Osseous Regeneration after Embolization of Mandibular Arteriovenous Malformation

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A mandibular arteriovenous malformation (AVM) is rare, and unawareness of this condition can lead to catastrophic complications. Embolization is an effective means to treat these lesions. The osseous changes and long-term outcomes among the pediatric patients after embolization treatment remain unclear. We present a 6-year-old patient with mandibular arteriovenous malformation who had sustained hemorrhagic shock after a tooth extraction. She was referred to us after stabilization of the hemodynamic condition. The patient received transarterial and direct intralesional embolization. Local infection occurred 3 months after embolization, but was well controlled. Serial computed tomography and panoramic radiographs were performed during follow-up examinations. The imaging studies showed gradual obliteration of the vascular space followed by normal bone regeneration and remodeling. The processes were complete during a period of 2 years. There was no recurrence of the vascular malformation and no abnormal growth of the right side of the mandible after 4 years and 4 months of observation. *(Chang Gung Med J 2003;26:937-42)*

Key words: mandible, arteriovenous malformation, bone regeneration.

Mandibular arteriovenous malformations are recognized as uncommon and potentially life-threatening vascular malformations.[1-3] Young female patients are predominantly affected. These lesions frequently appear as high-flow vascular malformations[4] characterized by congenital dysmorphogenesis of the arterial and venous structures in the region involved.[5] Their clinical presentations vary from onset of minor gingival bleeding while brushing the teeth to dental loosening, lower lip numbness, facial deformity, malocclusion and even hemorrhagic shock following extraction of teeth.[2,6-8] The radiographic appearances of these lesions are variable ranging from a small radiolucency to a markedly obvious osseous erosion of the alveolus with apparently floating teeth. Computed tomography (CT) scans and magnetic resonance imaging (MRI) are helpful imaging tools to assess the extent of the lesions into the bone, soft tissue, and major vessels. While conventional angiography has been an important procedure for the diagnosis of mandibular arteriovenous malformations,[9-11] MR angiography is a much safer and less invasive procedure for the effective diagnosis of a lesion.

There is still no consensus among researchers in the management of the mandibular arteriovenous malformations. Because most clinically symptomatic patients with arteriovenous malformations are adolescents or younger, the continuous growth and development of the facial bones should be considered in the management of the lesions. While primary segmental excision or radical resection of the involved bone after preoperative embolization has been reported, recently authors have described that direct transosseous puncture or distant transvascular methods of embolization are the treatments of

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choice.[7,9-12] Among pediatric patients, the long-term results of the management of these lesions remains an essential topic to be considered in terms of both functional and esthetic outcomes.

CASE REPORT

A 6-year-old girl presented with an expansile mass at the right mandibular area for 5 weeks. Originally, it grew slowly, but it became large during the 2 weeks prior to admission. One month prior to admission, she had a tooth extracted that was followed by massive bleeding which was stopped using digital compression. Three days after the extraction, severe active bleeding from the oral cavity was noted. She was then admitted to a local hospital for the management of shock. She was discharged with a stable condition 1 week after admission, although the mandibular enlargement persisted and progressively became more noticeable. Hence, CT scan and angiography were performed and a mandibular vascular malformation was impressed. The patient was subsequently referred to our hospital for further care.

The physical examination revealed an expanding hard mass on the right cheek with a palpable thrill (Figs. 1A and B). The overlaying skin and intraoral mucosal surfaces were slightly discolored. Malocclusion with an anterior open bite was noted. The panoramic radiograph showed multilocular radiolucent lesions within the body and the angle of the right mandible (Fig. 2A). CT scan and MRI disclosed the right mandibular lesion with bony destruction and soft tissue swelling (Fig. 2B). Arteriovenous malformation was considered. Digital subtraction angiography revealed a huge vascular anomaly with direct arteriovenous shunts in the right mandibular region. The main arterial supply originated from branches of the right internal maxillary artery. Embolization of the feeding arteries was planned. Under general anesthesia, embolization with supplementary injection of a mixture of Lipiodol and n-butyl cyanoacrylate was performed. Gradual resolution of the local swelling was observed following the embolization. Local infection was noted 3 months following embolization, and it was successfully treated with curettages and antibiotic therapy.

Consecutive CT examinations with 3-dimensional image reconstruction of the head were per-

Fig. 1 The patient with the right mandibular ramus arteriovenous malformation showing the right cheek swelling. She was 6 years of age prior to embolization treatment, in anterior (1A) and face-up (1B) views.

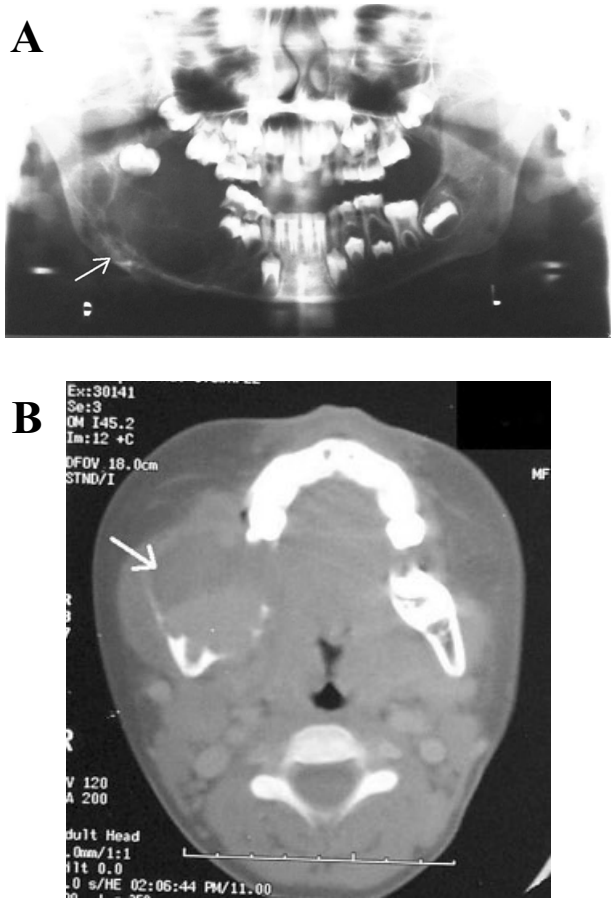

Fig. 2 Panoramic radiograph (2A) before embolization treatment showing enlarged right ramus with a space-occupying lesion. The teeth were missing and displaced at the lesion site. CT study (2B) at the same time showed destruction of the osseous structures and swelling of the peripheral soft tissue. The cortical bone was thin and eroded.

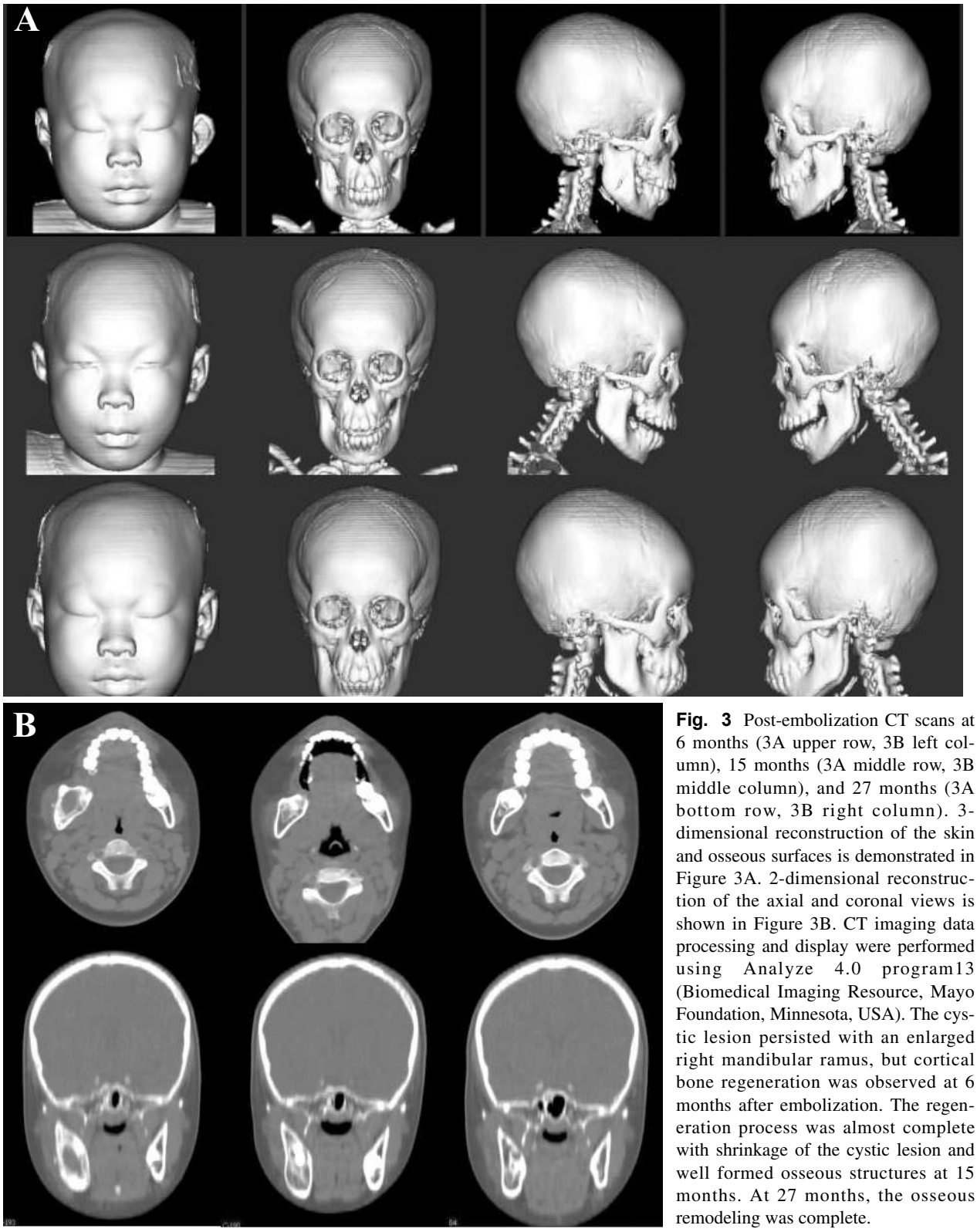

Fig. 3 Post-embolization CT scans at 6 months (3A upper row, 3B left column), 15 months (3A middle row, 3B middle column), and 27 months (3A bottom row, 3B right column). 3-dimensional reconstruction of the skin and osseous surfaces is demonstrated in Figure 3A. 2-dimensional reconstruction of the axial and coronal views is shown in Figure 3B. CT imaging data processing and display were performed using Analyze 4.0 program13 (Biomedical Imaging Resource, Mayo Foundation, Minnesota, USA). The cystic lesion persisted with an enlarged right mandibular ramus, but cortical bone regeneration was observed at 6 months after embolization. The regeneration process was almost complete with shrinkage of the cystic lesion and well formed osseous structures at 15 months. At 27 months, the osseous remodeling was complete.

formed at 6, 15, and 27 months after embolization for the evaluation of the vascular lesion and the osseous changes (Figs. 3A and B). At 6 months after embolization, enlargement of the right mandibular ramus was still present. Cortical bone regeneration was also observed. At 15 months after embolization, the right ramus had returned to close to normal shape. The cortical and cancellous bone structures were almost completely developed with only a small empty space left (Fig. 3B, middle column).[13] At 27 months after embolization, the osseous regeneration and remodeling of the right mandibular ramus were complete. There was no excessive osseous growth as compared with the previous CT images. Slight facial asymmetry with persistent mild right cheek swelling was noted (Fig. 3A, first column from the left). Serial panoramic radiographic examinations correlated with the changes observed on CT images (Figs. 4A and B). Four years after embolization, stable facial and mandibular growth and development were evident. There was no recurrence.

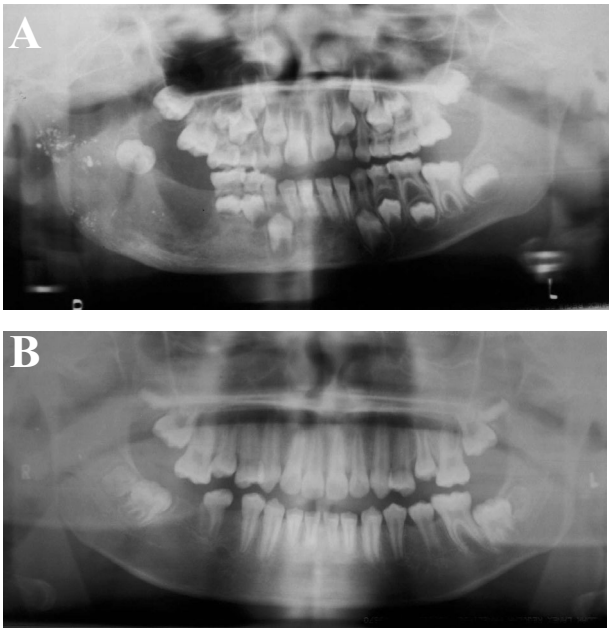

Fig. 4 Panoramic radiograph at 4 and a half months after embolization (4A) showing gradual resolution of the vascular lesions as in Figure 2A. Osseous regeneration was observed, but it was still incomplete with the appearance of a radiolucent area. The panoramic radiograph in Figure 4B was taken when the patient was 9 years and 8 months of age, 3 years and 8 months after the embolization. It showed complete osseous regeneration and remodeling of the right side of the mandible.

DISCUSSION

The initial clinical presentation of the current case was similar to those reported in the literature.[8,10,14] Mandibular arteriovenous malformations have usually been underestimated, sometimes leading to fatal complications.[15] Several differential diagnoses including cyst, ameloblastic fibroma, giant cell granuloma, fibrous dysplasia and malignant tumors might be considered.[16] Svan et al. suggested a puncture-test before biopsy to prevent disastrous complications.[17]

The management of mandibular AVM is variable. Ligature of the external carotid artery is a conservative treatment. However, it cannot prevent the recurrence caused by collateral circulation.[3,11] Partial or complete surgical resection of the affected bone with primary or secondary bone grafting has been reported as a radical treatment of these lesions.[18] Long-term postoperative sequelae such as scar contracture, dysphasia, facial asymmetry and malocclusion have been observed.[1] Partial resection or curettage following preoperative embolization have also been published.[1,3,8,10] Preoperative embolization eases the surgical procedure performed within 48 hours to 2 weeks after embolization.[19] However, the surgery often produces morphologic alterations in the growing face.[6,14]

The embolization method has gained popularity during the last decade for its efficacy in terms of functional anatomy preservation and aesthetic results. Different embolization approaches including transarterial, transvenous, and direct percutaneous intralesional puncture have been used.[6,9,11,12,14] The transarterial embolization sometimes fails to control blood flow from the collateral feeding vessels, thus, recurrent bleeding occurs causing difficulties in future treatments.[1,6,19] The transvenous technique must be complete, otherwise increased intravenous pressure with subsequent rupture and bleeding will follow.[20] Embolization through direct percutaneous intralesional puncture with precision of the needle placement in the site of the lesion have been reported.[6,9,12,14] Complete venous occlusion can be achieved right after the injection and this completely obliterates all feeders to the lesion.[12]

Possible complications of the direct percutaneous method are mucosa dehiscence with exposure of the embolic material and local infection as in this

case.[9,12] In our patient, the osseous regeneration, development, and remodeling on the involved ramus were completed in 2 years. At 52 months of follow-up, an acceptable facial profile and symmetry, along with excellent function of the mandible, and without signs of infection or recurrent bleeding were found.

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下顎骨動靜脈畸形栓塞治療後的骨骼再生

莊武帝 葛蘭妲 羅綸洲

下顎骨動靜脈畸形並不常見，臨床上如果疏忽這個疾病，可能會造成嚴重問題。應用經動脈和直接穿刺進行栓塞病灶，可以有效治療這個疾病，在小兒病人經栓塞治療後的骨骼變化以及長期效果並不清楚。我們報告一位6歲病人，因爲拔牙引起出血性休克，情況穩定後轉送我們治療，病人經過栓塞治療3個月後出現局部感染，病況經有效控制。追蹤期接受系列的電腦斷層和全口X光檢查，影像檢查顯示病灶逐漸縮小，同時有新骨生長和重造，這過程在兩年中完成，在往後追蹤沒有發現疾病復發或骨骼的異常生長。(長庚醫誌 2003;26:937-42)

關鍵字：下顎骨，動靜脈畸形，骨骼再生。

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