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Case Report

Ameloblastic fibro-odontoma located in the maxilla of a 3-year-old girl

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ABSTRACT

Ameloblastic fibro-odontoma (AFO) is a rare benign tumor which contributes to 1–3% of all odontogenic tumors. AFO is mainly localized in the posterior region of the mandible and typical patients are children who present to their dentists due to non-erupting teeth. We describe an unusual example of a large AFO located in the upper right maxilla and maxillary sinus of a 3-year-old girl. The aims of this case report are to strengthen the knowledge of and to stimulate the discussion about AFO and to point out, that a conservative surgical procedure with regular follow up is highly recommended.

1. Introduction

Contributing to only 1–3% of all odontogenic tumors [1–4] the ameloblastic fibro-odontoma (AFO) was first described by Hooker in 1967 and distinguished from the more aggressive ameloblastic odontoma (AO), also called odontoameloblastoma [5]. According to the revised World Health Organization (WHO) Classification of odontogenic tumors of 2005, the AFO is described as a rare mixed odontogenic tumor which is usually benign [6]. The AFO shares similarities with the ameloblastic fibroma (AF) and the ameloblastic fibrodentinoma (AFD) but may be distinguished from these two entities by the presence of dentin and enamel on histological examination [6–8].

A current study describes a mean age of 9.6 years which underlines the fact that AFOs are predominantly found in children, and comprise around 7% of all odontogenic tumors under the age of 16 [3,9]. Commonly, these lesions are found on routine radiography of patients with delayed eruption of teeth or in patients presenting with asymptomatic intraoral swellings [10].

Radiographs generally show well-defined radiolucent areas containing various amounts of radio opaque material of irregular size and form with the radiopaque areas dominating in most cases [4,5,10–12]. Earlier tumors may initially present

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as radiolucent areas only and could show calcification in later stages of development [13].

Even though there seems to be a clear definition after the WHO Classification of 2005 concerning AF, AFD and AFO, there are still many controversies regarding the etiology, histology, therapy and prospective outlook [4,14]. Our case report aims to further the discussion and classification of such tumors by presenting a patient with a large ameloblastic fibro-odontoma involving an unusual location, namely the maxillary sinus.

2. Case report

A three-year-old girl was referred to the Department of Pediatric Dentistry at the University of Zurich by her private dentist after the mother noted a swelling in the upper right maxilla and non-eruption of a tooth in the related area. Non-eruption of the first deciduous molar as well as a hard, tumorous entity in the described region was clinically seen by her private dentist and a large opacity was seen on the dental radiograph (Fig. 1). The dentist was concerned for an infection of the sinus cavity and referred the patient to the Pediatric Dentistry Department, University of Zurich.

The child was a healthy, well-nourished girl with an unremarkable social, medical and family history. Additionally there were no related histories or clinical signs of trauma or infection in the described area and oral inspection showed adequate oral hygiene.

Clinically there was a slight swelling in the region of teeth 54 and 55, with tooth 54 being absent. Palpatory examination however revealed a non-tender, bony hard high buccal swelling in the region of the right maxilla. The orthopantomograph revealed an erupted tooth 55 but instead of tooth 54 a complex of predominant opacities and more peripheral radiolucent areas could be detected. The lesion extended into the maxillary sinus with conforming tooth displacement (Fig. 2).

The differential diagnosis from the pediatric dentist included an odontoma and therefore the patient was sent to the Department of Cranio-maxillofacial and Oral Surgery of the University Hospital of Zurich for further examination.

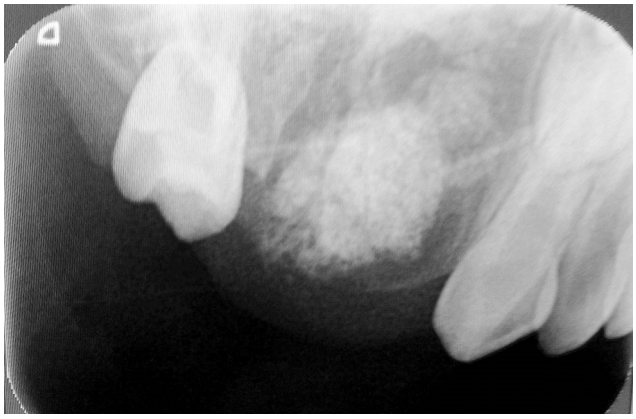

Fig. 1 – Pre-operative dental film showing absent tooth 54, central radio-opaque area surrounded by a radiolucent area.

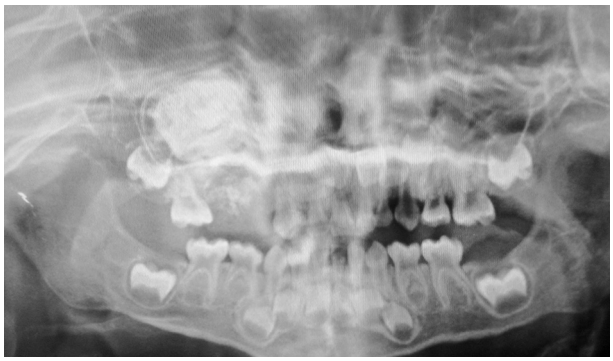

Fig. 2 – Orthopantomogram view showing the comparative obliteration of the right maxillary sinus.

Volume tomography (Fig. 3a, b and c) revealed a large calcifying tumor of the upper right maxilla involving the 54/55 area, reaching into the upper right maxillary sinus and filling out the latter nearly completely with conforming tooth displacement. A biopsy was performed under general anesthesia including bony-like fragments and firm white-gray tissue- fragments measuring 18 × 18 × 9mm.

Histopathological examination revealed a composite tumor of well-formed dentin and enamel lying in a cell rich myxoid stroma without mitotic activity consistent with the diagnosis of an ameloblastic fibro-odontoma (Fig. 4a, b).

The tumor was resected two months later under general anesthesia at the Children's Hospital of Zurich. Access to the tumor was obtained via an extended lateral buccal vestibular incision. Wide, sub periosteal exposure of the lateral and anterior wall of the maxillary sinus was achieved and the inferior orbital nerve was isolated and protected. The lateral wall of the maxillary sinus was removed and the tumor was identified from the maxillary alveolus up to the cranial extension thereof. Due to its large size, the tumor had to be segmented with a bur and chisel and then removed. Due to the clinical and radiological examination it can be presumed that the permanent tooth follicles were involved in the pathological process and partial anodontia will result in this area. The defect was not filled with any material and direct closure was achieved with resorbable sutures, avoiding the risk of an oro-antral communication.

The postoperative histological examination was identical to the biopsy done before and confirmed the diagnosis of AFO. Immediate and late post-operative healing was uneventful. Two months after the tumor ablation an additional low radiation dose volume tomogram was done, confirming complete removal of the tumor (Fig. 5a, b). In a follow up control by means of a low resolution volume tomogram six months later no recurrence was noted.

3. Discussion

This case study shows an unusual large and less common location of an AFO occurring in a three-year-old girl. In the literature only few studies are dealing with ameloblastic fibro-odontoma involving the maxillary sinus [13,15,16]. Buchner [9]