

An extremely rare pseudoaneurysm of posterior superior alveolar artery arising after orthognathic surgery

ABSTRACT

Le Fort 1 osteotomy is one of the most versatile techniques in orthognathic surgery employed for the correction of dentofacial deformities and is considered technically safe. Pseudoaneurysms (PAs) which can cause life-threatening hemorrhage are rare after corrective jaw surgery. Here, we describe a clinical case of delayed postoperative epistaxis secondary to an extremely rare PA of the posterior superior alveolar artery followed by Le Fort 1 osteotomy subsequently managed with endovascular selective embolization.

Keywords: Embolization, internal maxillary artery, Le Fort 1 osteotomy, orthognathic surgery, pseudoaneurysm

INTRODUCTION

Orthognathic surgery became the need of the hour for the patients with dentofacial skeletal deformities. Le Fort 1 osteotomy is considered to be one of the safest, reliable, and predictable surgical techniques across the globe for the correction of such dentofacial deformities.^[1,2] Le Fort 1 osteotomy also carries few uncommon serious postoperative vascular complications, resulting from unrecognized intraoperative vascular injury.^[3] Vascular complications as a result of this can range from common postoperative oronasal bleeding to very rare arteriovenous fistulas and pseudoaneurysms (PAs).^[4] Utmost care should be taken while performing osteotomies, especially the pterygomaxillary disjunction in order to prevent injury to the nearby vasculature, including the internal maxillary artery (IMA) and its branches.

Acute epistaxis after Le Fort 1 osteotomy in the immediate postoperative period is relatively common, but the diagnostic challenge lies when it persists further. True aneurysms are characterized by the expansion of all the three layers of blood vessel, whereas PA involves rupture of one or more layers followed by an incomplete injury of the vessel. This results in the leakage and collection of blood within the layers of blood vessels or formation of a capsule into the surrounding soft tissues.^[4,5]


The rupture of PA can lead to severe life-threatening hemorrhage, and the surgeon should be competent enough to diagnose and perform primary management in an emergency suite. PA can be asymptomatic most of the times or they may even present with symptoms such as pain, swelling, and epistaxis. Therefore, they can be undiagnosed for a period of weeks or months, which becomes a diagnostic challenge. The most vulnerable facial vessels for the formation of PAs are facial artery and superficial temporal artery because of their relative superficial anatomy^[2,4] and their nature of being more prone to maxillofacial trauma. The possibility of PA in the branches of IMA was reported in the literature,^[1-4,6] but they are relatively uncommon after Le Fort 1 osteotomy.

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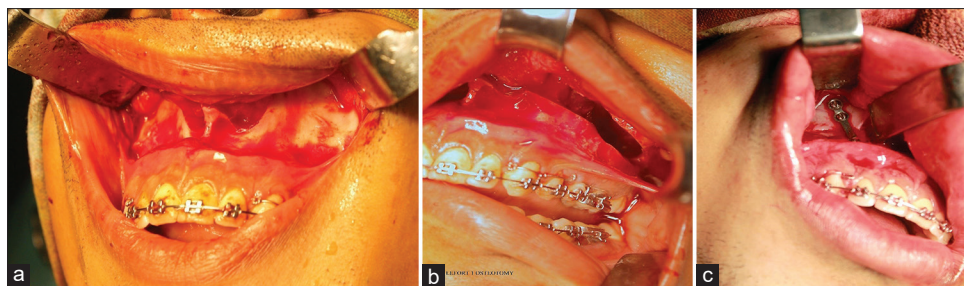


Figure 1: (a-c) Le Fort 1 osteotomy with miniplate fixation



Figure 2: Augmentation genioplasty with miniplate fixation

Here, we report an extremely rare case of PA of posterior superior alveolar artery (PSAA) followed by Le Fort 1 osteotomy treated with endovascular selective embolization using gelatin foam.

CASE REPORT

A 26-year-old gentleman reported to the department of oral and maxillofacial surgery for the correction of dentofacial deformity. He was diagnosed with vertical maxillary excess and hypogenia. The patient had no history of drug, alcohol, or tobacco intake, and his blood values were within normal limits. As per the proposed treatment plan, the patient underwent Le Fort 1 osteotomy with superior maxillary repositioning [Figure 1] accompanied by augmentation genioplasty [Figure 2] under general anesthesia. The postoperative course was uneventful, and the patient was subsequently discharged on the 4th postoperative day.

The patient was apparently normal, with no signs of infection and other relative illness up to 5 weeks after the surgery. By the 6th postoperative week, the patient reported to the outpatient department with mild pain and swelling over the right anteromedial cheek region. Clinical examination was inconclusive with nonspecific symptoms, and he was prescribed with analgesics for symptomatic relief. After

2 weeks, the patient was brought to the emergency department following an episode of epistaxis during sleep and was primarily managed with anterior and posterior nasal packing. A contrast computed tomography was performed, which did not reveal any abnormalities. Hemogram and bleeding parameters were within the normal range. Accordingly, the patient was monitored round the clock and discharged subsequently in the absence of any events.

By the 11th postoperative week, the patient presented with another episode of epistaxis accompanied by severe fall in hemoglobin level to 5.5 mg/dL, alarming an emergency. He was transfused with two units of packed red blood cells and intravenous maintenance with crystalloids. Hereafter, an interventional radiologist joined the team, and the patient was shifted to the radiological suite. Transfemoral angiogram (digital subtraction angiography) of the right external carotid revealed PA of the PSAA [Figure 3], which is a branch of the third division of IMA. Superselective transcatheter embolization of IMA with gelatin-foam implantation [Figure 4] was done after 72 h. The patient was then transferred to the intensive care unit after the procedure, and the subsequent postoperative period was uneventful. He remained asymptomatic with no further episodes of epistaxis during our 24-month-long follow-up.

DISCUSSION

Le Fort 1 osteotomy involves the mobilization and separation of the entire maxilla in order to correct various dentofacial deformities. During the procedure, pterygopalatine disjunction is coupled with the positioning of the osteotome and its manipulation in closer proximity to the branches of IMA, which makes it more vulnerable to vascular injury.^[7,8] The incidence of PA followed by orthognathic surgery still remains scarce with a few published literatures. However, PA of the PSAA after Le Fort 1 osteotomy is extremely rare with no evidence in literature till date. The foremost reason behind the rarity of PA in these areas is attributable to the smallest diameter of blood vessels, which makes their partial transection unlikely.^[9]

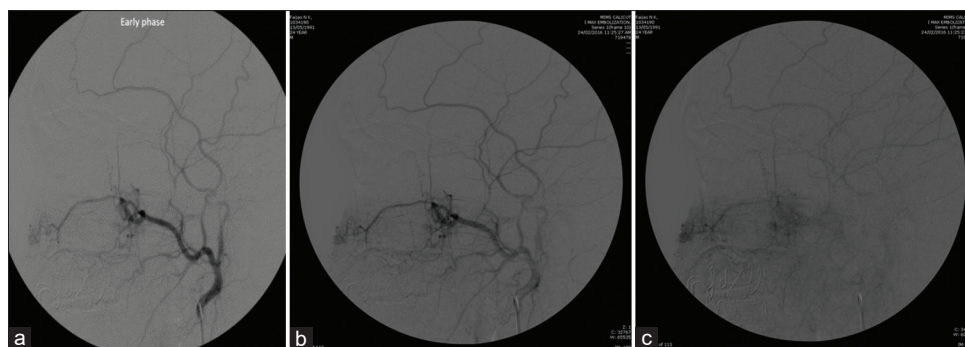


Figure 3: (a) Angiography (digital subtraction angiography) revealing pseudoaneurysm of the right posterior superior alveolar artery – early arterial phase, (b) mid-arterial phase, and (c) late arterial phase

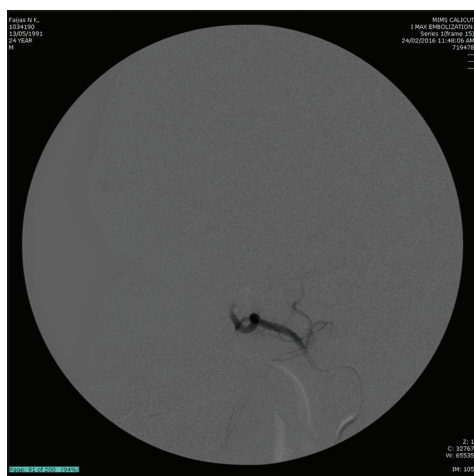


Figure 4: Postembolization showing the resolution of right, PSAA pseudoaneurysm

In the present case, the patient initially had mild diffuse swelling over the anteromedial cheek region, which was not pulsatile. This may be due to the small size of the injured vessel as well as being in a deeper location. The surgeon should always rule out the possibility of PA with the help of angiogram in patients with a history of persistent epistaxis even after the 2nd postoperative week. If PA is diagnosed during angiography, the treatment of choice is transcatheter selective embolization.^[10] One of the advantages of embolization over surgical intervention is that the more distal vessels that supply the bleeding source may be obliterated while sparing the more proximal vessels. This is particularly important following orthognathic surgery to avoid a further compromise of an already-diminished vascular supply, which could contribute to the development of aseptic necrosis of maxilla.^[11]

According to the literature, different embolization materials such as GELFOAM®, Gianturco coils, Dacron® fibers, detachable balloons, N-butyl cyanoacrylate, autologous clot, polyvinyl alcohol, and complex platinum coils have been employed with varying degrees of success for treating PAs.^[4,6,7] We opted for the implantation of GelFoam, which

was inexpensive and easily available. Transcatheter selective embolization technique showed superior results with no signs of recurrence in our 24-month-long follow-up.

CONCLUSION

Rupture of PA and life-threatening hemorrhage following orthognathic surgery is very rare. Oral and maxillofacial surgeons should be cautious and aware of the possibility of PA as a potential complication after Le Fort 1 osteotomy. The ideal investigation of choice in such clinical scenarios is best reserved for angiography.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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