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Case Report
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Pseudoaneurysm of the Ascending Pharyngeal Artery as a Complication of Orthognathic Surgery: A Case Report and Literature Review

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Abstract

Introduction: Pseudoaneurysms are caused by injury of the vessel wall leading to subadventitial dissection and subsequent dilatation of the vessel with a persistent communication between the true arterial lumen and the dissecting cavity. Pseudoaneurysm directly related to a surgical procedure such as the Le Fort I osteotomy is an exceedingly rare complication. Furthermore, a pseudoaneurysm related to the ascending pharyngeal artery has never been reported.

Case Report: A LeFort I osteotomy associated with a bilateral sagittal split osteotomy was carried out on a 15-year-old female patient for a surgical correction of Class II skeletal discrepancy associated with a deep bite. Five weeks after surgery, the patient was still experiencing significant pain in her right mandibule. The CT scan with contrast highlighted a liquid mass of 5.2 cm × 2.7 cm × 4.7 cm. An exploration of the operative site demonstrated a severe intra-oral bleeding controlled with conservative treatment. Due to recurrence of the pain and swelling, angiography was performed, revealing a pseudoaneurysm of the right ascending pharyngeal artery. This pseudoaneurysm might be related to a supraperiosteal positioning of the osteotome during the pterygomaxillary disjunction in the Le Fort osteotomy. An embolization was performed with coils placed into the distal ascending pharyngeal artery using a 3×14 Nester microcoil and the patient recovered well.

In this case report a literature review was performed and a description was made of this first-time seen case of a pseudoaneurysm of the ascending pharyngeal artery after a Le Fort I osteotomy of the maxilla.

Keywords: Pseudoaneurysm; Le Fort I osteotomy; Ascending pharyngeal artery; Angiography

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Abbreviations

BSSO: Bilateral Sagittal Split Osteotomy; PVA: Pieces of Polyvinyl Alcohol; n-BCA: n-Butyl Cyanoacrylate; ECA: External Carotid Artery; SARPE: Surgically Assisted Rapid Palatal Expansion

Introduction

The Le Fort I osteotomy (LFI) is a well-established method for correction of maxillary deformities. It may be used alone or in combination with mandibular surgery to correct dentofacial deformities.

During LFI surgery, as with any surgical procedure, a variety of intraoperative and postoperative complications may occur. Among intraoperative complications, bleeding is considered the most serious problem [1].

An exceedingly rare bleeding complication can be due to arterial damage leading to a development of a Pseudoaneurysm (PA). A pseudoaneurysm results from a subadventitial dissection of an arterial vessel leading to the formation of aneurysmal dilatation of the vessel [1,2]. The adjacent tissues containing the pseudoaneurysm in completely counterbalance the arterial pressure compressing the

defect and stabilizing the artery. The pseudoaneurysm is a result of a persistent leakage of blood in the aneurysmal dilatation. If this blood leak remains undiagnosed a massive post-operative bleeding can occur [1]. It usually takes between 1 to 8 weeks from the initial injury until the formation of a pseudoaneurysm that clinically translates into a pulsatile mass [3]. Patient commonly experience pain and present with an unresolved swelling, which is eventually associated with heartbeat pulsations. Depending on the size and the rapidity of the hematoma development, some patient also describes paresthesia due to compression of adjacent structures [1]. Most reported cases of PAs of the External Carotid Artery (ECA) occur in the facial artery and the superficial temporal artery due to their relatively superficial position and the bone structure that they pass [3].

Case Presentation

A 15-year-old Caucasian woman was referred to AZ Monica Hospital (Antwerp, Belgium) in 2020 for surgical correction of a dentoskeletal deformity. The patient also suffered from Systemic Lupus Erythematosus (SLE) and was treated with Plaquenil 300 mg a day. The initial examination revealed a Class II skeletal discrepancy associated with a deep bite with maxillary vertical excess, mandibular hypoplasia and dental midline discrepancy. After a thorough evaluation and 3D planning of the surgery, the following simultaneous surgical procedures were performed: Firstly, a Le Fort I maxillary impaction of 2 mm and advancement of 2.5 mm, midline correction and leveling of the occlusal plane. Secondly a bilateral sagittal split osteotomy for mandibular advancement of 7 mm and a rotation of 1 mm to the left.

There were no intraoperative complications. The LeFort I osteotomy was performed using a reciprocating saw and osteotomes and was fixed using four 1.4 mm titanium plates and mini-screws (Titamed, Kontich, Belgium). The mandibular osteotomies were then performed. After the osteotomy, a series of small to large osteotomes were used to initiate the split. Three bicortical screws ranging from 10 mm to 14 mm were used to stabilize the bony fragments (Figure 1). The patient was in a good condition after the surgery and was discharged home 24 h after surgery with prophylactic antibiotic treatment (amoxicillin 500 mg 4 times a day during five days).

After five weeks, she was still experiencing uncommon significant pain in the right temporal region. Although the swelling on the left side had resolved normally, swelling persisted on the right side at the level of her right cheek. The pain was described as pulsatile and associated with worsening headache. The patient was first treated as it was a postoperative reinfection with oral antibiotics (amoxicillin-clavulanic acid 875 mg 3 times a day for a total duration of 7 days). Two days after the beginning of the antibiotic therapy, a head and neck Computed Tomography (CT) scan with intravenous contrast was performed, revealing a liquid mass of 5.2 cm × 2.7 cm × 4.7 cm with a strong contrast uptake around the right mandibular ramus (Figure 2).

A first surgical approach was undertaken in order to drain the fluid mass. While no pus could be found in the mass, a profuse bleeding was encountered. The bleeding was controlled with local measures, including electrocautery, Spongostan™ (Absorbable Haemostatic Gelatin Sponge, Johnson and Johnson Ethicon Inc, USA), hemostatic agent Floseal® (Baxter) and packing. A week later, a recurrence of the same symptoms occurred, especially a pulsatile pain with the same localization as previously. A second CT scan with

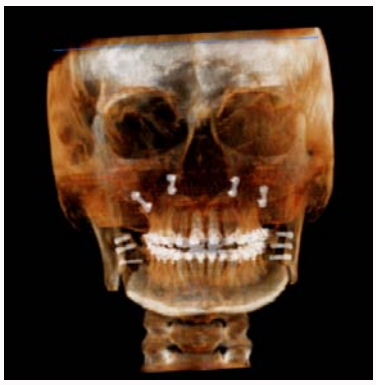

Figure 1: Post-operative three-dimensional reconstruction.

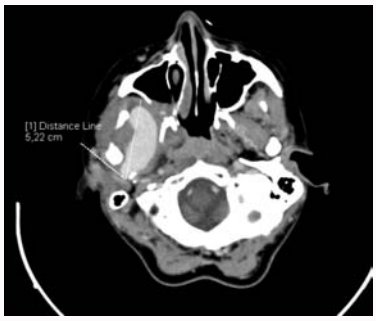

Figure 2: CT scan with contrast showing showed a liquid mass of 5.2 cm × 2.7 cm × 4.7 cm with a strong hyperdense aspect after contrast administration around the right ramus.

intravenous contrast demonstrated a leakage of contrast and a late filling of a 5.7 cm × 5.6 cm mass involving the medial and part of the lateral portion of the right mandibular ramus. An angiography was then performed and confirmed the presence of a pseudoaneurysm in the distal portion of the right Ascending Pharyngeal Artery (APA) at the level of the bifurcation of the maxillary artery and the superficial temporal artery (Figure 3). An embolization was performed with coils placed into the distal ascending pharyngeal artery using a 3×14 Nester microcoil (Figure 4). The post-embolization angiogram (Figure 5) showed no further patency of the pseudoaneurysm of the distal APA. The postoperative course was uneventful and the patient was discharged home the next day. After 5 months, no ischemic damage was observed in the maxilla or in any other part of the face and no relapse or recurrence of bleeding had occurred. The orthognathic treatment was successful and the patient remained satisfied with the results.

Review of the Literature

Pseudoaneurysm is a rare clinical entity caused by the rupture of arteries with extravasation of blood in the vessel wall due to a subadventitial dissection. Traumas such as penetrating lesions in the external carotid artery and its deeper branches or to the superficial temporal artery are the most important risk factors for the development of a pseudoaneurysm. Pseudoaneurysms rarely occur after head and neck surgical procedures such as orthognathic surgery [3,4]. In a series of more than 8000 aneurysms and pseudoaneurysm, only 21 pseudoaneurysm of the external carotid artery were described, among which 19 occurred after surgery in the region surrounding the carotid artery [5].

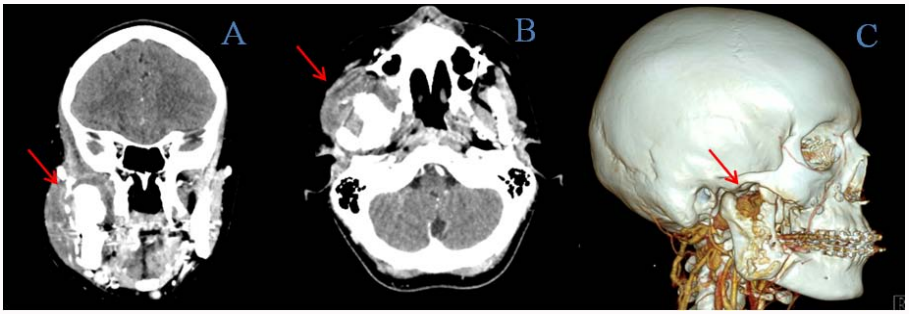

Figure 3: CT scan with contrast: A, coronal view, B axial views and C, 3 dimensions reconstruction revealed a well-defined radiolucent cavity (arrow), involving the medial and part of the lateral aspect of the right mandibular ramus, near the sigmoid notch.

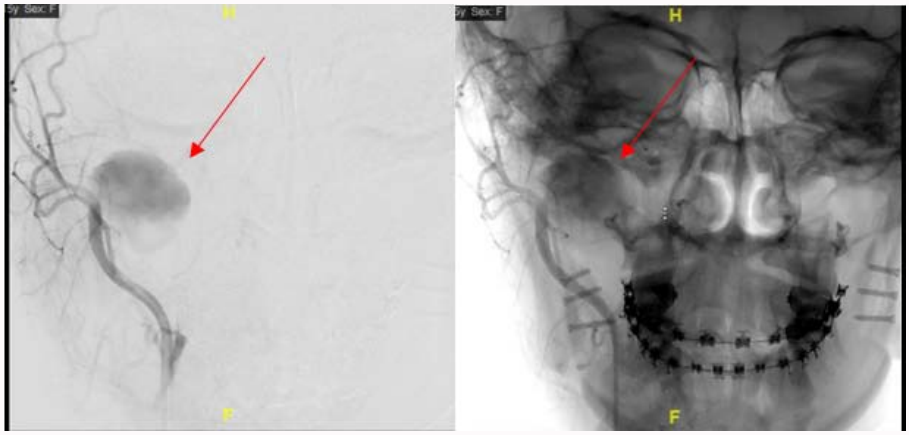

Figure 4: A: External carotid artery injection demonstrates a pseudoaneurysm (arrow) of the ascending pharyngeal artery, B: Frontal view of unsubtracted images of the same.

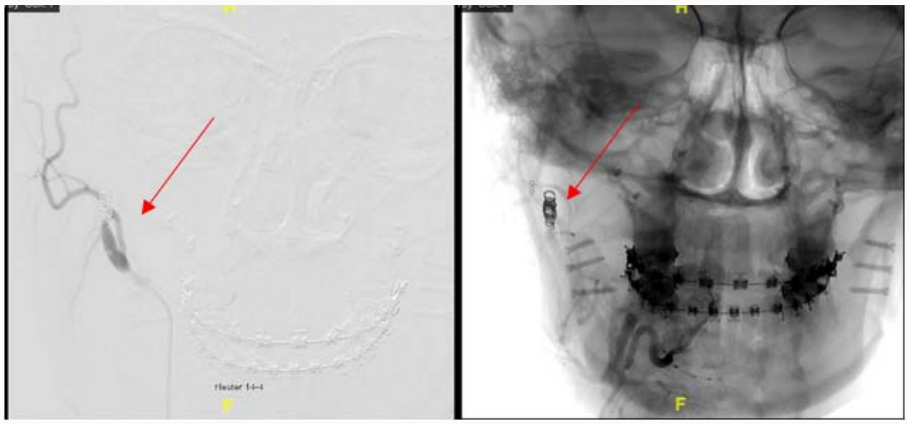

Figure 5: A: frontal view of external carotid artery injection after coil embolization (arrow) B: Unsubtracted view of the same after coil embolization of the pseudoaneurysm.

To further outline the occurrence of pseudoaneurysm following orthognathic surgery, as well as the clinical spectrum and treatment modalities, a search of the PubMed databank using the following search terms was performed: "osteotomy," "orthognathic surgery,""pseudoaneurysm," "false aneurysm," "aneurysm". These terms were used in every possible spelling and as synonyms, acronyms, and key or text words. The Boolean operators used were "OR" and "AND". Exclusion criteria were reviews, personal opinions, letters, and conference abstracts. Full text not available or not written in English, public communications or posters were also excluded.

The first phase of the selection highlighted 106 articles across the before mentioned electronic databases. After the revision process of the titles and the abstracts, 29 articles were eligible according to the inclusion criteria (Figure 6). One of these 29 articles was excluded because it did not report on a pseudoaneurysm but on an aneurysm.

In total, 31 patients who suffered from PA following orthognathic surgery (Table 1) are described in literature. The most common orthognathic surgery associated with PA is a Le Fort I osteotomy (67.7%), followed by osteotomy of the mandibula (29%) and finally, one case of PA after an osteotomy for a surgically assisted rapid

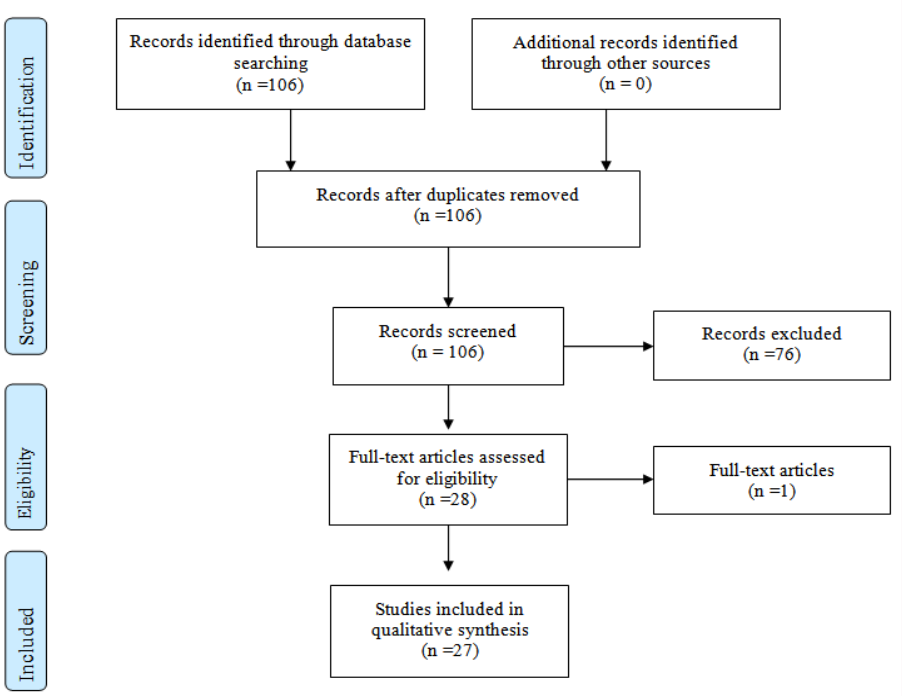

Figure 6: Prisma diagram.

Table 1: Reported cases of the formation of pseudoaneurysms after orthognathic surgery.

Author (Year)	Age, y	Sex	Surgery	localization	Duration of the lesion	Treatment
Hemmig et al. [11]	29	Female	Le Fort I	Sphenopalatine artery	2 weeks	Gianturco coil
Clark et al. [12]	15	Male	Subcondylar osteotomy	Maxillary artery	9 weeks	3-mm Gianturco coil
Solomons and Blumgart [13]	20	Male	Le Fort I	Maxillary artery	4 weeks	Large gel foam followed by 3-mm Gianturco coil
Lanigan et al. [14]	13	Female	Le Fort I	Sphenopalatine artery	6 weeks	Le Fort I associated to direct ligation of the artery
Lanigan et al. [14]	20	Female	Le Fort I	Sphenopalatine artery	3 weeks	Ligation of the carotid artery
Lustbader et al. [15]	17	Male	Le Fort I	Maxillary artery	12 days	Three guide wires
Albernaz and Tomsick [16]	40	Female	Le Fort I	Descending palatine artery	3 weeks	50 Pieces of PVA
Albernaz and Tomsick [16]	30	Female	Le Fort I	Maxillary artery	Several days	3 PVA sponge, a 3-mm platinum coil, and 0.1 mL n-BCA
Rogers et al. [17]	23	Male	Le Fort I	Maxillary artery	3 days	Fiber coils
Bradley et al. [18]	17	Male	Le Fort I	Maxillary artery	8 months	Coil
Procopio et al. [19]	37	Male	Le Fort I	Sphenopalatine artery	63 days	2 complex helical fibered platinum coils (3 mm × 30 mm)
Procopio et al. [19]	24	Male	Le Fort I	Sphenopalatine artery	10 days	4 Complex helical fibered platinum coils
Fernandez-Prieto et al. [20]	26	Female	Le Fort I (segment)	descending palatine artery	30 days	Platinum microcoils dcs-18 spiral
Manafi et al. [21]	42	Female	BSSO	Maxillary artery	20 days	300-Km Polyvinyl acetate
Silva et al. [1]	20	Male	BSSO	Mandibular branch of maxillary artery	6 weeks	Coils
Elton et al. [22]	41	Female	Subcondylar osteotomy	Maxillary artery	5 years	Ligation of ECA
Pappa et al. [23]	17	Male	BSSO	facial artery	7 days	Coils
chepla et al. [24]	17	Female	Le Fort I	Maxillary artery	1 week	platinum coils and n-BCA glue
Avelar et al. [3]	20	Male	Le Fort I	Sphenopalatine artery	9 weeks	Microcoil
Krishnan et al. [25]	37	Male	Le Fort I	Intern carotid artery	1 week	overlapping telescoping stents
Cohen et al. [26]	20	Male	Le Fort I	descending palatine artery	1 week	Proximal coiling
Precious et al. [27]	32	Male	BSSO	Facial artery	2week	Coil embolization

Precious et al. [27]	26	Male	BSSO	arterial branch of the external carotid artery	9wk	Coil embolization
Precious et al. [27]	22	Male	BSSO	facial artery	2 weeks	ultrasound probe
Hacein-Bey et al. [28]	18	Male	Le Fort I	Internal carotid artery	4 weeks	coil
Kim et al. [29]	31	Male	Le Fort I	Sphenopalatine artery	2 weeks	embolization with lipiodol and n-BCA
Bykowski et al. [30]	27	Male	Le Fort I	Maxillary artery	3 weeks	coilembolized
Laskarides et al. [31]	22	Female	Le Fort I	Sphenopalatine artery	2 weeks	coil
Niazi et al. [32]	?	Male	Le Fort I	descending palatine artery	1 weeks	placement of three Vortex Pushable coils
Neto et al. [4]	33	Female	BSSO	Facial artery	1 week	embolization

palatal expansion is described.

According to collected articles, the most common localization of the PA is the internal maxillary artery or one of its branches, with 77.4% of the PA occurring in this localization. In the specific context of an osteotomy of the mandibula, the most frequently affected artery is the facial artery, reported in 50% of the reported cases after an osteotomy of the mandibula.

So far in literature review no case of PA involving the ascending pharyngeal branch following an orthognathic surgery has been described. The ascending pharyngeal artery most commonly emerges from the posterior wall of the proximal external carotid artery, cephalad to the source of the occipital artery [6]. The ascending pharyngeal artery can also arise from the proximal occipital artery and in rare cases; it can alternatively arise from the internal carotid artery or from the ascending cervical artery [6]. The ascending pharyngeal artery has a width of 1.57 (0.9-2.3) mm and divides early into two major trunks, [7,6] the posterior neuromeningeal trunk that enters the posterior fossa through the foramen magnum and the anterior pharyngeal trunk. A third branch, the inferior tympanic branch arises between these two major trunks [6].

The ascending pharyngeal artery has a close relationship with the maxilla. The vascularization of the maxilla is ensured by a rich anastomotic network, including multiple arteries such as the sphenopalatine artery giving rise to the nasopalatine artery, the descending palatine artery, the ascending palatine branch of the facial artery, the alveolar branches of the internal maxillary artery and the anterior branch of the ascending pharyngeal artery [8].

The ascending pharyngeal artery plays an important part in the healing process of Le Fort I osteotomies, due to the vascular supplies attached to the posterior palatal soft-tissue pedicle [6,9]. Its anterior branch enters the soft palate more cephalad compared with the ascending palatine artery by passing over the tensor veli palatini and levator palatini muscles posterior to the pterygoid muscles [8].

The pseudoaneurysm develops after an incomplete tangential damage of the arterial wall, preserving its continuity, and its size increases progressively. Following orthognathic surgery, PA mainly concerns the maxillary arteries and sphenopalatine arteries. The risk occurs during the pterygomaxillary disjunction due to a poor positioning of the osteotome or during the "down fracture" of the maxilla or due to the sharp margins of the bony cuts nearby the stretched vessel. Because of the localization of the "neck" of the PA situated on the level of bifurcation between the internal maxillary artery and the temporal artery, we assume that the PA was caused due to supraperiosteal positioning of the osteotome during the pterygomaxillary disjunction.

PA is often suggested by a patients complaints, the most common being the report of a pulsatile mass in an area that was recently injured [3], confirmed by physical examination. Other common manifestations include unresolved swelling and pain, a thrill or a noise and paresthesia from compression of adjacent structures can be associated. This clinical entity is usually diagnosed after a few weeks due to its progressive development and therefore late clinical manifestation. It is often mistaken as a postoperative infectious collection [10]. A contrast CT is the recommended first diagnostic tool due of its ability to reveal anatomic details of vascular injuries [3], while the angiogram is the most specific exam, describing the pathway feeding the injury and locating the site of bleeding to allow precise treatment. The main objective of the embolization is to deposit a substance such as polyvinyl particles, microcoils or liquid adhesives within the aneurysmatic network [3]. The selective embolization is executed with a catheter introduced through the femoral artery until the ECA and its branches [3].

Conclusion

Pseudoaneurysms occur very rarely after orthognathic procedures. Late onset pain and facial swelling after these types of surgery should raise suspicion to the possibility of developing a pseudoaneurysm rather than assuming postoperative infection. Early diagnosis and angiographic embolization can minimize complications.

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