Intraosseous Epidermoid Cyst: a Case Report

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ABSTRACT

Background: Epidermoid cysts are benign lesions that occur throughout the body. Their development in the oral cavity is extremely rare. Intraosseous epidermoid cysts of the jaw are even rarer and difficult to distinguish from other lesions. For this reason, we would like to draw the attention of practitioners to this pathology as a differential diagnosis through the presented clinical case.
Methods: This study presents an unusual case of a type of epidermoid cyst in an edentulous maxilla. A 70-year-old man was referred to the Maxillofacial Surgery Division at the University Multiprofile Hospital for Active Treatment and Emergency Medicine "N. I. Pirogov", Sofia, Bulgaria, for a single radiolucent area in his anterior maxilla. The patient underwent surgery to extract the cyst.
Results: Based on the clinical and radiographic evaluation, a preliminary diagnosis of dentigerous residual cyst was made. The histopathological examination of the hematoxylin and eosin stained sections revealed an epidermoid cyst based on the observed thick keratin layer resembling epidermis together with the stratified squamous epithelium lining with many layers of sheaves of orthokeratin.
Conclusions: This report presents an uncommon case of an intraosseous epidermoid cyst occurring without a history of maxillary trauma. Although intraosseous epidermoid cysts are extremely rare in jaws, they should be considered in the differential diagnosis of radiolucent lesions.

Keywords: dermoid cyst; jaw cysts; maxilla; teratoma.

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INTRODUCTION

Epidermoid cysts are benign soft tissue neoplasms encountered throughout the body, particularly in areas where embryonic elements fuse together. Most (80%) are located in the ovaries and sacral region. Some (about 7%) can be found in head and neck region [1]. These cysts are cystic malformations lined with squamous epithelium. They develop out of ectodermal tissue. Epidermoid cysts represent the simplest expression of teratoma spectrum. Intraoral epidermoid cysts should not be confused with the more common epidermoid cyst of the skin, a nonteratomatous lesion that arises from the hair follicle. In oral cavity, these formations are benign slow-growing and painless and commonly found in the submandibular, sublingual, and submental regions [2]. Their development intraorally is unusual. They constitute 1.6% to 6.9% of all cysts in the head and neck region [3]. Cases of epidermoid cysts developed in the jaw bones are relatively fewer than those in soft tissues [4]. In this case report a unique case of intrabony epidermoid cyst involving the maxilla is presented.

CASE DESCRIPTION AND RESULTS

A 70-year-old male patient presented to the Maxillofacial Surgery Division at the University Multiprofile Hospital for Active Treatment and Emergency Medicine "N. I. Pirogov", Sofia, Bulgaria, with a swelling in the left anterior region of the maxilla of several weeks' duration. History revealed that the swelling grew without any significant functional impairments or complications. All of the patient's maxillary teeth had been removed over the years, uneventfully. There was no history of trauma to patient's maxilla or of any other major surgery.

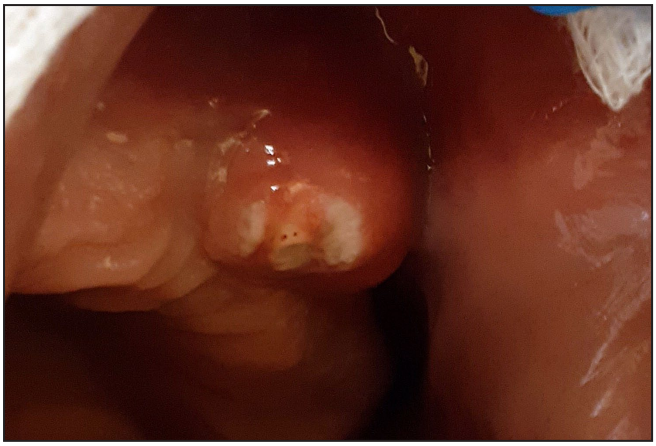

Figure 1. A preoperative intraoral view of the formation.

On physical examination, an intraoral swelling measuring about 2 cm in size was observed in the left anterior region of maxilla on the border area between the incisive bone and the palatine process of the maxilla. The swelling was localized, well demarcated from the surrounding bone, asymptomatic and firmly on palpation with initial dehiscention of overlying mucosa. No lymphadenopathy was clinically apparent (Figure 1). General physical examination was uneventful.

An orthopantomograph revealed an unilocular radiolucent area at his anterior left maxilla approximately 2 cm in size with well-circumscribed sclerotic border (Figure 2). Additionally, a cone-beam computed tomography scan (CBCT) in three projections (sagittal, axial and coronal) revealed the lesion to be approximately 3 x 2 cm in size (Figure 3).

Based on the clinical and radiographic evaluation, a preliminary diagnosis of dentigerous residual cyst was made. Differential diagnosis should be performed with other radiolucent lesions such as odontogenic keratocyst, globulomaxillary cyst, traumatic bone cyst, unicystic ameloblastoma and aneurysmal bone cyst [5].

Under general anaesthesia following elevation of a buccal mucoperiosteal flap the lesion was surgically completely enucleated and submitted for histopathological examination. The patient was prescribed non-steroid anti-inflammation drugs and amoxicillin/clavulanic acid 875/125 mg twice daily for five days. The postoperative period was uneventful.

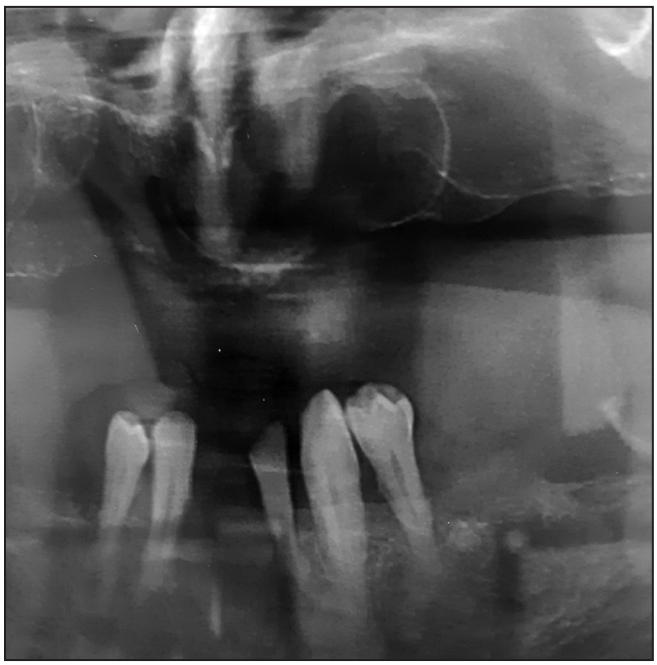

Figure 2. An orthopantomograph showing well-defined radiolucent lesion with a sclerotic margin.

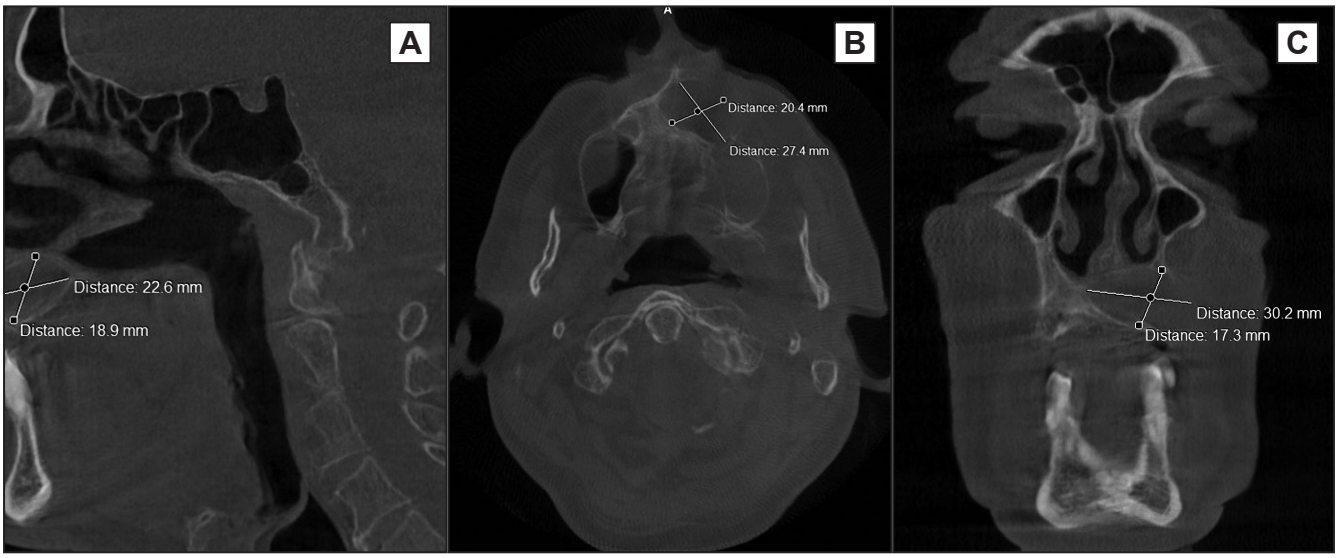

Figure 3. Cone-beam computer tomography scans in three projections (sagittal, axial and coronal) determining the size and location of the cystic lesion.

Gross examination of the specimen revealed a thick cystic sac measuring about 3 cm in size. The surface of the cystic lumen appeared smooth and contained thick creamy, cheesy like material. The histopathological characteristics indicated "epidermoid cyst" (Figure 4). Histopathological examination of the hematoxylin and eosin stained sections revealed a cavity that was lined with orthokeratinized stratified squamous epithelium resembling epidermis, with surface showing sheaves of keratin arranged in many layers. The epithelial lining was about 6 to 8 layers with numerous keratin scales in the lumen and a distinct granular cell layer underlying the stratum corneum, and the basal layer showed a low cuboidal morphology with no nuclear palisade. The epithelial-connective tissue interface was rather flat and devoid of rete ridges. The surrounding capsular tissue was made up of dense fibrous connective tissue, blood vessels, with scanty chronic inflammatory cell infiltration. There was absence of dermal appendages.

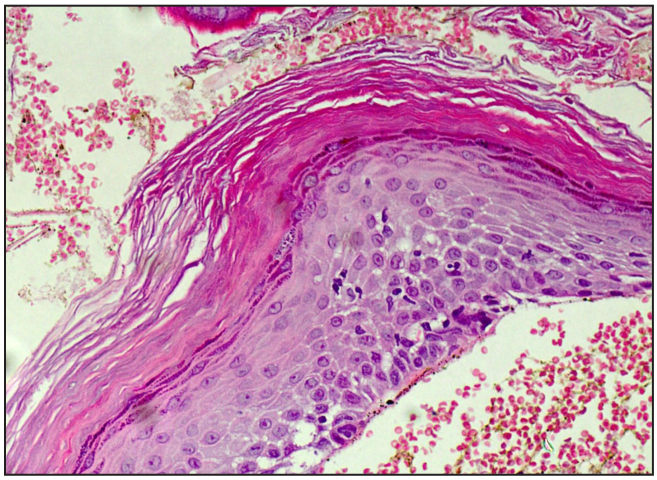

Figure 4. Histopathology showing thick keratin layer resembling epidermis.
Stratified squamous epithelium lining with many layers of sheaves of orthokeratin and low cuboidal morphology of basal layer cell with absence of palisading of nuclei (hematoxylin and eosin stain, original magnification x20).

DISCUSSION

Roser, in 1859 first described epidermoid cyst [6]. Depending on the pathogenesis, epidermoid cysts can be divided into congenital and acquired types. The former are called epithelial and develop from congenital inclusion of ectodermal tissue during embryological development, the latter type, first recognised by Werhner and originally referred to as 'implantation cyst' by Sutton in 1895 [7]. Since trauma is said to always precipitate in the formation of the implantation-type epidermoid cyst, King [8] termed it as 'post traumatic cyst'. There is usually a latent period after injury before the cyst is noticed clinically. Sometimes the injury is so slight, as in an insect bite that the trauma escapes unnoticed or is forgotten by the patient. When healing takes place, the implanted epithelial cells multiply, producing a central mass of keratin and lipid-rich debris [8].

Epidermoid cysts are non-odontogenic inclusion cyst lined by ectoderm [9]. These are rare benign conditions in the orofacial region derived from abnormally situated ectodermal tissue. Most reported cases have involved the floor of the mouth (sublingual dermoids), usually in the midline [10]. According to Toptas et al. [11], only 13 cases have been documented in literature, 7 in mandible and 6 in maxilla.

Some authors believed that these cysts are formed in much the same way as the epithelial germ that remained displaced within the maxillary bone during embryonic development [12]. They may also grow up from the tuberculum impar of His [10]. Histologically, they can be further classified as epidermoid cysts, dermoid cysts and teratoid cysts [12]. To be defined as a dermoid cyst, skin appendages such as hair follicles, sebaceous and sweat glands, and arrector pili muscles must be detected within the cyst wall. Because the specimen presented here does not contain these dermal tissues, it was determined as an epidermoid cyst.

Histologically, epidermoid cysts reveal cystic wall lined by keratinized stratified squamous epithelium filled with keratin. Epidermoid cysts are described as 'pearly tumour' due to shiny, smooth, waxy keratinous content of the cyst. Unlike dermoid cyst (compound type), they exhibit no adnexal structures such as hair follicle, sebaceous gland and sweat gland. Microscopic examination undoubtedly remains the primary means of diagnosing epidermoid cysts, as in presented case [1,8]. Thus, only histological differential diagnosis could determine the type of the cystic formation. Surgical management of these lesions is the same; it does not matter in their difference. All of them are very rare in bones, especially in jaws [14]. The prognosis of this kind of cyst is known to be fairly favourable and a successful surgical removal of it will not give rise to a relapse or aggravation. The epidermoid cyst rarely discloses malignancy [13]. Heidsieck [15], however, reported that of 5 cases of epidermoid cysts in the mandible, one relapsed and another one changed into malignancy.

CONCLUSIONS

Our case presents a rare epidermoid cyst located in the anterior maxilla. These lesions are interesting from the etiological point of view. They should be taken into account in the differential diagnosis of radiolucent lesions of the jaws, therefore during examination clinicians and surgeons should consider aspiration biopsy, ultrasonography and other advanced imaging techniques, as conventional radiographs are not sufficient for the differential diagnosis of cystic-like bone lesions. When treated surgically, epidermoid cysts have a favourable prognosis because they are non-aggressive lesions.

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The authors report no conflict of interest related to this study.

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