Osteoblastoma of the Hard Palate in a Child: A Rare Case

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Benign osteoblastoma is a rare bone tumour characterised histologically by the production of woven bone spicules, which are bordered by prominent osteoblasts. It mainly affects young adults. We report a rare case of benign osteoblastoma of the maxilla in a 7-year-old boy who presented with a painful swelling on the left hard palate. An incisional biopsy was interpreted as osteoblastic neoplasm most suggestive of osteoblastoma. After excision of the tumour there has been no recurrence for 2 years.

Key words: *bone tumour, child, maxilla, osteoblastoma*
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Benign osteoblastoma is a rare osteoblastic tumour of bone characterised by a proliferation of osteoblasts forming trabeculae set in a vascularised fibrous connective tissue stroma[1]. The lesion is more frequently seen in long bones and rarely involves the maxilla or mandible[2-4]. It has been estimated that benign osteoblastoma accounts for less than 1% of all tumours in the maxillofacial region[5]. The tumour is more common in males and it mainly affects young adults. In children under 10 years of age, benign osteoblastoma involving the maxilla is extremely rare. In a review of the literature, Jones et al[6] collected 43 well-documented cases of osteoblastoma of the maxilla and mandible and added 24 additional cases, but in the total series of 77 cases there were only six case reports relating to children, and in all of them the tumour was located in the mandible. We present here a very rare case of osteoblastoma arising in the maxilla in a 7-year-old male patient and discuss clinical features and the differential diagnosis of this benign tumour of bone. As far as we are aware, only five cases of osteoblastoma of the maxilla in children have been previously reported in the literature[7-11].

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Case report

A 7-year-old Caucasian male patient reported to the outpatients Department of the Oral and Maxillofacial Surgery of A Coruña University Hospital, with dull and intermittent localised pain in relation to a palatal mass of several months' duration. He had been previously healthy and denied any history of trauma or oral pathology. A physical examination revealed a tumour in the left hard palate approximately 2 cm × 2 cm in diameter, and of a hard consistency (Fig 1). The overlying mucosa was normal in appearance and colour. There was no regional lymphadenopathy.

The remaining physical examination was unrevealing. A computerised tomography (CT) scan revealed a well-circumscribed bone tumour of 2.3 cm × 1.2 cm, with intact cortical bone (Fig 2). The boy's chest radiograph was normal and results of all laboratory tests were also within normal limits. An incisional biopsy was performed under local anaesthesia. An area of mineralised tissue, with entrapped osteocytes in lacunae, lined by flattened to angular cells with a moderate to ample amount of cytoplasm and deep basophilic nuclei (osteoblastic rimming) was observed. Numerous discernible reversal lines were seen in the mineralised tissue. Multinucleated giant cells were seen in the resorption bays. The connective tissue was fibro-vascular with numerous proliferating angular cells. Areas of haemorrhage were also seen. Based on these findings, a diagnosis suggestive of osteoblastoma

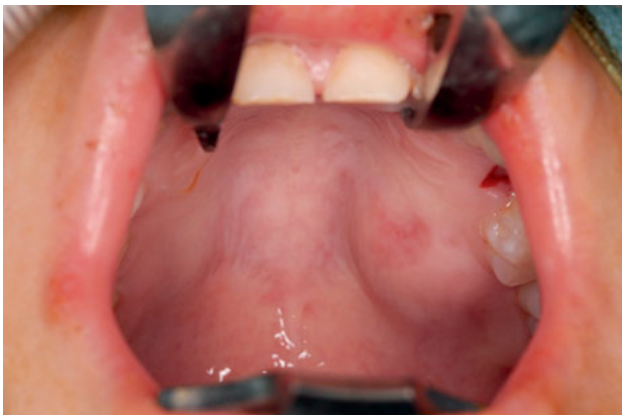

Fig 1 Tumour in the left hard palate; bigger than 2 cm and of a hard consistency.

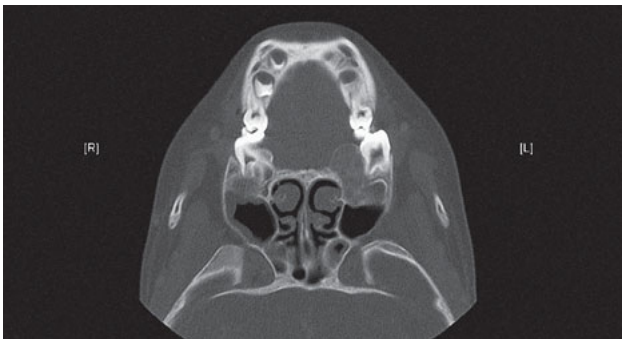

Fig 2 CT: Well-circumscribed bone mass of 2.3 cm × 1.2 cm in size, with an intact thin cortical, which is located in the inferomedial region of the left maxillary sinus, protruding towards the lumen of the sinus and into the oral cavity. Its radiological appearance is compatible with non-ossifying fibroma/fibrous dysplasia.

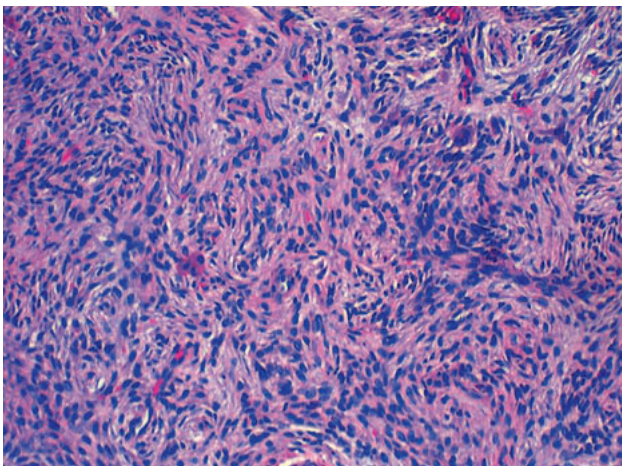

Fig 3 Osteoblastic proliferation forming trabeculae on a vascularised fibrous stroma.

was made (Fig 3). *En bloc* resection of the lesion was done under general anaesthesia. The excised specimen showed features indistinguishable from those seen in the incised specimen. A final diagnosis of benign osteoblastoma was given. There was no sign of recurrence after a follow-up of 2 years (Fig 4).

Discussion

Osteoblastoma arising in the maxilla is a very rare tumour, especially in childhood. Here we report an unusual case of benign osteoblastoma of the hard palate in a 7-year-old boy, who presented with the chief complaints of swelling and pain. The tumour was biopsied and histopathologic examination of the lesion confirmed it as a benign osteoblastoma. After complete mass excision, follow-up CT scans at regular intervals showed no recurrence for 2 years.

Previous descriptions of osteoblastoma in children are limited to five cases reports[7-11]. Shah et al[11] described a case of recurrent osteoblastoma in the maxilla of a 7-year-old male patient. Six months after initial surgical excision using the Caldwell-Luc procedure, the lesion recurred and the patient underwent mass excision with a subtotal left maxillectomy. Salmen et al[10] described the case of another 7-year-old patient with an aggressive osteoblastoma. The tumour fully involved the left maxilla, including the maxillary sinus and the nasal cavity. Following enucleation of the lesion, recurrent volume increase was observed after 2 months and an *en bloc* resection of the maxillary segment was performed. Lin et al[7] reported the case of a 10-year-old boy in whom an osteoblastoma of the hard palate recurred after two previous enucleations. Ismawati[8] described the case of a 7-year-old male patient with an osteoblastoma involving the left maxilla that was successfully treated with a partial maxillectomy.

These cases show that osteoblastoma in the maxilla in children is frequently characterised from a clinical point of view by locally aggressive behaviour, although histologically consisting of a relatively well-circumscribed mass of bone-producing epithelioid osteoblasts and occasional multinucleated osteoclasts characteristic of osteoblastoma. The typical osteoblastoma appears to be composed of small irregular bony trabeculae and osteoids separated by a vascular stroma[7]. The trabeculae of osteoids are usually delicate and often have a basophilic staining pattern. These abundant, irregular spicules of mineralised bony tissue are rimmed by osteoblasts that exhibit no significant nuclear atypia or mitotic activity. The rich vascularity of the fibrous stroma is responsible for the severe bleeding often observed during surgery[7].

The histopathologic differential diagnosis includes osteoid osteoma, osteosarcoma, cementoblastoma, and ossifying fibroma.

Although osteoblastoma is a benign tumour, complete removal of the tumour is usually mandatory because the recurrence rate after incomplete resection appears to be as high as 10% to 20%[7]. Of the five previous cases of osteoblastoma of the maxilla in children aged ≤ 10 years, recurrence of the tumour after initial enucleation of the tumour mass was reported in three cases (60%)[7,10,11]. In our patient, *en bloc* resection of the lesion was performed as the initial surgical procedure, and no recurrence has been observed after a follow-up of 2 years. The prognosis of osteoblastoma is good if the lesion has been removed completely. Although osteoblastoma is thought to exhibit benign behaviour, long-term follow-up is recommended to detect any recurrence at an early stage.

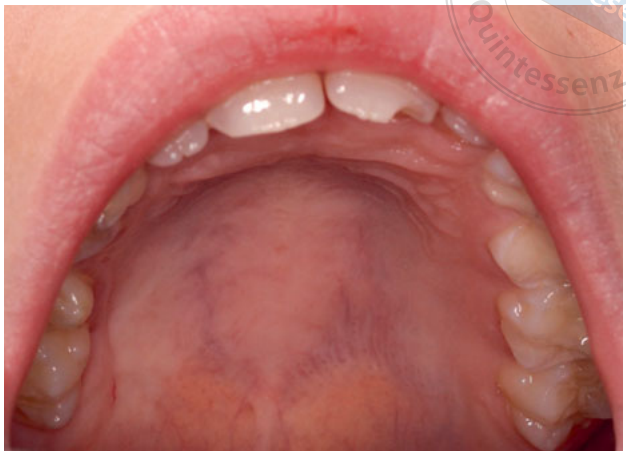

Fig 4 After a 2-year follow-up there is no evidence of recurrence.

Conclusion

Maxillary osteoblastoma is a very rare tumour especially in childhood. It is a benign lesion, even it is locally aggressive. Histopathologic confirmation is necessary for diagnosis. Treatment is surgical excision with free margins.

Conflicts of interest

The authors reported no conflicts of interest related to this study.

Author contribution

Dr Alicia González-Mourelle took the lead in writing the manuscript; Drs M. Pombo Castro and Dr B. Patiño Seijas contributed to the interpretation of the results; Drs I. Vázquez Mahia and J.L. López-Cedrún planned the surgery; All authors provided critical feedback and helped shape the research, analysis and manuscript.

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