REVIEW ARTICLE

Systemic review of global case reports on ankle pseudo aneurysm: Analysis of epidemiology, clinical presentation, diagnosis and treatment

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ABSTRACT

Introduction: Pseudo aneurysm of the ankle is a rare presentation and the management is complex due to the complex anatomy. The aim of this review is to examine the epidemiology, etiology, diagnostic modalities used and management. This is the first systematic review of this topic in literature.

Methods: We performed a systematic review in multiple databases (including PubMed, MEDLINE, EMBASE, and Scopus) from 1966 to May 18, 2019, to identify all case reports and case series describing patients with ankle pseudo aneurysm. This systematic review was performed in accordance with the Preferred Reporting Items for a Systematic Review and Meta-analysis (PRISMA). Our inclusion criteria included patients with ankle pseudo aneurysm of any age. We excluded reports in which the individual level data is not available. Patients demographic (gender, age), clinical characteristics (precipitating event, duration), diagnostic modality and treatment were recorded.

Result: There were in total 23 case reports identified on ankle pseudo aneurysm from 1966 until 2018. Among these twenty-three reports, 16 (70%) were male and seven (30%) patients were female. Age distribution showed higher number of reports among young adults, 15 patients (65%). Based on our systematic review trauma (48%), arthroscope (48%) and arthrodesis (4%) were the etiologies described in all these case reports. Ultrasound duplex and CT Angiogram has been used as a single modality in three reports each. In fifteen patients (65%) combination of imaging has been used for diagnosis. Anterior tibial artery is the most commonly injured vessel among the reported cases, comprised of 14 (61%) patients. Among these arthroscopes were the highest reported precipitating events, 9 (64%), followed by trauma in four patients (29%) and arthrodesis in one patient (7%). Treatment modalities described in all previous reports were excision and ligation, 10 (42%); excision of sac and primary repair, 4 (17%); excision of sac followed by reversed saphenous venous graft repair, 2 (8%); US guided compression, 2 (8%); US guided thrombin injection, 4 (17%); stenting, 1 (4%) and coiling, 1 (4%).

Conclusion: Ankle pseudoaneurysm is mostly preventable by detailed initial assessment following trauma or careful approach during arthroscope. Evolving diagnostic modality and treatment has shed some light into noninvasive management of pseudo aneurysm of ankle.

INTRODUCTION

False aneurysm, also known as a pseudoaneurysm, is when there is a breach in the vessel wall such that blood leaks through the wall but is contained by the adventitia or surrounding perivascular soft tissue. A direct communication of blood flow exists between the vessel lumen and the aneurysm lumen through the hole in the vessel wall. The risk of rupture is higher than that of a true aneurysm of comparable size due to poor support of the aneurysm wall and thus false aneurysms generally require treatment.

Pseudoaneurysm involving the ankle is rare and can be caused by trauma, arthroscopy, and arthrodesis. The incidence of pseudoaneurysm of pseudoaneurysm following arthroscopy has been reported as 0.008%.[1] Pseudo aneurysm of this region of the body of the body poses a management dilemma considering the complexities of anatomy of the anatomy of the ankle. An ankle joint forms the articulation between the leg and foot and it's primarily a hinged synovial joint. The joint is crossed by posterior tibial artery medially, anterior tibial artery anteriorly and peroneal artery laterally. The vessels in this region are superficial and short coursed posing a greater risk for injury compared to vessels in the thigh or knee.

The presentation of a pseudoaneurysm is either early or delay depending on the severity of the injury, size of the defect and associated coagulopathies or connective tissue diseases. In a large pseudoaneurysm, the physical examination is sufficient to make a diagnosis for further treatment. In other cases, ultrasound duplex remains an easily available modality for diagnosis. The Yin-Yang sign is pathognomic of a pseudoaneurysm.[2] However, ultrasound duplex is not a reliable tool in a huge pseudoaneurysm. Angiography and computed tomography computed tomography ((CT)) angiogram has the advantage of being able to show the size of the defect and also delineate the foot arch. A complete foot arch ensures the viability of the foot after ligating the proximal artery. Treatment in these cases is divided into excision followed by ligation or revascularisation.

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Revascularisation is complicated by the complex anatomy of the ankle joint. Ligation is a time saving procedure and has less reported post op complications. A few other methods have been described in the literature such as thrombin glue injection, stenting and ultrasound guided compression.

OBJECTIVE

This review is to understand the epidemiology of ankle pseudoaneurysm and describe the aetiology, and various management option.

MATERIALS AND METHODS

We performed a world-wide systematic review in multiple databases (including PubMed, MEDLINE, EMBASE, and Scopus) from 1966 until March 18, 2019, to identify all case reports and case series describing patients with an ankle pseudoaneurysm in accordance with the Preferred Reporting Items for a Systematic Review and Meta-analysis (PRISMA). (Figure 1) Our inclusion criteria included patients with ankle pseudoaneurysm of all age groups. We excluded reports where data of where data of the individual was not available. Patient's demographics (gender, age), clinical characteristics (precipitating event, duration), diagnostic modality and treatment were assembled. The age groups were grouped thus were grouped thus: <13 years old (children); 13-19 (teenagers); 20-59 (adult) and > 60 and above (elderly).

Methodology

This review was undertaken and reported in accordance with the preferred reporting items for systematic review and meta-analyses (PRISMA) guidelines.

Eligibility criteria

For inclusion, the published cases must have had documentation of (i) site of the pseudoaneurysm, (ii) the predisposing factors, (iii) the method(s) of diagnosis, (iv) management strategies, and (v) outcomes of management outcomes of management ((patient)). There was no age limit or time limit.

Search strategies and information sources

A systematic search using search strategies, comprising keywords and MeSH, was carried out in Ovid MEDLINE and Ovid EMBASE. Searches were limited to studies involving human cases published in English without any time limit. Reference lists of relevant articles were also searched.

Study selection

Search strategies were implemented with initial results imported and merged into reference management software, ENDNOTE® (version X7, Clarivate Analytics (formerly Thomson Reuters), Philadelphia, PA, USA). After removal of duplicates, the remaining titles and abstracts were assessed for inclusion. Full texts of relevant articles were retrieved and independently assessed.

Data collection process and data items

Using standardised data extraction forms, data were extracted independently by two authors and compared. Discrepancies were discussed. Data extracted included the year of publication, patient demographics, precipitating factor, duration of illness, the centre of management, diagnosis (including methods of diagnosis, artery involved), treatment modalities and outcome.

Summary measures and statistical analysis

Data were summarised using descriptive statistics, with means and standard deviations for continuous variables and frequencies and percentages for dichotomous variables.

RESULTS

Publication characteristics

In total there were 26 case reports on ankle pseudo aneurysm on ankle pseudo aneurysm identified from 1966 until 2019. Three of the case reports were excluded due to no available full reports. The remaining 23 case reports describing only individual patients. (Table I)

Quality appraisal

The overall quality of the cases was good. Most cases reported an adequate description of the chief complaint, centres involved, laboratory and imaging modalities, and treatments (91.0%). Accurate diagnosis with valid and reliable outcomes measures was reported for all cases.

Demographic

Among the 23 reports, 16 (70%) were in in males and seven (30%) were females. Age distribution showed a higher number of reports among adults, 15 patients (65%). Ankle pseudoaneurysm has been reported only three times in children (9%) compared to four (18%) in the elderly. There has been only one case (5%) reported in the teenage group.

Aetiology and presentation

Based on our systematic review, trauma (48%), arthroscopy (48%) and arthrodesis (4%) were the aetiologies described in all these case reports. (Figure 2) There is no gender or age predilection as contributing factors for the aetiologies. The time of presentation of patients did not differ among the precipitating events. Eleven patients (48%) presented within a week to a month of the precipitating event. Early presentation within a week was documented in four patients (17%) and late presentation after a month was reported in eight patients (35%).

Diagnosis

Diagnostic modalities documented in these reports were US duplex, CT Angiogram, Catheter angiography, and magnetic resonance (MR) angiography. In two reports there was no imaging used because of large aneurysm and concern of an impending rupture. Ultrasound duplex and CT Angiogram was used as a single modality in three reports each. In 15 patients (65%) combination of imaging was used for diagnosis. Ultrasound (US) duplex was used as a single modality or combination in 18 patients.

Anatomy

The anterior tibial artery is the most commonly injured vessel among the reported cases, comprised of 14 (61%) patients. Among these arthroscopy procedures were the highest reported precipitating events, in nine patients in nine

Table I: Demographic description and management of case reports on ankle pseudoaneurysm

Author	Etiology	Age	Gender	Duration of illn	Vessel involved	Diagnostic modality	Treatment
Ichiro Tonogai et al (3)	Arthroscope	19	Male	2 weeks	ATA	US dupplex, Angiography	Excision and Ligation
Hugo T C Veger aet al. (4)	Trauma	66	Male	12 weeks	ATA	US Dupplex, MRI	Excision and Ligation
Clay P. Wiske et al. (5)	Arthroscope	53	Male	8 weeks	DPA	None	Excision and primary repair
Olubusola A Brimmo et al. (6)	Arthroscope	36	Male	6 weeks	ATA	MRI, US Dupplex, Angiography	US guided thrombin injection
Subhajeet Dey et al. (7)	Trauma	48	Male	8 weeks	PTA	US Dupplex, MRI	Excision and Ligation
Simon Craxford et al. (8)	Arthrodesis	84	Female	13 weeks	ATA	Angiography, US Dupplex	Stenting
Daniela Battisti et al. (9)	Arthroscope	66	Female	12 weeks	Peroneal artery	CTA	Excision and ligation
Ashok L Ramavath et al. (10)	Arthroscope	36	Female	3 weeks	ATA	US Dupplex	Excision and primary repair
Ronit Wollstein et al (11)	Trauma	6	Male	4 weeks	PTA	US Dupplex	Ultrasound compression
Sujata Aiyer et al. (12)	Trauma	36	Male	2 weeks	PTA	CTA	Excision and ligation
Edwards P1 et al. (13)	Trauma	60	Male	5 weeks	PTA	CTA	Excision and ligation
Ammar Darwish et al. (14)	Arthroscope	70	Female	6 weeks	ATA	None	Excision and Ligation
Rahul S. Kotwal et al. (15)	Arthroscope	20	Male	10 days	ATA	US Dupplex, Angiography	Excision and repair with reversed saphenous vein graft
Pier Paolo Mariani et al. (16)	Arthroscope	50	Female	1 weeks	ATA	US Dupplex, MRI	Excision and repair with reversed saphenous vein graft
D. O'Farrell et al. (17)	Arthroscope	30	Male	1 weeks	ATA	US Dupplex, Angiography	Excision and primary repair
J.L. Yu et al. (18)	Trauma	60	Male	4 weeks	ATA	US Dupplex	US compression followed by ligation
C. J. Salgado et al. (19)	Arthroscope	12	Female	4 weeks	ATA	US Dupplex, Angiography	Excision and Ligation
Michael J Ramdass et al. (20)	Trauma	20	Male	2 weeks	Peroneal artery	US Dupplex, MRI	Excision and Ligation
Ichiro Tonogai et al. (21)	Arthroscope	58	Male	4 weeks	Peroneal artery	CTA, US Dupplex	Excision and primary repair
M C A M Melenhorst et al. (22)	Trauma	50	Male	4 weeks	ATA	MRI, US Dupplex, Angiography	US guided thrombin injection
Conor D Marron et al. (23)	Trauma	25	Male	2 days	ATA	US Dupplex, Angiography	US Guided thrombin injection, followed by Excision and Ligation
Stéphanie Elens et al. (24)	Trauma	43	Female	1 day	ATA	US Dupplex, Angiography	US guided thrombin injection, followed by DSA coiling
Aimanan K et al. (37)	Trauma	6	Male	3 weeks	PTA	US Dupplex, CTA	Excision and Ligation

ATA : Anterior tibial artery, PTA: Posterior tibial artery, DPA: Dorsalis pedia artery, CTA: CT angiogram

patients (64%), followed by trauma in four patients (29%) and arthrodesis in one patient (7%).On the contrary, the precipitating event in all posterior tibial vessel injuries was trauma. The peroneal vessel was documented injured in three patients (13%) of which following an arthroscopy in two (67%) patients and trauma in one (33%) patient. Dorsalis pedis was involved in one patient (4%) following an arthroscopy. (Figure 3)

Treatment

Treatment modalities described in all previous reports were excision and ligation, 10 (43%); excision of sac and primary repair, four (17%); excision of sac followed by reversed saphenous venous graft repair, two (9%); ultrasound (US) guided compression, two (9%); US guided thrombin injection, four (17%); stenting, one (4%) and coiling, one (4%). Single modality was successful in 20 patients (87%), in three (13%) patients a second modality was used. Among the three patients, it failed in two patients following a US guided compression and another patient following a US guided thrombin injection.

There were in total four patients who underwent US guided thrombin injection as the initial intervention and all involving anterior tibial artery.[6,22-24] In two patients a second intervention was needed following the failure of thrombin injection.[23,24] Bovine based thrombin was used in these two patients in comparison to recombinant thrombin in the other two patients with successful outcome. Other than that the two patients who failed the initial thrombin injection also had a short history of pseudoaneurysm of less than a week. There were no variations in the size of the pseudoaneurysm among all these patients.

Two reports described US compression as the treatment method for ankle pseudoaneurysm.[11,18] In this group there is one failure of compression requiring excision and ligation.18 The patient with successful compression had a small pseudoaneurysm 10mm^2 compared to size 2cm in the patient where compression failed.

DISCUSSION

We present here a large contemporary systematic review of published cases on ankle pseudoaneurysm, providing global insight into diverse epidemiology, aetiology, presentation and a spectrum of management strategy for ankle pseudoaneurysm. The current review was undertaken and reported using the PRISMA guidelines, including all cases of ankle pseudoaneurysms.

Demographics

The main difference in the demographics of a true aneurysm and pseudoaneurysm is the affected age groups. A true aneurysm is known as an elderly's disease. Even though it's multifactorial the prominent cause is a degenerative disease. On the contrary, pseudoaneurysm has no age limits and it's always acquired. Our systemic review revealed that it occurred in the adult age group, especially in those who are active and susceptible to trauma or ankle surgeries. Occurrence in children is quite rare and occurred mostly due to trauma.[11,19] Since the anterior tibial artery is anteriorly located and most commonly injured during arthroscopy procedure, pseudoaneurysm of ATA is frequently encountered than PTA and peroneal.

Aetiology

Trauma and ankle procedures are the two most common precipitating factors for pseudoaneurysm development. In patients with involvement of distal posterior tibial artery pseudoaneurysm, almost all cases were caused by trauma.[7,11-13] Karthigesu et al., described in his reports that these patients could have been better managed on the initial presentation to avoid the formation of pseudoaneurysm.[37] Primary care physicians should be aware of the possibility of pseudoaneurysm at ankle following blunt trauma, so a detailed examination is important and the injured vessels should be repaired in the first instance.[37]

Ankle arthroscopy is a known precipitating factor for anterior tibial artery pseudoaneurysm. With a thorough understanding of the anatomy surrounding the ankle, most

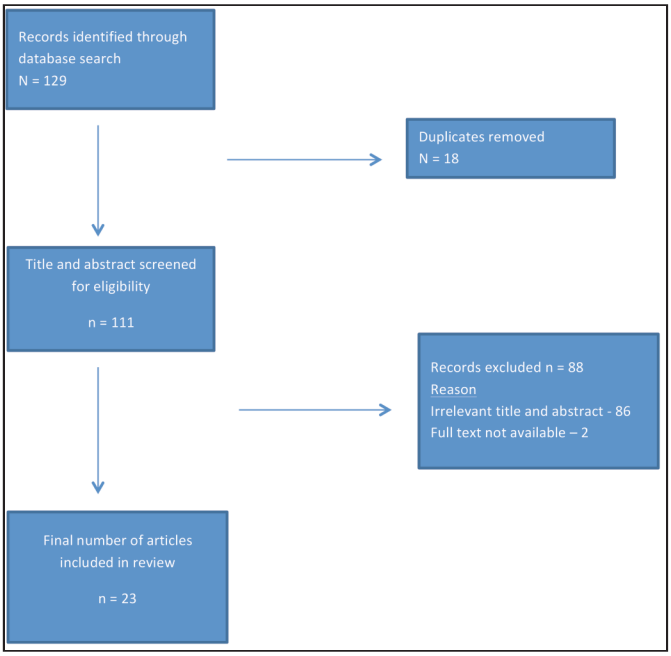

Fig. 1: PRISMA diagram depicting the case selection process.

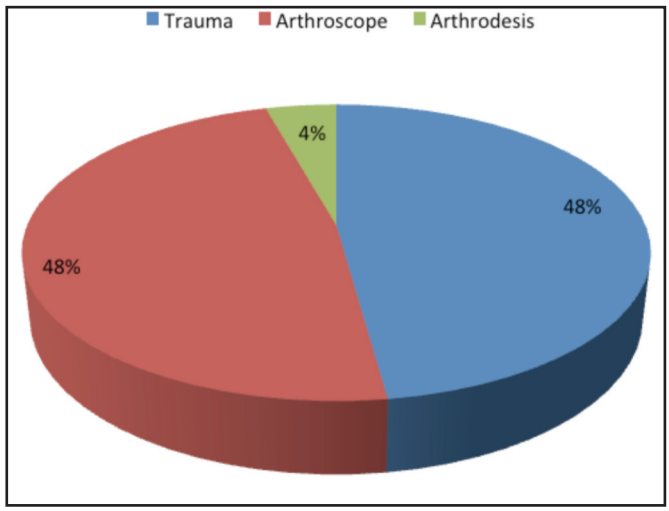

Fig. 2: Aetiology of pseudoaneurysm.

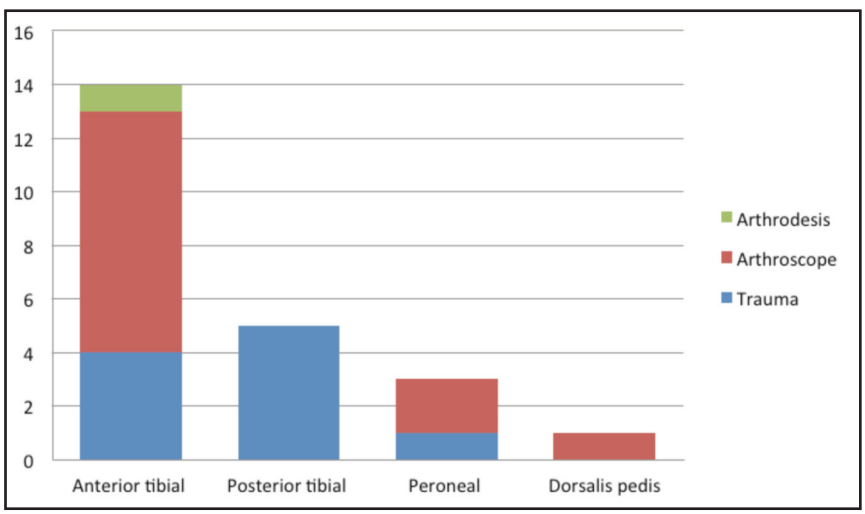

Fig. 3: Aetiologies according to the arteries involved.

arthroscopic complications can be avoided. Great attention to anatomical details must be exercised when portals are created and used, for it is then when complications are most likely to occur.[19] The anterior tibial artery runs deep to the superior and inferior retinaculum and is near the anterior ankle joint capsule at the level of the talar neck. The artery is most vulnerable to injury with the use of the anteroventral portal. However, injury to the artery has been reported with the use of the anteromedial and anterolateral approach.[25] Damage may be caused during portal insertion, instrumentation through the portals, or when exiting the capsule. Besides the technique of ports insertion, arthroscopists should bear in mind certain technical aspects of the procedure. Ofarrell et al., suggested that there are limits to the amount of talar neck exostosis that can be removed arthroscopically because some of the bony spurs are extracapsular and cannot be reached with the arthroscope.[17]

Physical examination and Role of imaging

A pulsatile mass in the ankle following an incident of trauma or previous ankle surgery is the usual presentation of an ankle pseudoaneurysm. However, in some cases, pulsations may be absent, too weak to be palpated, or masked by hematoma or thrombus formation. In such cases, making a diagnosis between aneurysmal lesions and other soft tissue masses becomes difficult. Thus, physical examination alone is not sufficient to diagnose a pseudoaneurysm.[26]

The role of imaging in an ankle pseudoaneurysm is to identify the affected artery, assess the size of the defect and ensuring a complete foot arch. These objectives can be achieved with Ultrasound duplex, CT angiogram, Catheter angiogram and MR angiogram as combinations or individually.

Duplex Doppler sonography is superior to other radiologic techniques because it is non-invasive, cheap, does not require the use of contrast agents, and is available at the bedside of patients. Coughlin et al., found that the pulsed wave Doppler method had a sensitivity of 94% and a specificity of 97% in the diagnosis of femoral artery pseudoaneurysms when compared with surgical findings.[27]

Colour Doppler sonography readily distinguishes pseudoaneurysms from other pulsatile masses because it permits the detection of a vascular mass connected to an artery by a neck or tract. During systole, anterograde flow passes into the pseudoaneurysm through its neck, and during diastole, the increased pressure in the pseudoaneurysm in relation to that in the underlying artery results in retrograde flow out of the pseudoaneurysm through its neck and back into the artery. This results in a characteristic "to-and-fro" pulsed Doppler pattern in the neck of the pseudoaneurysm and produces a characteristic "yin-yang" swirling colour flow pattern within its body.[28]

Although catheter angiography is the best method for detecting vascular pathologies, it is sometimes unable to differentiate an aneurysm from a pseudoaneurysm.[29] Based on our review it was never been used as a single modality in any previous reports on ankle pseudoaneurysm hence it's not ideal imaging for pseudoaneurysm. Besides, angiography is an invasive method, and patients are exposed to radiation.

Additional non-invasive diagnostic examinations such as contrast enhanced CT angiography or MR angiography may be performed to obtain a better diagnostic definition if information obtained by US duplex is insufficient. Both these modalities preferably used as an adjunct rather than the main investigation. Akita et al. described the use of MRI in a traumatic lateral plantar artery pseudoaneurysm secondary to a foreign body.[30] It was reported that a time-resolved contrast-enhanced 3D MR angiography is an accurate and effective modality in evaluating vascular pathology in the extremities.

Treatment

The most severe complication of an untreated pseudoaneurysm of the ankle is the rupture of this important vessel, given that the fibrous capsule of a pseudoaneurysm is devoid of the natural three-layered architecture of a true aneurysm and expands until it is confined by the limits of adjacent structures. This can be problematic, leading to haemorrhage into the soft tissue and hemodynamic instability, haemarthrosis of the ankle, and compartment syndrome in severe cases.[6]

The consensus on any pseudoaneurysm is conservative if thrombosed. However, none of the cases reported these patients were managed conservatively. These possibly due to imaging which showed an active contrast into the pseudoaneurysm sac.

Open repair is the standard method of treatment for false aneurysms of ankle pseudoaneurysm. Current treatment also includes non-surgical treatment approaches such as ultrasound-guided thrombin injection, ultrasound-guided compression, coated stents, and coil embolization. There are situations in which these methods may be either advantageous or disadvantageous. The choice of technique depends on various circumstances: the anatomical site and diameter of the false aneurysm, the patient's characteristics (general clinical condition, anticoagulant therapy, etc.), peripheral vascular bed, clinical complications (local pain, peripheral neurological symptoms, oedema due to venous compression).[31]

Excision of sac and ligation is the most commonly used technique for an ankle pseudoaneurysm as it is the least technically demanding. However, ensuring an intact foot arch is essential before proceeding with ligation to avoid ischaemia. In the event of incomplete foot arch excision followed by repair is paramount. Primary repair of pseudo aneurysm is an acceptable technique if the gap between the proximal and distal segments is not more than 3cm, otherwise interposition graft is an option. Patency of these vessels usually will be affected due to the smaller size. There were no reports on long term outcome in these patients following a primary or interposition repair to determine the exact benefit of this procedure.

There were key points highlighted in previous reports to improve the success rate of US guided compression such as combination of external compression with surgical stocking and repeated rounds of treatment.[32,38] Jang et al., suggested repeated rounds of compression may be an effective alternative to surgery for anterior tibial artery pseudoaneurysm in patients with an intact posterior tibial and plantar arch circulation without a coagulation deficit.[32] However, individual case selection is vital because compression is considered inappropriate for long standing injuries (i.e., >1 month).[33]

Ultrasound guided thrombin injections are relatively new and more commonly used in larger peripheral vessels such as the femoral artery, with reports of success rate in 60% up to 98%.[34] Its use in the distal arteries around the foot and ankle are less efficaciously supported, with some reports demonstrating positive results.[35] The recurrences following failure of this therapy usually detectable early and subsequent salvage therapy can be performed. There are two variants of thrombin available generally, human and bovine; bovine thrombin carries a small risk of inducing an allergic reaction as it is a foreign substance.[36] In this analysis bovine based thrombin associated with two unsuccessful outcomes involving anterior tibial artery pseudoaneurysm. Percutaneous endovascular coil embolization has been described in a handful of cases with good results, having the benefit of being performed along with the diagnostic angiography.

CONCLUSION

Ankle pseudoaneurysm is a preventable condition in many cases. Special anatomical consideration during ankle instrumentation and detailed assessment during initial trauma is essential for prevention. Newer approaches in surgery such as ultrasound guided thrombin injection and compression are widely used and show promising results.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest regarding the publication of this paper.

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