Bilateral Maxillary Pseudoaneurysms as a Complication of Craniofacial Fracture: A Case Report

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Patient: Male, 16-year-old
Final Diagnosis: The bilateral maxillary pseudoaneurysms
Symptoms: Pain • paresis • swelling
Medication: —
Clinical Procedure: Embolization
Specialty: Dentistry • otolaryngology

Objective: Rare disease
Background: Pseudoaneurysms of the head and neck region are relatively rare and consequently there are few reports in the literature. The main causes of pseudoaneurysms in the head and neck area are associated with blunt or penetrating injuries in this area or are iatrogenic, originating during surgical procedures.

Case Report: The authors illustrate a case of a 16-year-old boy who had an accident on a scooter. A maxillary artery hemorrhage occurred after multiple craniofacial trauma including bilateral fracture of the condylar processes and treatment with intravascular embolization. Due to the anatomical location of the injury and deep location of the maxillary artery, it was impossible to control the bleeding by direct pressure; the only option was vessel embolization using interventional radiology or surgical intervention. The authors describe symptoms of pseudoaneurysm: pain in the area of the temporomandibular joint on the left side, persistent bilateral swelling of the cheeks and parotideomasseteric region with a greater asymmetry on the left side, as well as peripheral paresis of the facial nerve on the left side. The diagnosis was based on magnetic resonance imaging, ultrasound, and angio-CT. A reduction in the clotted pseudoaneurysm size on the left side was confirmed after 6 and 10 months through monitoring ultrasound examination of the neck vessels.

Conclusions: No similar case of a patient has been reported in the available literature. The development of bilateral pseudoaneurysms after injury is very rare. However, doctors should be alert to this possibility in the case of massive post-traumatic or postoperative bleeding.

Keywords: Carotid Artery Injuries • Embolization, Therapeutic • Facial Injuries

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Background

True aneurysm is an outward bulging which results from localized weakness of a vessel wall, while a pseudoaneurysm is a lesion, which is connected with extravasation of blood from the artery as a result of an injury, with compressed soft tissue around the artery creating an aneurysmal sack [1]. Pseudoaneurysm involves a risk of heavy bleeding and the risk of rupture. Due to the deep anatomical location of the maxillary artery, it is impossible to control the bleeding in this artery by direct pressure, but only by using another treatment: vessel embolization via interventional radiology or surgical intervention [2,3]. Here, we present a rare case of bilateral maxillary pseudoaneurysm that occurred after facial trauma and surgical procedures.

Case Report

A 16-year-old boy was admitted to the Department of Otolaryngology, Head and Neck Surgery due to multiple craniofacial fractures, which had been caused by a scooter accident. Initially, multiple fractures within the craniofacial bone, right clavicle fracture, lung contusion, and pneumothorax were diagnosed. Craniofacial CT examination showed a bilateral, compound fracture of the mandibular condylar processes, with their dislocation and the lateral displacement of the mandibular ramus on the left side by about 5 mm, as well as a fracture of the body of the mandible in the region of the mandibular left first pre-molar with a luxation of the mandibular left canine. The upper jaw was fractured on both sides in the secondary maxillary right first pre-molar and secondary maxillary left second pre-molar and bilateral fracture of the floors and medial walls of the maxillary sinuses. After conducting the necessary medical consultations and placing upper and lower dental splints with flexible lifts during hospitalization, the patient was qualified for surgical treatment. Surgeons conducted, under general anesthesia, reposition and stable osteosynthesis of the mandible using 2 titanium plates. The permanent maxillary left first, second, and third molars were removed, along with a fragment of the maxillary bone and a fractured section of the pterygoid process and the palatine bone. The permanent maxillary right first, second, and third molars were also removed, along with bone fragments. Next, surgeons conducted stable bilateral osteosynthesis of the maxillary bones, also using 2 titanium plates. A Surgicel absorbable hemostat was placed into both maxillary sinuses. The wounds were tightly sutured with non-resorbable sutures.

Bilateral fractures of the condylar processes were left for conservative orthopedic treatment. Then, the patient was given a palatal plate with bite blocks and obturators.

Figure 1. Clinical status of the patient 6 days after surgery. The external clinical examination shows bilateral swelling of the soft tissues of the cheeks and parotidomasseteric regions.

Figure 2. Ultrasonography of the left salivary gland showing aneurysmal dilatation of the left maxillary artery with turbulent flow of blood inside.
On the first day after surgery, doctors found improvement of the mouth opening and proper occlusion in the anterior teeth. On the 6th day after surgery, the patient reported increasing pain in the area of the temporomandibular joint on the left side and persistent bilateral swelling of the cheeks and parotideomasseteric region, with the greater asymmetry on the left side (Figure 1). On the 10th day after the surgery, the clinical examination revealed symptoms of peripheral paresis of the facial nerve on the left side. The magnetic resonance imaging (MRI) examination revealed a structure with heterogeneous signals on the left side (4.6×3.1×2.7 cm), medially from the displaced mandible ramus. In the central part of this structure, we observed a signal-free area with a dilated maxillary artery, which was connected to the mandible ramus from behind. The described lesion modeled and narrowed the parapharyngeal space. The ultrasound examination of the salivary glands showed a fluid space with a
noticeable turbulent flow of arterial blood in the area of the left parotid gland (Figure 2). The patient was qualified for endovascular treatment. Angio-CT of the head and neck arteries showed hematoma, which was situated medially from the left mandibular ramus and the parotid gland (Figures 3, 4). There was also a smaller pseudoaneurysm of the right maxillary artery (1.2×0.7×1.8), in the region of the fracture of the mandibular ramus.

A catheter was placed into the left external carotid artery using the Seldinger method. After administration of the contrast

Figure 6. Followup orthopantomographic examination 6 months after surgery, showing radiopaque coils after embolization of the pseudoaneurysm on the left side.

Figure 7. Followup orthopantomographic examination 10 months after surgery, presenting radiopaque coils after embolization of pseudoaneurysm on the left as well as the right side.
agent, the pseudoaneurysm was visualized (Figure 5A, 5B). Subsequently, 3 embolization coils were used to restrict the blood supply to the pseudoaneurysm. Followup ultrasound examination of the neck vessels and angiography revealed the presence of a completely clotted left pseudoaneurysm of the maxillary artery without evidence of reperfusion. The pseudoaneurysm on the right side was left for observation.

Monitoring orthopantomogram X-ray after 6 months showed a lesion that might have indicated a resorption of the mandibular ramus on the right side, but without CT examination, we cannot confirm this hypothesis. Monitoring ultrasound examination revealed a slight increase in the size of the right pseudoaneurysm. The patient was qualified for the embolization of the lesion on the right side. Monitoring ultrasound examination of the neck vessels after 6 months and 10 months confirmed a reduction in the clotted pseudoaneurysm size on the left side. The orthopantomogram presents the presence of radiopaque coils, which were used for embolization of the pseudoaneurysms (Figures 6, 7).

Discussion

Pseudoaneurysms result from blunt or penetrating trauma that causes partial rupture of the inner layers of the artery walls [1,4,5]. A rupture of the artery wall causes an outflow of blood from the lumen of the vessel to the surrounding tissues, which leads to the formation of a hematoma. Bleeding from the artery will be stabilized when the pressure of a forming hematoma is equal to the arterial pressure [6,7]. The external carotid artery and most of its branches are well protected against external injury [8]. The 3 branches of the carotid artery that are particularly susceptible to external injuries and thus to the formation of pseudoaneurysms are the following: superficial temporal artery, maxillary artery, and facial artery [2]. The superficial temporal artery is susceptible to blunt and penetrating trauma due to its superficial position [7,9]. According to anatomical criteria, the maxillary artery is split into 3 sections: mandibular, pterygoid, and pterygopalatine [10,11]. The mandibular portion of the maxillary artery is located in the vicinity of the neck of the condylar process of the lower jaw, which makes it susceptible to injury as a result of fractures of the mandible [7,12]. The pterygoid portion of the maxillary artery is the deepest part, and it is susceptible to laceration as a consequence of type I Le Fort fractures [13]. In this case, the patient suffered from both types of fractures on the right and left sides, which brought about the growth of the pseudoaneurysms. Pseudoaneurysms develop immediately, as a result of the direct trauma, or secondarily, after surgical procedures [10]. Because of the extensive network of arterial anastomoses of the maxillary artery, deep localization, and difficult surgical access, bleeding in this area is difficult to control [7,14]. The main causes of pseudoaneurysms in the head and neck area are associated with injuries oriatrogenic action during surgical procedures. Orthognathic procedures, osteosynthesis of condylar fractures, and surgical removal of impacted upper third molars may lead to injury of the maxillary artery [7]. Most pseudoaneurysms of the head and neck area are asymptomatic. Clinical symptoms may appear immediately after an injury or weeks or months later [8,15]. A palpable pulsation symptom can be felt on average 1-8 weeks after the injury [7]. Initial symptoms include headache, pain in the face and neck area of the injury, and unexplained swelling, which may pulsate to the rhythm of the heartbeat [7,15]. Paresthesia of peripheral nerves may occur with the growth and development of an aneurysm. In our case, these symptoms appeared a week after the operation and increased gradually. We suspected that the pseudoaneurysm could have led to mandibular branch reperfusion; however, we needed to have performed CT examination to confirm this hypothesis. This complication could have been caused by pressure exerted by the pseudoaneurysm and/or it could have resulted from injury.

The first element of diagnosis is a physical examination, but diagnostic puncture is contraindicated because it could lead to strong bleeding [7]. Doppler ultrasonography examination is a basic, cheap, and non-invasive test that can reveal turbulent blood flow and vasodilation in blood vessels. Ultrasonography is limited to diagnosis of pseudoaneurysms of the head and neck area, because of deep localization and the presence of thick bone and a significant amount of adipose tissue, which limits the penetration of ultrasound waves [7,8,16]. Nowadays, CT with contrast and angiography is the criterion standard in diagnosing pseudoaneurysms of the head and neck area [15,17]. Angiography allows location of the site of bleeding and determination of the architecture of the blood vessels [7,14,18]. The differential diagnosis was an arteriovenous fistula, which is associated with continuous sound [19]. Pseudoaneurysm should also be differentiated from hematoma, abscess, and inflammatory lymph node, cyst, and neoplastic tumor [7,18].

According to most authors, treatment of maxillary pseudoaneurysm should be started immediately after diagnosis due to the unpredictable development of the lesion. Non-invasive methods include observation, manual pressure, or using ultrasound. These methods are aimed at turning a pseudoaneurysm into a hematoma, which will then dissolve. Some pseudoaneurysms can regress spontaneously [7,16]. Invasive methods include surgical removal or intravascular embolization of the lesion. Embolization is aimed at delivering an embolization substrate to the vascular network of the pseudoaneurysm, which will stop the bleeding but will not obstruct the main vessel [18]. This method is faster, more effective, and safer than surgical removal and is used to treat deeply located lesions [7,10,18].
Embolization of pseudoaneurysms in the head and neck region is also used in the treatment of epistaxis, post-traumatic hemorrhage, and preoperative revascularization of a tumor [7]. Materials for embolization of endovascular lesions in the head and neck include metal coils, polyvinyl alcohol, n-butyl cyanoacrylate (NBCA), polymers, or absorbable sponges [19]. Titanium coils are positioned proximally with regard to the pseudoaneurysm and distally to the normal vessel [5]. The negatively charged blood component is attracted to the positively charged titanium, which causes a thrombotic reaction, which contributes to occlusion of the abnormal part of the vessel, without obstructing the normal part. Afterward, post-embolization angiography should be conducted to verify the effectiveness of the treatment [13]. The last treatment method for facial pseudoaneurysms is surgical excision of the lesion, which is used in the case of failure of endovascular embolization, necrotic aneurysms, or a rapid expansion of the lesion [20]. Difficult access, the risk of damage to other tissues, and the presence of non-cosmetic postoperative scars on the face are the main drawbacks of the surgical method [10].

References:


Conclusions

Pseudoaneurysms of the head and neck region are relatively rare. Consequently, there are few reports in the literature. However, it is crucial to be vigilant in the case of massive post-traumatic or postoperative bleeding. Pseudoaneurysms that qualify for observation only (not requiring embolization) require regular and careful followup examination to avoid complications.

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