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Endovascular Management of Epistaxis Secondary to Dissecting Pseudoaneurysm of the Descending Palatine Artery Following Orthognathic Surgery

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Abstract

Orthognathic surgeries such as Le Fort I are widely used in clinical practice. Postoperative internal maxillary artery pseudoaneurysm is a very rare complication, which can lead to significant postoperative bleeding. In this article, we report a case of early postoperative bleeding secondary to pseudoaneurysm following Le Fort I surgery with a novel endovascular treatment approach.

Keywords

Embolization; internal maxillary artery (IMAX); postoperative complication; embolization; Le Fort surgery

Introduction

Vascular complications from orthognathic surgeries range from common postoperative oronasal mucosal bleeding to very rare arteriovenous (AV) fistula and pseudoaneurysm formation. Pseudoaneurysms in the external carotid artery (ECA) branches are commonly secondary to facial trauma and fractures leading to vascular injury. Other etiologies, albeit less frequent, include iatrogenic injuries during oral/facial surgeries, needle aspiration procedures, infections/abscesses, and radiation vasculitis. Le Fort I surgery is a procedure done to separate the pterygomaxillary junction by osteotomy. It is often performed to correct congenital/developmental facial and jaw anomalies.

Here, we report a unique combination endovascular management of a rare case of descending palatine artery pseudoaneurysm causing intermittent life-threatening delayed hemorrhage from orthognathic surgery.

Background and Objective

Fundamentally, pseudoaneurysms that occur within the territory of the ECA, specifically the internal maxillary artery (IMAX) and its branches, have been treated with either coil or glue embolization. Sometimes, when it is technically feasible the aneurysm itself is coiled, but in most cases, the IMAX from which an aneurysm arises is instead occluded. We describe a case of a novel combination approach in which we used coil-assisted glue embolization successfully to obliterate the pseudoaneurysm cavity arising from the descending palatine artery and the third portion of the IMAX. The rationale behind this technique is to direct the n-Butyl cyanoacrylate (n-BCA) into the neck of the aneurysm while also avoiding distal embolization by the metallic coils.

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Table 1. Summary of cases of pseudoaneurysm post Le Forte surgery

No	Study/year	Age/ gender	Presentation	Time interval	Artery	Size	Treatment
1	Hemmig *et al*. [4] 3473202	29 y F	Epistaxis	11 days	SPA	unknown	Coil embolization
2	Lanigan *et al*. [3], 2037912 (already there)	13 y F	Epistaxis	6 weeks	SPA	1 cm dia	Ligation and resection
3	Lanigan *et al*. [3] 2037912(already there)	21 y F	Epistaxis	3–11 weeks (multiple)	SPA	9 × 13 mm	Embolization × 2 unsuccessful followed by IMAX and then ECA ligation
4	Lanigan *et al*. [3] 2037912 (already there)	29 y M	Epistaxis	6 days	IMAX	Small	Coiling B/L IMAX
5	Bradley *et al*. [16] 12457086	17 y M	Facial asymmetric swelling	8 months	Distal IMAX	Unknown	Coiling
6	Procopio *et al*. [12] 12684974	24 y M	Epistaxis	5 days	SPA	unknown	Coil embo of IMAX
7	Procopio *et al*. [12] 12684974	37 y F	Epistaxis	21 days	SPA	unknown	Coil embo failed ×2 followed by ECA ligation
8	Avelar *et al*. [17] 21119417	20 y M	Epistaxis, swelling	9 weeks	SPA	2 × 2 mmInc. to 13 mm dia	7 platinum microcoils in sphenopalatine artery
9	Chepla *et al*. [18] 20613567	17 Y F	Epistaxis	1 week	IMAX	3.2 cm × 3.1 cm × 2.0 cm With AV fistula	35 bare platinum coils and n-BCA glueEmbolization of the venous drainage tract.
10	Cohen *et al*. [15] 22502912	20 y M	Oral bleeding	45 days	DPA	1.7 cm	Proximal coiling
11	Politis [13] 22480877	17 y F	Epistaxis	6 and 14 days	IMAX	Unknown	Coil embolization
12	Kim *et al*. (2013) 23601117	31 y M	Epistaxis and oral bleeding	2 weeks	SPA	2 cm dia	nBCA embolization

Method

We performed a PubMed literature search for original prospective, retrospective, observational studies, case reports, case series, and review articles on pseudoaneurysm formation and its treatment after orthognathic surgeries which were published in the English language. Keywords used included: “Le Fort I,” “pseudoaneurysm,” “orthognathic surgery,” “embolization,” “endovascular procedures,” “iatrogenic,” “postoperative complications,” and “epistaxis.” The authors reviewed full texts and reference lists from relevant papers to identify further articles. In some instances, relevant papers from outside the initial search parameters were included if there seemed to be a lack of more current evidence.

Result

10 reported publications regarding pseudoaneurysm as a complication of Le Fort I surgery were reviewed. A total of 13 cases were found as summarized in Table 1. We did not include posttraumatic or spontaneous cases of pseudoaneurysm in our review.

Clinical Presentation

A young patient underwent a Le Fort I osteotomy with advancement, impaction, and bilateral intraoral vertical ramus osteotomies of the mandible, in order to correct his mandibular hyperplasia and maxillary hypoplasia, with an uneventful immediate postoperative course. The patient was discharged two days after surgery. One week later, he started having multiple attacks of self-limiting intermittent oronasal bleeding. Anterior and posterior nasal packing was done and the patient was admitted to the hospital. No apparent coagulopathy was found. Despite the attempts to control the bleeding, the bleeding episodes continued and he was referred to endovascular neuroradiological surgery for angiographic evaluation based on high clinical suspicion of iatrogenic vascular injury/malformation. The patient was nasally intubated in the operating room in anticipation of airway difficulties due to his recent facial surgery. The patient was then transferred to the endovascular neurosurgical suite. The patient was suspected to have postoperative epistaxis secondary to underlying vascular injury. He was prepared for a diagnostic cerebral angiogram with intent to treat.

Digital subtraction angiography of the bilateral internal and external carotid arteries were performed, showing a saccular pseudoaneurysm of the right descending (major) palatine artery originating from the third (pterygopalatine) segment of the right IMAX measuring 7.8 mm × 11 mm with a 5-mm neck [Figure 1(A) and (B)]. The sphenopalatine artery was visualized to have connections with the anterior and posterior ethmoidal arteries of the ophthalmic artery through the septal arteries.

Endovascular treatment was then carried out by means of a 6F guide catheter (Envoy MPD Codman and Shurtleff-Inc., USA), placed in the right ECA after right internal carotid artery (ICA) angiographic assessment. Activated coagulation time of 200–300 seconds was maintained during the procedure by administering intermittent intravenous weight-adjusted heparin boluses. A Prowler Select Plus 150-cm 90° microcatheter (Cordis, USA) was advanced over Synchro 2 0.014-inch 200-cm soft microwire (Striker, USA) using roadmap technique into the distal third of the IMAX. Because of the above-

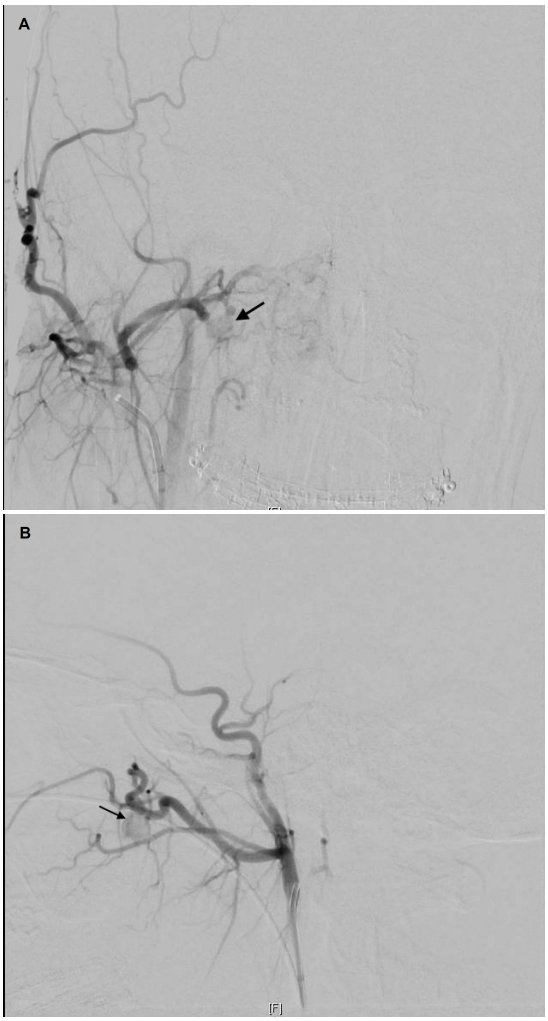

Figure 1. (A) Anterio-posterior projection and (B) lateral of the right ECA DSA angiography showing the right IMAX pseudoaneurysm (arrow).

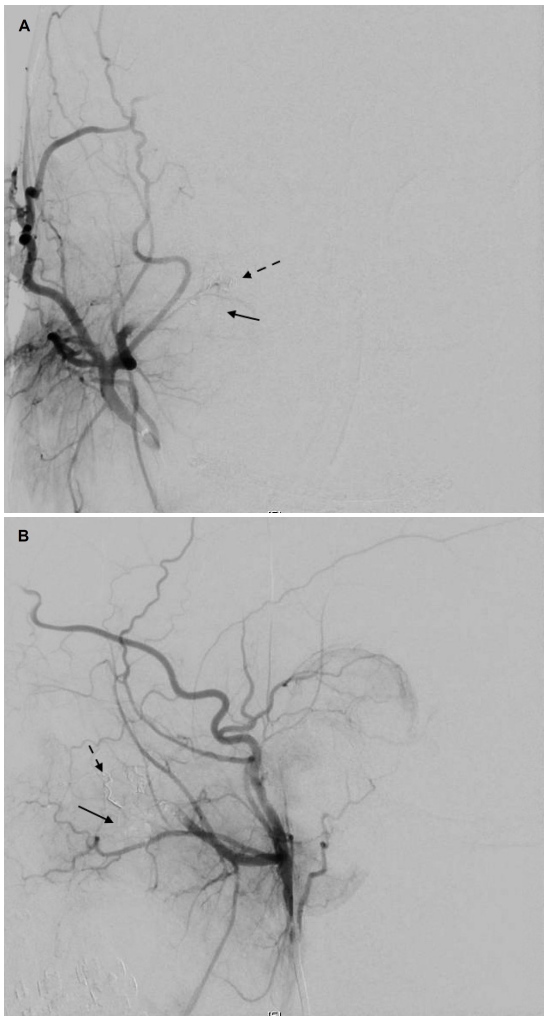

Figure 2. (A) Anterio-posterior projection and (B) lateral of the right ECA DSA angiography post embolization showing the resolution right IMAX pseudoaneurysm (arrow), and evidence of the embolization material coil (dotted arrow) and liquid embolic material.

mentioned ICA ECA connections the decision was made to deploy the coils in the proximal segment of the sphenopalatine artery in order to prevent n-BCA glue embolization of the ophthalmic artery.

The microwire was removed followed by sequential placement of three Vortex Pushable coils measuring 2 mm × 2.5mm, 3 mm × 2.5mm, and 4mm × 4mm into the IMAX distribution distal to the descending palatine artery origin (i.e., the sphenopalatine artery). Next, a mixture of n-BCA glue and ethiodized oil was prepared in a ratio of 2:1. Afterward, the microcatheter was placed just proximal to the origin of the pseudoaneurysm and flushed with D5 Saline. Under the continuous fluoroscopic mask, 0.5 cc of the n-BCA mixture was slowly injected through the microcatheter until penetration into the pseudoaneurysm was visualized [Figure 1(A) and (B)].

Control angiogram of the ECA after embolization demonstrated no contrast opacification of the pseudoaneurysm and complete occlusion of the third portion of the IMAX from the n-BCA cast proximally to the coils distally [Figure 2(A) and (B)].

After the procedure, no immediate complications were noted. The patient was observed in the intensive care unit for one day and was later successfully extubated. No recurrence of bleeding episodes was noted during the rest of the patient's hospital stay.

Discussion

Aneurysms, in general, are classified as either true or pseudoaneurysms. In true aneurysms, all three layers of the vascular wall, namely, the intima, media, and adventia are evaginated in the aneurysm. In pseudoaneurysms, only one or two layers of the vascular wall are evaginated [1]. Head and neck ECA distribution aneurysms are well defined but rarely occur. Among the branches of the external carotid arteries, the facial artery and superficial temporal artery are more prone to develop pseudoaneurysms secondary to trauma because of their tortuous, longer, and more superficial course [2]. Although the MAX along with its divisions is a deep terminal branch of the ECA and is less affected in trauma cases, it is the most common site of the development of pseudoaneurysms as a complication of orthognathic surgery secondary to an inadvertent partial tear in the arterial wall, as suspected in our case.

Regardless of whether the cause is trauma or iatrogenic injury, the mechanism of pseudoaneurysm formation remains the same. A partial tear in the vessel wall causes hemorrhage in the surrounding tissue, which leads to hematoma formation. Subsequent expansion of hematoma under arterial pressure occurs until the tamponade effect causes the hemorrhage to stop. Over the next 1–8 weeks, this hematoma liquefies and the internal lining gets endothelialized (pseudointima formation) [3]. It is at this point the blood starts flowing in and out of the aneurysmal cavity, leading to shear force and stress under arterial pressure causing pulsatile expansion, eventually rupturing and bleeding from the weakened portions of the aneurysmal wall. Because of the deep location, common clinical features of any pseudoaneurysm (i.e., pulsatile expanding painful mass with bruit and thrill, and/or neuropathy secondary to compression of local nerve) are not found in an aneurysm of the IMAX or of its branches, and this condition generally manifests as hemorrhage from the posterior nose. Due to the pathophysiologic reason, our patient, and most other patients, after Le Fort I surgery present with intermittent oronasal bleeding 1–8 weeks after surgery [4,5].

Technical Discussion

Open surgical treatment consists of ligation of the IMAX proximal and distal to the origin of pseudoaneurysm followed by en-bloc resection. The extensive nature of collateral and bilateral blood supply in the facial region renders the dissection and resection difficult and can be potentially complicated by blood loss. Since the development of endovascular techniques including embolization, stents, and stent grafts, the morbidity related to the treatment of pseudoaneurysms has significantly decreased. Embolization is most popular of these treatment modalities and is most widely used. Historically, different techniques and materials have been used for embolization of the IMAX including Gianturco coils, gel foam [4–6], gelatin, n-butyl cyanacrylate, polyvinyl alcohol, Dacron fibers, metallic coils, covered stents, and lately, electronically detachable coils [7]. Choosing among these methods of angioembolization is based on the size of the vessel, size of the aneurysm, accessibility, expertise of the surgeon, and whether the occlusion is needed temporarily or permanently [8].

Comprehensive understanding of the ICA and ECA connections and communications is key to avoiding catastrophic complications (i.e., strokes from endoembolization). Although safe and minimally invasive, endovascular embolization has the potential for complications, and sometimes causing significant morbidity and mortality. Minor complications include craniofacial pain, facial edema and paresthesia, nasopharyngeal ulceration, and trismus. Major complications, although rare, include intraoperative rupture of the pseudoaneurysm wall, embolic phenomena from aneurysm cavity thrombi or from embolization material to the brain from shunting via ECA/ICA collaterals causing strokes and other cranial neuropathy, ischemic necrosis of maxilla, loss of teeth, and inadvertent proximal branch occlusion [9,10].

Our case highlights the descending palatine artery pseudoaneurysm as a rare complication of Le Fort I procedure. Several key technical points are of paramount importance when providing endovascular treatment for it. Baseline angiograms of the ECA and ICA, both ipsilateral as well as contralateral, are required prior to embolization to assess for possible dangerous anastomoses. These include the various middle meningeal artery and inferolateral trunk variants with anastomoses with the retina and the ophthalmic branch of ICA, respectively [11]. Also, adequate positioning of the microcatheter in a nonwedged position and gentle injection are helpful in avoiding opening of anastomoses that may be initially invisible angiographically [11].

Another technical issue is adequate to control of distal embolic phenomena and the avoidance of inadvertent reflux of embolic liquid material proximally, which can cause branch occlusions. Our case very nicely demonstrates a novel combined embolization approach where we used pushable coils distal to the aneurysm neck, thereby avoiding the n-BCA from distal embolization and dilution. In addition, use of n-BCA injected into the IMAX proximal to pushable coils immediately obliter-

ated the pseudoaneurysmal cavity. With the first hint of reflux of contrast-enhanced n-BCA material, we stopped and pulled our microcatheter proximally. Check angiograms demonstrated not only lack of contrast enhancement of aneurysm but also patency of major proximal IMAX branches.

Overall, the technical advantages of this approach are a rapid, segmental, and complete occlusion of the desired area of the IMAX immediately proximal and distal to the pseudoaneurysm neck with the use of few relatively inexpensive distal coils and less than one vial of n-BCA. The limitations are a lack of complete control of the liquid embolic agent, potential embolization through an unopacified anastomosis, and the inability to reposition either the coils or n-BCA [11].

Comparing our technique with others reported in the literature, our technique is novel and is being described for the first time in the management of pseudoaneurysmal hemorrhage post-Le Fort I surgery. Most cases reported in the literature were treated with a single modality of successful coil embolization [3,4,12–17]. These cases do vary in the technique of coil placement, number of coils used, and unilateral versus bilateral IMAX occlusion as opposed to pseudoaneurysm obliteration. At least two cases were reported where coil embolization was attempted twice but failed to control hemorrhage; rescue ECA ligation was then performed via surgical exploration [3,12]. One case reported primary surgical ligation and resection of a pseudoaneusrysm and another case used a combination approach with dual coil and n-BCA embolization, but the n-BCA was injected in the venous drainage tracts, which arose from the aneurysm [3,18].

Limitations

As with any other case report, there are several inherent limitations that were unavoidable due to the retrospective design of the case report. The treatment methodology was based on the clinical judgment of operating physicians. Another major limitation is the absence of an untreated control group. This case does not allow the direct comparison with other potential embolization techniques but at the same time sheds light on a safe combination approach to consider. Finally, the small number of patients undergoing this therapy due to the infrequency of this condition and due to the potential life-threatening hemorrhage if left untreated limits the possibility of conducting a randomized control trial [11].

Conclusion

This case illustrated that combined endovascular embolization is a safe, minimally invasive technique for treatment of iatrogenic pseudoaneurysm of the descending palatine artery post-Le Fort I surgery.

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