Spindle cell lipoma of the floor of the mouth. A case report

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Received: 22-10-2005
Accepted: 4-03-2006

ABSTRACT
A sessile swelling in the floor of the mouth appeared three years ago in a 29-year-old Caucasian female located laterally to the opening of the right duct of Wharton. The mass covered by normal looking mucosa exhibited slight growth since then. After excision, histological examination revealed the presence of a tumor formed by areas of mature adipose cells interspersed with extensions of tightly disposed fusiform fibroblasts immunoreactive for vimentin and CD-34. There were foci of concentric fibroblasts forming dense whorls. When large these conglomerates exhibited chondrocytes in the center. Mast-cells were not rare throughout the fibroblastic areas. No signs of malignancy occurred. These features led to the diagnosis of a spindle cell lipoma with chondrous metaplasia. The relative rarity of such a tumor in this location, especially in the chondrous variety, was considered worth of presentation, while the differential diagnosis with other intraoral tumors, namely mucoceles, dermoid cysts, mesenchimomas, fibromas and myxomas of the floor of the mouth, is discussed.

Key words: Lipoma, spindle cell lipoma, intraoral tumor, floor of the mouth.

INTRODUCTION
Benign swellings in the floor of the mouth include mucoceles (1), dermoid cysts (2), mesenchymomas (3) and lipomas. In recent reviews of filed pathological cases Al-Naief et al (4) in the USA studied 164 oral lipomas, and Fregnani et al (5) in Brasil reported 46 tumors. The most common lesions were simple lipomas and fibrolipomas, and rarer variants included angiolipoma, intramuscular lipoma, pleomorphic lipoma and spindle cell lipoma. The spindle cell lipoma is a rather infrequent neoplasm, as shown by the fact that two recent surveys in the English language literature (5, 6) revealed only 25 cases. Of these only four were located in the floor of the mouth. Since spindle cell lipomas are composed of two tissues derived from embryonic mesenchyme, they bear some similarities to the also benign and rare mesenchymomas of which 21 cases have been reported so far in the oral cavity (3). However mesenchymomas usually contain more mesenchyme-derived tissues than the very cellular connective tissue and the mature adipose tissue occurring in spindle cell lipomas, and they lack the prominent fusiform fibroblasts of the latter.

We report a patient with a spindle cell lipoma in the floor of the mouth with conspicuous foci of chondrous metaplasia, which appears to be the fifth case described so far in this location.

CASE REPORT
A 29-year-old Caucasian female complained of a right-sided swelling in the floor of the mouth. The lesion had appeared 3-years previously, growing very slowly and without any accompanying symptoms. Oral examination revealed a sessile mass, 1.5 cm in maximal diameter, in the floor of the mouth lateral to the opening of the duct of Wharton (Fig. 1). It was firm and nonfluctuant on palpation, and not attached to the surrounding or deeper tissues. The overlying muco-sa was normal and there were no salivary changes. There
was no palpable cervical lymphadenopathy. The mass was dissected and excised. Upon formalin fixation and paraffin embedding, sections were treated with hematoxylin-eosine or the Giemsa stain diluted to 20%. Other paraffin sections were immunostained with primary antibodies against vimentin (monoclonal antibody raised in mouse provided by Dakoppatts, Glostrup, Denmark), CD-34 or the S-100 protein (both polyclonal antibodies from Labvision, Fremont, USA). A kit of ABC, from Labvision, followed the secondary antibodies. The reactions were developed with diaminobenzidine (DAB) and hydrogen peroxide.

Histological examination revealed areas of mature adipose tissue interspersed with areas of spindle shaped fibroblasts, the extent occupied by both tissues being similar (Fig. 2). Spindle cells were densely distributed in a fibrous matrix (Figs. 3 and 5) which also had some myxoid areas, as well as a few foci of concentric cells (Fig. 3), often arranged in whorls. The larger foci showed cartilaginous cells at the center (Fig.4). Extensions of mature cartilage were rare. Occasionally the spindle shaped cells occurred in palisading arrangements. Mast cells were consistently present in the spindle cell areas (Fig. 5). The mitotic index was low and there were no necrotic areas. Spindle cells were immunoreactive for vimentin and CD-34 (Fig. 6). Immunoreactivity for the S-100 protein occurred in cartilaginous cells and the periphery of some adipocytes. There were no recurrences during a 3 year follow-up.

![Fig. 1. The tumor after dissection before excision.](image)

![Fig. 2. Low power view showing the spindle cell (left) and the mature adipose (right) components of the tumor. Giemsa stain. (X 10)](image)

![Fig. 3. The background matrix was rich in fibrous connective tissue bundles. Asterisk marks a focus of concentric cells. Giemsa stain. (X 25)](image)

![Fig. 4. A larger nodule of concentric cells with chondrous metaplasia at the center. Giemsa stain. (X 25)](image)

![Fig. 5. Two mast cells among the spindle cells. Giemsa stain. (X 100)](image)

![Fig. 6. Spindle cells immunostained for the CD-34 protein. (X 25)](image)
DISCUSSION

Swellings of the floor of the mouth are often benign tumors. The mucous extravasations or mucoceles of major salivary glands, also called blue ranulas, may occur in this location and they may pluge down sending one extension to the submandibular space in the neck (1). Dermoid cysts may occur in the floor of the mouth but they may also produce another mass in the neck from where they originally arose due to the sequestration of cutaneous tissue along cervical embryonic lines of closure, later extending to the mouth (2, 7).

Fibromas and related entities caused by traumatic friction of prosthetic devices are common (8) and myxomas may arise from odontogenic rests (9). A small mass strictly circumscribed to the floor of the mouth with no signs of malignancy (no induration, adherence to deep plans, ulceration) is more likely to be an adenoma of the minor salivary glands if hard (8) or, if more tender, may be lipoma or a variant (5, 10) or a mesenchymoma (3).

The histopathology of our present case fits well with the prototype of the spindle cell lipoma, as described in the 38 (mostly cutaneous) cases reviewed by Fletcher and Martin-Bates (11). The distinction from the more common fibrolipoma was made by the greater density of fibroblasts in our tumor contrasting with the conspicuous collagen bundles intersecting the adipose tissue in fibrolipomas (5). The occurrence of a spindle cell liposarcoma (11) or a spindle cell carcinoma (12, 13) was excluded by the lack of mitotic cells and lipoblasts. Although a rare case of lipomatous neurofibroma has recently been reported in the mouth (14), this possibility was ruled out in our case by the lack of specific immunostaining for S-100 in the palisading cell arrangements. Finally it should be stressed that the chondroid nodules described here were consistent with different steps of a metaplastic process occurring in cell foci distinct from the areas of stable cartilaginous tissue present in mesenchymomas3.

In a case of the rare intramuscular variant of spindle cell lipoma, Fletcher and Martin-Bates (11) noted foci of chondroid and osseous metaplasia, while Nwaorgu et al (15) described striking chondroid metaplasia in a pharyngeal lipoma hence designated as chondroplipoma. However, to our best knowledge, no such data have been found in intraoral spindle cell lipomas (4, 6).

Oral spindle cell lipomas appear not to differ from the cutaneous variety, which is much more common. In both recurrences are rare and histological signs of malignancy are lacking. Nevertheless, their very rarity demands reporting of every new cases.

REFERENCES