Oral and Maxillofacial Surgery Cases 1 (2015) 15–18

Contents lists available at ScienceDirect

Oral and Maxillofacial Surgery Cases

journal homepage: www.oralandmaxillofacialsurgerycases.com

Case report

Spontaneous regression of a mandibular arteriovenous malformation

Scott B. Raymond, MD, PhD [a], Leonard B. Kaban, MD, DMD [b], James D. Rabinov, MD [c,*]

[a] Department of Radiology, Massachusetts General Hospital, Boston, MA
[b] Department of Oral and Maxillofacial Surgery, Massachusetts General Hospital, Harvard School of Dental Medicine, Boston, MA
[c] Interventional Neuroradiology, Endovascular Neurosurgery, Massachusetts General Hospital, Harvard Medical School, Boston, MA

ARTICLE INFO

Article history:
Received 22 January 2015
Received in revised form
16 April 2015
Accepted 21 May 2015

Keywords:
Arteriovenous malformation
Lucent mandibular lesions
Endovascular embolization
Mandible
Developmental
Bleeding

ABSTRACT

Mandibular arteriovenous malformations (AVMs) are rare lesions that may initially present as catastrophic bleeding during dental surgical procedures. Owing to the significant risk of bleeding, most mandibular AVMs are treated definitively by resection or embolization. In this report, we describe a mandibular AVM that spontaneously regressed after biopsy.

1. Introduction

Arteriovenous malformations (AVMs) are high-flow vascular anomalies that often involve the mandible and surrounding soft tissues [1,2]. They are present at birth and expand during childhood [3], classically presenting in adolescence or adults with massive hemorrhage during tooth extraction [4]. When intraosseous components expand, the mandible may be deformed resulting in disfigurement [5], tooth loosening [4], and gingival bleeding [6]. Compression of the inferior alveolar nerve may cause pain and paresthesia with sensory deficits [7]. Because of the significant risk of bleeding, current practice is to definitively treat mandibular AVMs with embolization, resection, or a combination of techniques [4,8,9].

2. Presentation of case

A healthy 17-year-old male with no significant medical problems presented to his dentist with chief complaints of paresthesia and pain in the right lower lip and chin. Physical examination revealed no skin discoloration or external swelling, no warmth to palpation, and no palpable thrill or audible bruit. On intraoral examination, there was slight fullness in the right mandibular sulcus with mild tenderness. The sensory examination was normal. A panoramic radiograph demonstrated a radiolucent lesion of the right mandible with no tooth displacement, root resorption of the overlying first and second molars, or cortical thinning. The mandibular third molars were not present, and the underlying bone was unremarkable. A subsequent contrast-enhanced computed tomographic (CT) scan showed an enhancing lesion of the marrow cavity of the right mandible without evidence of osseous erosion or soft tissue mass. By report, the radiologist offered a differential diagnosis including fibrous dysplasia, Langerhans cell histiocytosis, lymphoma, or leukemia and recommended biopsy. During the biopsy, the oral surgeon excised a subcentimeter portion of the buccal cortex of the mandible to unroof the lesion and attempted needle biopsy. Extensive arterial bleeding was encountered, and the biopsy was aborted. Hemostasis was eventually accomplished with Gelfoam and direct pressure.

Approximately 3.5 months later, the patient was then referred to the Oral and Maxillofacial Surgery (OMS) service at the Massachusetts General Hospital, where a panoramic radiograph was obtained, again showing a radiolucent lesion of the right mandible (Figure 1). Contrast-enhanced CT was performed, demonstrating a homogeneously enhancing lesion filling the marrow space of the

* Corresponding author. Interventional Neuroradiology/Gray 241, Massachusetts General Hospital, 55 Fruit Street, Boston, MA 02114. Tel.: +1-617-726-1767; fax: +1-617-726-3089.
E-mail address: JRABINOV@mgh.harvard.edu (J.D. Rabinov).

http://dx.doi.org/10.1016/j.omsc.2015.05.001

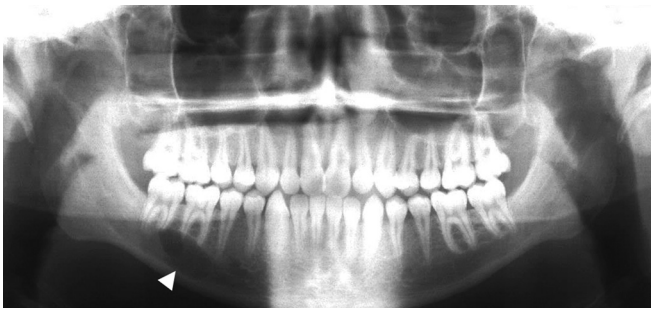

Figure 1. Panorex radiograph showing a rounded lucency in the body of the right mandible (*white arrowhead*).

right mandible, with venous drainage via a prominent mandibular vein (Figure 2). The CT appearance was unchanged compared with the prior study done 4 months earlier.

He then underwent diagnostic angiography which demonstrated a right mandibular AVM supplied by branches of the right facial artery and the mental branch of the left facial artery and drained by the right mandibular vein and external jugular system (Figure 3A). Early filling in the arterial phase confirmed that this was a high-flow lesion (arteriovenous [AV]), rather than a slow-flow vascular malformation. After multidisciplinary discussion with OMS and neurointerventional services, the patient and his mother elected that he should undergo endovascular arterial embolization of the AVM.

Approximately 2 months later, angiography demonstrated interval closure of the left mental branch feeding the AVM, leaving only a small residual AV shunt close to the angle of the mandible (Figure 3B). The draining right mandibular vein had decreased significantly in size compared with the prior angiogram. The OMS service was consulted during the procedure, and the consensus decision was to forego embolization at that time, with a plan to follow up the lesion over time.

Follow-up contrast-enhanced CT 4 months later showed near-complete thrombosis of the right mandibular AVM with partial trabeculation of the prior bony defect and normalization of the right mandibular vein. Angiography 11 months after the initial angiography showed no evidence of the right mandibular AVM previously seen (Figure 3C). Panoramic radiograph at roughly the same time point demonstrated trabecular bone filling in the prior defect in the right mandible (Figure 4).

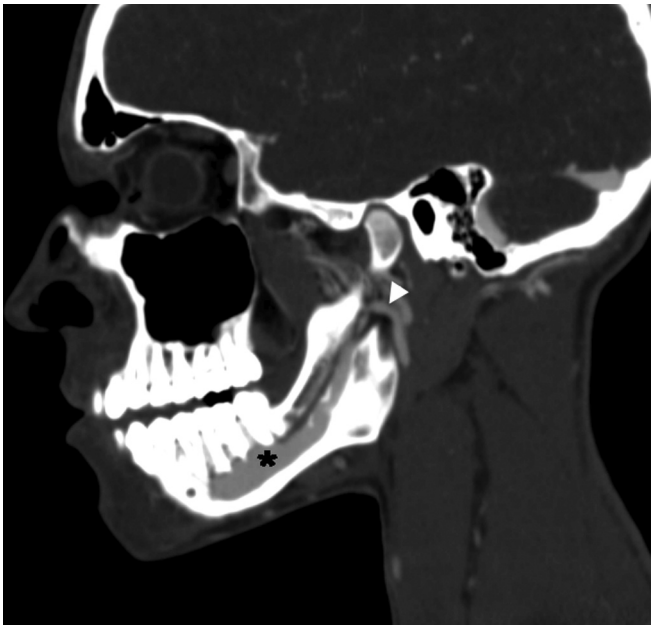

Figure 2. Curved reformat through the right mandible from a contrast-enhanced computed tomography scan showing a homogeneously enhancing lesion of the right mandible (*asterisk*) with prominent draining vein (*white arrowhead*).

3. Discussion

AVM of the mandible is a rare lesion that can present with arterial bleeding during a dental procedure. The exact incidence of mandibular AVMs is not known but, based on case series, is estimated at 1 in 50,000 [10-12]. Diagnosis can be difficult, given that the radiographic and CT appearances are variable and overlap considerably with other radiolucent mandibular lesions, including giant cell tumors, aneurysmal bone cysts, dentigerous and simple bone cysts, ameloblastoma, and keratocystic odontogenic tumors. Mandibular AVMs classically present as multilocular radiolucent lesions with cortical thinning and tooth displacement. The mandible can be expanded and demonstrates abnormal or absent trabeculation. In some cases, however, mandibular AVMs present as a more benign appearing unilocular radiolucent lesion.

In this case, diagnosis of an AVM of the right mandible was made by angiography after the patient presented with pulsatile bleeding during a surgical biopsy. This AVM had a particularly benign appearance on initial radiographs: it was nonexpansile, had a narrow zone of transition, no bony erosion, and no tooth root disruption. Contrast-enhanced CT demonstrated marked homogeneous enhancement in the marrow space with prominent draining veins, highly suggestive of a vascular lesion but not diagnostic. The final diagnosis was only confirmed after dynamic imaging by angiography.

Given the nonspecific and variable radiographic appearance of mandibular AVMs, it is important to correlate imaging findings with clinical symptoms such as pain, paresthesia, or other sensory deficits, which may be the only clue to the underlying diagnosis. Furthermore, when planning to biopsy a radiolucent lesion in the mandible, the oral surgeon should start with a fine needle aspiration first to exclude the possibility of an arterial lesion.

In this patient, the AVM regressed over 12 months and was replaced with trabeculated bone, with no active intervention beyond diagnostic transfemoral cerebral angiography. Spontaneous regression has been reported for intracranial AVMs and dural AV fistulas but, to our knowledge, has not been described in mandibular AVMs. Intracranial AVM regression is rare, occurring in $<$0.9-1.3% of all lesions [13,14]. The mechanism of regression is not well understood, although in the majority of cases, there is hemorrhage and/or thrombosis of a single draining vein [15,16]. Dural AV fistulas can also undergo spontaneous closure, which appears to be related to thrombosis of the vein or venous sinus at the confluence of the shunt [17,18]. Mandibular AVMs most closely resemble these lesions, with multiple AV shunts centered over a single vein or sinus.

In the case reported here, there was hemorrhage after a focal cortical unroofing and needle biopsy that may have caused partial thrombosis or altered hemodynamics within the AVM. However, subsequent computed tomographic angiography and angiography approximately 4 months after the biopsy showed no evidence of partial thrombosis or regression, making this hypothesis unlikely. Cerebral angiography is also unlikely to have resulted in thrombosis or regression of the feeding mental branch. Some have suggested that endothelial damage from intravenous ionic, high-osmolar iodinated contrast agents may cause endothelial damage leading to thrombosis [19]. However, the nonionic, low-osmolar contrast used at our institution is theoretically much less thrombogenic and unlikely to induce slow regression of the lesion over 12 months.