

Facial Arteriovenous Fistula following Orthodontic Treatment: Case Report

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Abstract

The arteriovenous fistula is an anomalous connection between an artery and a vein. The superficial temporal artery and the facial artery are the most affected in the head and neck regions. It can be congenital, acquired or a post-traumatic fistula. People with this anomalous connection have had a wide range of symptoms reported. It presents as a mass of soft tissue with pulsatility and audible murmur at auscultation. Clinical signs can be detected within hours or up to several years after the trauma, depending on the location and diameter of the vessels involved. The differential diagnoses should include pseudoaneurysm, true aneurysm, arteriovenous malformation, cyst, abscess and hematoma, because any non-vascular lesion can present pulsatility due to pulsation transmitted by adjacent arteries. Arteriovenous fistula of the maxillary, mandibular and preauricular areas may cause life-threatening hemorrhage from epistaxis or otorrhagia. The diagnosis is difficult and it should be supported by imaging studies. The angiography is the gold standard exam for diagnosis and it enables to simultaneously perform endovascular treatment.

Keywords: Acquired Arteriovenous Fistula; Embolization; Angiography; Trauma; Orthodontic Appliance

List of abbreviations: DMSO: Dimethyl Sulfoxide; FTA-ABS: Fluorescent Treponemal Antibody – Absorption; RPR: Rapid Plasma Reagin

Introduction

Arteriovenous fistula is an anomalous connection between an artery and a vein, and it can be congenital or acquired [1]. It is possible for an acquired fistula to be established immediately, after days or even weeks, and it may cause life-threatening hemorrhage, epistaxis or otorrhagia in places like maxillary, mandibular and preauricular areas [2,3]. The acquired fistula is usually caused by penetrating trauma or, less frequently, by trauma with firearm [1,4,5-8]. The iatrogenic injury is often caused by percutaneous intervention such as kidney biopsies, cardiac catheterization and orthopedic surgery [7]. The superficial temporal artery and the facial artery are the most affected ones in head and neck regions [2,3]. They may also be associated with extensive cervical infections, maxillofacial fractures, certain surgical procedures (such as orthognathic surgery), aneurysm rupture, fibromuscular dysplasia and arterial dissections [2,6,8]. The acquired arteriovenous fistula has a wide spectrum of clinical presentation and depends on location, size and duration [3-5]. It presents as a mass of soft tissues with pulsatility and audible murmur at auscultation or, less common, distal pulse deficit and nerve compression [4,6,9]. The angiography is the gold standard exam and, with it, it's possible to simultaneously perform endovascular treatment [2-12].

Case Report

A 25-year-old man presents at the Stomatology Department for a nodular lesion on the left cheek mucosa with an undetermined evolution time.

An year before, he had gone to the Emergency Department because of a genital ulcer. Simultaneously he presented an ulcer on the left cheek mucosa. Both were painless and without associated adenopathy. The medical team hypothesized a diagnosis of primary syphilis due to the presence of orogenital ulcers and the high-risk sexual behaviors of the young man.

The blood test indicated a reactive Rapid Plasma Reagin (RPR) (1:64) and a positive Fluorescent Treponemal Antibody - Absorption (FTA-ABS). The patient was medicated and referred to Infectious Diseases consultation. He returned to the General Emergency Department, about 4 months later, reporting persistence of the lesion in the left cheek mucosa, and the collaboration of the Stomatology Department was requested.

This time, the patient referred that the ulcerated lesion in the oral cavity was related to the placement of a fixed-appliance orthodontic treatment, which caused trauma due to its arch wire. He denied association with other precipitating factors, such as infections, direct trauma such as aggression or falls, insect stinging, physical, emotional or contact factors (a new toothpaste, mouthwashes, among others) or trips abroad. His personal history did not reveal any other diseases, with the exception of syphilis. Finally, he also denied food or medication allergies and the use of frequent medication.

According to the patient's clinical information, he started the fixed-appliance orthodontic treatment at the age of 22, with a duration of 2 years, ending treatment at 24 years old.

The treatment consisted of aligning and leveling dental class I malocclusion, with correction of canine rotations and midline deviation, using conventional brackets and nickel-titanium and steel arches. The patient stated that the lesion appeared shortly after the placement of the upper appliance and maintained a location overlapping the device's arch wire. Furthermore, he reported episodes during which the arch was attached to the lesion. Despite upper appliance removal about 15 months ago, the injury persisted. The patient also reported that, associated to this unresolved situation, he became more concerned with the recent appearance of facial flushing and inevitable aesthetic damage.

He denied associated symptoms, namely respiratory, gastrointestinal, neurological, auditory or ocular, changes in salivary or lacrimal secretion or general symptoms (such as fever, asthenia or weight loss).

On objective examination, the patient showed a good general condition, as he was nourished and with apparent age corresponding to the real one. Upon cervical-facial examination, flushing was observed at the transition from the middle third to the lower third of the left hemiface, distal to the ipsilateral labial commissure. We did not observe any facial dysmorphisms, depressions or scars and we did not palpate any adenopathy in the different ganglion chains. Neurological examination (sensitivity and motor function test) was performed, without important changes. The jaws had a normal shape, and on their palpation, they showed regular contours, with no exostosis or swelling. The flush was in direct relation with a nodular intraoral lesion, with an ulcerated center, with approximately 5 x 4 cm in dimensions, well-defined limits, regular edges, mobile in relation to the deep planes and skin, and painless on palpation (Figure 1). The presence of thrill was questioned. There were no traumatic intraoral factors (cusps with sharp edges) or other lesions in the oral mucosa, asymmetries, bulging or depressions; the salivary glands had normal salivary flow, with good drainage of clean, transparent saliva.



Figure 1: Nodular and ulcerated lesion

The team developed an extensive differential diagnosis, that included: reactive injuries, bacterial infections (e.g., syphilis, gonorrhea, tuberculosis), fungal infections (e.g., histoplasmosis), viral infections (e.g., herpes simplex), autoimmune or immune component diseases (e.g., Crohn's disease, erythema multiforme, granulomatosis, medication); neoplasms (e.g., squamous cell carcinoma) and vascular pathology (due to the presence of thrill).

To that extent, the following complementary exams were requested: blood count, biochemistry, serology, autoantibodies and tumor markers, as well as a simple chest X-ray, which revealed no changes. Subsequently, a soft tissue ultrasound was performed, which proved to be normal. Due to the presence of thrill, we decided not to perform any type of biopsy.

At this point, almost all clinical hypotheses were excluded (Table 1).

Only the hypothesis of traumatic injury by the fixed orthodontic appliance used in the past remained. Due to the presence of thrill, the hypothesis of vascular injury was raised, following the presumed trauma. At that time, the new differential diagnosis included: pseudoaneurysm, true aneurysm, acquired arteriovenous fistula, cyst, abscess and hematoma.

| Clinical hypotheses | Exclusion criteria |
|--------------------------|---|
| Secondary syphilis stage | <ul style="list-style-type: none"> - Complete resolution of the primary stage, without apparent progression to the secondary stage - Absence of typical signs, such as diffuse symmetrical macular or papular rash or lymphadenopathy - Normal serological study |
| Gonorrhoea | <ul style="list-style-type: none"> - No changes in the genito-urinary system - Negative bacteriological examination |
| Tuberculosis | <ul style="list-style-type: none"> - No respiratory symptoms - All tests (normal chest X-ray, tuberculin test) were normal |
| Histoplasmosis | <ul style="list-style-type: none"> - Denied travel abroad - No respiratory or general symptoms (fever, malaise, asthenia, anorexia) - Normal chest X-ray |
| Herpes simplex | <ul style="list-style-type: none"> - Absence of multiple and recurrent injuries |
| Multiforme erythema | <ul style="list-style-type: none"> - Without self-limiting lesions and with target pattern - No trigger association |
| Crohn's disease | <ul style="list-style-type: none"> - Absence of gastrointestinal symptoms - Normal analytical study |
| Squamous cell carcinoma | <ul style="list-style-type: none"> - Incompatible time, evolution and macroscopic characteristics - Young patient |
| Recent traumatic injury | <ul style="list-style-type: none"> - There were no traumatic intraoral factors (cusps with sharp edges) |

Initial differential diagnosis

Table 1: Initial differential diagnosis based on the inclusion of pathologies characterized by the appearance of red and/or ulcerative lesions

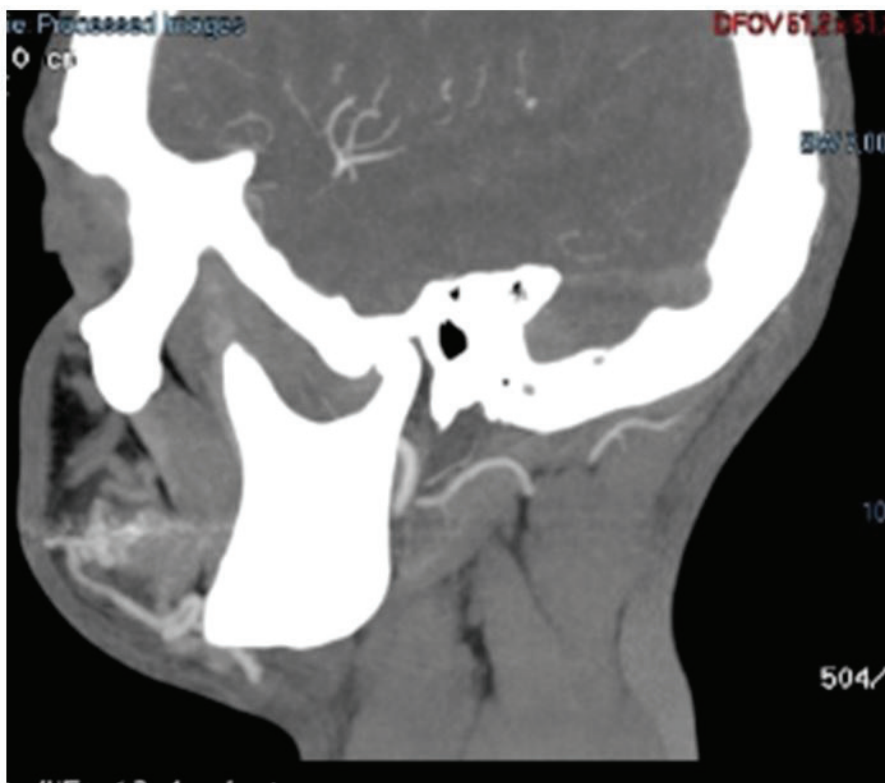


Figure 2: Angiography by CT maintained clinical suspicion of arteriovenous fistula. Vascular conglomerate with approximately 1.5-2 cm in the left hemiface

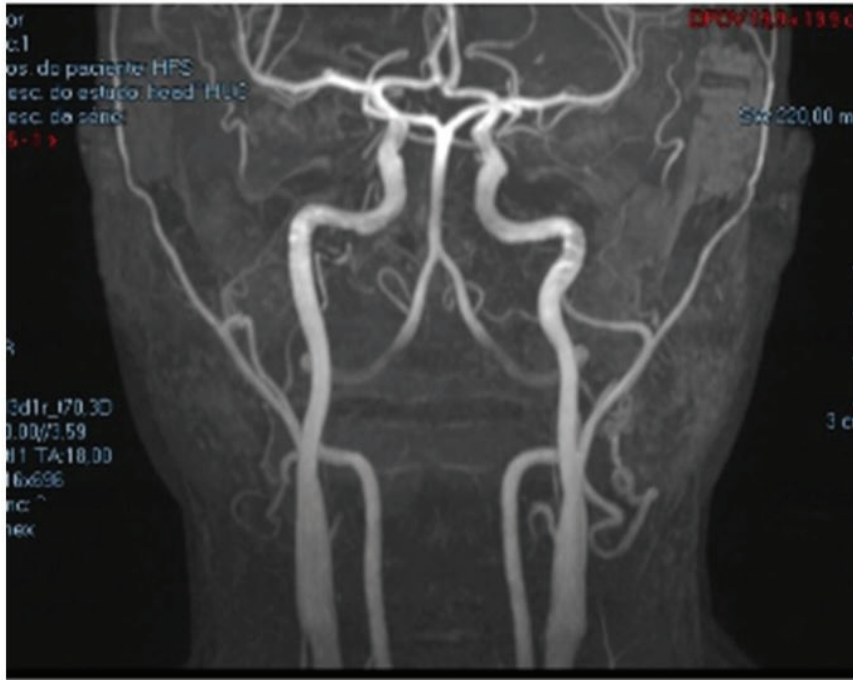


Figure 3: Angiography by MRI. Vascular conglomerate in the left hemiface

First, a doppler ultrasound was performed. The exam suggested 18 mm arteriovenous fistula. Then, the collaboration of the Neuroradiology Department was requested, which performed an angiography by Computed Tomography that revealed a vascular conglomerate with approximately 1.5-2 cm in the left hemiface, with arterial afferences through the facial/lower lip (Figure 2). The examination maintained clinical suspicion and an angiography by magnetic resonance imaging was performed, with confirmation of the diagnosis (Figure 3).



Figure 4: Selective Angiography from the facial artery of arteriovenous malformation nidus located in the upper jaw (arrow)

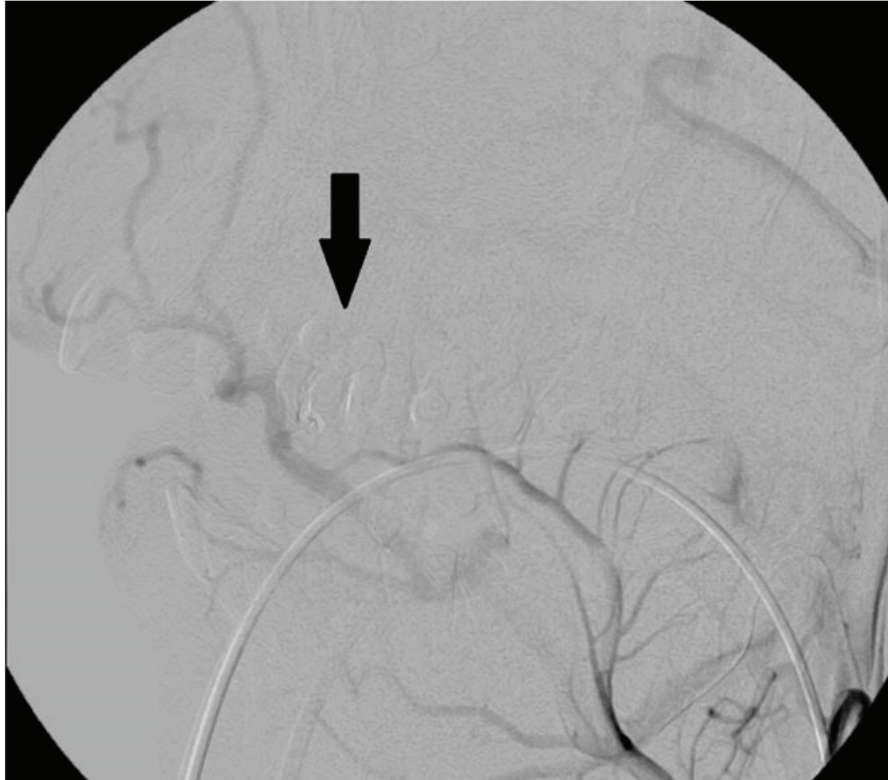


Figure 5: Postembolization selective angiography. Embolization exclusion of the fistula (arrow)

A selective Angiography was performed from the left facial artery of arteriovenous malformation nidus located in the upper jaw with Apollo Onyx Delivery Microcatheter (Ev3, Irvine, CA). The microcatheter was filled with dimethyl sulfoxide (DMSO) and the injection of ethylene-vinyl alcohol copolymer/Onyx® (Ev3, MA, USA) was continued, resulting in embolization exclusion of the fistula (Figures 4 and 5).

Four months after embolization, upon observation, the patient was asymptomatic, with significant reduction of the lesion and healing of the ulcerated lesion (Figures 6). The patient ended up leaving consultations, and further clinical follow-up was not possible.



Figure 6: Decrease of the lesion and healing of the ulcerated center

Discussion

Traumatic ulcers are very common in the oral cavity during orthodontic treatment and affect 76 to 81 % of patients, due to the repeated soft tissue trauma with arch wire, ligature wire, brackets and unsupported wire stretches [13,14]. Most cases correspond to minor and self-limited injuries, like erosions, contusions or desquamation; however, in a few cases involving severe traumatic ulcerations and with high duration and frequency, it can affect patients' quality of life [13,14]. In fact, several studies concluded that orthodontic appliances are one of the major causes of pain, discomfort and traumatic lesions concerning intraoral tissues [15]. Like any other type of right trauma, this iatrogenic injury of the oral mucosa has the potential to cause vascular injury, leading to an arteriovenous fistula [13-15].

In most cases, arteriovenous fistula does not resolve spontaneously [2]. An early diagnosis and an appropriate repair lead to a significant reduction in complications [2,3,6].

Despite the fact that most arteriovenous fistulas are asymptomatic, when significant and involving essential vessels, they can cause symptoms of distal ischemia [7-10]. Furthermore, it can produce venous and cardiac failure, infection; therefore, its treatment is essential [6,7,10].

The goal of the treatment is to close the fistula or fistulous nidus, with preservation of the patency of the various vessels [12]. The surgical option using the traditional technique is reliable; however, it may not be easily feasible due to grossly distorted and edematous tissue plans [5-8].

Regarding the endovascular technique, it is minimally invasive and allows for the precise location of the fistula with elimination of the vascular lesion, maintaining the normal patency of essential vessels [6,7,9,11,12]. It may include vessel embolization using coils, balloons or glue or stent placement, depending on the location and caliber of the vessels involved [7,9-12]. Embolization is used in cases where non-essential vessels are involved and it can be used in conjunction with surgical resection [6,7,9,12].

Two additional advantages of the endovascular options are rapid recovery and decreased pain [6,9]. As for complications related to the procedure, stent misplacement can lead to dissection or rupture of the vessel, subacute thrombosis, and hyperplasia of the intima layer. Furthermore, embolization can cause ischemic complications [6,7,11,12].

Nevertheless, the fistula may persist [13]. This fact can be explained by the presence of micro shunts (difficult to observe in angiography) or by technical failure, because of an incomplete penetration of the embolic material [2,3,8,13]. In these cases, embolization can be repeated, or it can be associated with a surgical approach [2,3,6,8,12].

Conclusions

The literature has not yet described any clinical case of arteriovenous fistula in the sequence of a repeated trauma due to arch wire of orthodontic treatment, which makes this case relevant.

This patient's case was successful due to its multidisciplinary approach and the development of a systematized and wide differential diagnosis.

Endovascular embolization with Onyx (ethylene vinyl alcohol, ev3) once again proves to be safe and effective, as it has a gradual pattern of deposition, which occurs from the outside to inside. [12]. The injection can be stopped as many times as necessary during the procedure so as to assess the extent and success of embolization [12,16]. For these reasons, Onyx was approved in 2005 for preoperative embolization of arteriovenous fistula, and has been globally used in order to address multiple vascular lesions and tumors [16].

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Conflict of Interest

The authors report no conflict of interest.

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