Profound facial bleeding after buccal mucosa graft harvest – A rare case of facial artery pseudoaneurysm

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A B S T R A C T

Facial artery pseudoaneurysms are exceedingly rare events that can occur as a complication of oral maxillofacial surgery or facial trauma. The management of such pseudoaneurysms following buccal mucosa graft harvest for urinary reconstructive indications has not previously been described. Here, we describe a facial artery pseudoaneurysm that presented as repeated, episodic facial bleeding episodes following buccal mucosal harvest for a patient undergoing urethroplasty.

1. Introduction

Pseudoaneurysms are a rare vascular pathology often resulting from trauma causing an incomplete tear of the vessel wall.1 Facial artery pseudoaneurysm is generally associated with facial trauma however, there have been rare, reported cases of facial artery pseudoaneurysms after tooth extraction, tonsillectomy, and mandibular osteotomy.2 To date, there have been no reported cases involving the harvest of buccal mucosal grafts. Here, we report a case of a facial artery pseudoaneurysm following buccal mucosal graft harvest for a patient undergoing urethral reconstruction.

2. Case presentation

Patient is a 43-year-old male with a history of hypospadias who had undergone previous repair as an infant. At presentation, he reported symptoms of incomplete emptying and recurrent urinary tract infections. Retrograde urethrogram showed anterior urethral stricture and covered a large portion of the penile urethra (Fig. 1). He elected to proceed with staged urethroplasty and underwent the 1st stage with harvest of a buccal mucosa graft from his left cheek performed by our reconstructive urology team. He was discharged home the day of surgery with a penile bolster dressing uneventfully.

On post-operative day 2, the patient developed of significant bleeding from his mouth and went to an outside hospital. He received packed red blood cell transfusions immediately upon presentation and had tranexamic acid (TXA) applied superficially on top of the harvest site in the mouth. The bleeding subsided and he experienced no recurrent bleeding for 24 hours. He was discharged with a stable hemoglobin.

He presented again to an outside emergency department greater than 1 week later for another episode of spontaneous, profuse bleeding. There were no inciting events or trauma to the graft site. At presentation, he was noted to be bradycardic and diaphoretic and was ultimately diagnosed with a pulmonary embolism (PE) managed with IVC filter placement. Due to the need for anti-coagulation for treatment of his pulmonary embolism and his recurrent episodes of bleeding, he was taken to the operating room for exploration. Intraoperatively, he was noted to have a bleeding vessel at the maxillary base of the left buccal graft harvest site which was oversewn with dissolvable suture and was further reinforced with an Alloderm tissue matrix. He was monitored for an additional 3 days after the procedure and discharged home on DVT prophylactic dosing of Lovenox.

One week later (postoperative day 20), the patient developed another spontaneous bleed from his buccal graft harvest site which self-resolved prior to presentation at our hospital. Given the recurrent bleeding, he was admitted for urology and otolaryngology evaluation with plans for an exam under anesthesia the following morning. A CT angiogram was performed prior to the operating room which demonstrated a 6mm focal dilation of a branch of the left facial artery compatible with a pseudoaneurysm (Fig. 2). Interventional radiology was consulted for elective embolization, and he was taken for emergent embolization of the left facial artery pseudoaneurysm by interventional radiology with microcoils when another spontaneous bleeding event occurred (Fig. 3). The patient was monitored for 4 days without additional bleeding and discharged home. He was seen in follow up and has had no recurrent episodes of bleeding greater than 2 months from the original operation. The patient has not developed any ischemic changes to his face or mouth because of the embolization. He has normal

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periorbital sensation and can masticate appropriately.

3. Discussion

Aneurysms are classified as true, false, or dissecting. True aneurysms are a dilation of all the layers of an intact vessel wall whereas false aneurysms – also known as pseudoaneurysms – occur when a blood vessel is injured and leaks blood into surrounding tissue. The leaked blood eventually results in tamponade or clot formation and ultimately forms an aneurysm sac. Such aneurysm sacs can gradually expand and may spontaneously rupture, resulting in hemorrhage. The growth and eventual rupture of a pseudoaneurysm can be delayed for weeks to months. Given the abnormal clinical presentation of this patient including recurrent bleeding without pulsatile findings CT Angiography was performed after surgical exploration and would have diagnosed the pathology sooner. Previously, the treatment of pseudoaneurysms involved surgical excision; however as interventional radiology and arteriography continue to become more refined, many pseudoaneurysms are now managed with angiographic embolization. This allows for confirmation of successful treatment via arteriography following the procedure and avoids the potential neurologic complications and surrounding tissue damage, all while reducing the risk of bleeding.

Pseudoaneurysms in the facial area are exceptionally rare and normally occur due to surgical procedures, maxillofacial trauma, dental extractions, or maxillofacial surgery. Facial artery pseudoaneurysms are rare due to the small diameter of the facial artery and its deep, protected location. Knowledge of key anatomic landmarks and ensuring harvesting of only the mucosal layer with avoidance of muscle when harvesting buccal mucosa is key to avoiding bleeding complications. The common presentation is painless pulsatile swelling with a palpable thrill, although occasionally they can rupture and cause hemorrhage. The gold standard for diagnosis is CT angiography or arteriography.

Treatment can vary from non-invasive methods such as compression...
and observation to progressively more invasive treatments such as percutaneous injection of thrombosing agents, embolization by interventional radiology, or surgical resection. Previously pseudoaneurysms underwent surgical resection however given advancements in endovascular approaches, surgical resection has become less common. Percutaneous embolization is generally performed by injecting thrombin or embolizing the pseudoaneurysm resulting in transformation into a hematoma which is resorbed over time. While facial artery pseudoaneurysms are rare, they should be considered in all types of surgery involving the oral cavity including buccal mucosal grafts.

4. Conclusion

Though bleeding from buccal harvest sites typically self-resolve, when a patient presents with repeat episodic spontaneous bleeding, a broader differential diagnosis must be formed, and more invasive diagnostics and interventions must be considered. This case represents the first known report of a facial artery pseudoaneurysm successfully treated with coil embolization following harvest of a buccal mucosal harvest graft.

References

1. Shetty NK, Shandilya R, Pawar S, Gadre PK, Gadre K, Singh D. Management of late post-traumatic facial artery pseudoaneurysmal cyst: review of literature. J Maxillofac Oral Surg. 2015;14(2):201–205. doi:10.1007/s12663-014-0678-9.
2. Jo HW, Kim YS, Kang DH, Lee SH, Kwon TG. Pseudoaneurysm of the facial artery occurred after mandibular sagittal split ramus osteotomy. Oral Maxillofac Surg. 2013;17(2):151–154. doi:10.1007/s10006-012-0339-4.
3. Madani M, Veznedaroglu E, Pazoki A, Danesh J, Matson SL. Pseudoaneurysm of the facial artery as a late complication of bilateral sagittal split osteotomy and facial trauma. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2010;110(5):579–584. doi:10.1016/j.tripleo.2010.03.021.
4. Day AT, Genther DJ, Hui F, Mydlarz WK, Griffith G, Desai SC. Successful facial artery pseudoaneurysm coiling and pedicle preservation following free tissue transfer. Am J Otolaryngol. 2017;38(1):103–107. doi:10.1016/j.amjoto.2016.09.017.
5. Zachariades N, Ralli G, Papademetriou G, Papakosta V, Spanomichos G, Soulem M. Embolization for the treatment of pseudoaneurysm and transection of facial vessels. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001;92(5):491–494. doi:10.1067/moe.2001.117453.