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Preauricular pilomatrixoma: An uncommon entity in a dental pediatric patient

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ABSTRACT

INTRODUCTION: Pilomatrixomas are benign follicular skin appendage tumors, commonly occurring in children and young adults. Most patients admit to dermatologists to seek treatment and are well known by them; however, dental professionals, especially pediatric dentists are not familiar with these tumors.

PRESENTATION OF CASE: This report presents a 16-year-old female with preauricular pilomatrixoma, located beneath the overlying skin of the temporomandibular region. Clinical examination revealed an asymptomatic lump, the overlying skin revealed no abnormalities. Patient was unaware of the lesion.

DISCUSSION: Pilomatrixomas are commonly encountered in the maxillofacial region, although not considered in differential diagnosis by dental professionals. They usually present as, asymptomatic, subcutaneous masses; although symptomatic cases have been reported. In literature, common differential diagnosis for head and neck pilomatrixoma includes sebaceous cyst, ossifying hematoma, giant cell tumor, chondroma, dermoid cyst, foreign body reaction, degenerating fibroxanthoma, metastatic bone formation, and osteoma cutis. We are of the opinion that temporomandibular joint disease should also be considered in differential diagnosis for preauricular pilomatrixoma.

CONCLUSION: Pediatric dentists should be aware of the condition and consider it in the differential diagnosis of pediatric conditions involving the temporomandibular joint.

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1. Introduction

Pilomatrixomas are benign follicular skin appendage tumors, initially described in 1880 by Malherbe and Chenantais [1]. They can occur at any age, although children and young adults are mostly affected, 60% of cases are reported to occur within the first two decades of life [1,2]. They are most commonly reported in the head and neck region with preauricular region being one of the most frequent locations and have a wide variety of signs, which often causes misdiagnosis [2–4]. They present as solitary or seldom multiple lesions, which are commonly asymptomatic, superficial, subcutaneous hard masses demonstrating variable degrees of calcification, often attached to the skin but mostly mobile over the underlying

tissues; as the tumor grows more superficial, a bluish color or ulceration of the overlying epidermis may be noted [1,2,5]. Treatment is by incision and curettage or by excision only [4].

In spite of high occurrence frequency, dental professionals have been rarely facing the lesion and pediatric dental literature publications are rare [1].

The aim of this paper is to present a case of preauricular pilomatrixoma located in the maxillofacial region in a 16-year-old female, along with an analysis of literature in order to make a contribution to the pathogenesis, treatment and differential diagnosis of the lesion.

2. Presentation of case

Our case is reported in line with the SCARE criteria [6]. A 16-year-old female patient from the Society for Protection of Children was brought for dental care, to our pediatric outpatient clinic by her legal guardian. The patient's general medical history was unremarkable. Intra-oral examination revealed multiple caries lesions and radix relicta in all quadrants. A panoramic radiograph showed a calcified mass, which was superimposed to right mandibular

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Fig. 1. Panoramic radiograph of the patient.

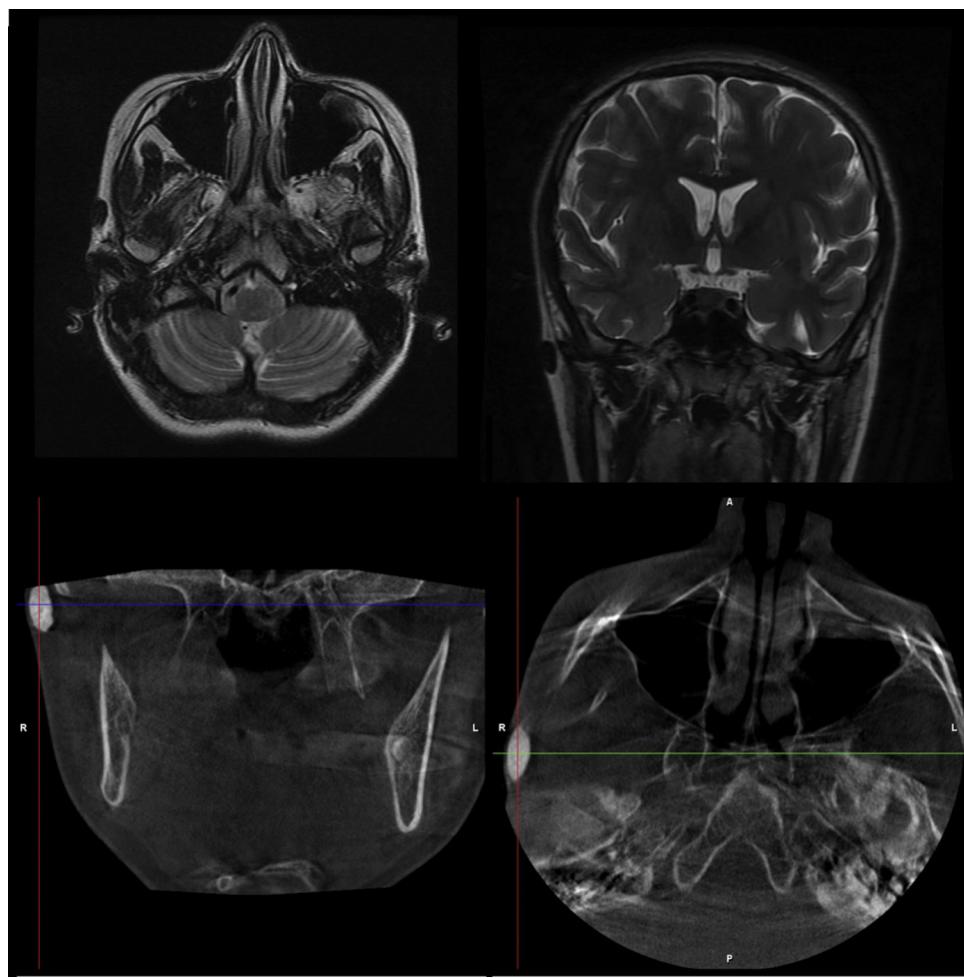


Fig. 2. Coronal and axial CBCT views and T1 axial and T2 coronal MRI views of the tumor.

condyle (Fig. 1). Patient was unaware of the lesion and had no complaints.

Head and neck examination revealed no evidence of adenopathy, paresthesia or motor nerve deficiency. However, physical examination revealed a hard, mobile mass measuring 1.5×1 cm, localized 1 cm anterior to the right crus of helix, in the overlying skin of the temporomandibular region. The skin covering the mass was normal.

In order to have a definition of the pathology and to minimize concerns of radiation to the child, initially, magnetic resonance imaging (MRI) was performed. MRI demonstrated signal void areas both on T1 and T2 images (Fig. 2a,b), which couldn't predict a differential diagnosis because of signal loss in the lesion. For this reason, it was decided to perform a cone beam computed tomography (CBCT) scan for obtaining a more precise location and definition of the pathologic features. CBCT demonstrated the calcified lesion lying

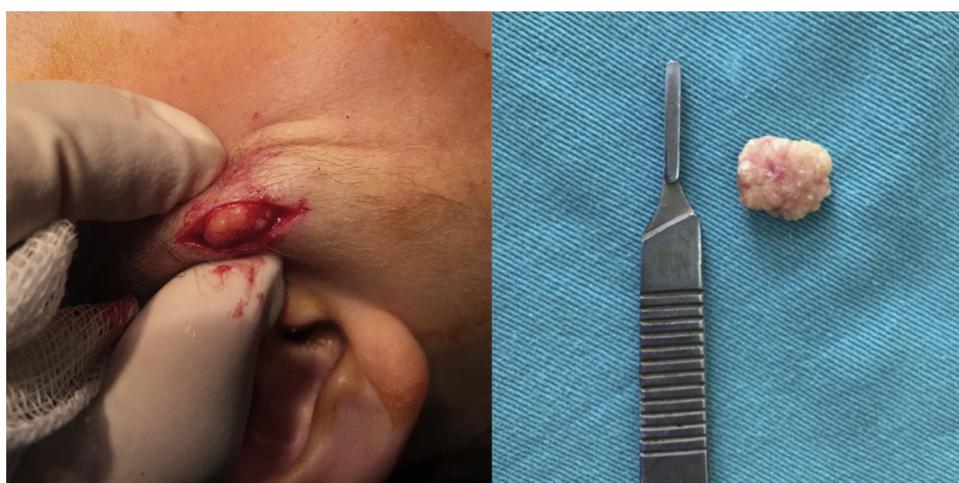


Fig. 3. View during surgery and macroscopic view of the tumor.

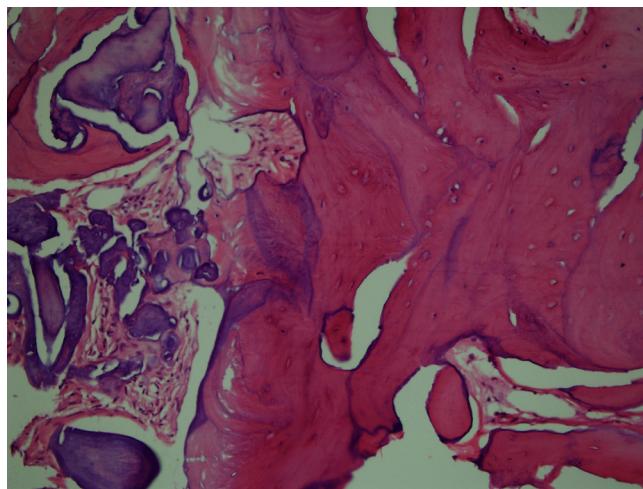


Fig. 4. Microscopic view of the tumor consisting of epithelial ghost cells. (HEx200).

just beneath the skin surface did not have any relation with the temporomandibular joint (TMJ) (Fig. 2c,d).

After clinical and radiographic examination initial diagnoses were made as chondroma, calcinosis cutis, osteoma cutis or foreign body reaction. Treatment and follow-up options were discussed with the patient and legal guardian who accompanied her. Patient wanted to have the lesion removed but insisted that the operation was made under sedation. Total excision under sedation was planned.

During surgery, care was given in order to protect the surrounding vital structures such as the facial nerve and a small incision was made on the overlying skin and soft tissue was dissected using blunt dissection scissors (Fig. 3a,b). The superficial lesion was easily removed and wound was sutured with 3.0 Vicryl® and 6.0 Prolene® (Ethicon, Johnson & Johnson, USA) sutures. Healing was uneventful.

Lesion was sent to histopathological evaluation and a tumor, consisting of epithelial cells demonstrating ghost keratinizations was observed. Tumor stroma consisted also of bone forming cells and foreign object type giant cells (Fig. 4). Histopathological evaluation confirmed the lesion as pilomatrixoma.

3. Discussion

The majority of pediatric head and neck masses are inflammatory, infectious or congenital and neoplastic conditions are

uncommon, however pilomatrixomas are commonly encountered in the head and neck region [2].

After initial description of the lesion by Malherbe and Chenatais in 1880, as a calcifying epithelioma, Forbis and Helwig proposed the term pilomatrixoma revealing the source of origin as the cortex of the follicle [7]. Over the following days beta-catenin gene mutation has been observed in pilomatrixomas and has been reported to play an important role in the formation of hair follicle-related tumors [8].

The lesion which originates from the dermis and extends into the subcutaneous fat, is histopathologically well defined and characterized with islands of tightly coherent epithelial cells with basophilic cytoplasms and a round vesicular nucleus (ghost cells) at the center of the lesion, and is occasionally accompanied by foreign body giant cells and calcifications. In tumors covered by vascular, atrophic skin, dilated lymphatic vessels fill the area between the lesion and the overlying epidermis, with numerous small blood vessels and chronic inflammatory cell infiltrate [6,9].

Pilomatrixomas are commonly encountered in the maxillofacial region (cheek%23, neck%22, eyebrow%18 and scalp%14), although they are usually not considered in differential diagnosis by dental professionals [1]. Although they usually present as slow enlarging, asymptomatic, subcutaneous masses; O'Connor et al. [1] reported that nearly%25 could be symptomatic. Surgery consisting of total excision with clear margins is the treatment of choice in order to alleviate possible symptoms and minimize the risk of malignant transformation; recurrence is reported to be rare [2,5].

The usual differential diagnosis for head and neck pilomatrixoma includes sebaceous cyst, ossifying hematoma, giant cell tumor, chondroma, dermoid cyst, foreign body reaction, degenerating fibroxanthoma, metastatic bone formation, and osteoma cutis [3]. In O'Connor et al.'s [1] study 201 cases were evaluated and they reported that a correct preoperative diagnosis was made in only 28% of all cases [1]. The clinical review study of 209 pilomatrixomas, by Julian and Bowers [4], also reported that the lesion is often misdiagnosed. They stated that a false diagnosis of malignancy or of an epidermoid cyst could be made [4]. Mundinger et al. [5] reported a case of a large pilomatrixoma involving the parotid gland and stated that the lesion should be considered in the differential diagnosis of large check tumors.

Temporomandibular joint disease also is a condition affecting children, and epidemiological studies report that signs and symptoms in children including joint sounds, impaired movement of the mandible, limitation in mouth opening, pre-auricular pain, facial pain, headache and earache to be as frequent as in adults

[10]. When pilomatrixoma is symptomatic signs like pain, tenderness and warmth on palpation may be present [11], all symptoms, which can be misdiagnosed for temporomandibular joint symptoms. Therefore, professionals have to be aware of preauricular pilomatrixoma and evaluate children who are admitted for temporomandibular joint symptoms in this manner.

4. Conclusion

Although pilomatrixoma is a lesion commonly encountered in the maxillofacial region, dental professionals commonly leave it out of differential diagnosis. Professionals should be aware of calcifications like this and for early diagnosis, they should carry out optimal clinical examination by means of radiographs and histopathology.

Conflict of interest

None.

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Ethical approval

Considering the retrospective nature of the report, Ankara University Faculty of Dentistry Ethics Committee has granted exemption in writing for the study.

Consent

Written and verbal consent were both obtained from the patient and legal guardian who accompanied her from the founding hospital.

Author contributions

PB has contributed in the study concept or design, data collection, data analysis or interpretation and writing the paper.

MEK has contributed in the study concept or design, data collection, data analysis or interpretation and writing the paper.

ÖG has contributed in the, data collection, data analysis or interpretation and writing the paper.

EE has contributed in the study concept or design, data collection, data analysis or interpretation and writing the paper.

KO has contributed in the study concept or design, data collection, data analysis or interpretation and writing the paper.

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