Arteriovenous Fistula of the Mandible Simulating an Odontogenic Cyst

A Case Report

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Summary

We describe a rare case of mandibular high-flow arteriovenous malformation (AVM) mimicking an odontogenic cyst in a young man.

The diagnosis of mandibular AVM was made by CT angiography and confirmed by digital subtraction angiography. CT scan showed the extent of mandibular bone alteration and a double enlarged mandibular canal on the same side. An AVM containing a large aneurysm was demonstrated by CT angiography. The mandibular AVM was successfully treated by endovascular therapy with Guglielmi detachable coils.

On panoramic radiogram, mandibular AVMs can appear as cystic lesions without pathognomonic features. Several benign and malignant tumours of this anatomical region must be considered in the differential diagnosis. We emphasize the radiological sign of double enlarged mandibular canal and the diagnostic role of CT, particularly CT angiography, to discriminate a mandibular AVM from neoplastic entities of this region, sparing the risks of a needle biopsy.

Introduction

Arteriovenous malformations (AVMs) involving the mandibular bone and the dental arcade are considered rare entities, representing about 5% of all vascular malformations [1].

Mandibular AVMs are usually of arterial type with high flow and potentially fatal because of frequent copious bleeding [1].

Traditional treatment includes surgical resection with mandibular reconstruction. Conservative treatments are direct injection therapy and endovascular embolization.

Case Report

A 23-year-old man with a history of blunt trauma (martial arts) was referred to the maxillofacial surgery department of our hospital with a previous diagnosis of odontogenic cyst of the mandible. The patient reported paresthesia of the lower lip for the last six months.

A panoramic radiograph disclosed a lytic lesion with benign radiological features in the right mandibular body, next to 4.4, 4.5 and 4.6 teeth roots. (figure 1A). These teeth had been endodontically treated by a dentist because of trauma to the teeth roots during surgery.

An intraoral incision was made in the 4.4-4.7 region. The vestibular mucosa was unstuck and the intraosseous lesion exposed. The malformation showed a vascular-like consistency. It was throbbing and easily removable from the bone. In the light of the intra-operative finding, the surgical procedure was interrupted.

A 16-row multidetector CT scan confirmed the extent of bone alteration in the right hemimandible and showed a double, enlarged mandibular channel on the same side (figure1 B,C).

CT angiography was performed and processed with maximum intensity projection (MIP) and the volume rendering technique (VRT).

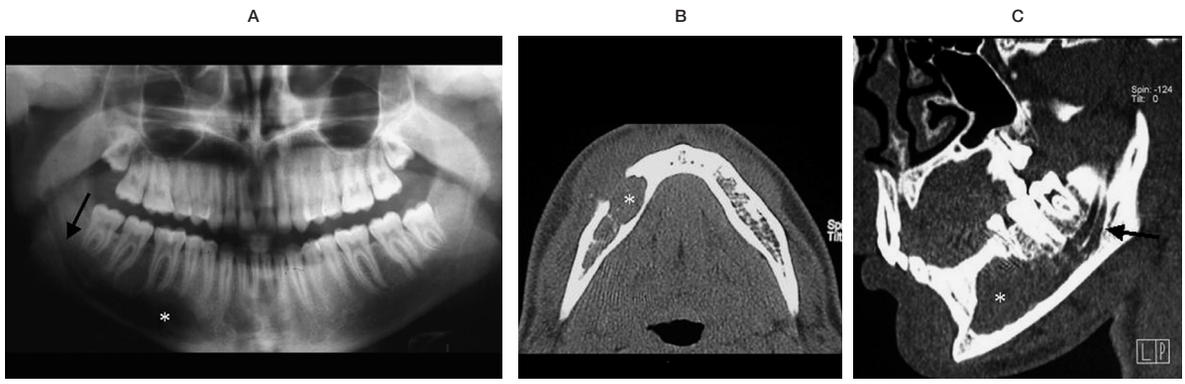

Figure 1 Panoramic radiograph (A), axial (B) and parasagittal (C) CT scan with bone algorithm. "Benign" lytic lesion in the right mandibular body (asterisk); double mandibular channel on the same side (arrow).

An aneurysmal dilatation of a vessel was demonstrated within the bone lesion (figures 2 and 3). A diagnosis of mandibular AVM was made and the patient underwent digital subtraction angiography.

Angiographic evaluation of the right external carotid artery was carried out by transfemoral catheterism. A venous aneurysm with a diameter of 2 cm was demonstrated within the bone lesion (figure 4). The aneurysm was the enlarged venous drainage of an arteriovenous fistula supplied by the inferior alveolar artery, a branch of internal maxillary artery. Moreover, the fistula was supplied by two terminal branches of the submental artery, anastomosed with the mental artery (figure 5).

Super-selective catheterization and embolization of feeding arteries was successfully performed with Guglielmi detachable coils (GDC) (figures 6 and 7).

CT angiography performed six months after endovascular treatment showed a complete disappearance of the arteriovenous fistula.

Discussion

Mandibular AVMs are rare conditions that could be fatal if left untreated as the result of massive blood loss after tooth extraction or attempts to remove or biopsy the lesion [1-4]. Mandibular AVMs can be acquired or congenital. The acquired forms are usually caused by a deep penetrating trauma involving any area of the jaw and affecting different ages [1-3].

The congenital forms occur as a result of error in vascular morphogenesis. This type of mandibular AVMs can be present at birth and enlarge with physical growth. This overgrowth is connected to hormonal factors, vasomotor disturbances or trauma. Most commonly, they occur in the second decade of life and involve the molar region [2-4]. Some authors have noted a predominance in women (2:1), while others have reported no sex prevalence [5].

Mandibular AVMs may be asymptomatic or show various signs and symptoms including bruit, dental loosening, erythematous and ble-

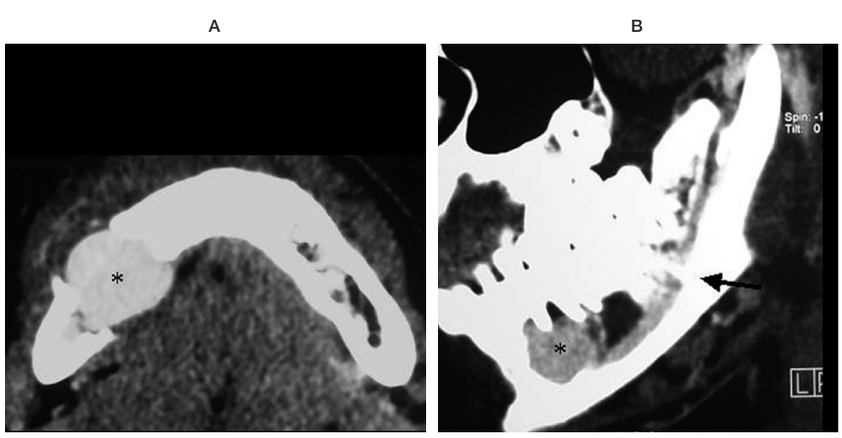

Figure 2 CT angiography axial (A) and parasagittal (B) images. Aneurysmal dilatation of a vessel is visible within the mandibular bone lesion (asterisks). An enlarged inferior alveolar artery and vein are also visible within the mandibular channel (arrow).

eding gingiva, swelling of the soft tissues of the face and dysesthesia of the lower lip or chin due to sensorial fifth nerve irritation [3].

Radiological diagnosis of mandibular bone pathologies is traditionally committed to panoramic radiography. Mandibular AVMs can appear as cystic lesions without pathognomonic features. However, evidence of an expanded (greater than 6 mm in diameter) or double mandibular channel on the same side as the lesion suggests a mandibular AVM. Bone expansion is probably due to intraosseous dilatation of the inferior alveolar artery and vein [2-4].

This essential radiological sign can be observed on panoramic radiography and, better, a CT scan. A double mandibular canal in the absence of a lacunar osteolysis is not clearly indicative of pathology, but an expression of an normal anatomical variation [5].

The role of CT in the diagnosis of a mandibular AVM has been greatly improved by the introduction of CT angiography. CT is the gold standard for detection of lytic changes in mandibular bone and the alteration of mandibular channel. Moreover, CT angiography will safely disclose vessels within the bone, discriminating a mandibular AVM from neoplastic entities of this region, sparing the risks of a needle biopsy.

Digital Subtraction Angiography is irreplaceable for a precise definition of a mandibular AVM. This technique accurately defines the vascular architecture of the lesion, particularly the number of feeding vessels from the ipsi- and contralateral facial, lingual and maxillary arteries. This vascular map is essential for planning surgical or endovascular procedures.

Mandibular AVMs have traditionally been treated by surgery. Surgical treatment requires

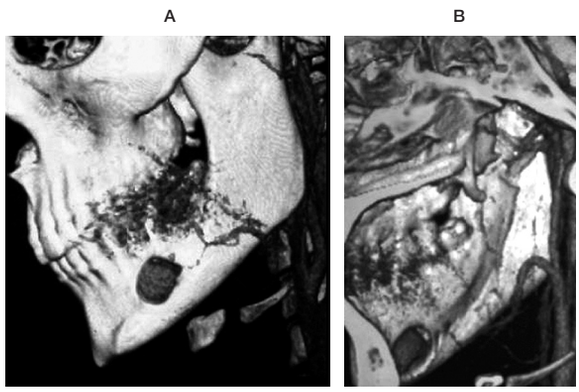

Figure 3 CT angiography parasagittal VRT images (A,B) allow a three-dimensional, concomitant evaluation of both vascular and osseous alterations.

Figure 5 Angiographic evaluation of the arterial supply through the inferior alveolar artery after releasing platinum GDC coils (arrow).

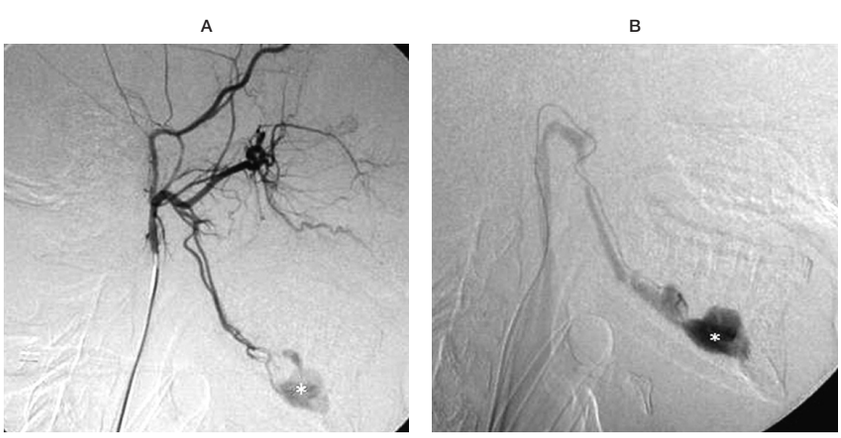

Figure 4 Digital subtraction angiography. A) Selective catheterization of the external carotid artery common trunk shows the arteriovenous malformation supplied by a split inferior alveolar artery. A 2 cm venous aneurysm is visible (asterisk). B) Superselective angiography of the inferior alveolar artery better shows the venous aneurysm (asterisk).

Figure 7 Post-embolization angiographic evaluation of alveolar and submental arteries by external carotid common trunk catheterization. The AV malformation is completely occluded.

Figure 6 Angiographic evaluation of the facial artery. Another arterial supply to the arteriovenous malformation is visible by retrograde flow from two terminal branches of the submental artery, anastomosed with the mental artery (arrow).

extensive bone resection with complex reconstruction to maintain the mandibular functions and prevent disfigurement [6,7,13].

Endovascular treatment of mandibular AVMs can be carried out by transfemoral catheterization or by direct puncture [8,9,10,11,12,14,15].

Either technique can be performed as a preliminary adjunct to excision and reconstructive surgery or as the sole definitive approach. In our case, endovascular treatment was preferred because the lesion was an arteriovenous fistula without an arteriovenular nidus: complete and durable treatment is difficult when a nidus is present [8,9,10,11,12,14,15].

In conclusion, we suggest the use of CT angiography in case of a well-circumscribed lytic lesion of the mandible combined with a double enlarged mandibular canal. Mistaking an arteriovenous malformation for a simple odontogenic cyst is very dangerous for the patient.

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