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Case Report

Soft tissue myxoma: report of an unusual case located on the gingiva

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Abstract

Background: Soft tissue myxoma of the oral cavity is rare. Only three cases of myxoma of gingiva have been reported in the literature. We present a case of soft tissue myxoma arising from the left maxillary adherent gingiva in a 42-year-old female patient.

Methods: Histological examination showed spindle-shaped cells in a myxoid stroma. Immunohistochemical stains with S-100 protein were negative, while those with vimentin were positive.

Results: Clinical examination revealed a soft tissue mass, with tense elastic consistency on palpation. The overlying mucosa was normal and healthy. A clinical diagnosis of fibroma was given. Histological examination showed spindle-shaped and stellate cells, arranged in a myxoid fibrous stroma, with collagen fibres distributed uniformly. Scattered islands or strands of inactive odontogenic epithelium were present. On the basis of the histological and immunohistochemical findings, the final diagnosis was soft tissue myxoma.

Conclusions: Further studies are necessary to clarify the origin and histogenesis of this lesion.

Key words: fibroma; gingiva; S-100 protein; soft tissue myxoma; vimentin

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Soft tissue myxomas of the oral and perioral tissue are rare and considerably less common than odontogenic myxoma of the jaws (Shimoyama et al. 2000, Chang et al. 2001, Curran et al. 2002). The intra-oral or perioral soft tissue myxomas are benign tumours, slowly growing, insidious and potentially infiltrative (Elzay & Dutz 1978, Ramaraj & Shah 2003); in contrast with osseous myxomas, they show a less-aggressive behaviour and rarely recur following conservative excision (Barnes 2001, Ramaraj & Shah 2003). However, not many details are available in the literature related to soft tissue myxomas, if compared with their counterpart, central odontogenic myxomas.

Tse & Vander (1985) reviewed 43 cases of soft tissue myxoma of the head and neck region. In their study, men were affected more than women (59–41%) and the most common location was the palate, followed by the parotid area. In other studies also other locations like the cheek and the floor of the mouth were reported (Elzay & Dutz 1978, Barnes 2001). These tumours may occur almost in every decade of life, with a peak occurrence in the fourth decade (Tse & Vander 1985, Regezi & Sciubba 1999). They present macroscopically as grey-white, mucoid masses with a smooth or multinodular external appearance and they are usually encapsulated or circumscribed (Barnes 2001). Although commonly soft tissue myxomas are hypocellular and hypovascular, hypercellular and hypervascular variants may exist (Remstein et al. 1996, Nielsen et al. 1998).

Several theories concerning the pathogenesis of this tumour were proposed. The prevailing opinion was that altered fibroblasts or myofibroblasts could produce an excess of mucopolysaccharides and were commonly incapable of forming mature collagen even if some cells could retain this capacity (Barnes 2001). Another theory attributed the origin of these tumours to mesenchymal elements derived from dental papilla, dental follicle or periodontal membrane (Tse & Vander 1985, Gunhan et al. 1991, Shimoyama et al. 2000, Chang et al. 2001). However, the histogenesis of these lesions remains obscure and further studies are necessary to clarify its origin. Pathologically, it may be difficult to differentiate from other tumours with a myxoid stroma and is occasionally misinterpreted as malignant (Ramaraj & Shah 2003).

Because of the rarity of this lesion, we report a case of soft tissue myxoma located in the left maxillary adherent

gingiva. To our knowledge, only three other cases have been reported previously (Tahsinoglu et al. 1975, Chang et al. 2001, Iezzi et al. 2001) with no radiographic evidence of erosion of underlying bone.

Case Description and Results

A 42-year-old male presented at the Outpatient Department of the Dental School of the University of Chieti-Pescara complaining of a mass, which had appeared a few months previously, located on the left maxillary adherent gingiva in the lateral incisive area (Fig. 1). The lesion was asymptomatic. Clinical examination revealed a soft tissue mass measuring about 1 cm, with a tense elastic consistency on palpation. The overlying mucosa was normal. Radiologically, neither the erosion of the underlying bone nor other signs of radiolucency were present. A periodontal probing was performed with no evidence of localized or general periodontal disease; the preoperative clinical diagnosis was fibroma. Excisional biopsy was performed following administration of local anaesthesia, and the tissue (1.4 × 1 cm) was sent for histological evaluation. Grossly, the lesion was round, well circumscribed and had a pale mucoid cut surface. The lesion presented a gelatinous nodule of about 0.8 cm diameter with a white-grey colour in the central area. Necrosis and haemorrhage were absent. Histological examination showed spindle-shaped (bipolar) (Fig. 2a) and stellate cells, arranged in a myxoid fibrous stroma (Fig. 2b), with collagen fibres distributed uniformly. No mitoses were present. Scattered islands or strands of inactive odontogenic epithelium (Fig. 3a), occasionally surrounded by zones of hyalinization, were present (Fig. 3b). Moreover, prominent capillaries were present in the lesion. Immunohistochemical stains with S-100 protein were negative, while those with vimentin were positive. On the basis of these histological and immunohistochemical findings, the final diagnosis was soft tissue myxoma. No recurrences were present at a 4-year follow-up.

Discussion

Many characteristics make this myxoma case unusual. First of all, the location: only three other cases of myxoma of gingiva have been reported in the literature (Tahsinoglu et al. 1975, Shimoyama et al. 2000, Chang et al. 2001); moreover, all these were located in the mandibular gingiva, and our case is the first located in the maxillary gingiva.

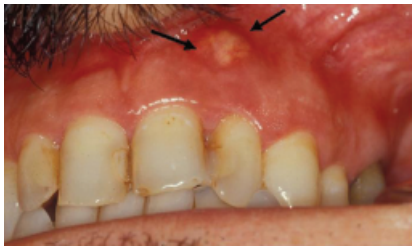

Fig. 1. Clinical appearance of the lesion (arrow) at first presentation.

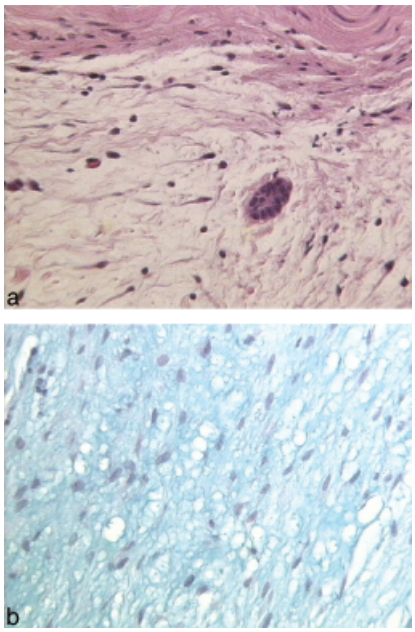

Fig. 2. (a) Soft tissue myxoma. Histological examination showed spindle-shaped cells in the central part of the tumour, haemotoxylin & eosin × 200. (b) Soft tissue myxoma. Microscopic analysis of the lesion showed spindle cells arranged in a myxoid fibrous stroma. Alcian blue staining × 400.

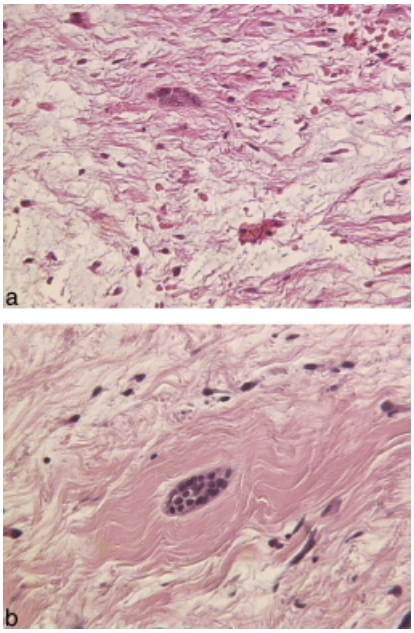

Fig. 3. (a) Soft tissue myxoma. histological examination showed scattered islands of inactive odontogenic epithelium with a moderate amount of collagen and some capillaries, haemotoxylin & eosin (H&E) × 200. (b) Soft tissue myxoma. Histological examination showed scattered islands of inactive odontogenic epithelium, occasionally surrounded by zones of hyalinisation. H&E × 200.

In our case, the clinical differential diagnosis was made in particular with traumatic fibromas (Regezi & Sciubba 1999), extra-osseous odontogenic fibromas, nerve sheath tumours and oral focal mucinosis (Lucas 1998, Barnes 2001). Traumatic fibroma is difficult to differentiate clinically; for this reason, it was our first clinical diagnosis. Microscopically, it is characterized by dense collagen fibres (Regezi & Sciubba 1999). Extra-osseous odontogenic fibroma contains strands of inactive odontogenic epithelium immersed in a stroma rich with collagen (Lucas 1998). Nerve sheath tumours are S-100 protein positive, whereas myxomas are usually (not always) negative for this marker (Barnes 2001). Lombardi et al. (1995) demonstrated that the spindle cells of two odontogenic myxomas were keratin, non-specific enolase, glial-specific protein, neurofilament and factor VIII-related antigen negative, but were strongly positive for S-100. This may cause difficulties in distinguishing myxomas from the above-mentioned tumour. By contrast, Moshiri et al. (1992) reported the spindle-shaped cells to be vimentin and actin positive but S-100 negative. However, odontogenic myxomas have a different pathogenesis from soft tissue myxoma, because they arise from primitive mesenchyme of tooth germ, after early induction of fibroblasts into odontoblasts. The majority of studies evaluated the expression of these markers in odontogenic, but not in soft tissue myxoma. Our study confirms the results of other studies (Moshiri et al. 1992; Chang et al. 2001) with regard to S-100 negativity and vimentin positivity. Oral focal mucinosis is

clinically indistinguishable, but, histologically the connective tissue is alcianophilic and with no reticulin fibres (Iezzi et al. 2001).

Awareness of potential diagnostic pitfalls and careful evaluation of clinical and radiological data are necessary to narrow the differential diagnosis.

Because soft tissue myxoma is a benign tumour, conservative surgical resection is the treatment of choice (Elzay & Dutz 1978, Tse & Vander 1985, Barnes 2001, Kumar et al. 2002, Ramaraj & Shah 2003). The recurrence rate is 3–8%, and the neoplasm is most likely to recur within 2 years; hence, close follow-up is required (Kumar et al. 2002). The prognosis of this soft tissue tumour is good (Tse & Vander 1985).

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Clinical Relevance

Scientific rationale: Not many details are available in the literature related to soft tissue myxomas, if compared with their counterpart, central odontogenic myxomas. Because of the rarity of this lesion, data collection on tumour behaviour and appropriate treatment are feasible only through case reports.

Principal findings: We decided to report a case of soft tissue myxoma, that could be of interest to a Periodontist for its location (maxillary gingiva) and its clinical differential diagnosis with lesions with a different behaviour, such as traumatic fibromas, extra-osseous odontogenic fibromas, nerve sheath tumours and oral focal mucinosis.

Practical implications: Awareness of this lesion, combined with a careful histological analysis, can help clinicians to avoid diagnostic pitfalls and to pursue the appropriate treatment.