Nodular lesion in the buccal mucosa

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A 55-year-old woman showing symptoms of a nodular lesion involving the left buccal mucosa with a history of approximately 5 years sought treatment at our dental clinic. The patient’s medical history revealed diabetes mellitus, hypertension, and arthrosis treated with metformin, enalapril, hydrochlorothiazide, and ibuprofen. The patient reported no alcohol use or tobacco consumption. The extraoral examination revealed no abnormalities. The intraoral examination revealed a single, well-circumscribed, submucosal, nodular lesion covered with normal epithelium, measuring approximately 1.0 centimeter in diameter (Figure 1A). On palpation, the lesion was asymptomatic, had a hard consistency, and appeared to be attached firmly to subjacent tissue. The panoramic radiograph revealed no bone alterations. On the basis of these findings, the clinical diagnosis was benign mesenchymal neoplasm. Excisional biopsy was performed. During the surgical procedure, the specimen was grayish, had fibrous consistency, and measured 1.0 cm in diameter (Figure 1B). The lesion appeared to be inserted into the adjacent tissues. No capsule was identified.

The specimen was submitted to histopathologic analysis, which revealed a well-defined circumscribed lesion with no well-defined capsule (Figure 2A). Ovoid, polygonal, and spindle cells were arranged in lobes, surrounded by dense connective tissue (Figure 2A). These cells were interspersed with myxoid matrix and chondroid matrix (Figures 2A and 2B). No local invasion, mitosis, necrosis, or minor salivary glands were observed. Some areas had foci with cellular atypia, exhibiting cells with nuclear hyperchromatism (Figure 2C) as well as the presence of binucleated and multinucleated cells. An immunohistochemical profile was performed including vimentin, S100 protein, glial fibrillary acidic protein (GFAP), CD57, desmin, smooth muscle actin, and pancytokeratin AE1 and AE3. The panel produced the following results: focally positive for vimentin (Figure 3A), S100 protein (Figure 3B), GFAP (Figure 3C), CD57 (Figure 3D), desmin (Figure 3E), and smooth muscle actin (SMA) (Figure 3F), and negative for AE1 and AE3.
Figure 1. A. Clinical aspect revealed a nodular lesion covered with normal epithelium situated in buccal mucosa. B. The macroscopic appearance of the specimen was grayish and white, had an irregular surface, and measured 1.0 centimeter in diameter.

Figure 2. A. Low-power photomicrograph showing no well-defined continuous capsule. Matrix composed of myxoid and chondroid zones associated with tumor cells can be seen (hematoxylin and eosin stain, x40 original magnification). B. High-power photomicrograph demonstrating ovoid, polygonal, and spindle cells related to chondroid areas (hematoxylin and eosin stain, x200 original magnification). C. Focal area of chondroid tissue exhibiting cellular atypia with nuclear hyperchromatism (hematoxylin and eosin stain, x400 original magnification).

Figure 3. A. Tumor cells showing focal positive in cytoplasm for vimentin in chondroid and connective tissue area (streptavidin-biotin complex [sABC] method, x100 original magnification). B. S100 protein positive in cytoplasm distributed in some chondroid and connective cells (sABC method, x100 original magnification). C. Focally positive cytoplasm reaction for glial fibrillary acidic protein in chondroid and connective tissue area (sABC method, x200 original magnification). D. Cytoplasm tumor cells were positive for CD57 in chondroid and connective tissue area (sABC method, x100 original magnification). E. Focal positive cytoplasm cells for desmin in chondroid and connective tissue area (sABC method, x200 original magnification). F. Cytoplasm tumor spindle cells were focally positive for smooth muscle actin in connective tissue area (sABC method, x200 original magnification).

**CAN YOU MAKE THE DIAGNOSIS?**

A. extraskeletal chondroma
B. pleomorphic adenoma
C. soft-tissue chondromyxoid fibroma
D. ectomesenchymal chondromyxoid tumor
Ectomesenchymal chondromyxoid tumor (ECT) is an uncommon, benign, mesenchymal, soft-tissue neoplasm of the oral mucosa. ECT was first described in 1995, with 52 cases reported in the English-language literature: 50 cases were in the tongue; 1 was in the hard palate; and 1 was in the buccal mucosa. The cases described in the literature reported that ECT most commonly affects adults in the third to sixth decades of life. Clinically, ECT presents as a slow-growing, painless, firm, well-circumscribed, submucosal nodule, covered by normal mucosa, that appears to involve only the oral cavity and usually is located on the anterior dorsum of the tongue. Such tumors range in size from 0.3 to 2.0 cm. The diagnosis of ECT is grounded on clinical characteristics, light microscopy, and immunohistochemistry.

The microscopic features (low-power microscopic analysis) reveal lobular sheets of cells surrounded by skeletal muscle fascicles and connective tissue. The lesion is not encapsulated, but loose connective tissue may be found at the interface between the tumor and normal tissue. Three different microscopic patterns are generally seen: cellular, myxoid, and chondroid matrix. Cellular areas are characterized by the proliferation of oval, polygonal, and spindle cells. ECT may exhibit focal areas of cellular atypia, including pleomorphism and mitotic figures. The immunopathologic characteristics indicate a tumor of mesenchymal, neurogenic origin with the expression of vimentin, S100 protein, and GFAP. ECT presents variable immunoreactivity for CD57, desmin, and SMA, as we observed in the case described in this article. Ki-67 protein labeling is low, indicating the benign nature of the tumor. The presence of biphasic myxoid and chondroid patterns and the positivity of tumor cells for vimentin, S100 protein, and GFAP are important findings for making the diagnosis of ECT.

The treatment for this tumor is conservative surgical excision. The recurrence rate of ECT is 7%. In the follow-up of the case described in this article, the patient remained free of disease after 12 months, but the intraoral affected area exhibited paresthesia.

DIFFERENTIAL DIAGNOSIS

Extraskelatal chondroma. Extraskeletal chondroma is a benign, soft-tissue, cartilaginous tumor usually located in the hands and feet, but it occasionally occurs in the head and neck region, in which the most reported sites are the paranasal sinuses, nasal cavity, larynx, and tongue. Investigators have described extraskeletal chondroma as a cartilaginous choristoma owing to the presence of a tumorlike mass composed of “normal tissues” in an “abnormal location.” The histologic analysis reveals a well-circumscribed, lobulated mass composed of mature hyaline cartilage surrounded by condensed collagenous tissue. Some cellular atypia occasionally is observed. In the immunohistochemical analysis, the cells are positive for S100 protein and vimentin as well as negative for pancytokeratin (AE1 and AE3), myoepithelial markers, and GFAP. The treatment of choice is local excision, and the recurrence rate is 10% to 15%.

Pleomorphic adenoma. Pleomorphic adenoma is the most common salivary gland tumor, accounting for approximately 60% of all salivary neoplasms. The mean age of presentation is 40 years, but the age of patients with this condition can range from the first to the ninth decades of life. Approximately 80% of pleomorphic adenomas arise in the parotid gland. Clinically, these lesions are usually painless, slow-growing tumors with a hard consistency, measuring 2 to 5 cm in diameter. There is a proliferation of epithelial and myoepithelial cells consisting of glandlike structures, ducts, cell nests, cords, spindle-shaped, and plasmacytoid cells, intermingled with a mesenchymal or chondromyxoid component with frequent metaplastic changes. The arrangement of these tissues varies from predominantly cellular to predominantly myxochondroid. The tumor cells are positive for pancytokeratin and are variably positive for S100 protein, a-SMA, GFAP, calponin, CD10, and muscle-specific actin. Cells in the chondroid areas are positive for both vimentin and pancytokeratin. Treatment is complete surgical excision. The recurrence rate as a carcinoma ex pleomorphic adenoma is nearly 3%.

Soft-tissue chondromyxoid fibroma. Soft-tissue chondromyxoid fibroma is a rare, benign, cartilaginous tumor that mostly arises from the metaphysis of long bones and rarely is seen in the head and neck region. Investigators have reported the extension of the tumor to soft tissue. However, to the best of our knowledge, investigators have described only 2 cases of a primary tumor with no bone involvement. In the histologic analysis, soft-tissue chondromyxoid fibroma exhibits a lobular pattern with stellate or spindle-shaped cells on a myxoid or chondroid background. The presence of mitosis is uncommon. Giant cells often are found at the periphery of the tumor. Immunohistochemically, the cells are positive for S100 protein, and the periphery of the chondroid area demonstrates positive staining for...
SMA, muscle-specific actin (known as HHF-35), and CD34. The prognosis of soft-tissue chondromyxoid fibroma has not been reported in the literature, likely owing to its rarity.

In addition to the neoplasms described previously, other important differential diagnoses for ECT include myoepithelioma, myxoid neurofibroma, and myxoid chondrosarcoma. Myoepithelioma and ECT have some similar characteristics, but the clinical location and absence of salivary glands adjacent to ECT are important features that differentiate these 2 lesions.

Furthermore, myoepithelioma is usually positive for α-SMA and does not present chondromyxoid-exuberant areas. Myxoid neurofibroma is a tumor with cells that contain wavy nuclei. Atypia or chondroid areas are not found in myxoid neurofibromas; nevertheless, both lesions express vimentin, S100 protein, and GFAP.

Extraskelatal myxoid chondrosarcoma typically exhibits more areas of cellular atypia and more aggressive clinical behavior than ECT.

CONCLUSIONS

In this article, we described the case of a patient with a nodular lesion on the left buccal mucosa who had a histopathologic diagnosis of ECT. A dental clinician can make the diagnosis of this type of rare oral neoplasm only through the combined analysis of clinical, histopathologic, and immunohistochemical characteristics. It is important to perform an accurate diagnosis of this type of lesion, because it may be confused with other lesions that have similar clinical features but a different prognosis.

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