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CASE REPORT

Locally aggressive central odontogenic fibroma associated to an inflammatory cyst: a clinical, histological and immunohistochemical study

Iñaki Cercadillo-Ibarguren[1], Leonardo Berini-Aytés[1], Vicente Marco-Molina[2], Cosme Gay-Escoda[3]

[1]*Oral Surgery and Implantology Department, Barcelona University Dental School;* [2]*Service of Pathology, Catalonia General Hospital, Sant Cugat del Vallés;* [3]*Oral and Maxillofacial Surgery Department, Teknon Medical Center, Barcelona, Spain*

The case describes a 38-year-old woman presenting a multilocular radiolucency affecting the entire right half of the lower jaw, with an unerupted third molar displaced to the region of the coronoid process. The histological study showed the presence of fibroblasts, focally with pleomorphic nuclei, dense collagen and an odontogenic epithelium with dystrophic calcifications. A cyst with an important inflammatory infiltrate was, moreover, observed.
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Case report

A 38-year-old woman had been referred us by her dentist following the casual X-ray identification of a radiolucent area occupying the right hemimandible. For the previous 4 years the patient had suffered sporadic gingival swelling in the distal region of the lower right second molar, accompanied by the drainage of fluid into the mouth. Orthopantomography and computed tomography (CT) revealed the presence of a multilocular radiolucency measuring 4.5×6.0 cm^2 and extending from the root zone of the lower right first molar to the coronoid process, in relation to a third molar displaced to that position. Slight reabsorption of the distal root of the lower right first molar was observed and both bucal and lingual bone cortexes were preserved (Figs 1–3). Exploration revealed expansion of the bucal and lingual bone cortexes of hard consistency accompanied by suppuration in response to probing distal to the lower right second molar.

Correspondence: Dr Cosme Gay-Escoda, Centro Médico Teknon, Vilana 12, 08022 – Barcelona, Spain. Tel: +34 9339 33170. Fax: +34 9339 33070. E-mail: cgay@ub.edu
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The lesion was resected under general anaesthesia, and the lower right first and second molars were removed, together with the unerupted third molar displaced to the coronoid process. The surgery was completed by curettage of the remaining bone bed. Three irregular tissue fragments were obtained, globally measuring 6×6 cm^2 in size, of a bright whitish colour, elastic-rubbery consistency and fibrous appearance (Fig. 4).

Sections were stained with haematoxylin/eosin and immunohistochemistry was performed using avidin–biotin complex technique, with LSAB2 peroxidase kit in a DAKO Autostainer (DAKO Corp., Carpinteria, CA, USA). The immunohistochemistry panel included antibodies against cytokeratin AE1/AE3 (DAKO, prediluted), p63 (DAKO, 1/50), Vimentin (DAKO, 1/200), CD34 (DAKO, prediluted), c-kit (DAKO, 1/50), Desmin (DAKO, prediluted), smooth muscle actin (DAKO, 1/50), CD 68, anti-human macrophage (DAKO, prediluted) and S100 (DAKO, 1/500). Microscopic examination showed a tumour mass formed by partly hyalinized dense collagenous tissue with proliferation of fibroblasts. The fibroblasts showed medium-sized fusiform nuclei; focally, large pleomorphic nuclei were noted, some with stellated cytoplasm. Multiple clusters of odontogenic epithelium were present in the tumour, some showing dystrophic calcification. The surface epithelium was invaginated in the tumour forming an inflammatory cyst, which was lined by focally ulcerated hyperplastic squamous epithelium. The epithelium-surrounded eosinophilic material (Rushton bodies) and foreign bodies, including vegetal fibres and amalgam material. The immunohistochemical study showed the fibroblasts to be positive for vimentin and c-kit, while desmin and smooth muscle actin proved negative. Some fibroblasts showed S100 positivity. CD68 highlighted the presence of macrophages. The odontogenic epithelium and squamous epithelium lining the inflammatory cyst showed positivity for both cytokeratin AE1/AE3 and p63 (Fig. 5).

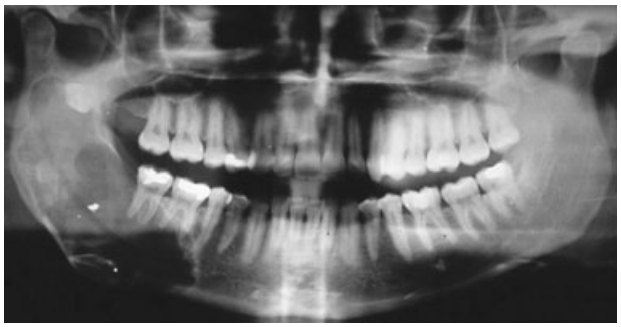

Figure 1 Orthopantomographic view showing a multilocular radiolucency occupying almost the entire right hemimandible, with the presence of a third molar displaced to the coronoid process. Slight reabsorption of the distal root of the lower right first molar is noted, along with the presence of a radiopaque foreign body in the centre of the lesion.

Comments

Histologically, central odontogenic fibromas (COFs) are regarded as benign fibroblastic neoplasms characterized by the presence of mature collagen with numerous interposed fibroblasts and variable amounts of apparently inactive odontogenic epithelial nests (1). This type of neoplasm has been described 68 times in the latest reviews in the literature (2), and in only one case was it seen to present pleomorphic fibroblasts (3) with association to an inflammatory cyst – as in our patient (4). Although the lesion has been diagnosed in individuals ranging from 5 to 80 years of age, it is more frequent in the third and fourth decades of life. A clear predilection for the female sex has been observed (5, 6). In general, the most common radiological finding of COF is a uni- or multilocular radiolucency with well-defined margins, related to the adjacent teeth and displacing them in 55% of cases or causing root resorption in 29% of patients (6).

As already suggested by Handlers et al. (5), despite the existence of the same histological pattern, those lesions apparently formed from the dental follicle may exhibit a greater growth potential than tumours arising from the periodontal ligament. In any case, we consider that this hypothesis should be contrasted with the age of the patient at the time of diagnosis of the lesion, as in younger patients evolution of the tumour has been briefer, and the attainment of a large size in such individuals would imply a greater growth potential.

One of the most interesting aspects of the tumour described in our study was the observation of an extremely dense collagen stroma reminiscent of the

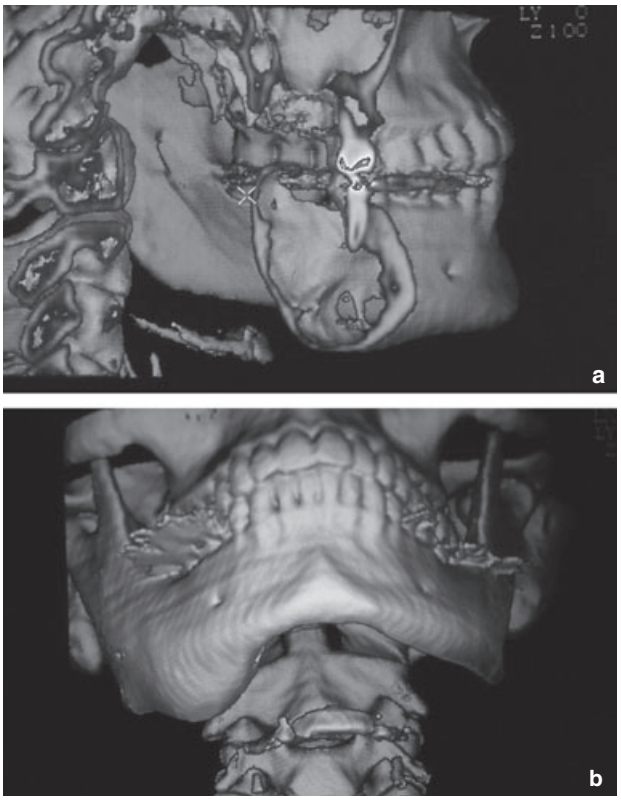

Figure 3 Three-dimensional computed tomography (CT) reconstruction showing expansion of the right-side mandibular base (a) and the extent of the lesion with preservation of the cortex (b).

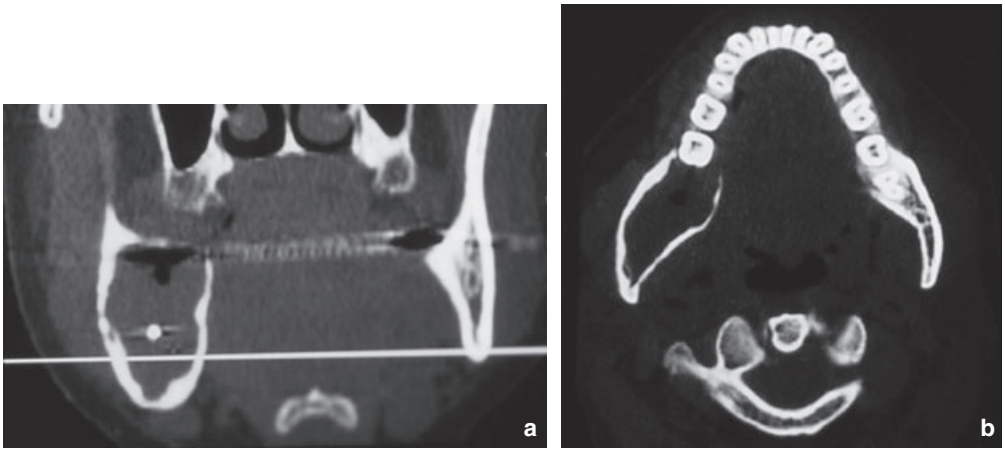

Figure 2 Computed tomography (CT) view showing occupation of the mandible and important expansion of the preserved bone cortex. (a) Coronal view. (b) Sagittal section.

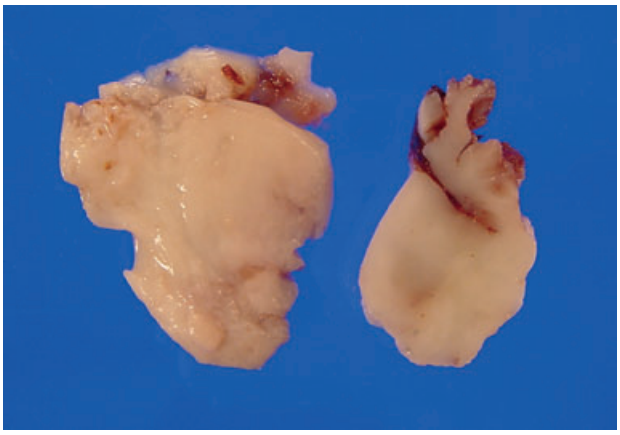

Figure 4 Macroscopic view of the lesion, of a whitish colour, fibrous appearance and rubbery texture. The cut surface shows homogeneous, glistening tissue. A concave upper surface corresponds to the wall of the inflammatory odontogenic cyst.

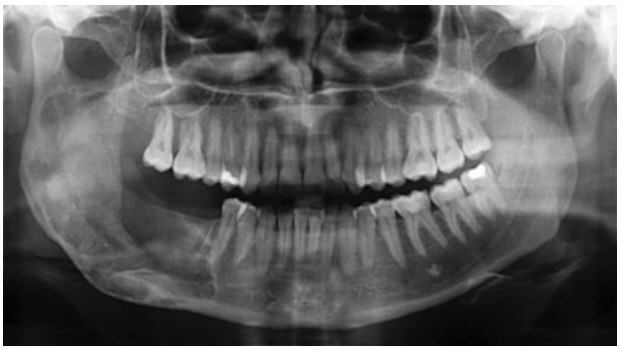

Figure 5 Control panoramic X-ray view 18 months after surgical treatment.

histological pattern found in desmoplastic fibroma. However, the variable amounts of odontogenic epithelial nests, the numerous dystrophic calcifications and proximity of an unerupted third molar, clearly indicate an odontogenic origin of the lesion. Nevertheless, the latter could be more aptly described as being desmoplastic, because of the greater than usual presence of collagen. The association in our patient between COF and an inflammatory cyst may provide clues to the concrete aetiopathogenesis involved – suggesting that the cyst may have been the primary lesion that stimulated COF development. The presence of an important inflammatory infiltrate in the cyst, and the enveloped silver amalgam and plant fibres, support this hypothesis – indicating the presence of a cyst lesion prior to the actual COF.

Another observation of note was the presence of large pleomorphic fibroblasts with a strongly manifest nucleolus. Considering the unusual association of COF and pleomorphic fibroblasts, it is difficult to assess the

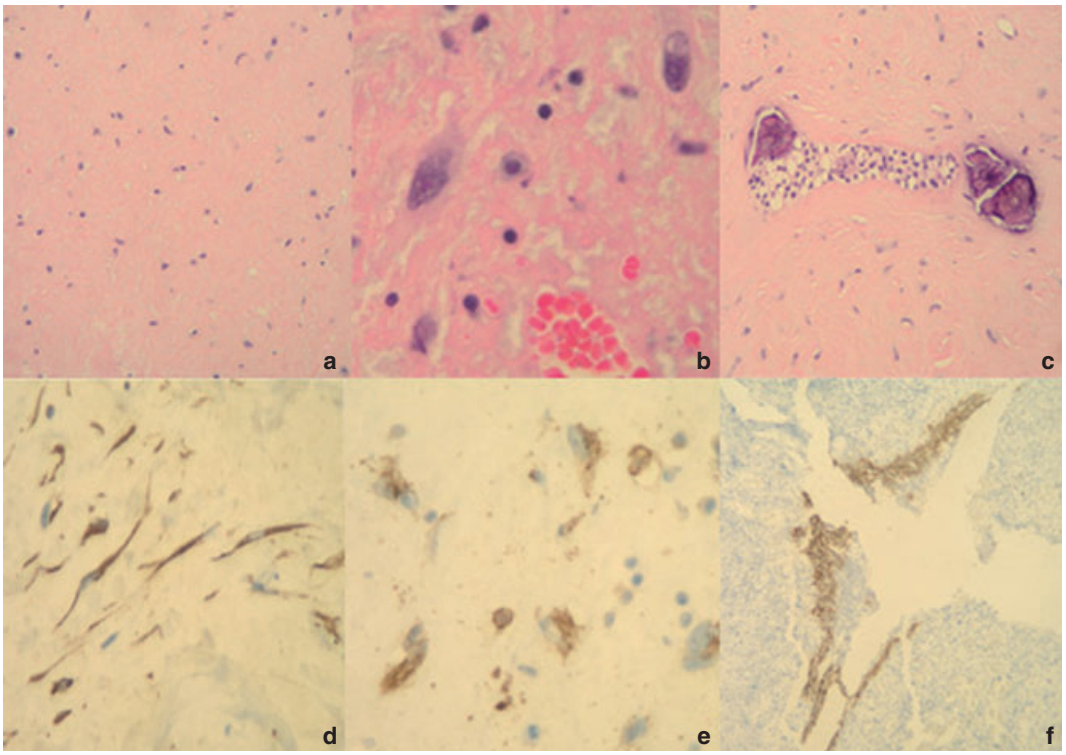

Figure 6 (a) Presence of a fibroblastic proliferation and dense collagen stroma (H&E, 40×). (b) Presence of pleomorphic fibroblasts and inflammatory cells, including mast cells (H&E, 400×). (c) Odontogenic epithelial nests and dystrophic calcifications (H&E, 200×). (d) The fibroblasts show positive staining for vimentin (Immunostain, 200×). (e) The cytoplasm of fibroblasts and mast cells shows c-kit positivity (Immunostain, 200×). (f) The odontogenic cyst appears surrounded by a squamous epithelium with positive staining for cytokeratin AE1/AE3 (Immunostain, 40×).

biological behaviour of this histological variant. However, in the only similar case described in the literature by Günhan et al. (3), no recurrences were observed 5 years after conservative surgical treatment in the form of lesion exeresis and curettage. This would support the idea that this histological variant does not imply a difference in biological behaviour of the tumour. In our case, follow-up during 18 months after surgery proved normal with good bone healing and no signs of recurrence (Fig. 6). The five cases of tumour recurrence described in the literature appear to have no common features that would allow the definition of an aggressive pattern or the prediction of recurrence (4). However, preoperative histological diagnostic error and the application of an inadequate surgical technique are the two factors most often described as possible causes of lesion recurrence. In this context, conservative surgical management of COF is recommended, with tumour resection and curettage of the residual bone bed – provided the lesion has been shown to be non-aggressive, with the absence of histological evidence of malignancy. Although the tendency towards recurrence is very low, postoperative patient follow-up for 5 years after surgery is advisable.

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