Case report

Mandibular tuberculosis fortuitously discovered after surgical resection of an ameloblastoma: A case report

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ABSTRACT

Introduction: Mandibular localization of tuberculosis is rare and represents less than 2% of skeletal locations. Its clinical and radiological features are not specific. In this paper, we report a case of fortuitous discovery of mandibular tuberculosis after a histopathological analysis of the surgical resected specimen during surgical management of an ameloblastoma.

Presentation of case: A 50-year-old female patient was admitted to our department with a 2 years history of left cheek swelling, the clinical examination revealed a left cheek swelling, extending from the mandibular angle to below of temporomandibular joint, measuring approximately 5 cm in diameter. The swelling was firm to hard in consistency, and cervical lymphadenopathy of submandibular region was noticed.

Computed tomography (CT) scan revealed a large multiloculated osteolytic expansive lesion measuring 56 * 48 * 53 mm.

An interrupting hemimandibulectomy, was performed from the left parasymphys opposite to 33 tooth, extending to the left temporomandibular joint.

The histopathological findings confirmed the diagnosis of ameloblastoma, with negative free margin. A mandibular and lymph node tuberculosis were associated with giant cells and caseating necrosis.

The patient was successfully treated with a standard anti-tuberculosis therapy.

Discussion: Ameloblastoma is a benign odontogenic tumor, 80% of these tumors are found in the mandible. Primary mandibular tuberculosis is an extremely rare entity. Its clinical presentation is not specific. Radiologically, tuberculosis has no characteristic appearance. However, it is possible to evoke it in case of a lytic image of the mandible. The positive diagnosis is based on histology. The treatment is medical, but surgery is necessary for some cases.

Conclusion: The association between ameloblastoma and mandibular tuberculosis represents an extremely rare entity. Mandibular tuberculosis is rare and should be considered as a possible diagnosis in pandemic areas.

1. Introduction

Primary tuberculosis of the oral cavity is very rare. The mandibular localization of tuberculosis is rare and represents less than 2% of the skeletal locations [1–3]. The lesions of primary orofacial tuberculosis could be the only presentation of the disease; however the orofacial presentation is usually associated with systemic manifestations of tuberculosis [4]. Mandibular tuberculosis presents often a difficulty of diagnosis because of the rarity and non-specificity of clinical presentation and the absence of pathognomonic signs. [5]

The treatment of this disease is medical, but surgery is required for some cases. Medical treatment should be started as soon as the diagnosis is made and; it is based on the use of multidrug therapy. [6,7]

We report a case of 50 year old patient admitted for the management of mandibular ameloblastoma, and a mandibular tuberculosis was discovered incidentally on the anatomopathological examination. To the best of our knowledge; this association has never been reported in the literature. This case report was written according to the SCARE criteria. [8]

2. Presentation of case

A 50-year-old female patient was admitted in our department of oral and maxillo-facial surgery with a 2 years history of left cheek swelling,
progressively increasing in volume. There was no clear history of tuberculosis contagion and no personal or family history of any chronic disease. The patient reported a history of dental avulsion during this period. Clinical examination revealed a left cheek swelling, extending from the mandibular angle to below of temporomandibular joint, measuring approximately 5 cm in diameter, with normal overlying skin.

On palpation, the swelling was firm to hard in consistency. It was non-compressible, non-fluctuant, and slightly tender, with hypoesthesia in the chin area (Fig. 1).

The oral examination of the patient showed a good mouth opening with a poor oral health, and the swelling was found to present a vestibular expression. In addition a cervical lymphadenopathy of sub-mandibular region was noticed.

The patient reported a history of dental avulsion during this period. Clinical examination revealed a left cheek swelling, extending from the mandibular angle to below of temporomandibular joint, measuring approximately 5 cm in diameter, with normal overlying skin.

The oral examination of the patient showed a good mouth opening with a poor oral health, and the swelling was found to present a vestibular expression. In addition a cervical lymphadenopathy of sub-mandibular region was noticed.

The viral serologies (HIV viral hepatitis B and C) and syphilis were negative.

The orthopantomogram (OPG) showed a multilocular osteolytic lesion involving the left body of mandible, the angle and the ramus.

Computed tomography (CT) scan revealed a large multiloculated osteolytic expansive lesion arising from the left body of mandible to the ramus measuring 56 * 48 * 53 mm. A submandibular lymphadenopathy was noted measuring for the most voluminous 12 * 10 mm (Fig. 2).

The chest X-ray was normal.

The surgical management was insured by a qualified professor with the aid of residents. It consisted of a resection of mandible and an interrupting hemimandibulectomy was realized by extraoral approach, from the left parasympathic to 33 tooth, extending to below the left temporomandibular joint. The reconstruction was planned for a second time (after the end of medical treatment).

The histopathological analysis of excised piece confirmed the diagnosis of ameloblastoma, with negative free margin associated with mandibular and lymph node tuberculosis consisting of caseating necrosis and presence of giant cells (Fig. 3). The postoperative period was simple and uneventful. The patient was discharged from the service at the 5th postoperative day and was referred to start the anti-tuberculosis therapy.

The patient received a two months intensive therapy of isoniazid (300 mg/24 h), rifampicin(600 mg/24 h), pyrazinamide(1,6 g/24 h) and ethambutol(1 g/24 h) (ERIP-K4*), followed by maintenance treatment with four months of rifampicin(600 mg/24 h) and isoniazid(300 mg/24 h).

To date (the 5th post-operative month), the patient is under regular follow-up without complications (Fig. 4).

3. Discussion

Ameloblastoma is a benign odontogenic tumor and it is most commonly diagnosed between the fourth and fifth decades of life, with no gender predominance. [9]

Ameloblastoma is usually asymptomatic but when it attains considerable size, it can present with jaw expansion. Radiologically, ameloblastoma has an osteolytic pattern.

Approximately, 80% of these tumors are found in the mandible, but the maxilla is infrequently affected. [10]

A wide surgical excision with safe margins is the treatment of choice. [10]

However, ameloblastomas are well-known for their recurrence. Malignant transformations are rarely seen and account for less than 1% of cases. [11,12]

Our patient was admitted for ameloblastoma, mandibular tuberculosis was diagnosed incidentally on the anatomopathological examination, to our knowledge this association has never been reported in literature.

The lesions of primary orofacial tuberculosis could be the only presentation of the disease; however orofacial tuberculosis is usually associated with systemic manifestations of tuberculosis. [4]

Primary orofacial tuberculosis represents 0.1% to 5% of all tuberculosis infections; primary tuberculosis of the mandible is an extremely rare entity which represents less than 2% of all skeletal localizations. [1,2,3]

The mandible is affected more than the maxilla with the angle and the alveolus of the mandible being the commonly affected areas [5].

The mechanism of extension of the infection to the mandible can be by direct inoculation through dental extraction or lesions of mucosa or perforation of an erupting tooth [13]. Other routes for the occurrence of infection can be by extension from a nearby soft tissue lesion which involves the underlying bone. Hematogenous seeding has also been suggested by other authors [14].

In our case, there was a history of dental avulsion during the development of ameloblastoma, and possibly the site of entry might have been gingivitis as the patient’s dental hygiene was not maintained.

Mandibular tuberculosis presents often a difficulty for diagnosis because of the rarity and non-specificity of clinical presentation and the absence of pathognomonic signs. In a few cases, it appears as an acute inflammatory swelling [5].

In developing countries, where tuberculosis stills endemic, it must always be considered in the differential diagnosis of many infectious or tumoral lesions.

Radiologically, there is no characteristic appearance of tuberculosis. Most lesions of the jaw and alveolar bone are identical to those caused by pyogenic organisms [15]. Mandibular tuberculosis begins as an area of rarefaction with trabecular blurring. Gradually erosion of cortical bone occurs, which is then replaced by soft granulation tissue and subsequently a sub-periosteal abscess formation takes place culminating into a visible painful swelling. Pathological fractures of the mandible or sequestration have also been reported [16].

The Treatment of mandibular tuberculosis is medico-surgical. In the literature, cases of minimal destructive lesions are only managed by medical intervention of antitubercular therapy to achieve resolution. The medical treatment is based on anti-tubercular therapy of four conventional drugs using rifampicin, isoniazid, pyrazinamide and...
ethambutol initially as an intensive regimen followed by rifampicin and isoniazid as a maintenance for a period of 6–9 months with clinical and biological monitoring [6,7]. Surgical treatment may be indicated to excise the cold abscess developed in the soft tissue and extract the necrotic bone sequestrates.

Decortication of bone is indicated for moderately destructive lesions

Fig. 2. Computed tomography (CT) scan revealed a large multiloculated osteolytic expansive lesion (a) 3D computed tomography, (b) CT scan coronal views.

Fig. 3. Histology showing cystic cavity lined by a typical ameloblastomatous epithelium associated with caseating necrosis and the presence of giant cells confirming mandibular tuberculosis.

Fig. 4. Postoperative photograph of patient.
perforation [2].

Our case has been admitted for ameloblastoma which was managed by radical surgical approach, involving elective hemimandibulectomy.

4. Conclusion

In this case we reported an extremely rare association between ameloblastoma and mandibular tuberculosis. Mandibular localization of tuberculosis is a rare diagnosis but it deserves special attention in endemic areas. Clinical presentation is not specific which makes its diagnostic a challenge for clinicians. Radiologically, tuberculosis has no characteristic appearance and it is possible to evoke it in case of lytic image of the mandible. The positive diagnosis is still based on the histopathological analysis.

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Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Dr. Zainab Elzouiti wrote the manuscript and analysed the literature research, Pr. Adil Eabdenbi, Pr. Fahd Elayoubi supervised the writing of manuscript and performed the scientific validation. All authors read and approved the manuscript.

Guarantor

Dr. Zainab Elzouiti.

Declaration of competing interest

The authors have no conflicts of interest to declare.

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