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Surgical technique for the treatment of high-flow arteriovenous malformations of the mandible

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SUMMARY. **The high-flow intraosseous arteriovenous malformation is a problematic vascular lesion which may affect bone and the dentition. Variable clinical presentations of this anomaly have resulted in a gamut of treatment modalities being reported ranging from simple curettage, resection, radiotherapy, sclerosing injections, and various forms of embolization, to immediate replantation of the resected segments. Embolization techniques alone have not been universally successful and have often resulted in rapid development of collaterals from surrounding vessels. Definitive treatment has usually involved complete surgical resection (when feasible) either alone, or in combination with other modalities such as embolization. Jaw resection, however, is deforming and leaves a defect often requiring subsequent reconstruction of the hard and soft tissues and replacement of any teeth lost with the resected segment. We report a surgical technique to treat mandibular arteriovenous malformations, which permits ligation of the feeding vessels and provides access allowing for complete removal of the intraosseous lesion. At the same time it not only prevents facial deformity by preserving the mandibular bone and oral soft tissue, but also, and more importantly, may preserve the dentition as well. © 2000 European Association for Cranio-Maxillofacial Surgery**

INTRODUCTION

Intrabony arteriovenous malformations of the mandible are considered to be very rare (*Babin* et al., 1983; *Laskin*, 1985; *MacIntosh*, 1992; *Perrott* et al., 1994; *Fathi* et al., 1997; *Behnia* and *Motamedi*, 1997). In their review of the literature *Gallagher* et al. (1983) found 124 published cases. The lesion is comprised of a tangled mass of small confluent interconnecting arteries and veins without interpositioning arterioles (*Gallagher* et al., 1983; *Babin* et al., 1983; *Laskin,* 1985; *MacIntosh*, 1992; *Perrott* et al., 1994; *Behnia* and *Motamedi*, 1997; *Regezi* and *Sciubba*, 1999); 35% involve bones (*Perrott* et al., 1994). In contrast to the haemangiomas, they affect bone more often and do not involute (*Regezi* and *Sciubba*, 1999). The intraosseous arteriovenous malformation may be confined to the bone, but there often is involvement of soft-tissue components as well. It has no recognized association with trauma or other defined aetiologic factors and is usually present at birth already. It may have high-pressure draining veins (varices) and involve significant portions of the venous and external carotid arborizations (*MacIntosh*, 1992; *Regezi* and *Sciubba*, 1993; *Perrott* et al., 1994; *Behnia* and *Motamedi*, 1997).

Vascular jaw lesions in general have an overall 2 : 1 female to male ratio with a peak incidence in the second decade (*Laskin*, 1985). All vascular malformations are essentially similar in that blood passes from the arterial to the venous system without the normal interposition of a capillary bed and thus, usually have a high blood flow (*Laskin*, 1985; *Regezi* and *Sciubba*, 1993). However, arteriovenous malformations are frequently devoid of overt signs or symptoms until they expand causing facial asymmetry, bone destruction, tooth mobility, and bleeding. Hypertrophy of the buccal mucosa may become apparent (*Perrott* et al., 1994). Radiographs may demonstrate irregular bony rarefactions within the normal confines of bone, with expansion or cortical disruption, or a vague poorly circumscribed radiolucency mimicking other intraosseous lesions (*Laskin*, 1985; *MacIntosh*, 1992; *Behnia* and *Motamedi*, 1997; *Regezi* and *Sciubba*, 1999). Root resorption, and loss of lamina dura may ensue (*Laskin*, 1985; *Regezi* and *Sciubba*, 1993; *Perrott* et al., 1994; *Behnia* and *Motamedi*, 1997). Patients may also experience pain, pressure, toothache, earache, ocular pain, epistaxis, pulsations, spontaneous haemorrhage around mobile teeth, and compressibility of the teeth into their sockets (*Laskin*, 1985; *MacIntosh*, 1992, *Regezi* and *Sciubba*, 1993; *Perrott* et al., 1994; *Behnia* and *Motamedi*, 1997). Auscultation often reveals bruits, and palpation a pulse-synchronized beat or thrill (*Gallagher* et al., 1983; *Laskin* 1985; *Perrott* et al., 1994; *Behnia* and *Motamedi*, 1997; *Regezi* and *Sciubba*, 1999).

Microscopically, the lesion consists of numerous arterial and venous vessels without muscular support, endothelial cell proliferation, neovascularization and giant cells (*Gallagher* et al., 1983; *Laskin*, 1985; *Perrott* et al., 1994; *Behnia* and *Motamedi*, 1997). Capillary and lymphatic channels may be present. Lesions of purely one type of vessel have also been

encountered (*Gallagher* et al., 1983; *Regezi* and *Sciubba*, 1993; *Behnia* and *Motamedi*, 1997).

Needle aspiration and angiography as well as ultrasound are of diagnostic value in both pre- and postoperatively. They also help differentiate these lesions from other haemorrhagic lesions such as the aneurysmal bone cyst, which it may mimic (*Motamedi* and *Khodayari*, 1993; *Motamedi*, 1998). The latter does not demonstrate filling of the lesion with angiography (*Motamedi* and *Khodayari*, 1993; *Motamedi* and *Yazdi*, 1994; *Motamedi*, 1998).

Like all vascular lesions of the jaws and dentoalveolar structures, arteriovenous malformations (AVM) are potential threats to life and deserve special consideration. Many surgical and nonsurgical methods have been used alone or in combination to treat symptomatic and haemorrhagic arteriovenous malformations. Nonsurgical methods have included radiotherapy, injection of sclerosing solutions, and various types of embolization. *Rodesch* et al. (1998) reported a 34% cure rate using acrylic glue in 12 lesions of the dental arches. The primary surgical techniques for accessible lesions has been some form of excision or resection. Simple curettage alone although reported to be curative (*Fathi* et al., 1997) can be dangerous since it may result in exsanguinating haemorrhage in high-flow lesions. It also may carry a high incidence of recurrence if the peripheral vessels of the lesion are not adequately sealed. Excision of the lesion with safety margins has been advocated as the goal or treatment of choice (when the extent and site of the lesion permits (*Coleman*, 1973; *Laskin* 1985; *Regezi* and *Sciubba*, 1993; *Perrott* et al., 1994; *Watzinger* et al., 1997; *Behnia* and *Motamedi*, 1997). This ablative procedure, however, may necessitate future surgery for reconstruction and dental rehabilitation. It may also require reconstruction plates, splints, or space maintainers to preserve form and jaw function until reconstruction is performed. Harvesting bone grafts and immediate reconstruction in the same operation may well prove to be inconvenient, increases blood loss as well as operating time, and bone harvesting and grafting add to postoperative morbidity. Previously we reported treatment of a mandibular AVM by resection and immediate replantation of the cortical shell (*Behnia* and *Motamedi*, 1997). Although this technique was successful, preserved the basal bone, and prevented a facial defect with asymmetry, it required subsequent restoration of the dentition. The best modality would be to treat the lesion definitively (when accessible) whilst preserving the mandibular dentition. This report describes the technique which we have used successfully in two patients with mandibular lesions and compares the advantages of this technique with other treatment modalities.

CASE REPORT

A 13-year-old girl was referred to our Medical Centre on 17 July 1996 complaining of bleeding from the gums and a swelling of the right side of the face which she had noticed approximately one month earlier. The patient was in good general health, and did not have a history of facial trauma. Clinical examination was normal except for a non-tender facial swelling over the right mandibular body. On palpation, the swelling was soft, warm, and pulsatile. Bruits could be auscultated using a stethoscope.

Intraoral examination revealed visible mobility of the premolars and molars of that side. The teeth were depressable upon application of vertical pressure and marginal gingival bleeding was evident around these teeth. Engorgement of the submucosal buccal vessels was apparent when the cheek was retracted (Fig. 1).

Panoramic radiographic examination revealed a vague radiolucency and rarefaction in the body of the mandible extending posteriorly from the first premolar to the first molar on the right side. Loss of lamina dura and displacement of the first and second molars was evident (Fig. 2). Computed tomography scans of the lesion demonstrated central bone destruction, bilateral cortical expansion, and perforation of the lingual cortex (Fig. 3).

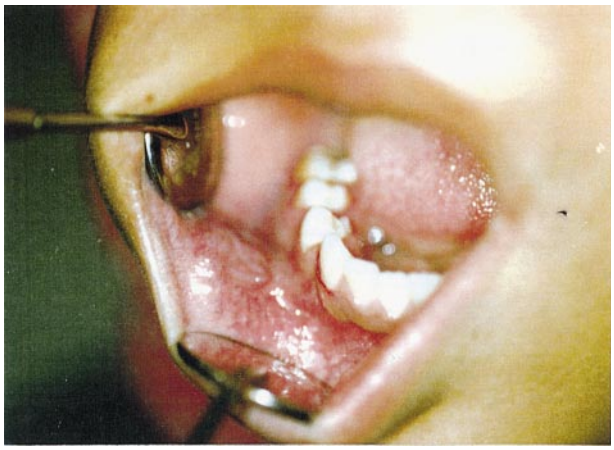

Fig. 1 – Intraoral view. Note engorgement of the submucosal blood vessels and bleeding of the gingival margin.

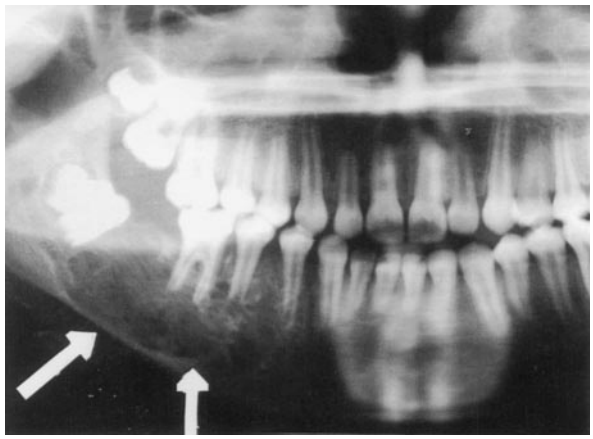

Fig. 2 – Panoramic radiograph of the patient demonstrating only a vaguely defined radiolucency and bone resorption in the right mandibular body region (arrows) and displacement of the first and second molars and loss of lamina dura.

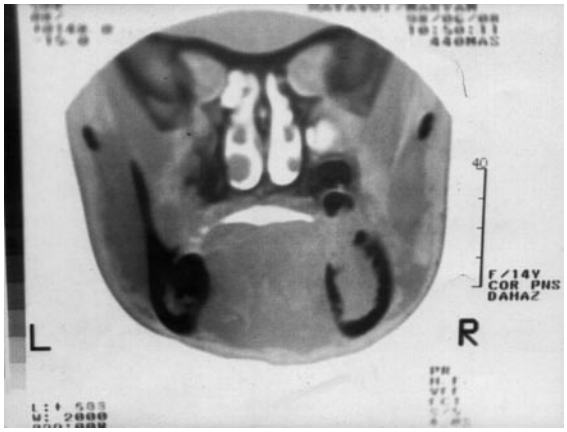

Fig. 3 – CT scan of the lesion demonstrating central bone destruction and bicortical expansion of the right mandibular body.

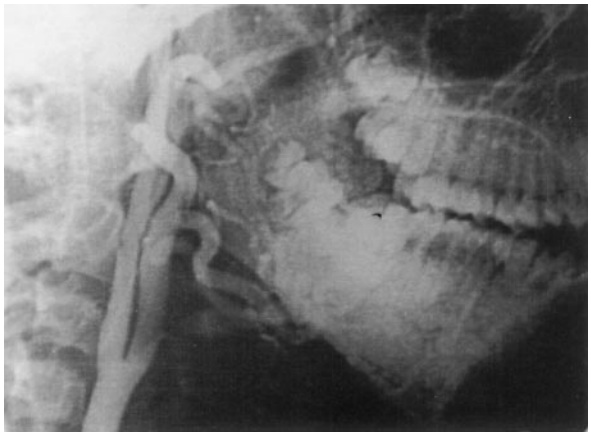

Fig. 4 – Right carotid angiogram revealing engorged vessels and central vascular filling of the lesion.

Right carotid angiography revealed enlargement and engorgement of the feeding vessels and vascular filling of the lesion in both the hard and soft-tissues (Fig. 4). It was decided to undertake surgical treatment for this high-flow vascular lesion.

Surgical procedure

Under general anaesthesia via nasoendotracheal intubation and hypotension (systolic blood pressure of 80 mm Hg), submandibular skin was incised and the marginal mandibular branch of the facial nerve was identified and retracted with the soft-tissues. The large, engorged convoluted vessels and varices were exposed, ligated, and transected. The lingual and facial arteries, and the facial and external jugular veins were also ligated and transected to reduce the peripheral blood supply of the lesion. After exposing the mandible, a longitudinal incision was made in the periosteum along the inferior border and the periosteum was gently reflected. The cribriform-appearing cortical bone had numerous, minute bleeding points which were effectively controlled using electrocautery as the periosteum was elevated. Two small buccal and lingual intraoral incisions were made distal to the right canine. After extracting the canine, the mandible was cut through the extraction site using a saw. Then the proximal segment of the mandible was retracted laterally with a bone hook, the intramedullary lesion was curetted and a large bur was used to obtain clean bony margins within the segment (Fig. 5). Intramedullary oozing was controlled by electrocautery. It was not necessary to section the mandible posteriorly as the bleeding had ceased. The mandible was then repositioned and fixed with two titanium miniplates (Fig. 6). The extracted canine tooth was replanted in its extraction socket and an Essig tension-band splint was placed on the adjacent teeth. Intermaxillary fixation was not retained. A suction drain was placed and the wound was closed in layers.

Postoperative course

The patient's postoperative course was uneventful. The pathology report confirmed the diagnosis of a mandibular arteriovenous malformation (Fig. 7). The follow-up panoramic radiograph demonstrates complete bone consolidation of the right mandible almost 2 years later (Fig. 8). All the previously mobile teeth were completely firm and functioning. The replanted canine and the first premolar were nonvital and were treated endodontically. There was no paraesthesia of the lower lip or signs of recurrence.

DISCUSSION

When resection is required to treat a benign mandibular lesion, the maintenance of facial symmetry, occlusion and aesthetics postoperatively is an important concern. This lead to the concept of immediate reconstruction introduced by *Weaver* and *Smith* (1973). He and others made use of the patient's own frozen mandibular bone to reconstruct the surgical defect (*Weaver* and *Smith* 1973; *Sakoda* et al., 1992). *Behnia* and *Motamedi* (1997), reported treatment of a vascular malformation by resection of the segment and immediate replacement (without freezing) after curetting the lesion from the bone and removing the teeth within the segment. Both techniques, although effective in restoring bone continuity and facial symmetry, were associated with some bone resorption (common with free non vascularized bone grafts). They also undertook dental rehabilitation of the edentulous hemimandible. The main advantages of our technique lies not only in preservation of bone and prevention of bone resorption but in this case it was possible also to preserve the patient's own dentition which quickly firmed up despite preoperative mobility. At the same time, the feeding vessels of the lesion were addressed prior to osteotomy and adequate margins in normal bone were achieved.

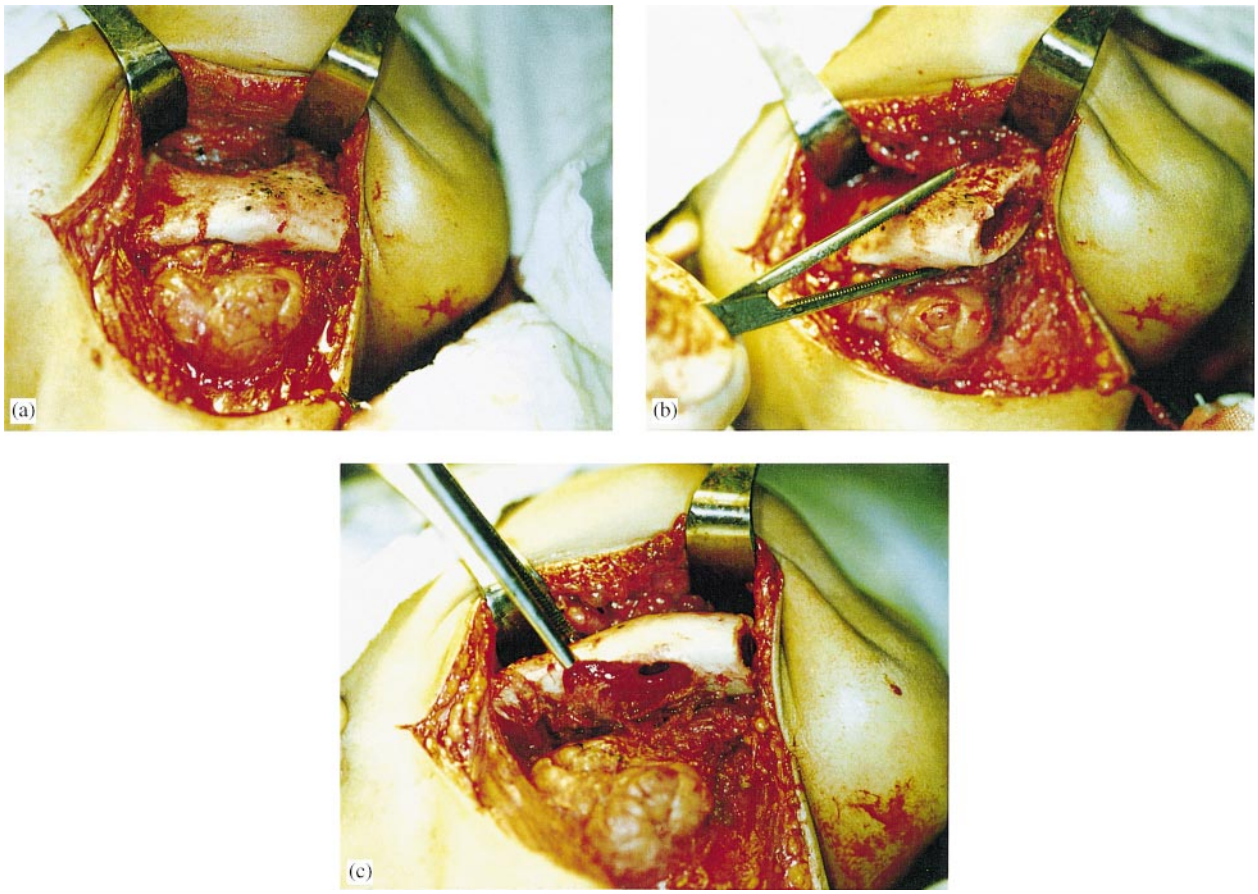

Fig. 5 – Intraoperative view of the mandible. (A) Note cribriform-like cortex which has been cauterized. (B) Bleeding effectively controlled after osteotomy, hollowing-out the bone, and cauterization inside the bone. (C) Perforated lingual cortex.

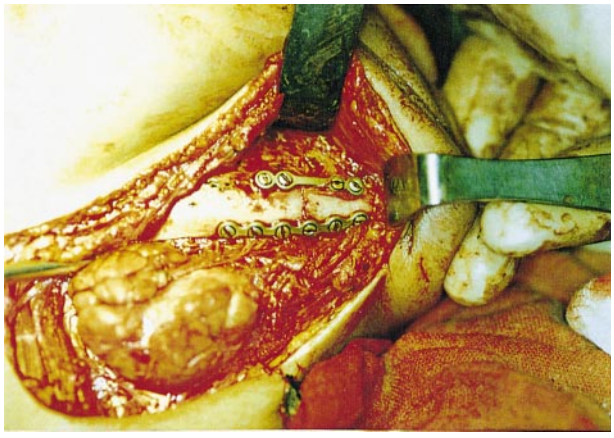

Fig. 6 – Intraoperative view after fixation with titanium miniplates.

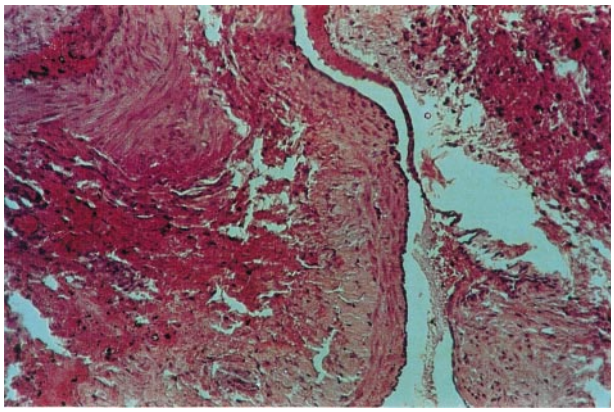

Fig. 7 – Thick-walled vessels of varying caliber, endothelial cell proliferation, neovascularization and an admixture of arterial and venous channels (H&E × 1000).

Additional advantages include: preservation of jaw function and facial symmetry, the option of preserving the alveolar nerve, no necessity for maxillomandibular fixation, and no secondary operation for reconstruction or dental rehabilitation.

Preoperative arterial embolization is used in an attempt to decrease haemorrhage intraoperatively. However, in arterialized (high-pressure) venous systems, the vascular pressure may not be reduced significantly by transarterial embolization alone (*Perrott* et al., 1994).

Depending on the site and the feeding vessels of the mandibular lesion, the osteotomy may be made anteriorly or posteriorly and it may then be removed by lateral distraction of the mandible. This can also be performed bilaterally.

Immediate replacement of the canine served several functions: 1. It prevents mandibular collapse which

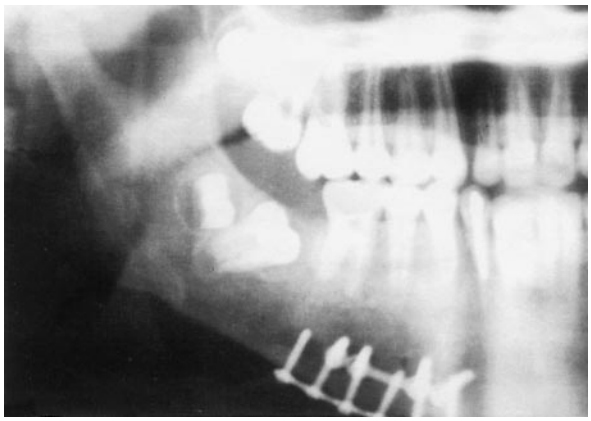

Fig. 8 – Postoperative panoramic radiographs showing bone consolidation and healing.

may occur when using compression plating. 2. It acts as a space maintainer to prevent migration of adjacent teeth into the extraction site until definite dental restoration can be done. 3. It restores function and aesthetics in the immediate postoperative period especially in preadolescence when jaw growth is continuing and definitive prosthodontic work should be delayed. 4. It preserves the site for future implant placement. If the root is gradually resorbed, it is replaced by bone and an implant can be placed immediately without having to wait for an extraction socket to fill with bone.

Extraction of the mobile teeth within the lesion may not be necessary if the roots and furcations are not involved as these teeth firm up and become functional postoperatively provided the crown to (embedded) root ratio is acceptable. *Perrot* et al. (1994) reported a case which required extraction because of repeated embolization during the 6-year course of the lesion which resulted in collateralization, expansion of the lesion, root resorption, and more mobility of the teeth. They reported having to extract three teeth because of life-threatening haemorrhage. Extraction resulted in 1100 cc blood loss despite the insertion of 228 Hilal coils for embolization through the sockets. Apparently, the lesion had extended as much that it could no longer be treated definitively.

It seems wise thus, to treat these lesions expediently after diagnosis and presurgical embolization as they tend to enlarge rapidly once activated, making surgical treatment more difficult or impossible with time. The single-site osteotomy technique with intramedullary curettage as described may be indicated in accessible jaw lesions without extensive bone destruction or dental involvement. It should be noted that the intraosseous component should be accessed only after all extraosseous feeders and soft-tissue components have been dealt with. The intrabony blood supply must then be tackled. In the event that single-site osteotomy and curettage proves insufficient to control haemorrhage intraoperatively, a proximal osteotomy will then be required. We would then prefer to reposition the segment. Long-term follow-up is mandatory in these patients. We have so far followed the two patients treated this way for 7 and 2 years respectively.

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