VERIFIABLE CPD PAPER

IN BRIEF

- Both arteriovenous malformations and solitary bone cysts can present in the mandible.
- Mental nerve paraesthesia can be a presenting feature with both types of lesions and diagnosis may be difficult.
- Arteriovenous malformations of the mandible can have potentially life-threatening complications.

Arteriovenous malformation of the mandible – A case report

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Both arteriovenous malformations (AVMs) and solitary bone cysts of the mandible are uncommon lesions. The latter can be considered fairly innocuous but AVMs require careful management. The following is a description of a case where an arteriovenous malformation of the mandible presented with mental nerve paraesthesia. However, radiographically the features appeared to be consistent with a solitary bone cyst. It is important for clinicians in both a primary and secondary care setting to be aware that this type of lesion can have life threatening complications.

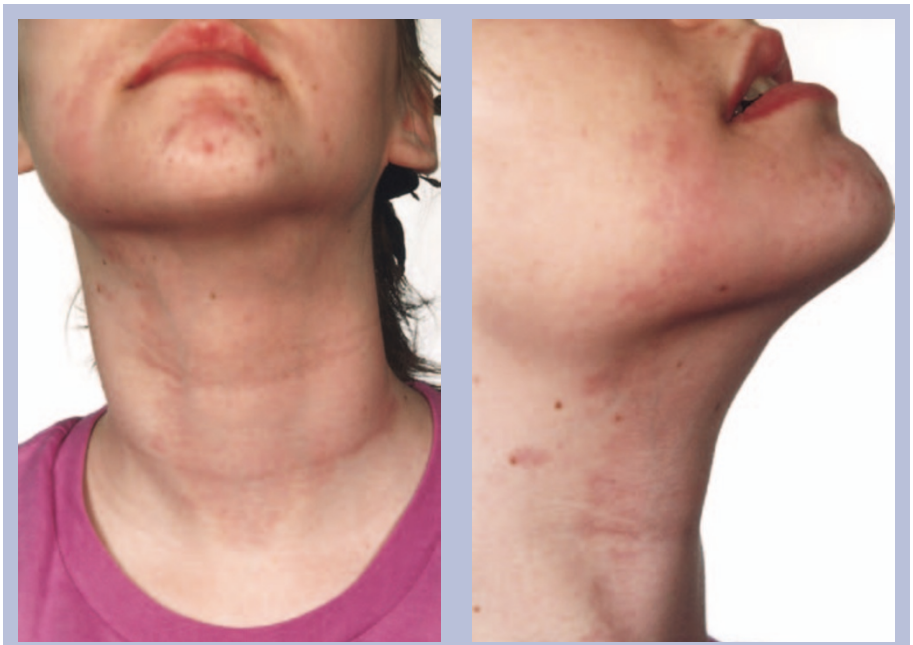
Arteriovenous malformations (AVMs) are rare and unusual lesions. It has been stated that AVMs of the mandible are very rare, although 50% of all AVMs within bone occur in the maxillofacial region.[1] Trauma induced by a biopsy or an extraction in the region of an AVM can lead to serious intra-operative bleeding, which could be life-threatening. Solitary bone cysts are also uncommon lesions. Hoffmeister and Härle reported only 19 cases of this type of cyst in their series of 3,353 reported jaw cysts. This corresponds to a frequency of 0.6%.[2] Howe[4] reported a review of 60 cases, in which the incidence of these cysts was greater in males and almost 78% of solitary bone cysts presented in the second decade of life. Solitary bone cysts of the jaws are usually detected as an incidental finding during a radiographic examination of the mandible and are not a risk to human life.

The following is a description of a case in which an arteriovenous malformation of the mandible presented with mental nerve paraesthesia but radiographically mimicked a solitary bone cyst.

CASE REPORT

A 14-year-old Caucasian female was referred to Manchester Dental Hospital by her GDP. Her presenting complaint was numbness affecting the lower right lip. This altered sensation had been first noticed three months earlier. In the history, the patient claimed there had been no history of trauma to the region, no swelling and no recent dental treatment.

The patient's general health was good, other than asthma that was well controlled by a salbutamol inhaler, and irritable bowel syndrome.



Figs 1a and 1b Clinical photograph showing diffuse bilateral vascular lesion affecting the perioral tissues and neck.

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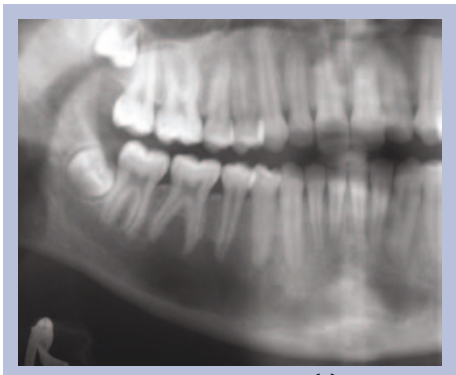
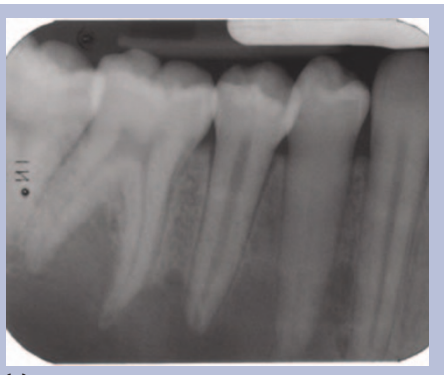




Figs 2a and 2b Dental panoramic (a) and periapical (b) radiographs of the patient at the time of presentation. There was a radiolucency in the right mandible extending from the distal root of the LR6 to LR3 (46-43). It arched up between the roots of the teeth with the loss of lamina dura but no visible root resorption. LR4 (44) showed sclerosis of the pulp.

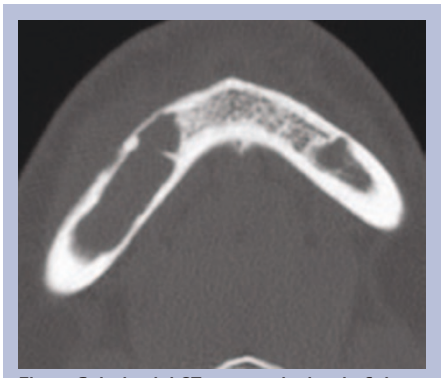


Fig. 3 Spiral axial CT scan at the level of the mandibular lesion, revealing the mandibular cavity and thinning of the buccal and lingual cortical plates.

Extra-orally, no lymph nodes were palpable and there was no tenderness in the submandibular regions. However, a diffuse bilateral vascular lesion was noted affecting the perioral tissues and neck (Figs 1a and 1b). According to the patient's mother this 'birthmark' had been present throughout life, but had become less conspicuous over time.

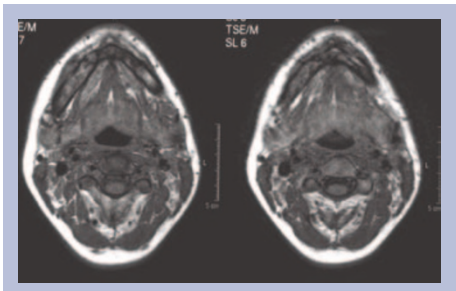
On intra-oral examination, there was no evidence of pathology in the lower right quadrant. The patient indicated that the paraesthesia extended from the midline to the lower right premolar region, which is consistent with the right mental nerve distribution.

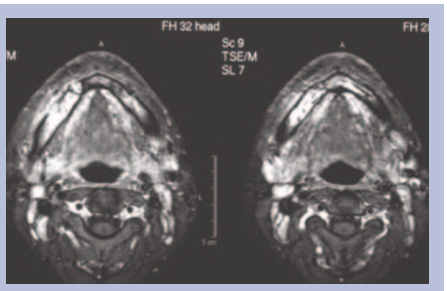
The following special tests were conducted:

1. Vitality testing was performed on the teeth in the lower right quadrant and no teeth (LR7-LR1) (37-31) responded to electrical pulp stimulation.
2. Two-point discrimination was used to determine the level of paraesthesia. Both the overlying skin and mucosa in the lower right quadrant were assessed for light touch sensation, using a cotton wool roll and crude touch via a blunt probe. The paraesthesia appeared to be more profound intra-orally to both light and crude touch.
3. Periapical radiographs of the LR7-LR1 (37-31) region and a 1/2 DPT of the right-hand side were taken (Figs 2a and 2b).

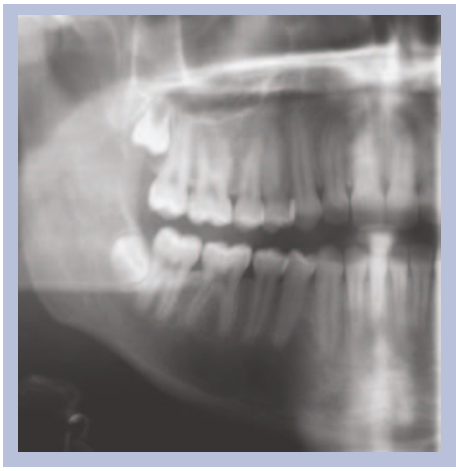
On the basis of the radiological findings, the provisional diagnoses were made:

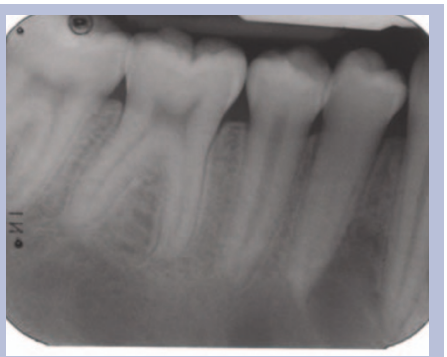
1. Solitary/haemorrhagic bone cyst (as indicated by the radiographic features)
2. Odontogenic keratocyst.





Figs 4a and 4b T1-weighted MR images show the bony cavity contains material of heterogenous signal intensity (a) that enhances markedly on administration of intravenous contrast (b). The soft tissues immediately adjacent to the buccal cortex also show contrast enhancement.





Figs 5a and 5b Dental panoramic (a) and periapical (b) radiographs of the patient at review showed a reduction in size of the radiolucency and more ill-defined margins, consistent with partial

After discussion with both the patient and her parents, it was felt that a treatment plan involving a surgical intervention would be required, in order to confirm the diagnosis. Due to the invasive nature of the procedure, it was decided that general anaesthesia would be appropriate.

A buccal mucoperiosteal flap was raised extending from the LR6-LR3 (46-43) region. A window of bone (7 mm x 5 mm) was cut below the apices of the LR6-LR5 (46-45). On elevation of this window of bone, spontaneous rapid haemorrhaging occurred with 500mls collected immediately. Both oxycell and swabs were used to control the bleeding with difficulty. Once the bleeding had arrested, bone wax was placed on the window and multiple vicryl sutures used for wound closure. It was noted that the initial diffuse vascular lesion (birthmark) present affecting both the mandible and neck bilaterally had become more prominent. The patient made an uneventful postoperative recovery and was discharged the following day.

At a one week post-operative review, the patient was healing well and reported no other symptoms. There had been no further postoperative bleeding. Taking into account the intraoperative findings it was felt that the most likely diagnosis was that of an arteriovenous malformation of the mandible. Preventive advice including oral hygiene instructions was provided for the patient emphasising the importance of reducing the need for future surgery to the lower right quadrant of the mandible. Spiral CT (Fig. 3) and MR scans (Figs 4a and 4b) with intravenous contrast were performed and a referral arranged for a neuroradiological opinion.

At a six-month postoperative review the patient informed us that the paraesthesia of the right side of her jaw had resolved and that normal sensation had returned. This was confirmed by two-point

discrimination; however, the teeth still gave a negative response to vitality testing. A DPT and periapical radiographs were taken. These showed an obvious decrease in the size of the radioluency between the roots of the LR6–LR3 (46-43) when compared to earlier radiographs (Figs 5a and 5b).

After consultation with the neuroradiologist it was decided that any definite treatment should be deferred and a period of monitoring should be instigated, given the clinical and radiological improvements and the risks, including that of cerebral vascular accident, should arterial embolisation be undertaken.

DISCUSSION

In the literature both AVMs and solitary bone cysts of the mandible can present with paraesthesia.[3,4] Diagnosis of AVMs cannot be solely achieved based on radiographic features. It has been stated that there are no distinct radiographic features to characterise AVMs on panoramic films.[5] However, Gelfand[6] described the following possible radiographic features: sunray appearance, soap bubble or honeycomb and an ill-defined radiolucency. In contrast, a classical radiographic feature associated with haemorrhagic cysts is 'scalloping', observed as a series of well-defined radiolucent extensions between the roots of the teeth,[7] as demonstrated in this case (Fig. 2).

Based on the radiographic findings in this case, it was reasonable for the investigators to assume a provisional diagnosis of solitary bone cyst. The standard approach for the surgical treatment of this cyst involves raising a buccal mucoperiosteal flap adjacent to the cyst, removing a window of buccal bone, removing the contents for histopathological examination and suturing the wound. In most cases this surgical procedure is curative. Curettage of the bony walls of the cyst has also been advocated.[7,8]

As described above, the patient reported that her paraesthesia had resolved at six months and radiographically the radiolucency in the LR6-LR3 region appeared to be regressing. These findings are consistent with a diagnosis of solitary bone cyst. However, the degree of haemorrhage encountered at surgery did not support this diagnosis. The intraoperative haemorrhage and the increased prominence of the birth mark, suggested rather that the lesion was an AVM. This was later confirmed by a MR scan of the mandible. Historically, AVMs have been treated with bone wax or by injecting sclerosing agents.[9] Current concepts of management include interventional radiology techniques such as arterial embolisation or transcutaneous transosseous embolisation and/or a surgical approach which includes either enucleation or resection of the AVM.[10]

It is not unreasonable to assume that the apparent bony in-filling of the AVM may have been stimulated by the placement of bone wax. In addition, it would be unwise not to consider that both lesions may have possibly co-existed in the same region of the mandible.

This case describes a situation in which a classical radiographic appearance of a lesion misled the clinicians and resulted in a potentially life-threatening complication. The case highlights the importance of performing an extra oral examination, as the observation of the skin lesion contributed to the development of the final diagnosis.

1. Unni K K, Ivins J C, Beabout J W, Dahlin D C. Hemangioma, hemangiopericytoma and hemangioendothelioma of the bone. *Cancer* 1971; **27:** 1403-1414.
2. Hoffmeister B, Härle F. Cysts in the maxillofacial region — A catamnestic study on 3,353 cysts. *Dtsch Zahnarztlz Z* 1985; **40:** 610-614.
3. Kelly D E, Terry B C, Small E W. Arteriovenous malformations of the face scalp. *J Cadiovasc Surg* 1968; **9:** 109-140.
4. Howe G L. Haemorrhagic cysts of the mandible-I. *Br Dent J* 1965; **3:** 55-76.
5. Woods N K, Goaz P W. *Differential diagnosis of oral lesions.* 4th edn. p 285. St.Louis: CV Mosby Co, 1991.
6. Gelfand G. Central cavernous hemangioma of the mandible. *J Oral Surg* 1975; **33:** 448.
7. Shear M, Seward G R. Solitary bone cyst (traumatic, simple, haemorrhagic bone cyst). *In Cysts of the oral regions.* 3rd edn. pp171-176. London: Wright, 1992.
8. Kaffe I, Littner M M, Buchner A. Traumatic bone cyst. *Quint Int* 1982; **13:** 469-472.
9. Martis H. Central hemangioma of the mandible. *J Oral Surg* 1973; **31:** 613.
10. Giaoui L, Princ G, Chiras J *et al.* Treatment of vascular malformations of the mandible: a description of 12 cases. *Int J Oral Maxillof Surg* 2003; **32:** 132-136.