Journal of Oral Science, Vol. 52, No. 2, 329-332, 2010

Case Report

An estimate of the rate of growth of a juvenile aggressive ossifying fibroma in a 15 year old child

Clarence Pace[1)], Richard Crosher[1)], Donald Holt[1)] and Audrey Pace[2)]

[1)]Oral and Maxillofacial Surgery, Rotherham Hospitals NHS Trust, Rotherham, UK
[2)]Rotherham Community Dental Clinic, Rotherham NHS, Rotherham, UK

(Received 23 October 2009 and accepted 8 March 2010)

Abstract: Juvenile ossifying fibroma (JOF) is an uncommon fibro-osseous lesion occurring in the facial bones. It has been recognised as a separate histopathological entity among the fibro-osseous group of lesions. Fibro-osseous lesions of the craniofacial bones are difficult to classify and treat however a common factor to all is the conversion of the bone to benign fibrous tissue with a varying degree of mineralised tissue. This case describes a 15 year old patient with a juvenile aggressive ossifying fibroma (JAOF) and an estimate of the rate of growth of such a rare lesion. (J Oral Sci 52, 329-332, 2010)

Keywords: juvenile aggressive ossifying fibroma; estimated rate of growth.

Case Report

A 15 year old male presented with a 4 month history of a rapidly growing, painless swelling of his mandible. He had a bony hard expansion of the right body of his mandible, which measured approximately 5 × 3 cm. Intra-orally marked expansion of the alveolus in the pre-molar region was noted (Fig. 1). The first and second premolars were displaced. He had a posterior lateral and anterior open bite. The overlying oral mucosa and skin was normal. There was no lymphadenopathy.

The panoramic radiograph showed a radiolucent lesion with a sclerotic margin in the right premolar region. The tooth displacement noted clinically was confirmed.

Correspondence to Mr. Clarence Pace, Oral and Maxillofacial Surgery, Rotherham Hospitals NHS Trust, Rotherham, UK
Tel: +44-1709304648
Fax: +44-1709307639
Email: clar.aud@hotmail.com

An axial computed tomogram (CT) and a 3D reconstruction showed an expansile lesion with evidence of cortical perforation and extension of the lesion into the soft tissues (Figs. 2 and 3A,B).

Blood investigations including bone biochemistry were within the normal range.

The lesion was biopsied under local anaesthetic and sent for histopathological examination. The report was of numerous small, rounded mineralised collagenous foci (psammomatoid ossicles) within a relatively cellular stroma consisting of uniform stellate and spindle cells. No mitotic figures were seen (Fig. 4). The appearances were consistent with a diagnosis of aggressive or juvenile ossifying fibroma.

The treatment plan was to resect the tumour with reconstruction with a deep circumflex iliac flap. However the patient refused surgery on several occasions and there was a delay of 35 months between diagnosis and surgery. Over this time the lesion increased in size considerably,

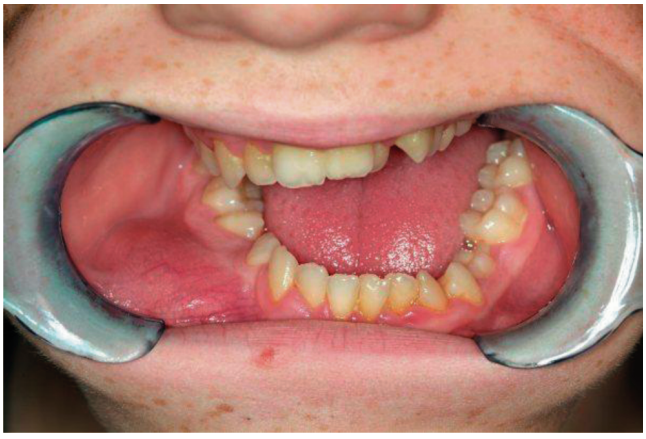


Fig. 1 Clinical photograph showing a lower right buccal sulcus swelling in the region of the premolar.

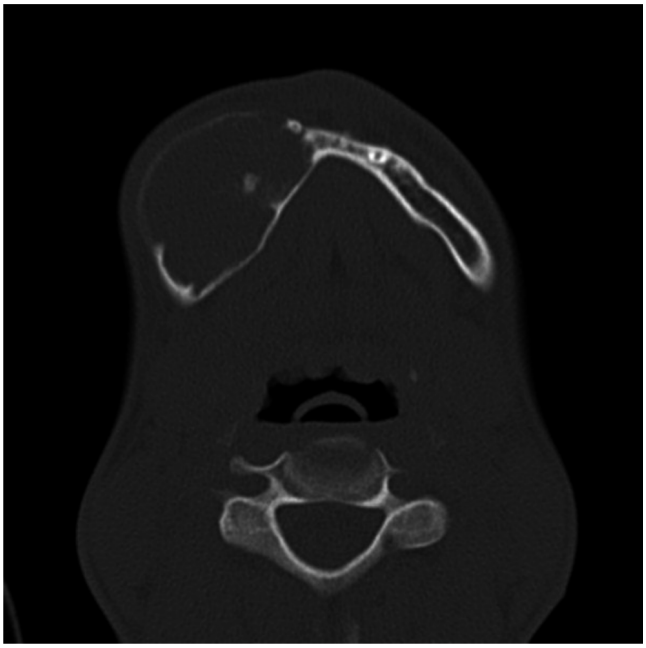


Fig. 2 Axial CT image showing the extent of the lesion at the time of diagnosis.

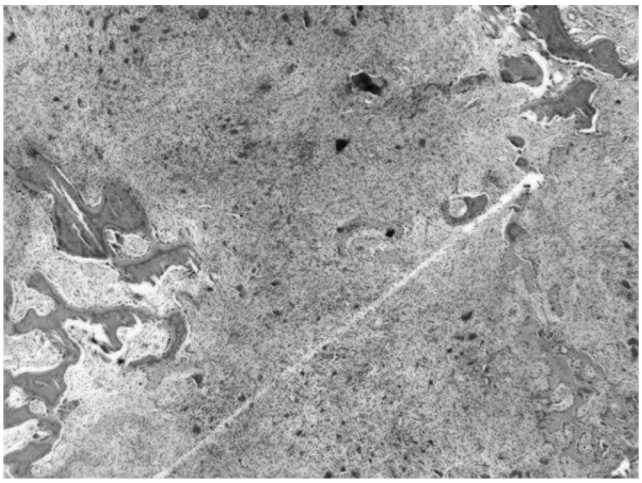
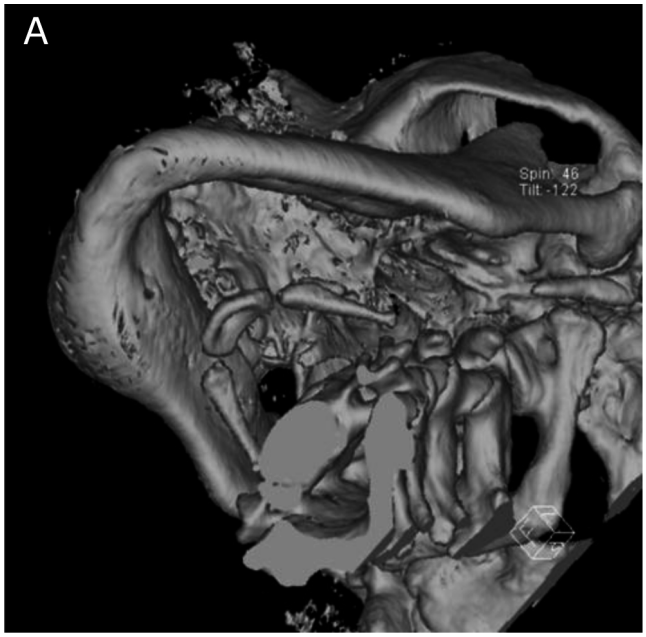


Fig. 4 High power histology section clearly showing bone, spindle cells and giant cells.



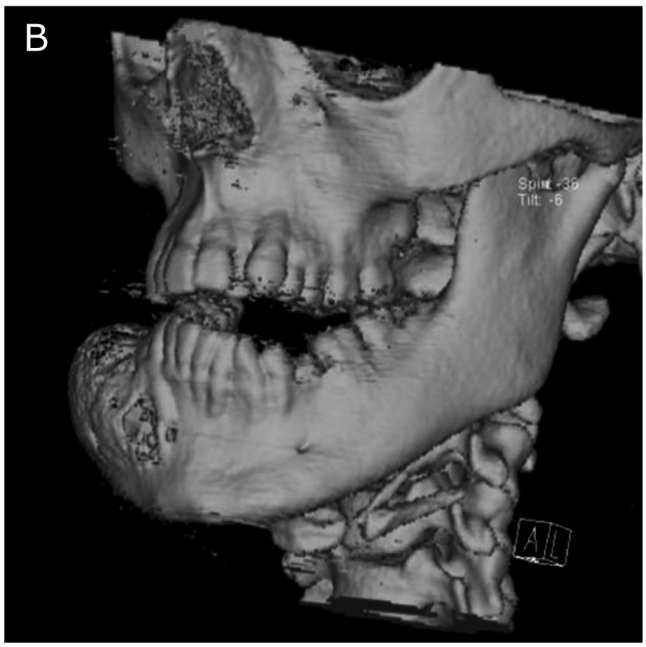


Fig. 3 A and B Initial 3D CT reconstruction showing the JAOF of the right body of mandible with moderate lingual expansion.

with near complete effacement of the buccal sulcus. The extent of growth was confirmed by a panoramic radiograph and CT scan which showed extension of the lesion across the midline towards the lower left canine region (Figs. 5 and 6). The lesion was measured at 6.2 × 4.2 cm in the transverse plane and 5 cm in the supero-inferior plane. The difference in size was very evident on the 3D CT taken after the delayed presentation (Figs. 7A and 7B). The treatment plan remained resection of the tumour but the plan for reconstruction was changed to a free fibula flap which could reconstruct both the both the body and symphysis of the mandible (Fig. 8)

Discussion

JAOF is a rare benign tumour. The two histological variants are the psammomatous and trabecular type (1). They occur most often in children under 15 years old (2). There is no gender and racial predilection. JAOF is a well defined histological entity that is now considered separately

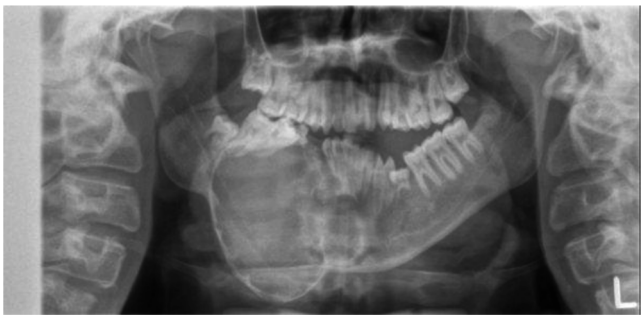


Fig. 5 Panoramic radiograph showing the increase in size of the lesion 35 months following the diagnosis.

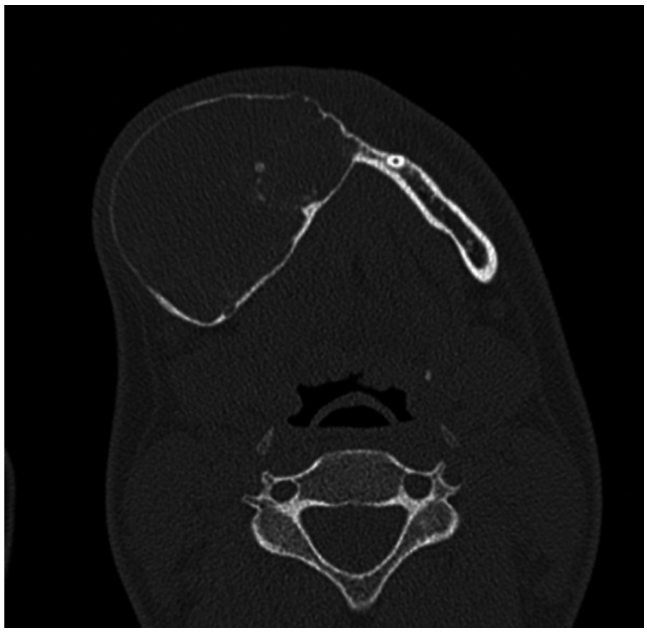


Fig. 6 Axial CT image confirming the increase in size of the lesion 35 months following the diagnosis.

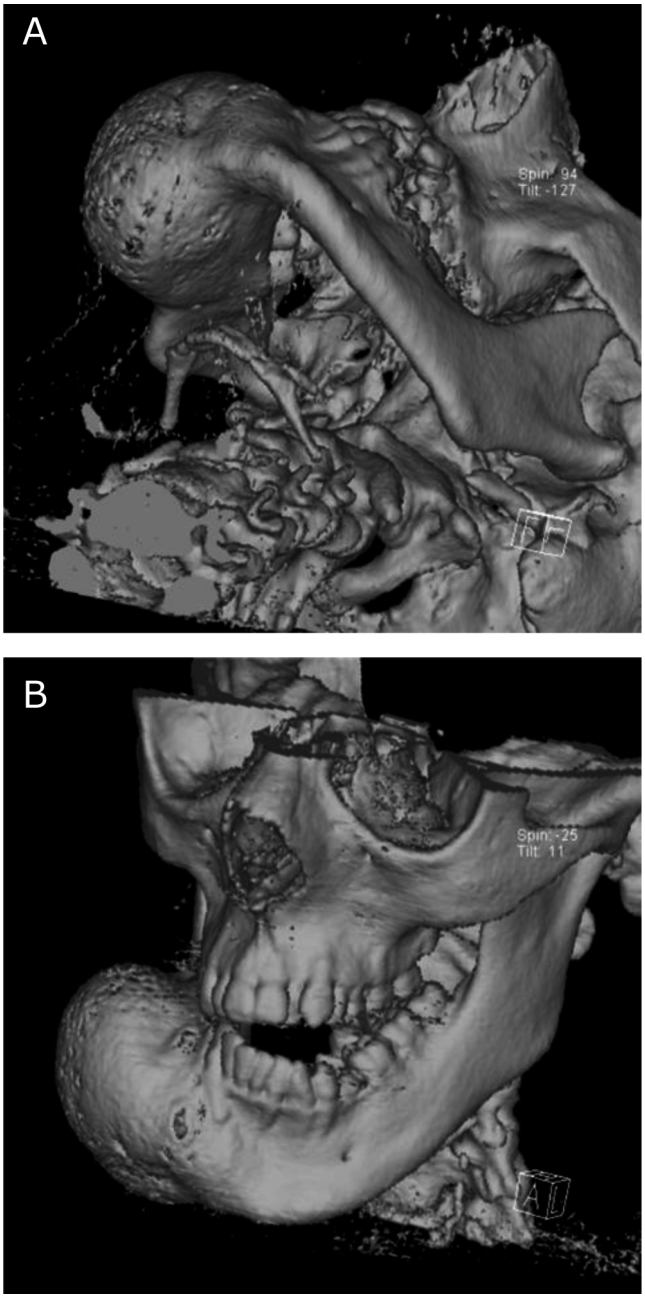


Fig. 7 A and B 3D CT reconstruction 35 months later showing extension of the lesion across the midline with marked bony expansion.

from other fibro-osseous lesions, including cemento-ossifying fibroma. The JAOF occurs in the craniofacial skeleton in 90% of cases, with the maxilla, paranasal sinuses, orbital and fronto-ethmoidal bones being the most common sites. Only 10% occur in the mandible (3,4).

The recommended treatment of JAOF is surgical excision with a 5 mm margin (5). Complete excision is curative. Recurrence rates for JAOF vary, with reports as high as 30% to 56% (6,7). Malignant transformation has not been reported.

In the case presented there was a delay between diagnosis and treatment of 35 months, which gave an opportunity to estimate the rate of growth of the tumour as, measured on CT scans using specialised software. The initial dimensions were 29×38 mm with an area of 571mm^2 when

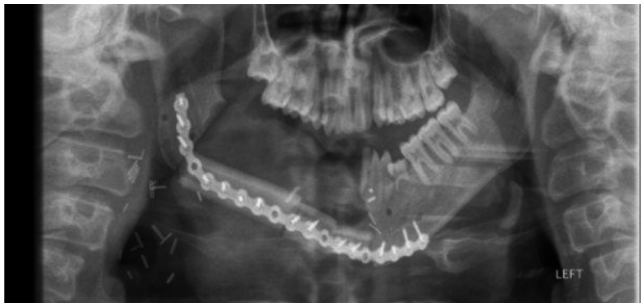


Fig. 8 Post-operative panoramic image showing reconstruction with a fibula free flap and a reconstruction plate.

calculated on axial imaging. The equivalent measurements 35 months later were 60 × 45 mm, with area of 2,089 mm^2. This indicates that the lesion had doubled in length and the cross-sectional area had increased more than 3.5 times.

To our knowledge this is the first report of an estimate of the rate of growth of JAOF. There are obvious limitations to this method of measurement based on two dimensional images. Also, we cannot assume that the rate of growth in this single case will predict the rate of growth of other such tumours. As with most tumours, there is likely to be a range of growth rates dependent on a number of unknown factors. However this case might provide a pointer to the potential rate of growth of these tumours which could be modified if further information from other tumours becomes available. In any event the rate of growth in the case reported seems to justify the term 'aggressive' of this particular pathology.

References

1. El-Mofty S (2002) Psammomatoid and trabecular juvenile ossifying fibroma of the craniofacial skeleton: two distinct clinicopathologic entities. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 93, 296-304.
2. Kramer IR (1992) The World Health Organization: histological typing of odontogenic tumours: an introduction to the second edition. J Dent Assoc S Afr 47, 208-210.
3. Leimola-Virtanen R, Vähätalo K, Syrjänen S (2001) Juvenile active ossifying fibroma of the mandible: a report of 2 cases. J Oral Maxillofac Surg 59, 439-444.
4. Lawton MT, Heiserman JE, Coons SW, Ragsdale BD, Spetzler RF (1997) Juvenile active ossifying fibroma. Report of four cases. J Neurosurg 86, 279-285.
5. Smith SF, Newman L, Walker DM, Papadopoulos H (2009) Juvenile aggressive psammomatoid ossifying fibroma: an interesting, challenging, and unusual case report and review of the literature. J Oral Maxillofac Surg 67, 200-206.
6. Noffke CE (1998) Juvenile ossifying fibroma of the mandible. An 8 year old radiological follow-up. Dentomaxillofac Radiol 27, 363-366.
7. Johnson LC, Yousefi M, Vinh TN, Heffner DK, Hyams VJ, Hartman KS (1991) Juvenile active ossifying fibroma. Its nature, dynamics and origin. Acta Otolaryngol Suppl 488, 1-40.