

Isrogenic Arteriovenous Fistula between the Middle Meningeal Artery and Pterygoid Venous Plexus Secondary to a Le Fort I Osteotomy: A Case Report

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Abstract

Le Fort I osteotomy is a common surgical technique for correcting jaw deformities. Although it is generally a safe procedure, there have been cases of postoperative vascular complications, which include arteriovenous fistulas. Published reports describe the development of arteriovenous fistulas that originate from the maxillary artery after this procedure; nevertheless, no cases of arteriovenous fistulas arising from the middle meningeal artery (MMA) have been reported. We present the case of a 19-year-old woman who developed pulsatile tinnitus immediately following a Le Fort I osteotomy. Angiography with proximal flow control of the external carotid artery showed an arteriovenous fistula between the extracranial portion of the MMA and the pterygoid venous plexus. The fistula was successfully treated using coil embolization, preserving the maxillary artery, which results in tinnitus resolution. In this report, the effectiveness of endovascular treatment for arteriovenous fistulas of the MMA that occur following a Le Fort I osteotomy was demonstrated. Angiography with proximal flow interruption was effective in shunt location detection.

Keywords: Le Fort I osteotomy, arteriovenous fistula, surgical complication, middle meningeal artery

Introduction

Le Fort I osteotomy is jaw corrective surgery that is associated with a few intraoperative or postoperative complications.¹⁾ Several cases of postoperative arteriovenous fistulas of the maxillary artery have been reported, with successful outcomes obtained using endovascular treatment to occlude the maxillary artery.²⁻⁵⁾

We describe the treatment of an arteriovenous fistula of the middle meningeal artery (MMA) that occurred following a Le Fort I osteotomy, which was initially assumed to originate from the maxillary artery. However, angiography with proximal flow control revealed that the shunt point was limited to the MMA. Coil embolization was successfully performed, which preserves the maxillary artery.

Case Report

A 19-year-old woman, who had undergone multiple facial surgeries for cleft lip and palate since birth, presented with pulsatile tinnitus. Eight months earlier, she had undergone a Le Fort I osteotomy and sagittal splitting ramus osteotomy performed by plastic surgeons. According to the medical record, 500 mL intraoperative blood loss had occurred; nevertheless, no significant abnormality was found in the intraoperative results. Figure 1 shows her osteotomy line. She noticed tinnitus immediately postoperatively. On the 1-month postoperative follow-up, she reported to have been experiencing tinnitus. She was then referred to the neurosurgical department and underwent cerebral angiography on the 3-month postoperative follow-up. The initial angiography suggested an arteriovenous fistula that drains into the left pterygoid venous plexus from the left maxillary artery, which was suspected to be the source of her

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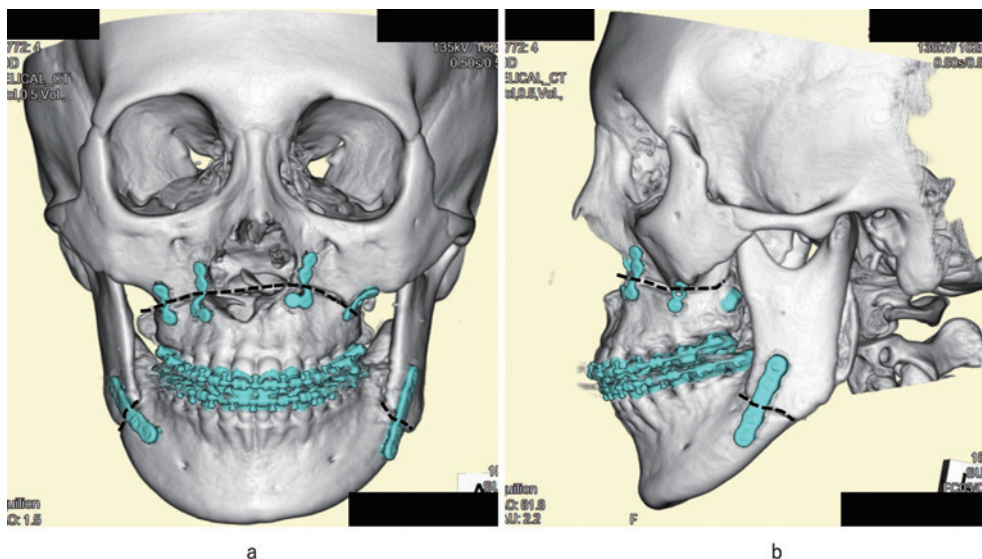


Fig. 1 Reconstructed three-dimensional computed tomography images after Le Fort I osteotomy, shown in anteroposterior (a) and lateral (b) views. We can determine the osteotomy lines (black dotted lines) fixed with plates.

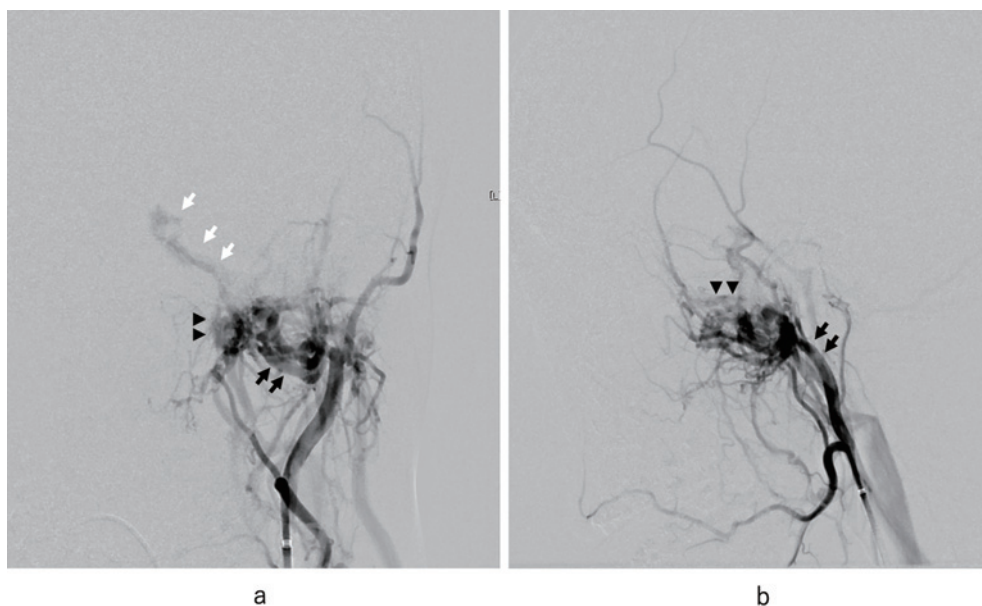


Fig. 2 Preoperative left external carotid artery angiography, shown in anteroposterior (a) and lateral (b) views. A suspected arteriovenous fistula between the left maxillary artery (black arrows) and the pterygoid venous plexus (black arrowheads), as well as the limited retrograde flow of blood into the left cavernous sinus (white arrows).

symptoms (Fig. 2). Her hearing was impaired because of the severity of the tinnitus, requiring prompt intervention to treat the arteriovenous fistula.

The procedure was performed under general anesthesia, with a 7 F OptimoEPD (Tokai Medical Products, Inc. Aichi, Japan), guiding catheter with a balloon inserted into the left external carotid artery via a transfemoral approach. Angiography of the left external carotid artery carried out with proximal blood flow interruption showed a pseudoaneurysm in the extracranial portion of the MMA and

an arteriovenous fistula that drains from the pseudoaneurysm to the pterygoid venous plexus. The pseudoaneurysm formed distal to the osteotomy line, as shown by a reconstructed image from a three-dimensional rotational angiography (Fig. 3). A microcatheter was guided into the MMA, and the aneurysm and MMA were embolized with nine coils (Fig. 4). Angiography demonstrated complete occlusion of the shunt. Her tinnitus resolved immediately, and no symptoms recurred 6 months postoperatively.

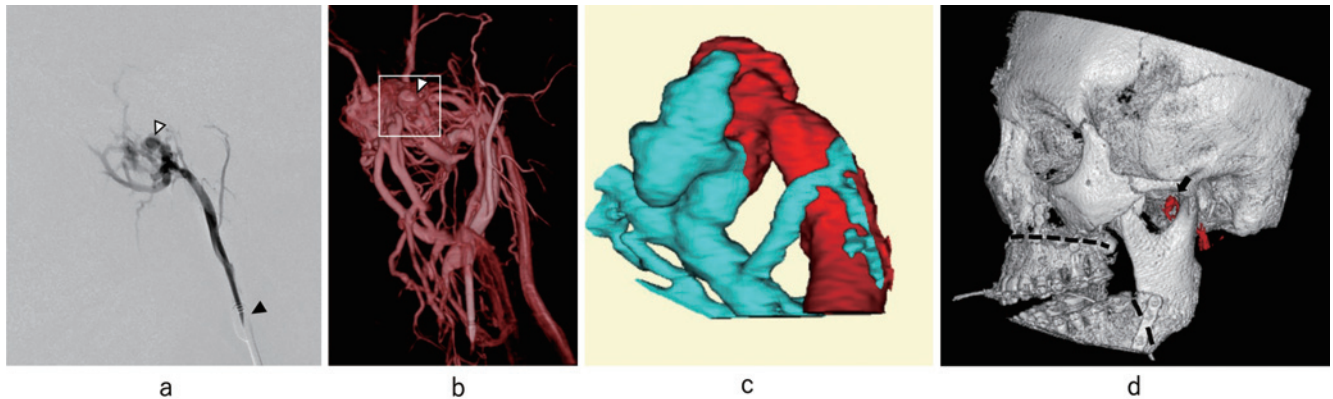


Fig. 3 External carotid arteriography under proximal flow control via an inflated balloon (black arrowhead). A pseudoaneurysm of the middle meningeal artery (white arrowhead) is shown on the lateral view (a) and reconstructed image of three-dimensional rotational angiography (b). Magnified image (c) of the shunt point, with the red-colored vessels being the middle meningeal artery with the pseudoaneurysm, and the turquoise-colored vessels are dilated veins contiguous with the pseudoaneurysm. The magnified image (c) is enlarged within the white square in (b).

Reconstructed three-dimensional rotational angiography before embolization (d) shows that the pseudoaneurysm (black arrow) has formed away from the osteotomy line (black dotted lines).

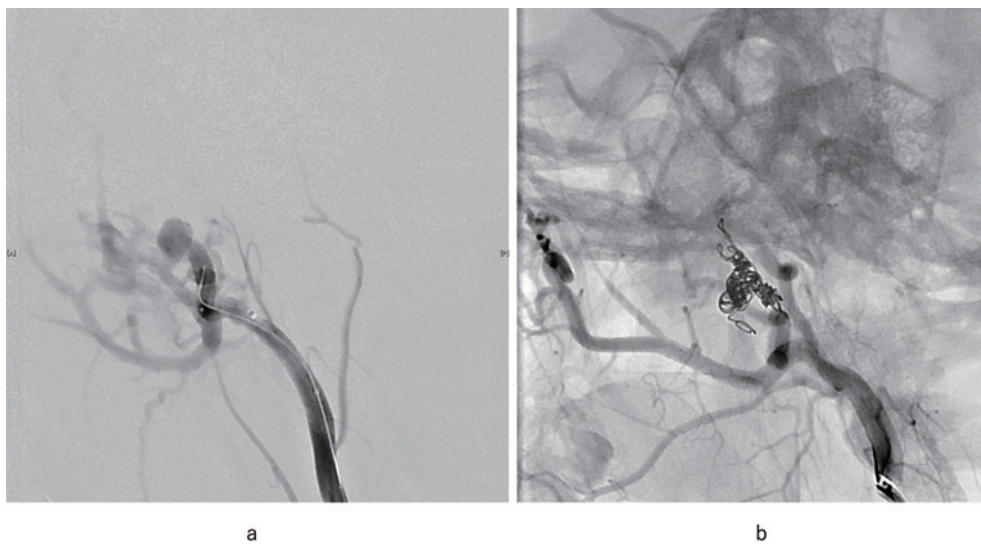


Fig. 4 Embolization of the fistula with detachable coils.

Discussion

Le Fort I osteotomy is a surgical technique that is employed to separate the facial bones from the skull base and reposition them anteriorly. The complication rate for this procedure is reported to be approximately 6.4%. Nevertheless, the risk for complications increases to 25.2% in patients with anatomical anomalies, such as cleft lip and palate or other vascular malformations, in comparison to 3.9% in those without these conditions.¹⁾ Among these are vascular complications such as hemorrhage from injury of the maxillary artery, pterygopalatine artery, pterygoid venous plexus,⁶⁻¹⁰⁾ and pseudoaneurysms.^{7,11-13)}

Several cases of arteriovenous fistulas of the maxillary

artery following a Le Fort I osteotomy due to maxillary artery injury have been reported.²⁻⁵⁾ However, these reports fail to provide accurate details on the pathogenesis or procedures that may have caused them. In these cases, the number of days between surgery and the onset of disease varied from a few days to 3 months,²⁻⁵⁾ and endovascular treatment was utilized, which results in the immediate resolution of symptoms. Detachable coils,^{3-5,11)} polyvinyl alcohol,⁴⁾ n-butyl-2-cyanoacrylate,^{2,4)} and various other materials have been utilized as embolic agents. As with traumatic arteriovenous fistulas, there was often a single shunt point, and complete occlusion of this point appeared to effectively cure the condition, irrespective of the embolic material used.

In the current case, a pseudoaneurysm was determined in the extracranial portion of the MMA. Additionally, an arteriovenous fistula drained into the pterygoid venous plexus adjacent to the pseudoaneurysm, which was observed on proximal flow control angiography. Since the osteotomy line and the pseudoaneurysm were far apart and there was no unusual intraoperative bleeding or vascular damage, the pseudoaneurysm was probably formed by a minor traction injury to the MMA during the osteotomy; this then ruptured into the adjacent venous plexus, which creates an arteriovenous shunt.

Angiography under proximal blood flow control was helpful for accurately determining the shunt point. The MMA has a rich collateral blood pathway, and there is a low risk of neurologic manifestations owing to proximal embolization. The maxillary artery supplies a large area from the inferior temporal fossa to the nasal cavity and palate, and we were able to treat the patient while preserving the maxillary artery.

To our knowledge, this report is the first to describe a case of postosteotomy arteriovenous fistulas of the MMA. Other reports were not able to provide a detailed examination of the shunt point of the maxillary arteriovenous fistulas under proximal flow control, which leads us to assume that some of these cases might have been arteriovenous fistulas of the MMA and misdiagnosed as maxillary artery fistulas due to the inability to identify the shunt point because of its high flow. Other cases might have been undiagnosed or unnoticed because of the relatively minor symptoms of the patients, such as tinnitus.

To identify the shunt point and assure safe treatment, a thorough evaluation using angiography under blood flow control is required for cases of high flow shunt in iatrogenic or traumatic arteriovenous fistulas, in addition to postoperative problems following osteotomy.

List of Abbreviation

MMA, middle meningeal artery

Conflicts of Interest Disclosure

All authors declare no conflicts of interest.

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