus content, antigravity flow, and the long, tortuous course

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Fig. 1. Clinical view of firm mass presenting on the left side of the floor of the mouth.

of Wharton's duct.⁴ Most submandibular stones are found either at the distal portion of the duct or at the hilum. Occasionally, they are associated with intermittent painful swelling of the affected gland, a classic feature that is often most notable during mealtimes. Although total obstruction of salivary flow may be noted in some cases, enlargement and tenderness can occur with only partial obstruction of the affected gland. When visible intraorally, sialoliths often present as a nodule of normal or yellowish color.⁴ A radiopaque mass can usually be identified within the affected soft tissues by occlusal or panoramic radiographs; however, a small percentage of sialoliths are not detectable by plain films. In addition, small calculi positioned in the posterior one third of the duct or in the parenchyma of the gland may also not be visualized on routine radiographs.⁵ Sialography may be helpful, but this procedure is often uncomfortable for the patient and has the potential to introduce infection.⁵

The floor of mouth is an unusual location for minor salivary gland tumors, representing only 3-5% of total cases. The majority (80-88%) of these, however, are malignant.^{6,7} Both mucoepidermoid carcinoma and pleomorphic adenoma could present as circumscribed tumors similar to the current case.⁷ While obstruction of the submandibular duct is not a common feature of minor salivary gland tumors, a single case of intraductal papilloma of the submandibular gland was found in the literature.⁸ Such a presentation could explain both features of mass and obstruction seen in our patient.

An asymptomatic, circumscribed submucosal mass in the floor of the mouth could represent a benign connective tissue tumor. The yellowish color of the present lesion is consistent with a lipoma, which can appear as a smooth-surfaced, sessile, or pedunculated

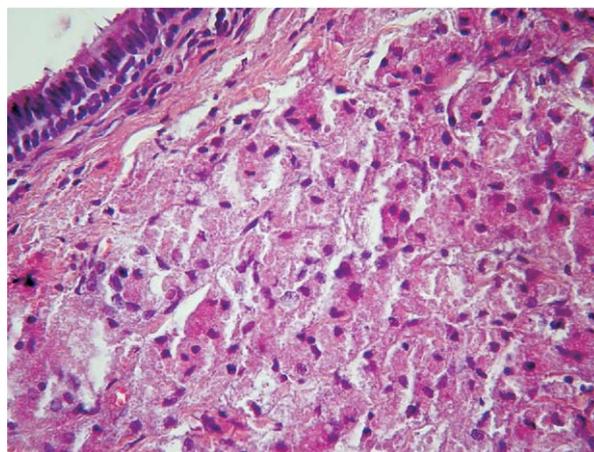


Fig. 2. Photomicrograph depicting sheets of large cells with abundant granular, eosinophilic cytoplasm and round, eccentrically placed dark nuclei (hematoxylin-eosin, original magnification $\times 100$).

tumor.⁹ It comprises 0.1% to 5% of all benign tumors of the mouth and may arise in the floor of the mouth of adults.⁹ The consistency of a lipoma is usually soft to fluctuant. While osseous or cartilaginous metaplasia may be seen in large or long-standing tumors (benign mesenchymoma), this entity has not been described in the floor of the mouth.¹⁰ Finally, enlargement and tenderness of the submandibular gland have been reported in association with cases of granular cell tumor occurring in the floor of the mouth.^{11,12}

The clinical presentation of the lesion was considered most consistent with a sialolith of the submandibular duct that was not identifiable in the panoramic radiograph due to low inorganic content. Further inves-

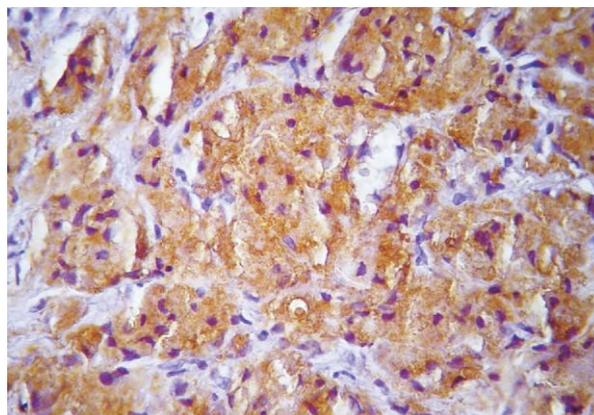


Fig. 3. Immunohistochemical probe analysis showing strong cytoplasmic expression of S-100 protein by the lesional cells (avidin-biotin-peroxidase, original magnification $\times 100$).

Table I. Main clinical features of previously reported cases of granular cell tumors on the floor of the mouth

Reference	Age	Sex/Race	Submandibular signs	Treatment	Recurrence
Bjorkroth ¹¹	49	M	diffuse swelling and fluctuation	blunt dissection	12 months
Templeton et al. ¹²	55	F/B	progressive and painful swelling	excision in continuity with the gland and duct	no - 6 months FOD
Horn and Stout ¹⁴	35	M	n/s	n/s	n/s
Caine and Mesa ¹⁵	23	F/B	n/s	excisional biopsy	no
Collins and Jones ¹⁶	38	F/B	n/s	none	n/s
present case	48	F/C	slight enlargement and tenderness on palpation	excisional biopsy	no - 12 months FOD

M, Male; F, Female; B, Black; C, Caucasian; n/s, not stated; FOD, free of disease.

tigation (occlusal radiograph, CT, sialography) was considered unnecessary and a decision was made to proceed with excisional biopsy.

MANAGEMENT

With the patient under local anesthesia, an incision was made through the oral mucosa along the course of the duct in order to locate and retrieve the stone. Instead, the lesion was found to consist of a soft tissue mass attached to both the overlying mucosa as well as Wharton’s duct. Local excision of the mass was performed. Because this procedure required removal of a portion of the duct as well, a new orifice was created. On the first postoperative day the patient experienced swelling of the floor of the mouth and some difficulty in swallowing, but the symptoms gradually subsided. Fifteen days later the surgical wound had healed completely. The decayed roots were later extracted by her dentist.

DIAGNOSIS

The surgical specimens were fixed in 10% buffered formalin. Grossly, two pieces of solid, gray-white tissue were seen. Microscopic examination showed that the lesion was composed of sheets of large, polyhedral cells with abundant granular, eosinophilic cytoplasm and round, eccentrically placed dark nuclei. They were mainly arranged in small clusters or cords that were infiltrating collagen bundles, muscle fibers, and salivary glands, and were surrounding a large excretory duct (Fig. 2). Superficially, they were in contact with the overlying epithelium, while small epithelial islands were dispersed among them. The granular cells were strongly positive for S-100 protein (Fig. 3).

Final diagnosis: Granular cell tumor.

DISCUSSION

The granular cell tumor (GCT) is an uncommon benign soft tissue lesion with a predilection for the oral cavity.³ The tongue is the single most common site of GCT, accounting for one third to one half of all re-

ported cases, followed by the buccal mucosa.³ GCT usually affects adults in the fourth to sixth decades of life and a female predilection has been noted. GCT usually manifests as a solitary, sessile, rubbery nodule that can vary from pink to yellow in color.³ The histogenesis of GCT has not been established conclusively, but morphological, immunohistochemical, and ultrastructural observations support a neural origin. Similarly, the neoplastic versus reactive nature of the tumor has not been completely resolved. GCTs are usually not encapsulated masses and often show a pseudoinvasive growth pattern with tumor cells spreading between adjacent connective tissue elements, especially muscle fibers and nerve bundles.³ However, conservative surgical excision is curative and recurrences are uncommon.³ GCTs with malignant biological behavior are extremely rare.³

Since Abrikossoff¹³ first mentioned the presence of GCTs on the floor of the mouth in 1931, six cases have been reported in the English literature.^{11,12,14-16} The main clinical features of those cases are summarized in Table I. Two more cases were included in a review of 377 GCTs by Peterson.¹⁷ The case of Horn and Stout¹⁴ is included, although the description of tumor cells is suggestive of a rhabdomyoma. The patient reported by Collins and Jones¹⁶ had eight GCTs in the head and neck region, two of them on the floor of the mouth. One of them, near the orifice of the submandibular gland, had a clinical appearance similar to the present case.

Bjorkroth¹¹ and Templeton et al¹² reported the association of GCTs on the floor of the mouth with submandibular swelling, described as “marked”, “progressive”, or “painful”. The first case was clinically diagnosed as a retention cyst of the sublingual gland,¹¹ while Wharton’s duct was extrinsically obstructed by tumor in the other case.¹² A case arising adjacent to the orifice of Stensen’s duct was not associated with parotid enlargement.¹⁷ It is noteworthy that signs of excretory duct obstruction have also been reported in a case of granular cell tumor of the pancreas.¹⁸

In the present case, microscopic evaluation con-

firmed that the connective tissue surrounding the excretory duct of the submandibular gland was diffusely infiltrated by tumor cells. It is suggested that replacement of the normal periductular tissue by granular cell tumor impaired the ability of the duct to expand, leading to signs and symptoms of mild, intermittent chronic obstruction of the affected gland.

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