

Intraosseous epidermoid cyst of the mandible: Diagnosis and management

Kyste épidermoïde intra-osseux mandibulaire: Diagnostic et traitement

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ABSTRACT:

Objective: To describe clinical, radiological, and therapeutic features of a mandibular intraosseous epidermoid cyst.

Case report: We present the case of an adult male patient presenting with a painless swelling of the left cheek, which appeared two years ago. Imaging found a well-circumscribed osteolytic lesion of the mandibular ramus that was first thought to be a malignant tumor. Surgical removal of the lesion was performed. Histological examination confirms the diagnosis of an epidermoid cyst. No complications or recurrence were noted during the follow-up period.

Conclusion: Epidermoid cyst is an uncommon condition that presents as a slow-growing mass. Intraosseous localization is rare. Clinical presentation may mimic malignant bone tumors. The treatment is based on surgical resection.

Keywords: Epidermoid Cyst; Intraosseous; Mandible; Surgery

RÉSUMÉ

Objectif: Décrire les caractéristiques cliniques, radiologiques et thérapeutiques du kyste épidermoïde intra-osseux mandibulaire.

Observation: Nous présentons le cas d'un patient présentant une tuméfaction indolore de la joue gauche, apparue il y a deux ans. L'imagerie a révélé une lésion ostéolytique, bien circonscrite de la branche montante de la mandibule, faisant suspecter une tumeur maligne. Une exérèse chirurgicale de la lésion a été réalisée. L'examen histologique a confirmé le diagnostic d'un kyste épidermoïde. Aucune complication ou récurrence n'a été notée pendant le suivi.

Conclusion: Le kyste épidermoïde est une lésion rare d'évolution lente. La localisation intra-osseuse est peu fréquente. La présentation clinique peut donner le change à des tumeurs osseuses malignes. Le traitement est basé sur la résection chirurgicale.

Mots clés: kyste épidermoïde ; Intra-osseux ; Mandibule ; Chirurgie

INTRODUCTION:

Epidermoid cyst (EC) is a benign soft tissue neoplasm that can be encountered throughout the body. Localization in head and neck region is rare and accounts for 7% of all EC [1]. The intraosseous location is uncommon. Most described sites were the hand phalanges [2, 3]. Few cases of mandibular location have been described [4]. Through this manuscript we try to illustrate the clinical presentation of EC, presumed etiologic factors and therapeutic approach.

CASE REPORT:

we report the case of a 52 year-old-man, with no medical past history, who presented with a slow growing, painless, swelling of the left cheek that appeared two years ago (figure 1). There was no history of facial injury or dental extraction.



Figure 1: A left cheek swelling with no cutaneous inflammatory signs

Clinical examination revealed a 6 cm well-circumscribed mass of the left cheek and the periauricular area with a regular surface. No inflammatory signs of the skin were noticed. There was no facial palsy or numbness

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of the cheek. The oral cavity was tumor-free. There was no evidence of cervical lymph nodes. Computed tomography (CT) scan showed an osteolytic mass of the mandibular ramus, measuring 25 mm, causing a thinning and rupture of the cortical bone. The masseter muscle was infiltrated. There was no periosteum reaction or osteosclerosis (figure 2). Two similar osteolytic lesions of the contralateral mandibular ramus were observed.

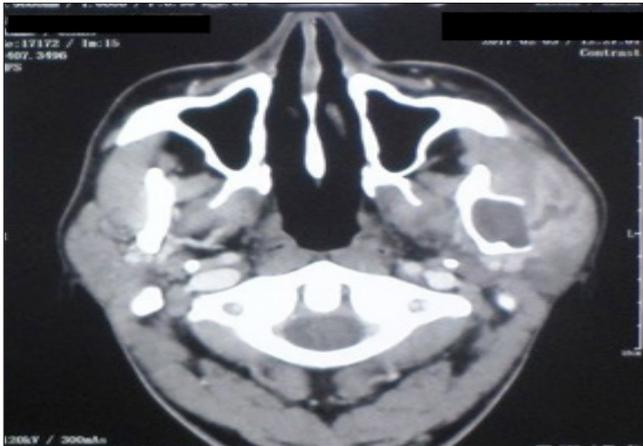


Figure 2.a

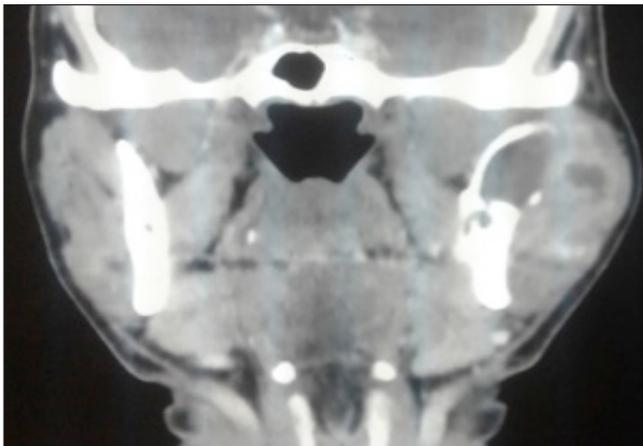


Figure 2.b

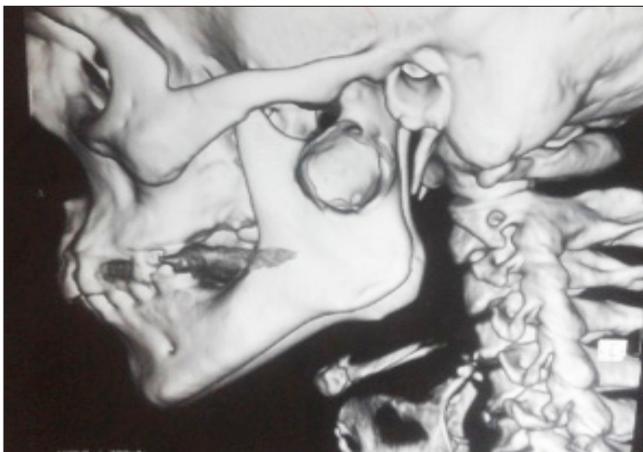


Figure 2.c

Figure 2: Axial (Figure 2.a), coronal (Figure 2.b) and 3D reconstruction CT scans (Figure 2.c) showing a hypo dense osteolytic well-defined mass of the left mandibular ramus

The clinical and radiological presentation suggests the diagnosis of an aggressive giant cell tumor or an ameloblastoma. The patient was operated by Risdon approach. The tumor was totally removed. Curettage of osseous lesions was also performed. There was no need to perform a grafting since the basilar border of the mandible was preserved. No complications have been noticed in the post-operative period. The histological examination concluded to an EC. There was no recurrence during the three years follow-up.

DISCUSSION:

EC is a rare benign epithelial lesion which is also known as epidermal cyst and keratin cyst. Tumor is usually described in the skin. Intraosseous localization is uncommon. Reported cases are generally found in the skull and hand phalanges [5]. Occurrence of EC in the oral cavity is extremely rare. Few cases have been reported especially on the floor of the mouth [6]. Other sites such as foot phalanges, sternum, ulna, tibia and maxilla are rarely encountered [7]. According to Toptas, 13 cases have been documented in literature, 7 in the mandible and 6 in the maxilla [8]. EC, generally, occurs in male adult patients more than female (sex-ratio: 3) [5]. It may be congenital or acquired [2]. We reported in this manuscript the case of an intraosseous EC of the mandible that occurred in a male patient.

There are many theories to explain the presence of ectopic epidermal tissue in the bone. For some authors, it is thought to be a migration of an epidermal cyst from soft tissue into the bone below [3]. For others, it might be due to the growth of ectopic ectodermal cells during embryological development. The embryologic accident occurs between 3 and 5 weeks of gestation [8]. The most relevant theory is related to trauma; it may be an external trauma, a surgery or a tooth extraction that causes a disruption of bone integrity so that epidermal cells migrate to deeper mesenchymal structures [9]. Implanted epithelial cells multiply and produce a mass of keratin during the silent period following injury [10]. Among traumatic etiologic factors reported in literature, we found molar extraction [10], impacted mandibular wisdom teeth [8], and a gunshot incident [4]. In 1923, Sonntag described a case of a young female patient that was amputated of hand phalanx after an injury. Twenty-four years later, she developed a lesion at the remaining phalanx that was thought to be a sarcoma. Histological examination after excision of the lesion confirms the diagnosis of EC [11]. An EC of the calvarium was also described after a head injury. In our patient, we didn't find a history of an injury or a dental extraction.

Clinically, the EC presents as a nodular, fluctuant, and painless slow-growing lesion [1].

Multiple sites of the tumor can be observed even though it's a rare condition. Kranz reported multiple incidence of intraosseous EC in the mandible and maxilla [12].

The most frequent complications associated with EC are compression, rupture of the cyst and infection.



Infection occurs especially in long bones and may mimics a chronic osteomyelitis [13]. A case of facial paralysis related to an infected EC of the parotid region was described [14].

Imaging is necessary to assess the diagnosis and to plan the therapeutic management. On ultrasonography, EC presents as a hypo-echoic, or iso-echoic, homogeneous, unilocular mass with an internal echo [15]. CT scan shows a well circumscribed unilocular osteolytic lesion. Surrounding sclerotic bone may be associated. Radiological characteristics of intraosseous EC are similar to those of malignant bone tumors [7]. Magnetic resonance imaging, when performed, shows a hypointense T1-weighted sequences and hyperintense T2-weighted sequences mass [15]. In our patient, a CT scan was performed and showed an osteolytic mass of the mandibular ramus, associated with a thinning and a rupture of the cortical bone.

Histological examination is essential to confirm the diagnosis of EC and to exclude differential diagnosis such as ameloblastoma, giant cell tumors, malignant tumors (sarcoma, chondrosarcoma, metastasis...) and osteomyelitis [7]. The tumor is lined with squamous cell epithelioma and contains keratinized substance of non-structure layers stainable by eosin [12]. EC should be differentiated from dermoid cyst which contains skin appendages such as hair follicles, sebaceous glands or sweat glands. Dermoid cyst may be found in the eyelid,

peripheral cornea or orbit. A malignant transformation of squamous epithelium should be eliminated [15].

The treatment consists of surgical "en bloc" excision of the lesion with the surrounding soft tissue [13]. The cyst wall should be completely removed to avoid recurrence. Autologous or synthetic grafting can be performed in case of large osseous loss [7]. The prognosis is favourable with no recurrence unless the tumor is removed completely [5]. In our case, a total removal of the cyst was performed. There was no need to an osseous grafting. No recurrence was reported during the follow-up.

CONCLUSION:

Even though it may present as a malignant tumor, intraosseous EC should be considered especially in a context of a trauma. Only histological examination can confirm its benignity. The treatment is limited to complete surgical excision associated with grafting if needed.

Compliance with ethical standards

Conflict of interest: The authors stated that there is no conflict of interest.

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REFERENCES:

1. Ertem SY, Uckan S, Ozdemir H. An unusual presentation of an intraosseous epidermoid cyst of the anterior maxilla: a case report. *J Med Case Rep.* 2014;8:262.
2. King E. Post-traumatic epidermoid cysts of hands and fingers. *Br J Surg.* 1933; 21: 29-43.
3. Zadek I, Cohen HG. Epidermoid cyst of the terminal phalanx of a finger; with a review of the literature. *Am J Surg.* 1953;85(6):771-4.
4. Noffke CE. Implantation-type epidermoid cyst of the mandible. *Dentomaxillofac Radiol.* 1999;28(6):383-5.
5. Alimoglu Y, Duman C, Ogreden S, Cansiz H, Dervisoglu S. Mandibular Ramus Epidermal Inclusion Cyst. *J Craniofac Surg.* 2010;21(5):1647-9.
6. Koca H, Seckin T, Sipahi A, Kazanc A. Epidermoid cyst in the floor of the mouth: report of a case. *Quintessence Int.* 2007;38(6):473-7.
7. Ozdemir HM, Senaran H, Ogun TC, Oltulu P. Intracortical epidermoid cyst of the tibia. *Skeletal Radiol.* 2004;33(8):481-4.
8. Toptas O, Akkas I, Tek M, Ozan F, Boran C. Intraosseous epidermoid cyst associated with impacted mandibular wisdom teeth: an uncommon entity. *J Clin Diagn Res.* 2014;8(7):ZD31-2.
9. Carroll RE. Epidermoid (epithelial) cyst of the hand skeleton. *Am J Surg.* 1953;85(3):327-34.
10. Jayade BV, Upadya VH, Gopalkrishnan K, Shirganvi MS. Epidermal inclusion cyst of the mandible after extraction of a third molar: case report. *Br J Oral Maxillofac Surg.* 2012;50(5):e72-4.
11. Sonntag F. Traumatic Epithelial Cyst in Tip of Bone in a Finger Stump Following Injury, München. *med. Wchnschr.* 1923;70:1055-6.
12. Takigawa T, Onuma M, Kodama M, Tanaka H. Case report of epidermoid cysts developed in the anterior and ramus regions of mandible. *J Nihon Univ Sch Dent.* 1967;9(4):180-7.
13. Mimura T, Yamazaki H, Hayashi M, Kato H. Intraosseous Epidermoid Cyst of the Radius. *J Hand Surg Am.* 2019;44(11):996 e1- e5.
14. Streppel M, Thomas JP, Stennert E, Guntinas-Lichius O, Wagner M. Infected epidermoid cyst as cause of peripheral facial palsy. A case report. *Laryngorhinootol.* 2001;80(10):617-9.
15. Erol B, Osma U, Selcuk OT, Suren D, Eyigor H, Gurses C, et al. Submandibular lateral epidermoid cyst: imaging findings of a rare case. *B-ENT.* 2014;10(2):165-9.