

CURRENT CONCEPTS REVIEW

Enormous Articular Hemorrhage Following Arthroscopy, Total Joint Replacement and other Surgical Operations in Hemophilic Patients due to Arterial Pseudoaneurysms: Diagnosis and Treatment

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Abstract

Pseudoaneurysms in hemophilic patients are unusual. We must have a suspicion when the patient has suffered an arterial trauma (following arthroscopy, total joint replacement and other surgical operations). Pseudoaneurysms may take place in any anatomical zone where there has been trauma. The diagnosis must be verified by duplex ultrasonography (US), standard angiogram, computed tomography angiogram or magnetic resonance angiography. There are diverse alternatives for the management of arterial pseudoaneurysms. Small pseudoaneurysms can be solved with conservative noninterventional management. It includes outside pressing, US probe pressing or US-guided thrombin injections. In larger pseudoaneurysms, endovascular methods, such as coil embolization, are now preferred. If the aforesaid methods are unsuccessful, standard surgical management with simple ligation or arterial reconstruction must be carried out. We must suspect a pseudoaneurysm when, following an arterial trauma, there is severe bleeding that does not stop with appropriate management with intravenous injection of the insufficient coagulation factor. The diagnosis has to be verified by imaging. Endovascular methods, such as coil embolization, are now preferred by the majority of physicians. The diagnosis and management of an arterial pseudoaneurysm must be carried promptly to avert adverse events.

Level of evidence: III**Keywords:** Arterial embolization, Arterial pseudoaneurysms, Arthroscopy, Diagnosis, Hemophilia, Joint arthroplasty, Treatment**Introduction**

Hemophilia is an X-linked disease caused by the deficiency of clotting factors VIII (FVIII) or IX (FIX). It is associated with repetitive bleeding into articulations, muscles, soft tissues, and organs. A pseudoaneurysm is blood that concentrates between the two external layers of an artery, the tunica media, and the tunica adventitia. It is commonly due to a penetrating trauma to the artery that bleeds into a space between the two aforesaid layers, instead of out of it. Pseudoaneurysms can occur in any artery in the body.

Pseudoaneurysms are uncommon in hemophilic patients. We must have a suspicion when the patient has suffered an arterial trauma. Pseudoaneurysms can occur anywhere there is trauma. That is, they can be found after implantation of arterial catheters, dull trauma, or penetrating injury. The diagnosis must be verified by duplex ultrasonography (US), computed tomography (CT) angiogram or conventional angiogram. There are several options for the management of pseudoaneurysms. Arterial surgery was the management of choice in the

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past (surgical ligation with or without distal bypass), other less invasive treatments are now preferred, such as covered stenting, US probe compression, and US-guided thrombin injection (1). In the case of small pseudoaneurysms, conservative noninterventional treatment with external pressing can be effective. Customary surgical management with simple ligation or arterial reconstruction can be indicated in great pseudoaneurysms. Recently, endovascular methods, such as coil embolization, have also been described (2).

Prompt diagnosis and management of this adverse event are crucial. Endovascular technique is a good minimally invasive treatment alternative. If arterial embolization does not solve the pseudoaneurysm, open vascular surgery with surgical ligation with or without distal bypass must be carried out. Whichever the method, precise surgical hemostasis must be accomplished by injection of factor concentrate (recombinant or plasma-derived). The availability of activated prothrombin complex concentrates (APCCs) and recombinant factor VII (rFVIIa) has made it plausible to perform invasive procedures in hemophilic patients with high-titer inhibitors (1).

Pseudoaneurysms in the upper limbs

In 1978, Lokey et al. communicated a case report of a traumatic pseudoaneurysm in the hand of a patient with hemophilia following a puncture wound; 17 days later, surgical repair of the pseudoaneurysm and the digital nerve in the thumb was achieved and the problem was solved without complications (3).

In 1997, Fields et al. reported a child who suffered a pseudoaneurysm in the radial artery after an arterial perforation who was subsequently diagnosed with hemophilia. In this child, conservative treatment was chosen, with the pseudoaneurysm slowly decreasing in size (4).

In 2002, Harkin et al. presented a case of radial artery pseudoaneurysm in a 57-year-old hemophilic patient after radial artery cannulation. Surgical repair by means of ligation of the radial artery and resection of the pseudoaneurysm was performed, with a satisfactory result (5).

In 2014, Clerico et al. published the case of a 42-year-old hemophilic patient who had a painful ganglion where the radial pulse is taken. After infusion of an anti-fibrinolytic drug, arthroscopic drainage of the ganglion was performed. Two weeks later, the patient developed a pseudoaneurysm of the radial artery that required emergent intervention (6).

In 2014, Vora et al. reported the case of a 3-week old hemophilic child who presented an expanding mass on the left wrist after radial artery puncture. Doppler US demonstrated that the mass was a pseudoaneurysm of the left radial artery that was removed with primary end-to-end anastomosis (7).

In 2017, Ferreira et al. published the clinical case of a 35-year-old hemophilic patient who had a perforating trauma to the palmar side of his right hand that had occurred 3 weeks earlier and had been sutured at the local hospital. Since then, he

had noticed a pulsating inflammation in the palm, and paresthesia and reduced sensation in the index finger. A pseudoaneurysm of 2 x 3 cm was observed, partially thrombosed, and with probable origin in the palmar arch or in the common digital artery. Angiography showed patency of the palmar arch but without perfusion of the pseudoaneurysm; the duplex scan showed a complete thrombosis of the pseudoaneurysm. After 2 months of follow-up, the duplex scan was repeated and the repermeabilization of the pseudoaneurysm was verified. The patient was then treated with a Doppler-guided percutaneous thrombin injection. An immediate thrombosis of the lesion was found, without evidence of ischemic complications. The patient remained asymptomatic under clinical surveillance. In this particular case, given the risk of hemorrhage, a minimally invasive percutaneous treatment was chosen, achieving clinical and imaging success, and without any complications. Percutaneous treatment with an echo Doppler-guided thrombin injection can be an effective and safe treatment, especially in pseudoaneurysms associated with surgical risk factors (2).

In 2018, Thomas et al. reported two patients who had traumatic pseudoaneurysms of the ulnar artery. In 1 patient, the aneurysm was removed and the artery was ligated; in the other case the artery was grafted because of poor hand perfusion (8).

Pseudoaneurysms in the lower limbs

Arterial pseudoaneurysms of the lower limbs are rare. In the majority of patients, a common femoral artery pseudoaneurysm developing after percutaneous access and management with US-guided thrombin injection accomplishes a success percentage of approximately 98%. However, treatment of a pseudoaneurysm of the vessels of the distal part of the leg is more complicated and may need supplementary endovascular and/or surgical techniques (9).

In 2006, Kickuth et al. reported a 40-year-old hemophilic patient and high response inhibitors who presented with repetitive knee bleeding following bilateral total knee arthroplasty (TKA) despite appropriate hematological management. He had two events of intense postoperative articular hemorrhage needing extensive hematological treatment. Digital subtraction angiography revealed a pseudoaneurysm of the lower lateral geniculated artery, and arterial embolization was carried out with success (10).

In 2007, Kotwal et al. presented a case of pseudoaneurysm of the tibialis anterior artery found 10 days following ankle arthroscopy on a hemophilic patient. The diagnosis was verified with a duplex US scan. Evacuation of the hematoma, removal of the damaged segment of the artery and reconstruction with a reversed long saphenous vein interposition graft was performed. The patient recovered without problems after the second surgery (11).

In 2008, Sadat et al. presented the treatment of uncommon ruptured metachronous aneurysms in a hemophilic patient with FVIII inhibitor who

experienced endovascular and open operations (12). In 2010, Saarela et al. presented the case of a hemophilic patient in which a pseudoaneurysm of a synovial artery, 2.5 x 2 x 3 cm in size, was diagnosed after revision TKA and was treated with local thrombin injections. Three months after surgery, the problem had been solved and knee range of motion (ROM) was normal (13).

In 2010, Park et al. described the case of a woman with hemophilia who presented with repetitive hemarthroses 9 days following revision TKA. The hemarthrosis did not solve with hematological treatment. Five weeks after surgery, angiography detected a pseudoaneurysm of the left upper genicular artery. Percutaneous embolization was successful (14). In 2011, Kumar et al. reported the case of a 14-year-old hemophilic patient with a pelvic pseudoaneurysm that was successfully solved with coil embolization (15).

In 2011, Drew et al. reported two hemophilic patients with pseudoaneurysms that were managed with factor substitution and observation, without the need for more aggressive intervention (16).

Gopal et al presented a case of recalcitrant pseudoaneurysm affecting the distal peroneal artery that appeared following blunt trauma in a hemophilic patient. The patient failed US-guided thrombin injection and needed 2 coil embolization interventions to be resolved (9).

In 2014, Rodriguez-Merchan et al. presented the case of a 46-year-old hemophilic patient who suffered severe hemarthrosis 6 days after a TKA. After aspiration of the hemarthrosis by arthrocentesis (120 mL), a pseudoaneurysm was suspected. CT angiogram and digital subtraction arteriography verified the diagnosis of a pseudoaneurysm in an upper lateral geniculated artery. An embolization of the pseudoaneurysm was promptly carried out, utilizing a helical microcoil, with a satisfactory outcome (17). In 2016, Chamseddin et al. presented the case of a pseudoaneurysm of the tibialis anterior artery after ankle arthroscopy in a hemophilic patient (18).

Table 1 summarizes how we achieve the diagnosis of a pseudoaneurysm in hemophilia. Table 2 shows the various treatments that we can apply for a pseudoaneurysm in hemophilia. The diagnosis and management of pseudoaneurysms must be carried out promptly to avoid adverse events (excessive bleeding

Table 1. Diagnosis of an arterial pseudoaneurysm

Penetrating arterial injury, which then bleeds
Duplex ultrasonography
Conventional angiogram
Computed tomography (CT) angiogram
Magnetic resonance angiography (MRA)

Table 2. Treatment options of an arterial pseudoaneurysm

Noninterventional treatment with external compression
Ultrasound probe compression
Ultrasound-guided (percutaneous Doppler-guided) thrombin injections
Endovascular techniques such as coil embolization
Conventional surgical treatment with simple ligation and resection of the pseudoaneurysm or arterial reconstruction with a reversed long saphenous vein interposition graft

with secondary anemia or infection of the hematoma secondary to the bleeding, which can be catastrophic, especially if there is an orthopedic implant, as in the postoperative period of a TKA [Figure 1].

Conclusion

We must have a suspicion of an arterial pseudoaneurysm when, following arterial trauma (arthroscopy, total joint replacement, other surgical procedures), there is severe articular hemorrhage that does not respond to management with intravenous injection of the insufficient clotting factor. The diagnosis must be verified by imaging methods. The treatment can be more or less invasive, depending on the size of the pseudoaneurysm, although it is advisable to move from the least invasive (US probe compression) to the most aggressive (standard surgical management with simple ligation or arterial reconstruction), passing through more aggressive techniques such as US-guided thrombin injections and endovascular methods such as coil embolization. The majority of authors now prefer endovascular techniques. If not diagnosed and treated rapidly, pseudoaneurysms, in addition to producing excessive bleeding with consequent anemia, can increase the risk of infection in patients with hemophilia, especially those operated on for TKA. We cannot forget that blood is an excellent breeding ground for bacteria and that a prosthetic infection is a catastrophic complication in hemophilia that is difficult to solve satisfactorily.

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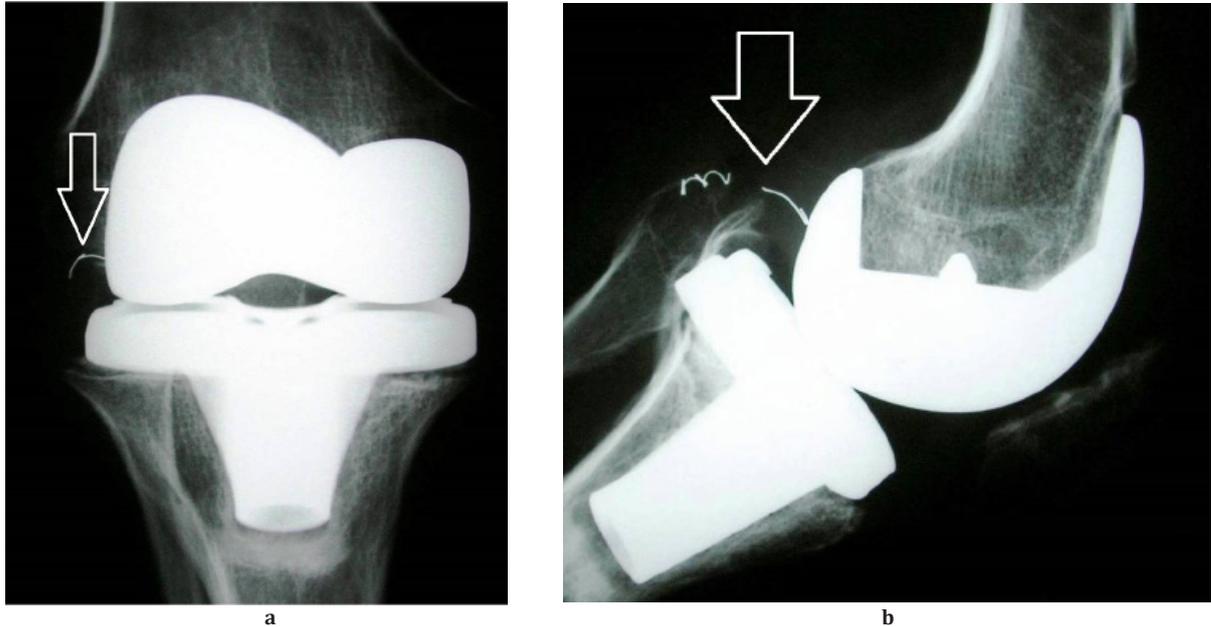


Figure 1 (a-b). Arterial pseudoaneurysm of a geniculate artery as an adverse event of total knee arthroplasty (TKA) in a hemophilic patient. In the postoperative stage there was continuous and uncontrolled bleeding in spite of adequate hematological management. Selective angiographic embolization solved the problem. Notice the vascular stent utilized for the arterial embolization as an arrow in part (a) of the figure, and as another arrow in part (b).

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