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Paediatric oral surgery: endoscopic approach in ameloblastic fibroma management. A preliminary report

ABSTRACT

Background *Ameloblastic fibroma (AF) is a rare benign odontogenic tumor of slow-growing behaviour characterised by proliferation of both epithelial and mesenchymal odontogenic tissues. Treatment of AF is usually conservative by means of enucleation, curettage and long term follow-up.*

Case report *This report describes a case of AF in a 12-year-old boy. Examination of the oral cavity revealed absence of tooth 3.7 and mild swelling in the same mandibular area. Orthopantomography and Cone Beam Computed Tomography (CBCT) showed a wide multilocular radiolucent lesion at the left mandibular angle, extending from the first molar to the lower part of the mandibular branch; agenesis of tooth 3.7 and displacement of tooth 3.8. Surgical excision was performed under general anaesthesia. A fiberscope was used to perform a more conservative bone removal and extraction of tooth 3.8. Furthermore, an accurate curettage of the bone site was performed. The histological investigation showed an AF pattern: an epithelial cell component arranged in nests and tubules immersed in a stroma of low differentiated mesenchymal tissue. No sign of recurrence has been observed during the 6-month follow-up period.*

Conclusion *Although AF is a rare tumor, it is more prevalent in children's jaw. A new intra-operative visual technique could provide a conservative treatment by minimising bone deformities and permitting an accurate bone curettage.*

Keywords *Ameloblastic fibroma; Endoscopic surgery; Odontogenic tumors.*

Introduction

Ameloblastic fibroma (AF) is a rare, benign neoplasm of odontogenic origin, comprising about 1.5–4.5% of all odontogenic tumors [Philipsen et al., 1997; Chen et al., 2007; Melo et al., 2015]. AF is characterised by the proliferation of both epithelial and mesenchymal odontogenic tissues without the formation of hard tissue structures [Kramer et al., 1993]. As described by the World Health Organization (WHO), it is "composed of odontogenic ectomesenchyme, reminding of the dental papilla and epithelial strands and nests, reminding dental lamina and enamel organ: there are no hard tissues appearance" [Barnes et al., 2005].

AF occurs with a slightly higher prevalence in males than females (1.4/1) [Cohen and Bhattacharyya, 2004]. Moreover, this tumor is frequently seen in the first and second decades of life; 77.7% of cases are diagnosed before the age of 20, and it is primarily considered a tumor of childhood and adolescence [Philipsen et al., 1997; Cohen et Bhattacharyya, 2004], although in literature a wide age range (0.5–62 years) is reported [Tozoglu et al., 2016]. AF is frequently encountered in the posterior mandible, with 80% of cases in the second primary molar or first permanent molar region and 75% associated with an impacted tooth; moreover, it is rarely diagnosed in the upper jaw [Ealla et al., 2015].

As above mentioned, AF has no dental hard tissue [Barnes, 2005]; if in the lesion is found dentinoid tissue without or with enamel formation, it could be ameloblastic fibrodentinoma (AFD), ameloblastic fibro-odontoma (AFO), or fully mineralised odontoma (complex and compound), which are mixed odontogenic tumors [Praetorius, 2009].

The AF is usually discovered during routine oral examination or an orthopantomography evaluation: it is characterised by an asymptomatic, unilocular, small radiolucent lesion. Larger lesions usually have a multilocular and radiolucent x-ray pattern, and may cause painless swelling [Peter and Hans, 2004].

Histopathological features of AFs are as follows: a tissue made of both epithelial and connective components and characterised by plump fibroblasts and delicate collagen fibrils similarly to the dental papilla [Kobayashi et al., 2005]. The epithelial component,

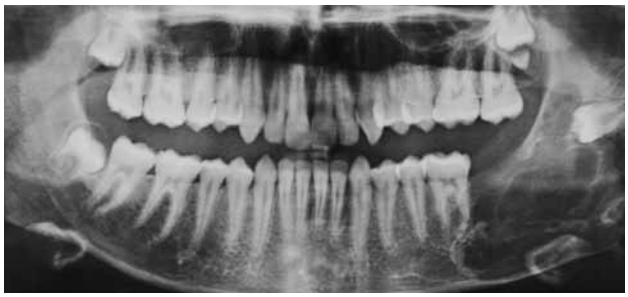


FIG. 1 Orthopantomography at baseline clinical situation.

which resembles embryonic dental lamina, is arranged in various thin branching cords or strands with double or triple layers of cuboidal cells as well as small nests or islands with scanty cytoplasm and basophilic nuclei, while stellate reticulum-like cells are common in larger nests [Peter and Hans, 2004].

Treatment of AF is surgical enucleation, usually involving a conservative approach [Ferrazzano et al., 2014; Ferrazzano et al., 2017], with a curettage of the surrounding bone, but sometimes extraction of the involved tooth is unavodable [Melo et al., 2015].

In this case treatment entailed a new approach to reduce the bone loss necessary to enucleate the lesion: through a fiberscope and a monitor (Video-rino-laringoscope CCD model 11101 VP 3.7 mm diameter; Karl Storz, Switzerland), the surgeon was able to visualise the intra-bone alveolar cavity, after removal of the main tumoral mass, in order to check for the presence of peripheral nuclei of the lesion and to assess the exact position of the included molar.

This new approach is not feasible in case of aggressive lesions, which require a radical approach [Nelson and Folk, 2009]. There are also conflicting reports regarding the recurrence and malignant transformation rates of AF [Kobayashi et al., 2005; De Oliveira et al., 1997]. Most of the reported cases underscore the need for a long-term follow-up to exclude the risk of tumor recurrences [Martín-Granizo López et al., 2003; Vasconcelos et al., 2009; Melo et al., 2015].

Case report

LS, a 12-year-old boy, was examined at the Department of Paediatric Dentistry of "Federico II" University of Naples on November of 2016, referred by his private dentist for a radiolucent lesion at the left mandibular angle, observed on an orthopantomography.

The remote medical history was non contributory. The extra-oral clinical examination did not reveal any skin affection or swelling. The intra-oral examination showed agenesis of tooth 3.7 and a mild swelling in the same mandibular area. The orthopantomography and Cone Beam Computed Tomography (CBCT) showed a wide multilocular radiolucent lesion at the left mandibular angle (Fig. 1-2), extending from the first molar to the lower part of the mandibular branch. The lesion probably displaced tooth 3.8, which was impacted in the mandibular branch. In addition, agensis of tooth 3.7 was noticed. The analysis of the sagittal frames of CBCT showed a thin residual cortical bone which allowed to

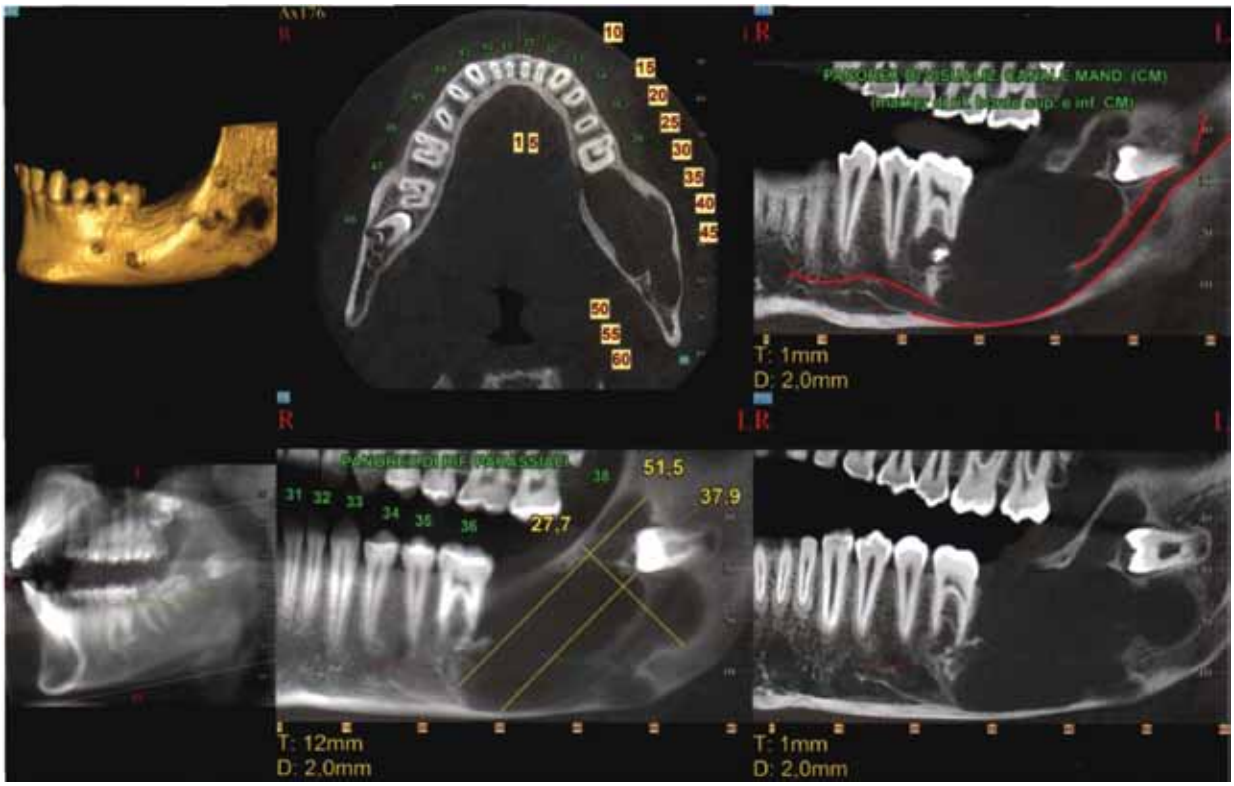


FIG. 2 CBCT scans of mandibular bone.

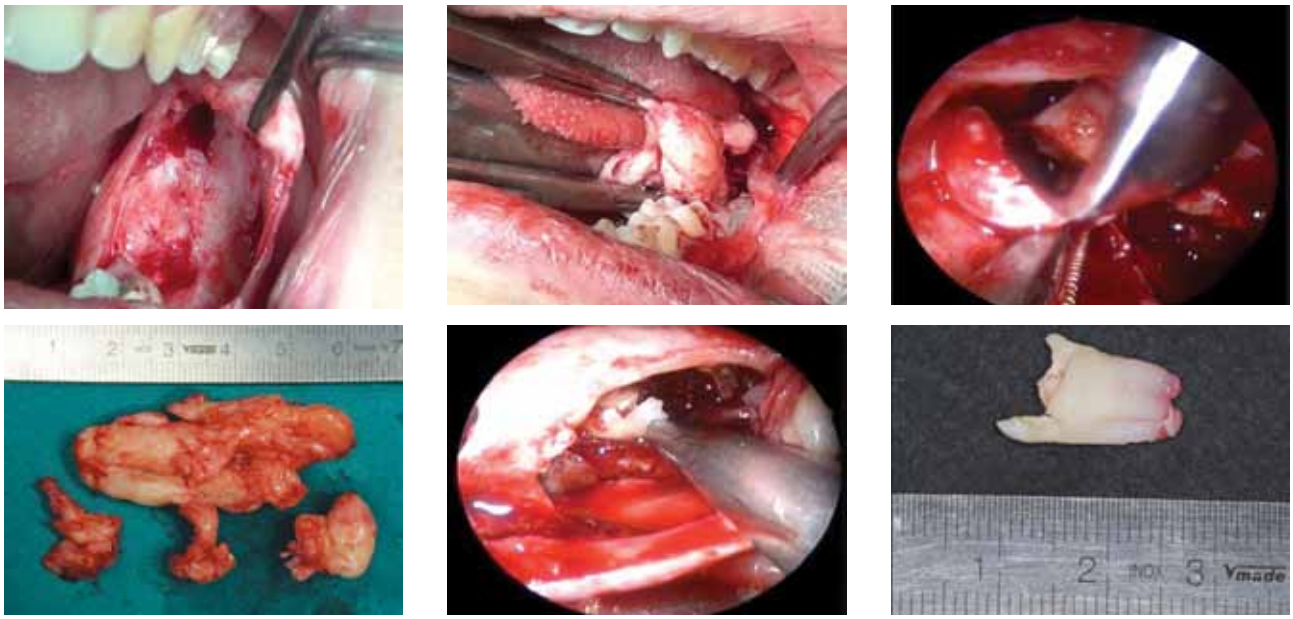


FIG. 3 surgical procedure and sample measurement.



FIG. 4 Video-rino-laringoscope CCD model 11101 VP 3,7mm diameter – Karl Storz (Switzerland)



FIG. 5 Macroscopic inspection of the tumor specimen.

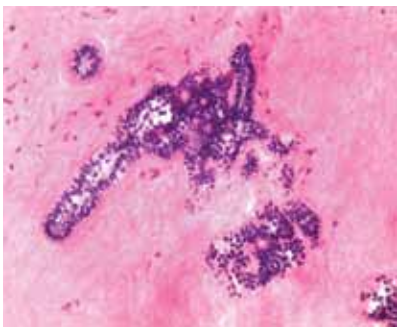


FIG. 6 Histological investigation (20x) that showed an ameloblastic fibroma pattern.

plan a new conservative surgical approach.

Surgical excision was performed under general anaesthesia, by means of intra-alveolar access and enucleation of the primary lesion (Fig. 3), without removing wide areas of vestibular cortical bone; moreover, through a fiberscope (Video-rino-laringoscope CCD model 11101 VP 3.7 mm diameter; Karl Storz, Switzerland) (Fig. 4) connected to a monitor, the intra-alveolar cavity was visualised in order to verify the presence of peripheral nuclei of the lesion and the exact position of the impacted 3.8 molar, performing a more conservative bone removal and tooth extraction; then, through the fiberscope, an accurate curettage of the bone site was performed.

Macroscopic inspection of the tumor specimen, which measured 60×40×40 mm, revealed an irregular surface and fibroelastic consistency (Fig. 5).

Then the lesion tissue was submitted to histo-pathological examination. The histological investigation showed an ameloblastic fibroma pattern: a stroma of low differentiated mesenchymal tissue, in which an epithelial cell component arranged in nests and tubules was immersed (Fig. 6). At the peripheral areas of the lesion, it was observed an undifferentiated bone-like tissue with scanty epithelial cells (Fig. 7).

No clinical complications were observed during the postoperative period. The patient was followed-up monthly over a period of 6 months and no clinical or radiographic signs of recurrence were observed: after 6 months the ortopantomography showed the formation of new bone in the left posterior region of the mandibular body.

Discussion and conclusion

This case report showed an AF involving the body, angle and branch of the mandible in a 12-year-old child.

During the clinical examination, the patient exhibited intraoral swelling extending from the left first molar to the mandibular branch on the same side. According to Melo et al., [2015] this swelling is the result of the gradual expansion of the cortical bone during growth of the lesion. In addition, these authors have reported that AF shows slower growth than other odontogenic tumors, such as ameloblastoma, and does not tend to

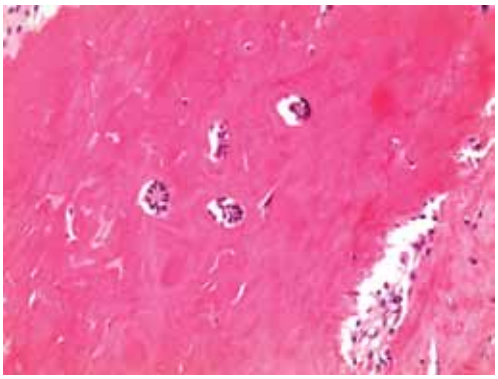


FIG. 7 Histological investigation of peripheral areas of lesion (10 x), where undifferentiated tissue similar to bone tissue was observed in which little nests of epithelial cells were immersed.

infiltrate the bone. As above mentioned, the differential diagnosis of AF includes dentigerous cyst, odontogenic keratocystic tumor, myxoma and ameloblastoma, due to the clinical and, above all, radiographic similarities between these lesions. In fact, the clinical intra-oral examination of the case reported showed mild swelling in the mandibular angle and the radiographic multilocular pattern was very similar to that of other referred lesions [Ozkan et al., 2014; Sasaki et al., 2012].

According to Nelson and Folk [2009], despite many similarities, it is crucial to differentiate AF from other mixed odontogenic tumors in view of its neoplastic potential, possibility of recurrence and malignant transformation potential. The histopathological features of the present case met the criteria for AF [Slootweg, 2005]: an epithelial cell component arranged in nests and tubules immersed in a stroma of low differentiated mesenchimal tissue. In the peripheral areas of the lesion, it was also observed an undifferentiated bone-like tissue, with little nests of epithelial cells. Other authors [Usubütün et al., 2002] also found extensive cystic formation as a result of a degenerative process, but this was not observed in the present case.

The patient was submitted to conservative treatment considering that the lesion was a primary tumor and to preserve as much bone as possible in order to avoid aesthetic deformities. Using this new technique with fiberscope, to visualise the bone cavity, surgeons could minimise buccal cortical resection and safely remove the peripheral lesion entirely. Moreover, en bloc resection results in significant morbidity and facial deformity and should be reserved for extensive tumors invading soft tissue and for recurrent cases [Vasconcelos et al., 2009]. In our case, 6 months after enucleation of the tumor the surgical cavity showed a radiographic pattern of sound regenerated bone.

AF is an odontogenic benign neoplasm that can be safely removed in a conservative manner. Using a new intra-operative visual technique, it is possible to minimise bone deformities, and ensurie an effective vision of the operating site in order to perform an accurate bone curettage.



FIG. 8 Control orthopantomography after 6 months.

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