Transarterial embolization of a high-flow intraosseous arteriovenous malformation of the mandibule causing cerebral manifestations

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Vascular anomalies often affect the soft tissues though primary intraosseous lesions and are uncommon. Arteriovenous malformations (AVM) of the mandibule are a rare lesion, which are probably hamartomas. We report a case of 8-year-old girl who presented with exanguinating hemorrhage after the extraction of the mobile right mandibullary second molar. The diagnosis of mandibular AVM was made by CT scan and confirmed by angiography. CT scan showed osteolytic picture at the right mandibular ramus blowing discreetly bone opposite the second molar. An AVM containing a large aneurysm was demonstrated by CT angiography. The mandibular AVM was successfully treated by endovascular therapy complicated by cerebral embolus migration with a left hemiplegia.

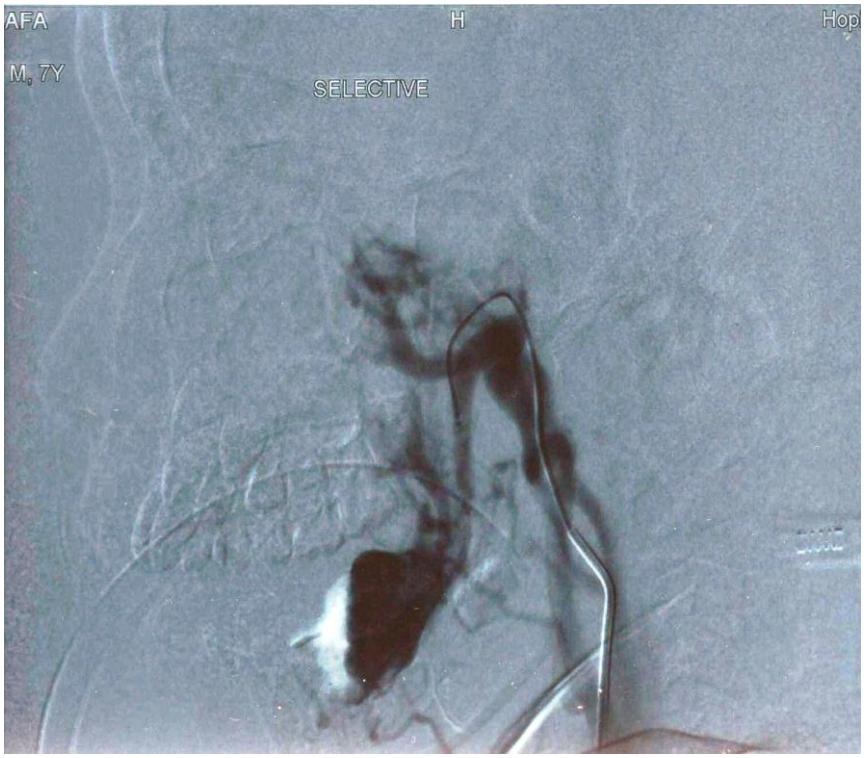
Introduction

Arteriovenous malformations (AVMs) involving the mandibular bone and the dental arcade are probably hamartomas. There are considered as rare entities, representing about 5,5% of all vascular malformations [1]. Mandibular AVMs are usually of arterial type with high flow and potentially fatal because of frequent copious bleeding. AVMs can either be congenital or acquired. Congenital AVMs occur as a result of lack of differentiation of arteries, veins, and the capillaries during vascular development, there is a persistent communication between them resulting in short circuiting of blood. The acquired malformations are usually associated with a previous history of surgery or blunt trauma [3]. Traditional treatment includes surgical resection with mandibular reconstruction, conservative treatments are direct injection therapy and endovascular embolisation but the literature does not mention any one particular modality as the treatment of choice.

Case report

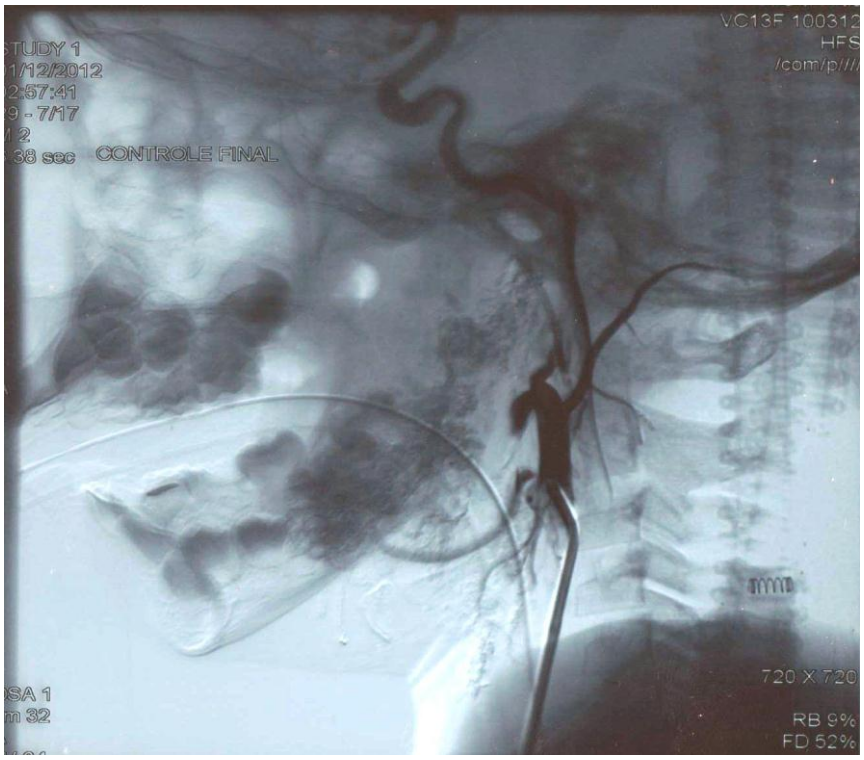
An 8-year-old girl with no past and family history of bleeding and coagulative disorders presented with massive bleeding from the oral cavity after the extraction of the mobile right mandibullary second molar. Immediately after extraction, a significant amount of bleeding was noticed through the socket which unusual to a routine extraction. When she reported to our department, she was in a low general condition due to prolonged bleeding with poor hemodynamic status and quick deglobulisation. On examination of the socket, there a pulsatile clot present. The patient was managed by blood transfusion, antibiotics and palliative measures. The bleeding was arrested with a ribbon gauge packed into the socket and biting pressure. All hematological investigations performed were within normal limits. A CT scan was performed which showed a multilocular radiolucent lesion involving the right ramus of the mandible. Finally an angiogram of the right external carotid artery was performed and revealed a large intraosseous arteriovenous malformation with multiples feeders from the right external carotid artery (Figs. 1a,b). The patient was planned for embolisation of feeding arteries under general anesthesia, during embolisation a cataclysmic hemorrhage occurred and generated a deglobulisation and shock requiring intensive care in the ICU for 48 hours. After extubation, we discover flaccid left hemiplegia. An MR angiography demonstrated a large high-signal intensity area in the right frontal and parietal parenchyma (Figs 2a,b). This accident was supported by sweet physiotherapy with a motor recovery after three weeks.

Figure 1a



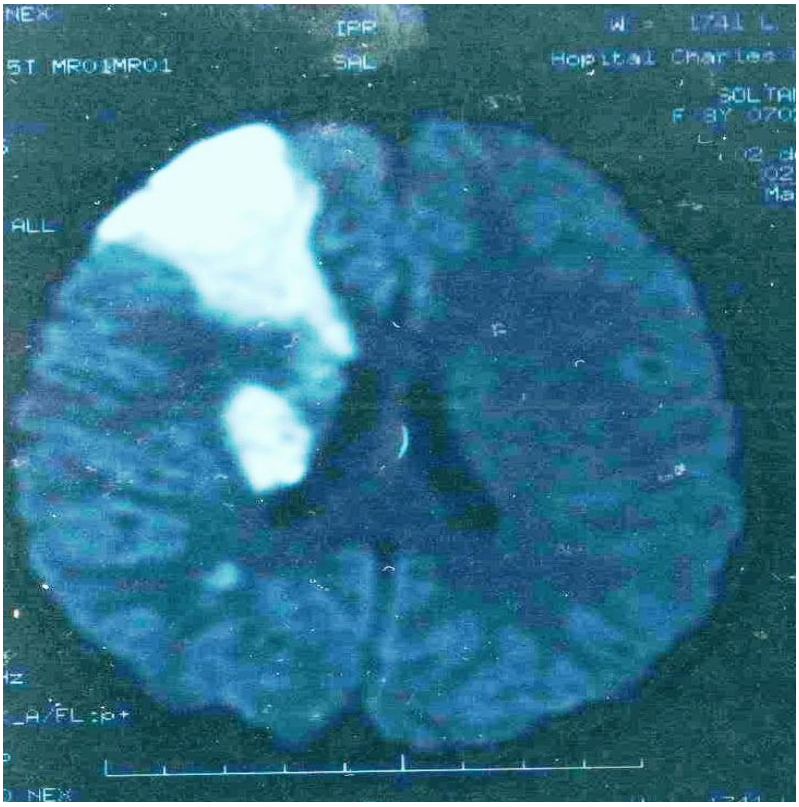
Right carotid angiogram revealing engorged vessels and central vascular filling of the lesion

Figure 1b



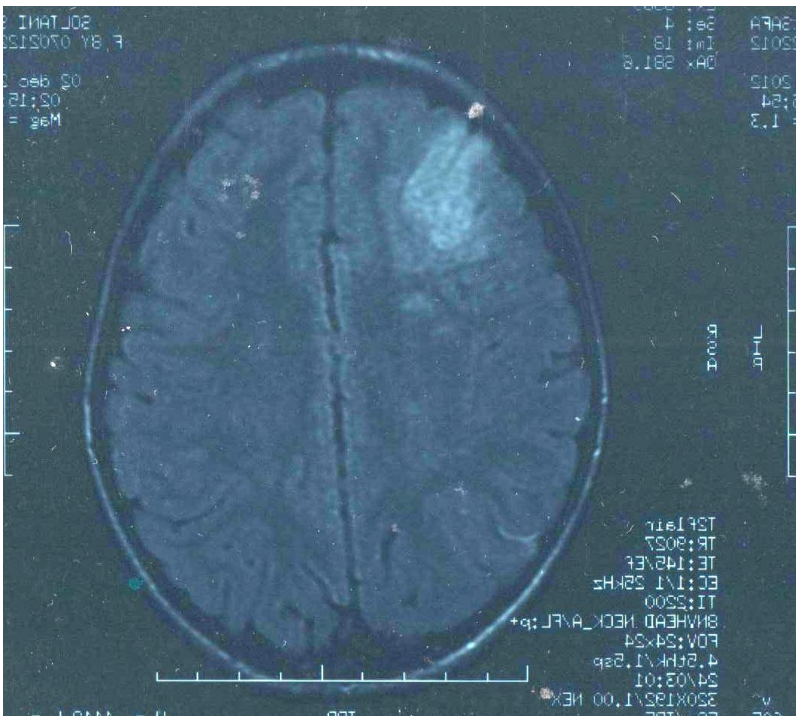
Right carotid angiogram after endovascular treatment of the AVM

Figure 2a



Brain MRI showing multiple lesions of temporo-parietal and frontal ischemia straight

Figure 2b



Brain MRI showing multiple lesions of temporo-parietal and frontal ischemia straight

Discussion

Mandibular AVMs are rare conditions that could be fatal if left undertreated as the result of massive blood loss after tooth extraction or attempts to remove. The 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. 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]. Some authors have noted predominance in women [2]. In contrast to hemangiomas, they never proliferate or involute. Mandibular AVMs may be asymptomatic or show various signs and symptoms including bleeding gingiva, dental loosening, swelling of the soft tissues of the face and dysesthesia of the lower lip due to sensorial fifth nerve irritation [3].

Radiological diagnosis of mandibular AVMs is tradionally committed to panoramic radiography that shows a cystic lesion without pathognomonic features. The essential radiological sign can be observed better with CT scan which reveals a multilocular radiolucent lesion involving the right ramus of the mandible with bone erosion. CT scan is the gold standard for detection of lytic changes in mandibular bone. The role of CT scan in the diagnosis of mandibular AVM has been greatly improved by the introduction of CT angiography. CT angiography will safely disclose vessels within the bone, discriminating a mandibular AVM from neoplastic entities of this region [4]. CT angiography is irreplaceable for a precise definition of a mandibular AVM and shows the exact angioarchitecture of the lesion, particularly the number of feeding vessels. This vascular map is essential for planning surgical or endovascular procedures. Our patient was planned for selective arterial embolisation, followed by direct injection of the intraosseous nidus and draining veins with tissue adhesive results in obliteration of the intraosseous AVM. While embolisation is the preferred treatment option it is not without risks. Potential complications of embolisation include puncture site hematoma, arterial dissection, and migration of the embolisation material, stroke and infection [5]. In the present observation endovascular embolisation was preferred. Which marks the originality of our observation is that it was complicated by concomitant cataclysmic hemorrhage requiring intensive care and embolisation material migration generating an ischemic stroke.

Conclusion

AVMs are a rare condition but potentially life threatening. It could be fatal because of frequent copious bleeding. It is usually present with exanguinating hemorrhage and best treated by a combination of transarterial and direct embolisation.

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