Clinical Notes
Delayed Presentation of Pseudoaneurysm after Le Fort I Osteotomy

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Although the osteotome is positioned in close vicinity to the maxillary artery and its branches during ptergomaxillary separation in a Le Fort I osteotomy, postoperative complications from vascular injuries are rare. The following report describes an unusual occurrence of a maxillary artery pseudoaneurysm following a Le Fort I and bilateral sagittal-split osteotomies for correction of mandibular and maxillary asymmetries in a patient with Goldenhar syndrome. This was recognized 8 months after the procedure when the patient developed acute facial swelling and required an emergent angiogram for uncontrolled bleeding. Vascular anatomy in the ptergomaxillary area is reviewed. A level of suspicion of occult vascular injuries in patients with sudden onset of unilateral facial swelling after orthognathic surgery, even months after the procedure, is recommended.

Key Words: Le Fort I, pseudoaneurysm, late complication

The surgical treatment of congenital facial skeletal deformities is fraught with a number of potentially life-threatening complications. The developmental nature of these deformities predisposes to alterations from established anatomic norms, further increasing the chances of an untoward result. Despite careful preoperative planning and meticulous surgical technique, unexpected outcomes in the extended postoperative period can occur.

Traumatic aneurysms to the tributaries of the external carotid artery, particularly the internal maxillary artery (IMA), are exceedingly rare occurrences.1–3 The diminutive caliber of the vasculature and its deep location in the mid-face have proven protective from blunt and superficially penetrating injuries.4 However, the proximity of osteotome positioning and manipulation commonly used for procedures to mobilize and reposition the mandible and maxilla introduces the potential for vascular trauma.5–7 The entire course of the IMA from its retro-mandibular origin to its termination within the pterygopalatine fossa is theoretically vulnerable to iatrogenic injury.

The following report describes an unusual complication following a Le Fort I and bilateral sagittal-split osteotomies for correction of an occlusal cant and facial asymmetries in a patient with Goldenhar syndrome. Eight months after this procedure the patient developed acute facial swelling and required emergent embolization for a ruptured pseudoaneurysm of the distal internal maxillary artery. Delay in recognition of a vascular injury after a Le Fort I osteotomy has not been documented in greater than a 30-day perioperative period.

CASE REPORT

A 17-year-old male was diagnosed with Goldenhar syndrome shortly after birth. His right craniofacial microsomia was manifested by right macrostomia, right mandibular hypoplasia, irregularities of the right tragus, and right preauricular skin tags but patent external auditory canals. His facial nerve function and muscles of mastication were mildly affected. He also had diplopia and strabismus. As a child he underwent a repair of his right macrostomia with Z-plasty transposition flaps. He also had corrective extraocular muscle realignment surgery for his strabismus.

When examined at skeletal maturation the patient had a significant occlusal cant with some over-
growth of the contralateral side (Fig 1). He underwent orthodontic preparation for correction of severe maxillary and mandibular asymmetries. A Le Fort I osteotomy, bilateral sagittal-split osteotomy, correction of occlusal cant, and asymmetric genioplasty were then performed.

Eight months after the procedure the patient visited the emergency room with sudden onset of right cheek swelling (Fig 2). Intraoral aspiration revealed thick, dark liquid and the patient was placed on antibiotics. Persistent swelling led to operative exploration. When nonlocalized bleeding was encountered, the wound was packed with hemostatic agents and closed. The patient was immediately sent to the angiography suite, while intubated and sedated.

The angiography demonstrated a pseudoaneurysm of the distal internal maxillary artery (Fig 3). Selective embolization with a coil was used to control the bleeding. The patient did well after this procedure. His did not rebleed and his facial swelling resolved (Fig 4). He has not had recurrent facial swelling or other problems in 20 months since this episode.

**DISCUSSION**

Arterial pseudoaneurysm can occur as a result of blunt or, more commonly, sharp trauma to the vessel wall. Transmural injury to the vessel sidewall allows for the development of an adjacent hematoma that remains in continuity with the arterial lumen. Liquefaction and cavitation rapidly follow
with the concurrent development of an endothelial lining at the periphery of the hematoma. A direct communication between the vessel of origin and the hematoma cavity is established and blood flow occurs in a closed circuit. The outermost layer of the pseudoaneurysm is the fibrous product of inflammation. Subsequent organization of surrounding tissues results in a structure which effectively halts further aneurysmal expansion. The entire process of pseudoaneurysm formation is usually completed within 1 to 8 weeks after blood vessel injury. This variable time period dictates the temporal sequence of events leading to the clinical manifestations of eventual rupture and hemorrhage.

Pseudoaneurysms in the head and neck region are usually encountered in large-diameter vessels.
The majority arise from either the common or internal carotid arteries. False aneurysms of the external carotid territory disproportionately involve the temporal and facial arteries because of their large vessel diameter and long, superficial course. Conversely, the internal maxillary artery (IMA) and its tributaries are rarely involved because of their relatively small caliber and deep location.

Anatomic review reveals that the cephalic portion of the external carotid artery divides within the substance of the parotid gland to give rise to two terminal branches; the superficial temporal and internal maxillary arteries (Fig 5A). The IMA is classically divided into three segments: mandibular, pterygoid, and pterygopalatine (Fig 5B). The mandibular portion of the IMA passes posterior to the mandibular condyle to the sphenomandibular ligament. The second part, the pterygoid segment, encompasses the vessel as it veers anteromedially into the infratemporal fossa. The terminal portion, the pterygopalatine segment, lies in the pterygopalatine fossa lateral to the facial nerve.

The mandibular segment of the IMA gives rise to deep auricular, tympanic, middle meningeal, accessory meningeal, and inferior alveolar branches (Fig 5B). The first four branches travel superiorly, but the inferior alveolar branch passes caudal and anterior to enter the mandibular foramen and canal. This first portion of the IMA, particularly the inferior alveolar branch, is vulnerable to damage from mandibular osteotomies.

Next, the pterygoid segment of the IMA gives off branches that supply the muscles of mastication and the buccinator muscle before passing through the pterygomaxillary fissure into the pterygopalatine fossa. Osteotomy during pterygomaxillary separa-

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**Fig 5** Drawings of the anatomic course of the internal maxillary artery related to the craniofacial skeleton. (A) Lateral view of the large arteries of the head. The external carotid artery’s terminal branches include the maxillary artery and superficial temporal artery. (B) This closer view of the mandibular ramus and condyle shows the three segments of the maxillary: the mandibular, pterygoid, and pterygopalatine. A segment of the mandibular ramus and coronoid process have been removed for better visualization of the maxillary artery branches (1) and pterygopalatine fossa.
tion risks injury to the IMA and branches at this juncture. Acute bleeding from a transected vessel in this location usually tamponades or is controlled surgically. Only rarely is the side vessel wall injured resulting in a hole and formation of a pseudoaneurysm.

The pterygopalatine segment of the IMA lies in the pterygopalatine fossa. This acts as a distribution center where each branch leaves the fossa through a canal or foramen that shares the same name. Branches include the posterior superior alveolar, infraorbital, greater palatine, lesser palatine, pharyngeal, artery of pterygoid canal (vidian artery), and sphenopalatine (giving rise to the posterior nasal). This site, particularly the sphenopalatine artery, can also be injured during a Le Fort I osteotomy.

Most aneurysms of the face and temple may be treated with vascular ligation or direct arterial repair. However, surgical approaches to the IMA and its branches can be difficult. Pseudoaneurysm of the deep branches of the internal maxillary artery are often better treated with embolization. Miller et al. reported embolization of the internal maxillary artery false aneurysm from trauma with the patient’s own clot. Since then major advances have occurred in angiographic techniques, including the use of variable-stiffness microcatheters for superselective injections, flow control techniques, and an array of embolization materials. These newer techniques allow successful treatment of small, deep vessels in a less invasive fashion than surgical exploration. Embolization may obliterate the exact bleeding source and spare more proximal vessels. In the postoperative period after orthognathic surgery, a good remaining blood supply to an osteotomized segment is important for healing.

In summary, our patient with right craniofacial microsomia underwent successful embolization of a ruptured distal IMA pseudoaneurysm 8 months after a Le Fort I osteotomy and bilateral sagittal-split osteotomy of the mandible. We recommend maintaining a level of suspicion for occult vascular injuries in patients with acute facial swelling after orthognathic surgery, even beyond the perioperative period.

REFERENCES