Abstract. The aim of this work was to describe a case of unicystic ameloblastoma located in the left posterior mandibular body and angle of a 14-year-old Caucasian male. The tumour was described at clinical and histological level; moreover, its histopathological phenotype was depicted. Finally, the problems of differential diagnosis with odontogenic or congenital cyst and the most appropriate therapeutic procedures are discussed.

Ameloblastoma is an epithelial tumour of enamel-organ types of tissue. In the classification of head and neck tumours by the World Health Organisation (1), it has been classified within the benign odontogenic tumours with the odontogenic epithelium associated with mature fibrous stroma without odontogenic ectomesenchyme. It represents 10% of all odontogenic tumours (2). The benign ameloblastomas may be classified into four clinical types: solid or multicystic (A-S/M), unicystic (A-U), desmoplastic (A-D) and extraosseous-peripheral (A-E/P) (1-3). Ameloblastoma originates in the mandible in 80% of all cases and nearly three quarters of these cases occur in the ramus and the posterior body of the mandible (2). Males and females are affected almost equally, with a male-to-female ratio of 1:1.3 (2). A-S/Ms are common during adulthood, while A-U is common in paediatric age and during adolescence. The ameloblastoma grows insidiously as a central lesion of the bone, being slowly destructive (3). It is common for an ameloblastoma to persist for many years without any clinical sign (1, 2, 4).

There are two most common histopathological types of ameloblastoma, the follicular and the plexiform types. The follicular type has columnar hypercromatic and vacuolated cells lined up in the cystic-like lesion and in which the central cellular groups may resemble the stellate reticulum of the enamel organ. The plexiform type is characterized by a tenuous stroma with columnar cells forming regular strands and less defined images of stellate reticulum (1, 4-6). The therapy of choice is surgical excision and its target is to prevent local recurrences (7, 8).

The A-U type of benign ameloblastoma is more common in paediatric age and during adolescence. It may be observed casually or after the appearance of non-specific symptoms, such as dentigerous cysts clinically and radiologically associated with unerupted teeth, or odontogenic cysts (3, 4, 9). Calretinin is an immunomarker for ameloblastoma that is useful for the differential diagnosis with keratocystic odontogenic tumour (10-12). Among neoplastic immunomarkers, PCNA and Ki-67 do not have a clear role for diagnosis of ameloblastoma (13). Therefore, the histological examination is the most certain tool for diagnosis of ameloblastoma.

Case Report

A 14-year-old male was referred to the Oral Surgery Department of the Dental School of the Second University of Naples (Naples, Italy) in July 2008 for the treatment of a cystic lesion. The patient presented with indolent swelling of the left cheek approximately two months prior to the hospital admission. After panoramic and dental check-up, a lesion was found located in the left posterior mandibular body and angle. The previously unknown lesion did not cause other symptoms and did not cause any problem in mastication. The panoramic radiograph (Figure 1A) showed a large unilocular radiolucent area between the first and second molars with involvement of the periapical area, clearly defined margins and the presence of the second unerupted molar. Expansion of both the buccal and the lingual plates was noted after a clinical intra-oral examination and computerised tomography (CT) dental (Figure 1B) and 3D (Figure 1C) scans. A unicystic appearance and perforation of the buccal cortical plate was also observed, but no resorption of the roots was noted. Clinically, the plate was slightly knobby, the overlying mucosa appeared normoechoich, normotrophic and not

Unicystic Ameloblastoma of the Mandible

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bloody, and the area was not compressible and was painless on palpation. Regional lymph nodes were not involved. The therapy of the lesion consisted of surgical conservative treatment. First, intra-oral incision was performed and, after adequate surgical exposure that permitted exposure of the lesion, an enucleation of the cystic lesion was performed (Figure 1D). On the lingual side, the periosteum was integral. A meticulous curettage of all surrounding bone was performed to ensure complete removal of any remaining tumour. The bone cavity was then examined for any remaining tissue and this was followed by copious irrigation with antibiotic solution. The involved teeth, without mobility, were not extracted. The flap was sutured primarily and the surgical wound healed without any complications. The patient was disease-free at one-year follow-up.

The surgical specimen measured 2×1.7 cm (Figure 1E). The excised biopsy tumour specimen was fixed in 10% buffered formalin and was paraffin-embedded. Five μm thick sections were stained with haematoxylin-eosin, haematoxylin-van Gieson and PAS-haematoxylin. Microscopic examination showed a papillary proliferation of metaplastic squamous epithelium. The proliferative epithelium resembled the hyperplastic lining of inflammatory odontogenic cysts and consisted of interconnecting cords supported by a delicate vascular connective tissue stroma (haematoxylin-eosin staining; original magnification ×20). G: Panoramic radiograph after one year, showing no recurrence of the lesion.

Discussion

In the presence, on a panoramic radiograph, of a unilocular radiolucent suspicious lesion of the mandible with or without associated teeth, it is necessary perform a CT examination. In the case of a large cystic lesion resembling an A-U, a biopsy may be insufficient to make a histological diagnosis for the presence of normal epithelium in the A-U. From a histological perspective it is possible to find, in all types of ameloblastoma, a basal layer of columnar pre-ameloblastic cells with polarisation of hyperchromatic basal cells, nuclei with a vacuolation of clear basal cell cytoplasm and a more superficial loose stellate reticulum-like epithelium. Basal palisades and intercellular oedema are features that characterize the lesion in this case report (1).

An A-U does not require radical surgery as is the case for dentigerous cysts (14). If the surgeon suspects that the lesion may be a real ameloblastoma, the area can be treated with local resection (en bloc resection) as a preventive measure; however, such procedure is disfiguring and distressing. This procedure may be chosen if the lesion has a small diameter or if it resembles a non-dentigerous cyst. When the clinical and radiological appearance is that of a suspicious A-U, then, on removal of the lesion, the surgeon should request a careful histological examination of more sections to avoid misdiagnosis of an A-U case. However, enucleation is the treatment of choice for cyst-like lesions, followed by a careful curettage of the remaining bone cavity, with additional means, such as Carnoy’s solution. The surgical excision with peripheral osteotomy (15) depends on the
surgeon's operative planning. Moreover, peripheral osteotomy (1-1.5 cm around the A-U) may not be performed in order to preserve bone continuity. Nevertheless, osteotomy around the tumour can be performed carefully by using rotatory bone-instruments 2-4 mm around the A-U. The wellbeing of the mandibular cortical bone is considered an important anatomic barrier for advancing tumour. When the A-U involves nervous structures in the bone, such as the mandibular nerve, a piezo-surgery technique (16) that is able to cut the bone with minimum tissue damage, may be indicated.

The postoperative management of A-U, as has been suggested by several reports (1, 4, 8, 10, 11, 13, 14, 16, 17), is a routine clinical and radiological follow-up that is required for many years.

A number of odontogenic cysts may be confused with A-U. Indeed, in the case presented, a differential diagnosis with odontogenic cysts as keratocyst, odontogenic tumours as ameloblastoma, non-odontogenic cysts as dermoid cysts, and cyst-like lesions as traumatic bone pseudo-cysts was proposed. The lesion was located between the second and third molar, without resorption of the roots and CT imaging showed a large radiolucent area on the left mandibular posterior region. The A-U was a possible diagnosis considering the young age of the patient and the clinical-radiological features. A complete surgical excision was conducted preserving the unerupted tooth, because it was presumed that the clinical features of the lesion were those of a benign lesion with less aggressive form. The literature regarding A-U suggests that most recurrent tumours are of mural type, where the connective tissue invasion is considered to be an important histological sign of recurrence (3). A histopathological diagnosis before a surgical treatment is not possible in all cases. Therefore, the choice of the treatment varies with: the clinical and radiological features (that may permit the initial diagnosis of a dentigerous cyst), the age of the first presentation and whether there is an association with an impacted tooth that occurs earlier and more frequently than in isolated A-U. It is, therefore, suggested that because the diagnosis of A-U may be made in a postoperative step by a careful histopathological examination of more sections, and because some A-Us may be derived by changes of the mural or lining epithelium in pre-existing dentigerous cysts, every odontogenic cyst of any dimension and all itsenucleated fragments should be referred to a pathologist (16, 17). In the case of a histological diagnosis of ameloblastoma, a long period of follow-up is recommended.

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