

Rupture of pseudoaneurysm of the posterolateral nasal artery associated to Le Fort I osteotomy: a case report

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Background: It is unusual to find a life-threatening hemorrhage as a postoperative complication of a Le Fort I osteotomy. However, there are reports related to the maxillary artery, and its branches, including the terminal branch, the sphenopalatine artery. Therefore, every surgeon performing this procedure must know this type of complication and the appropriate workflow to resolve it. This case is unique in terms of elucidating the clinical signs of symptoms that can be evidenced with a ruptured pseudoaneurysm after orthognathic surgery and how minimally invasive non-surgical procedures can resolve this complication.

Case Description: In this paper, we present the case of a 26-year-old male with no pertinent past medical history who presented with three postoperative episodes of epistaxis after a Le Fort I osteotomy for correction of class III craniofacial anomaly; this was secondary to a ruptured pseudoaneurysm of the posterolateral nasal artery, a branch of the sphenopalatine artery; the patient underwent embolization as definitive treatment. No bleeding episode was reported after definitive treatment with embolization, the patient continued with post-op follow-up appointments with no complications.

Conclusions: It is considered that making this type of reports contributes to the formation of the international community, enhances knowledge, and facilitates what to do when a hemorrhagic complication associated with a surgical procedure of orthognathic surgery occurs.

Keywords: Le Fort I osteotomy; epistaxis; pseudoaneurysm; maxillary artery; case report

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Introduction

Background

The use of maxillomandibular osteotomies for correction of craniofacial deformities had a significant increase in the 70s. However, a lot of surgeons were not confident about the procedure due to the possible risk of hemorrhage (1).

Vascular complications, in general are common during

surgical procedures, as well as vascular lesions secondary to penetrating injuries, such as gunshot wounds, or sharp objects trauma (2). However, these vascular lesions do not represent a frequent complication in orthognathic surgery (3); a systematic review from Almofreh *et al.* included 20 studies that reported a total of 21 patients presenting pseudoaneurysms after orthognathic surgery (4). Converse *et al.*, reported a severe hemorrhage in only one patient out of 50 craniofacial

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surgeries performed (5), Westwood and Wilson reviewed 400 Le Fort I osteotomies performed at University of Washington reporting only three significant bleeding complications in the post-op (6), and Panula *et al.*, in a 655 orthognathic surgeries review reported hemorrhagic complications of 0.91% (6 patients) and one of these patients required blood transfusion and Internal maxillary artery embolization; none of these patients presented a fatal outcome (7).

Rationale and knowledge gap

Pseudoaneurysms are rare complications in orthognathic surgery; when it happens, the most involved vessels are the internal maxillary artery and its sphenopalatine branch (8-10). Usually, during presentation of pseudoaneurysm after a Le Fort I osteotomy, an initial episode of epistaxis presents within the 2 first weeks after a straightforward post-op recovery process. The computed tomography-angiography (CTA) as a diagnostic tool and embolization are usually enough to resolution of pseudoaneurysm in the maxillofacial region after orthognathic surgery (11).

Objective

This report aims to present a case elucidating the clinical

signs of symptoms that can be evidenced by a ruptured pseudoaneurysm after orthognathic surgery and how minimally invasive non-surgical procedures can resolve this complication. This article is written following the CARE reporting checklist (available at <https://fomm.amegroups.com/article/view/10.21037/fomm-23-16/rc>).

Case presentation

The authors present a case review with IRB-exempt approval by the University of Valle Ethics Institutional Review Board. All procedures performed in this study were under the ethical standards of the institutional research committee and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

A 26-year-old male with no pertinent past medical history underwent a Le Fort I advancement osteotomy to treat a maxillary retrognathism with no complications during surgery. Pre-op complete blood count (CBC) showed: Hb: 18.3 g/dL (13.8–17.2 g/dL) and hematocrit: 50.6% (41–50%). Two hours after surgery, a mild epistaxis from the left nostril was documented and controlled with pressure, with no need for nasal packing, patient was hospitalized for monitoring and post-op evaluation. On postop day 1, the patient was hemodynamically stable, with surgical site hemostatic and intact, and the patient was discharged.

New onset of epistaxis was documented 13 days after surgery that required intravenous (IV) fluids resuscitation with 1,000 mL bolus of normal saline and maintenance IV fluids with normal saline at 120 mL/h and afterward an exam under anesthesia with a posterior and anterior nasal packing for bleeding control. Vital signs on admission were blood pressure: 110/70 mmHg, heart rate 130 bpm; post op CBC showed Hb 6.0 g/dL (13.8–17.2 g/dL), hematocrit 20.2% (41–50%), and patient received blood transfusion with 2 PRBC units. The nasal packing was removed five days later under IV sedation with no signs of persistent bleeding, the patient was monitored for 48 hours and discharged later with outpatient clinic follow-up appointments at 1 and 4 weeks.

A third onset of epistaxis was documented 38 days after initial surgery; the patient was treated with anterior and posterior nasal packing, admission CBC showed Hb: 6.7 g/dL (13.8–17.2 g/dL), hematocrit 20.4% (41–50%), 2 units of PRBC were transfused and his post transfusion CBC showed Hb: 7.6 g/dL (13.8–17.2 g/dL), hematocrit

Highlight box

Key findings

- Pseudoaneurysm is a rare complication of orthognathic surgery.
- The maxillary artery or its branches are frequently associated with pseudoaneurysm in Le Fort I surgery.
- Angiogram confirms the presence of pseudoaneurysm and is a diagnostic and therapeutic tool.

What is known and what is new?

- Vascular complications are common during surgical procedures, however, these vascular lesions do not represent a frequent complication in orthognathic surgery.
- Pseudoaneurysm are rare complications in orthognathic surgery; usually presenting after Le Fort I osteotomy, an initial episode of epistaxis presents within the 2 first weeks after a straightforward post-op recovery process. The computed tomography-angiography as a diagnostic tool and embolization is usually enough to resolution of pseudoaneurysm after orthognathic surgery.

What is the implication, and what should change now?

- This type of reports contributes to formation of international community, enhances knowledge, and facilitates what to do with hemorrhagic complications after orthognathic surgery.

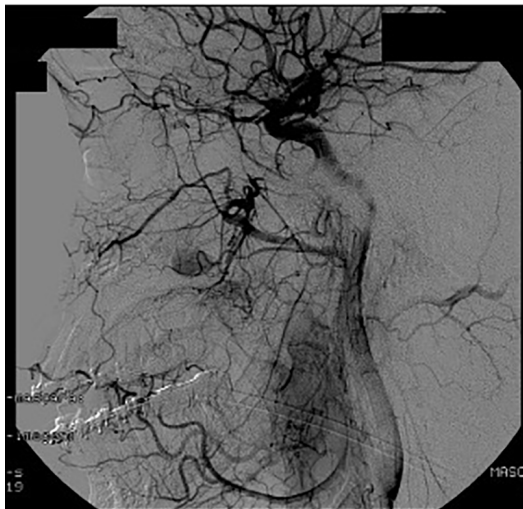


Figure 1 Carotid artery arteriography.

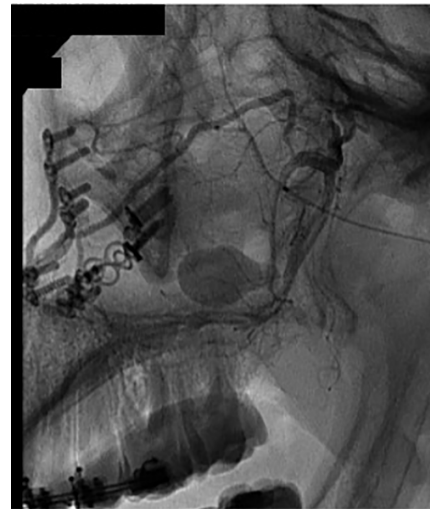


Figure 2 Angiography showing pseudoaneurysm of sphenopalatine artery branch.

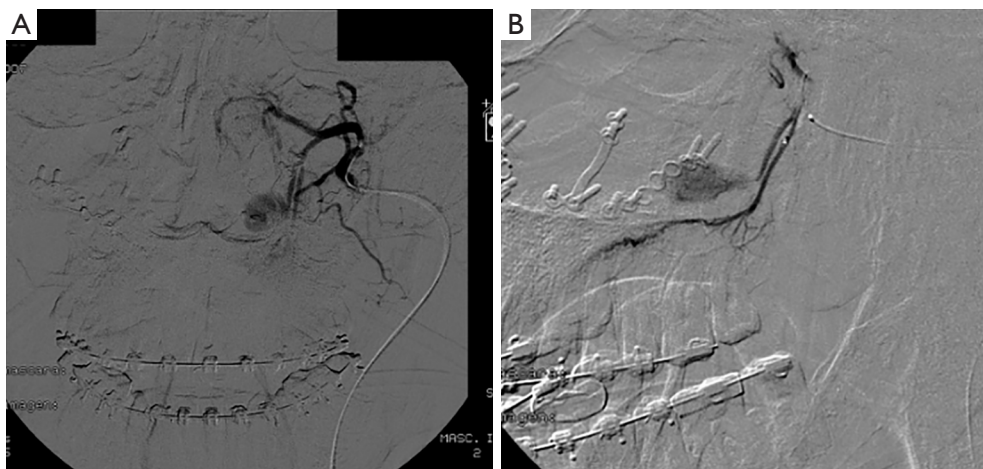


Figure 3 Coronal (A) and sagittal (B) view of pseudoaneurysm on left posterolateral nasal artery located posterior to the inferior turbinate.

22.7% (41–50%), a CTA carotid was obtained (*Figure 1*), that reported a pseudoaneurysm of the sphenopalatine artery, branch that runs on the lateral wall of the nasal cavity [posterolateral nasal artery (PLA)] (*Figure 2*) and is located posterior to the inferior turbinate (*Figure 3A,3B*). The decision was made against surgical clipping of the pseudoaneurysm due to challenging access to the posterior area of the inferior turbinate and the fact that on post-op day 38, the presence of scar and fibrous tissue would difficult the exposure and ligation of the vascular entity. The PLA was embolized using Coils + Histoacryl + microparticles (*Figure 4*). The patient tolerated the procedure without any

complication and was then transferred to the Intermediate care unit for surveillance. The nasal packing was removed 24 hours after the embolization with no evidence of new bleeding.

No bleeding episode was reported after definitive treatment with embolization, the patient continued with post-op follow-up appointments with no complications.

Discussion

Key findings

Anatomically, the maxillary artery branches at the end



Figure 4 Angiography after embolization of pseudoaneurysm.

to the sphenopalatine artery, which goes out to the nasal cavity through the sphenopalatine foramen, which is posterior between middle and upper turbinate; posteriorly, the sphenopalatine artery is divided into the PLA and posteromedial nasal artery (PMA) also known as septum artery. The PLA irrigates the middle and inferior turbinate with two small branches and the PMA after giving branches to the upper turbinate runs obliquely on the nasal septum and anastomoses in the palatine conduct with the descending palatine artery that runs from posterior to anterior from the soft palate (12,13).

The sphenopalatine artery represents the main vessel irrigating the nasal mucosa, massive hemorrhages of this artery can arise from endoscopic sinus surgery, secondary to injury on any of its branches, especially during removal of perpendicular lamina of the palatine bone in medial antrostomies (14). During Le Fort I osteotomies, the cuts are done on the lateral nasal wall and nasal septum below the inferior turbinate, located about 14.2 mm from the nasal floor (15).

Research published by Lee *et al.*, where surgical anatomy of the sphenopalatine artery was analyzed in 50 cadavers, the sphenopalatine foramen was located 90% of the time between the middle turbinate and the posterior horizontal lamina of the superior turbinate in 10% of the cases, this was located above the superior turbinate, what indicates that the sphenopalatine foramen was not identified below the middle turbinate. The PLA descends 1 cm anterior to the tail of the middle turbinate and 1/5 cm anterior to the tail of the inferior turbinate; the diameter is between 1.89 ± 0.23 mm. In 42% of cases (21/50), the PLA runs slightly posterior to

the posterior wall of the maxillary sinus; in 20% of cases (10/50), the superior part runs somewhat posterior to the posterior wall of the maxillary sinus; in 20% (10/50) runs intimately close to the posterior wall of the maxillary sinus and anterior to this in the inferior portion, in 18% (9/50) it runs anterior to the posterior wall of the maxillary sinus. It was also found that only in one case was the PLA at the end of the sphenopalatine artery; it was shown that the inferior turbinate artery is divided into two types, above the inferior turbinate and anterior to the middle portion of this (16). Similarly, there are other reports of variations in the arteries of the nose (17,18). This means that the possibility of direct damage to the sphenopalatine artery or one of its branches during Le Fort I osteotomies is considered a rare complication.

Strengths and limitations

The case report of a pseudoaneurysm following orthognathic surgery holds several strengths in the medical field. Firstly, such a report sheds light on a rare serious complication. By detailing the patient's clinical presentation, diagnostic methods, and management, the case report serves as a valuable educational resource for other healthcare professionals, improving their awareness and early detection of similar complications in the future. Furthermore, the report offers insights into risk factors associated with pseudoaneurysm development after orthognathic surgery, guiding surgeons in optimizing their surgical techniques and postoperative care.

Nevertheless, case reports also have inherent limitations. As isolated instances, they lack the statistical power to establish causality or generalize about the incidence of such complications in the broader population. Moreover, the absence of a control group hinders a direct comparison of risk factors or treatment outcomes. Despite these limitations, the case report remains a valuable contribution to medical knowledge and serves as an essential stepping stone for generating hypotheses and prompting further investigations into this specific complication.

Comparison with similar research

Pseudoaneurysms have been rarely reported after orthognathic surgery (9). Lanigan is the one who has investigated the most regarding the presence of lesions to the maxillary artery and specifically the presence of false aneurysms after orthognathic surgery, consisting of Le Fort

I osteotomy (8,9).

Yin reports in his 1994 article the compilation of cases from 1963 to 1991, where injuries to the maxillary artery were found to be related to sagittal and vertical mandibular osteotomies and temporomandibular joint surgeries, differing from this case where the pseudoaneurysm was associated with the LeFort osteotomy (10). Hemmig *et al.* 1987 reported the rupture of a pseudoaneurysm of the sphenopalatine artery postoperatively in a 29-year-old woman who underwent a Le Fort I osteotomy with impaction to correct vertical maxillary excess. In the postoperative period, the first episode of epistaxis was on the eleventh day; she developed recurrent epistaxis that was not controlled with anterior and posterior nasal packing. Angiography showed a pseudoaneurysm of the sphenopalatine artery that was successfully embolized. This case elucidates a similar management of the pseudoaneurysm with minimally invasive intervention. Additionally, they refer to the importance of considering anomalous vascular anatomy, such as arteriovenous malformations or aneurysms of a congenital, traumatic type or that have been caused by surgery (19).

Clark *et al.* 1987 reported the case of a 15-year-old man who underwent sub condylar vertical mandibular osteotomies to correct mandibular prognathism. A severe intraoperative hemorrhage occurred, the bleeding was controlled with pressure, and the patient's initial postoperative period was uneventful until approximately 2 months after surgery, the patient developed a pulsatile swelling in the preauricular area. Angiography confirmed the presence of a pseudoaneurysm of the maxillary artery, which was successfully embolized (20). Finally, Silva *et al.* 2007 reported this complication after performing sagittal osteotomies of the mandibular ramus, directly related to the medial osteotomy, completed near the sigmoid notch (21).

Explanation of findings

An explanation for the presence of pseudoaneurysm of the sphenopalatine artery or its branches would be vessel injury during the down fracture procedure, where unexpected microfractures could be generated towards the top with damage to the intima of the sphenopalatine artery or its branches, but not from direct trauma during the procedure.

Another explanation for the generation of pseudoaneurysm would be the presence of possible anatomical variations related to the structure involved in the pseudoaneurysm, examples of which would be a more significant number of branches, a lower-than-normal location described,

generating proximity to the surgical site or the considerable increase in the caliber of an artery from which major bleeding is not expected.

Implications and actions needed

It is probable that the small size and the difficult accessibility to detect the same bleeding site of most of the blood vessels in the facial region cause them to be partially sectioned or lacerated unintentionally; this must be the main reason these rare pseudoaneurysms occur in the facial area.

The management of the pseudoaneurysm is based initially on if it is stable or ruptured and its location. Spontaneous resolution has been reported in some cases in the head and neck area (22). However, it is essential to consider each patient's specific circumstances and clinical presentation, especially when experiencing recurrent episodes of hemorrhage and anemia, making spontaneous resolution an unviable option. The initial treatment includes immediate bleeding control and local pressure following Advanced Trauma Life Support (ATLS) guidelines in hemodynamic instability and hypovolemic shock (4). Surgical ligation is indicated when the pseudoaneurysm is accessible. However, in cases where the location of the pseudoaneurysm is challenging for surgical access, it is suggested to perform angiography and embolization (23).

Conclusions

For the diagnosis and adequate management of the pseudoaneurysm, it is suggested to obtain a CTA as a complementary diagnostic tool to a CT with IV contrast, and for definitive treatment embolization or as a last option, ligation of the external carotid artery or the supplying branch.

The anatomical study of any surgical procedure or complication is essential. The accuracy in knowing the anatomical structure involved can allow for defining the site and the causes of bleeding with greater precision.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at <https://fomm.amegroups>.

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://fomm.amegroups.com/article/view/10.21037/fomm-23-16/coif>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were under the ethical standards of the institutional research committee and with the Helsinki Declaration (as revised in 2013). IRB approval was exempted by the University of Valle Ethics Institutional Review Board. Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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