Case report

Successful surgical repair of an eminently rare case of an incidental idiopathic Common Femoral Artery Pseudoaneurysm in a 6-year-old female child - A Case Report

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ARTICLE INFO

Keywords:
Case Report
Idiopathic Pseudoaneurysm
Incidental Finding
Common Femoral Artery Pseudoaneurysm
Pediatric Vascular Emergency
False Aneurysms

ABSTRACT

Introduction and importance: A Pseudoaneurysm is an abnormal outpouching of the arterial wall which progressively enlarges and could lead to catastrophic consequences. Ensuing damage could culminate in the loss of the affected extremity due to dissection, exsanguination, thromboembolism, or infection. Some presentations are symptomatic, whereas others are incidental findings. Timely surgical mediation is vital to eliminate the potential morbid sequelae.

Case presentation: We present the case of a 6-year-old female, who was referred to the clinic due to an incidental finding of a pulsatile bulge in her right groin. We confirmed the presence of a visible pulsating bulge in the right groin associated with impalpable Popliteal and Pedal pulses. Preoperative imaging revealed an isolated Common Femoral Artery Pseudoaneurysm and hence, surgical intervention was successfully accomplished.

Clinical discussion: Surgical repair was achieved by pseudoaneurysmectomy and utilizing an autologous Saphenous Vein graft. From the proximal side, a primary end-to-end anastomosis was accomplished between the venous graft and the right Common Femoral Artery (CFA). Whereas from the distal side, a direct end-to-end anastomosis was completed between the right Superficial Femoral Artery and the veinous graft.

Conclusion: Arterial pseudoaneurysms constitute immensely rare vascular emergencies. The pediatric population is particularly vulnerable because of the rarity of occurrence of this pathology in children. Surgical intervention is the gold standard approach. Meticulous follow-up protocols ought to be carried-out to limit the possibility of recurrence. Documentation is the main building block in our profession. Physicians should possess high sense of clinical awareness when presented with such a pathology.

1. Introduction

A Pseudoaneurysm is a pathological entity defined as an outpouching of an arterial wall having a tight neck. It is commonly compared to a true aneurysm in that both diseases arise due to pathological irreversible dilations of arterial vessel lumens. Histologically, a Pseudoaneurysm has fibrous disorganized vessel wall components, while a true aneurysm is organized and involves the entirety of the three layers of the arterial wall [1].

Pseudoaneurysms are scarce diagnoses in the adult population and are even rarer occurrences in the pediatric age groups. The classical etiology for them is localized damage done to the arterial wall by an iatrogenic intervention, such as catheterizations to diagnose or treat a certain underlying condition. To a much lesser degree, pseudoaneurysms arise at several venipuncture sites (i.e., installment of arterial central access lines and after an impending trauma) [2].

The documented incidence rate of pseudoaneursms following such iatrogenic interventions is between 0.05 % and 1.2 % [3].

The chief complaint of presenting patients is classically a visible bulging pulsating compressible mass arising in a short period of time.

Abbreviations: CFA, Common Femoral Artery; DUS, Duplex Ultrasound; MSCT, Multi-Slice Computed Tomography; CTA, Computed Tomography Angiography; MRI, Magnetic Resonance Imaging.

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https://doi.org/10.1016/j.ijscr.2022.107362
Received 22 May 2022; Received in revised form 9 June 2022; Accepted 24 June 2022
Available online 27 June 2022
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Furthermore, it may be accompanied by pain, overlying skin changes, such as red discoloration, hotness and swelling. This culminates in misdiagnoses. The primary differentiating factor which results in the correct diagnosis is when such a mass is pulsating and has a vivid bruit upon auscultation [1].

On the other hand, there has been a degree of neglect to this rare pathology by being a profoundly rare diagnosis in the pediatric population. As a result, incidence and prevalence rates remain unknown and the resultant morbidity and mortality are thus far in need of documentation to alleviate the survivability for such a delicate population group [4].

The work has been reported in line with the SCARE criteria and the revised 2020 SCARE guidelines [5].

2. Presentation of case

2.1. Patient information

In this report, we demonstrate the unprecedented case of a 6-year-old Middle Eastern female child, who was referred to our university hospital’s Vascular Surgery clinic and brought by her parents from the Pediatrics Department. The chief complaint was the incidental finding of a pulsatile bulging in the patient’s right groin. The patient was regularly visiting a pediatrics hospital as she was treated for an infective endocarditis which was diagnosed 4 months prior to her referral to our clinic.

During her last visit to the Pediatrics clinic one day before her referral to us, to undergo regular treatment for her condition, her treating physician noticed a pulsatile bulge in her right groin. The bulge was painless as the patient did not complain from any pain upon palpation. Moreover, the patient did not report any “pins and needles” sensation in her ipsilateral extremity. Her parents reported documented palpation. Moreover, the patient did not report any sensations which revealed mild tachycardia, mild tachypnea, and a sub-coldness or bluish discoloration of the extremities was reported.

Her past medical history included an established diagnosis of infective endocarditis with marked vegetations, and Aortic Valve stenosis demonstrated on cardio-echogram four months ago. Blood cultures at the time of her diagnosis with infective endocarditis was done and yielded positive growth for methicillin sensitive Staphylococcus Aureus. As a result, the patient was being treated for infective endocarditis with the proper antibiotics. The Pediatrics Department used (Intravenous Oxacillin 200 mg/kg/day) every 6 h for 6 weeks. Since our patient weighed 20 kg, she was given 4000 mg per day. This means she was given 1000 mg of Oxacillin every 6 h for 6 weeks. In addition, intravenous Gentamycin 3-6 mg/kg/day every 8 h for 3–5 days. This means she was given 30 mg every 8 h for 5 days).

She was then referred to the Cardiac Surgery clinic for a consultation to resolve her underlying heart valve pathology.

Her family history, social history, allergic history, surgical history, and history of past similar complaints were all negative.

2.2. Clinical findings

We commenced our clinical examination via vital signs measurements which revealed mild tachycardia, mild tachypnea, and a sublingually measured low-grade fever. When inspecting the patient’s extremities, there was a vivid pulsatile bulging in the patient’s right groin. No erythema nor overlying skin discoloration and/or ulceration was noted. No peripheral cyanosis or pallor was seen. Via palpation, the patient’s right Femoral pulse was pounding. Nonetheless, neither her right Popliteal pulse nor her right Pedal arterial pulses were palpable. Moreover, a painless pulsatile bulging was examined, and it wasn’t characterized by any overlying palpable thrill.

The patient’s left lower extremity’s main arterial branches were all palpated and their pulses were within normal criteria. Via auscultation of the visible bulge, a soft bruit was demonstrated.

A conclusive laboratory panel was done, and the sole anomalies were a notable leukocytosis (white blood cell count was 21,000/μl) and a C-Reactive Protein value of 50 mg/dl. The rest of her investigations were all normal. We justified these laboratory values due to her previous infective endocarditis condition.

Prior to our surgical intervention, blood culture was done and yielded negative results of microbial growth.

2.3. Diagnostic assessment

Duplex Ultrasound (DUS) imaging demonstrated a frank pseudoaneurysm of the patient’s right Common Femoral Artery (CFA). Swirling arterial flow was vivid and the pathognomonic yin-yang sign was marked. Said pseudoaneurysm measured approximately (3.1 × 3.3 cm).

To fully visualize the presenting pathology and to meticulously depict its features, a Multi-Slice Computed Tomography (MSCT) of the patient’s lower extremity arterial tree was accomplished and established a right CFA Pseudoaneurysm accompanied by the occurrence of intramural thrombi formations. It was estimated to measure (3.4 × 3.6 cm) (Fig. 1).

Preoperative preparation, aside from a full blood panel, involved the administration of suitable preoperative intravenous antibiotics, in addition to blood analysis for crossmatch.

A noteworthy obstacle was the dearth of an interventional endovascular device in that hospital during the period of patient presentation.

2.4. Therapeutic intervention

Based on the merits of the clinical picture, urgent open surgical repair of the right CFA Pseudoaneurysm was deemed the modality of choice for treatment. The surgery was performed at our tertiary university hospital. It was seen through by a Vascular Surgery consultant and a senior first assistant with 17 years and 4 years of Vascular Surgery experience, respectively. General anesthesia was the modality of anesthesia used and it was carried-out without any perioperative complications. A longitudinal right groin incision was done to ensure adequate field of exposure. During the operation, our findings confirmed those of the preoperative imaging in that there was a right CFA Pseudoaneurysm measuring approximately (3.5 × 3.7 cm) with friable lumen (Fig. 2A).

Surgical repair was achieved by pseudoaneurysmectomy and applying an autologous Saphenous Veinous graft. From the proximal side, a primary end-to-end anastomosis was accomplished between the veinous graft and the right CFA. Whereas from the distal side, a direct end-to-end anastomosis was completed between the right Superficial Femoral Artery and the veinous graft. In addition, the Profunda Femoris Artery was anastomosed to said veinous graft via a distinct veinous graft segment (Fig. 2B).

A biopsy from the affected right Common Femoral Arterial wall was sent for histopathological and culture and sensitivity analyses. The rationale was to rule-out any infective or connective tissue etiology behind this presentation.

Histopathological analysis demonstrated an elastic fibrous vascular wall inclusive of several thrombi, in addition to microscopic mild suppurative inflammatory signs with mild calcifications and fibrosis (Fig. 3A-B).

Sensitivity and culture of the same specimen yielded a sterile result with no signs of microbial involvement. From a vascular surgery standpoint, postoperative physical examination results were immediately improved as the patient’s right extremity’s main arterial pulses were all palpable.

We administered anticoagulatives agents in the form of subcutaneous injections of Low Molecular Weight Heparin 1 mg/kg/every 12 h postoperatively while she was in the hospital. It was then bridged by...
Warfarin to keep the INR value between 2.0 and 3.0. Nevertheless, our patient had complete postoperative surgical recovery hence, she was discharged within 6 days of her operation. To aid in that endeavor, the patient was provided with several lifestyle aiding habits to help maintain adequate graft patency and help reach a full recovery. Said directives involved the integration of a balanced diet rich in vitamins and minerals far away from any complex carbohydrates. Furthermore, regular sterile wound dressings by a medical provider were done. In addition, suitable analgesics to help accommodate with any residual pain were prescribed. Nonetheless, we referred the patient back to the Pediatrics Department in conjuncture with the Cardiac Surgery Department for continued treatment of her cardiac situation. We followed-up our patient for two months now as she was assigned a specialized regimen of scheduled visits to our Vascular Surgery clinic, where thorough examinations were performed, and regular DUS imaging studies were done to ensure graft patency and adequate blood flow to the patient’s lower limb.

3. Discussion

According to the published literature, a Pseudoaneurysm is defined as a pulsating vascular cavity which is encircled by adventitia and vascular soft tissue. This cavity has an anatomical defect in its concomitant arterial vessel wall, via which, blood leaks through and maintains a pathological communication with the vascular system. Conventionally, it is perceived that such a pathology occurs following a poorly performed localized iatrogenic intervention and this is demarcated by an incidence rate estimated to be between 0.88 % and 8 % in the adult population [6,7].

With regards to lower extremities, it is astoundingly rare to witness an occurrence of a pseudoaneurysm of the arterial tree [8–11]. Furthermore, the scarcity of such a pathology in the pediatric age group is even more overwhelming. Moreover, we could find merely 4 cases reported in the literature about this pathology in said age population [12–15]. After reviewing the published research articles surrounding this pathology, we firmly believe that our case may be the first documented one of its kind. To the best of our knowledge, no previous similar case was documented.

Overall, peripheral arterial pseudoaneurysms are eminently rare [16]. Resembling true aneurysms, documented incidences have been categorized by percentage from high to low; Popliteal Artery (70 %), Femoral Artery (20 %), and the remaining 10 % were divided indiscriminately to different other arterial sites [16].
When we intend to classify the possible etiological conditions which could to the formation of pseudoaneurysms, the most prevalent underlying risk factors are localized trauma and different infections [17]. Nonetheless, pseudoaneurysms could have a congenital origin [18]. In addition, distinct connective tissue disorders could play a major role in weakening the vascular wall and result in such a pathology [19].

Diving deeper into the specifics of our case, CFA pseudoaneurysms majorly supervene upon an iatrogenic cause. Diagnostic and therapeutic cardiac catheterizations are prime examples [20,21].

There has been a multitude of other distinct etiologies for the development of such pseudoaneurysms, such as the installment of a large-bore catheter, the attempt to cannulate an artery and a vein in the ipsilateral side, the utilization of anticoagulation drugs, atherosclerotic blood vessels, high body mass index, short time manual compression after the removal of a cannulation sheath, Hypertension, and patients who are suffering from end-stage renal disease and who are put on hemodialysis [22].

The overwhelming scarcity of this pathology has resulted in lack of specifically-structured approach matrix [11].

Regardless of which, different diagnostic modalities for the establishment of an ironclad diagnosis of pseudoaneurysms exist. However, Doppler Ultrasound has been proposed by the latest guidelines and the published literature as the cornerstone diagnostic method for multiple reasons. Of which, we can mention its cost-effectiveness, readily available, and does not require or expose the patient to the usage of radiation nor anesthesia in the pediatric population. Nevertheless, we must not ignore the fact that it is ponderously operator dependent. Unfortunately, this could, at times, result in misdiagnoses. Otherwise, there are several other modalities, such as Magnetic Resonance Imaging (MRI), Computed Tomography Angiography (CTA), Magnetic Resonance Angiography, or Arteriography. Said complex diagnostic modalities enable physicians to accurately demarcate the pathological lesion in question, exclude embolic causes and diverse vascular anomalies [23–25].

As a conclusive standpoint, DUS is the current gold standard where it has an established sensitivity and specificity in detection of femoral arterial pseudoaneurysms of 94 and 97 % respectively [26]. Additionally, it can offer medical providers vital information, such as the diameter, anatomical neck of the lesion, and its overall morphological features.

In certain cases where we can still question the pathology at-hand, we can resort to the utilization of the previously mentioned complex diagnostic methods, such as CTA and MRI [8,9,11,27–29].

When discussing treatment approaches, there are a variety of proposed techniques to treat arterial pseudoaneurysms. However, in the pediatric age population, there are merely two recommended surgical approaches: Either we perform adequate excision of said pseudoaneurysm along with the affected arterial segment in addition to restoring arterial continuity via direct end-to-end vascular anastomosis, or we achieve complete vascular remodeling and reconstruction via venous autologous graft [30]. Current guidelines are in favor of performing arterial repair with direct end-to-end anastomosis as it is preferable on the premise that it won’t ensue any unwanted tension or tissue damage to the surrounding collateral blood vessels [30].

Conservative treatment approaches have been proposed but have proven their success in the adult population group rather than the pediatric one. These modalities include ultrasound-guided manual compression to obliterate the pseudo-aneurysmal cavity and localized thrombin injection. Those two methods are highly unrecommended in pediatrics due to the immaturity of the anatomical structures and the ensuing catastrophic complications which might occur [31].

Disease-specific complications of such a pathology include sudden rupture and acute limb ischemia which could lead to amputation. These complications have higher tendencies to occur in males rather than females in an estimated incidence of 63 %, whereas the average age where pseudoaneurysms could take place is 50 years of age [32].

4. Conclusion

We believe this is the first documented case of an incidental idiopathic CFA Pseudoaneurysm in a 6-year-old Middle Eastern female. Pseudoaneurysms constitute immense rare vascular emergencies. If not timely discovered, they lead to calamitous consequences. The pediatric population is particularly vulnerable because of the rarity of occurrence of such a pathology, and because it’s poorly studied in children. Surgical repair in a time-efficient manner diminishes the life-threatening complications, such as limb ischemia and amputation. Scrupulous follow-up protocols must take place to weaken the possibility of recurrence.

Documentation is the core building block in Medicine. Similar cases must be documented and applied in the context of contemporary clinical research to culminate in the better understanding this disease.

**Abbreviations**

| Abbreviation | Description |
|--------------|-------------|
| CFA          | common femoral artery |
| DUS          | Duplex ultrasound |
| MSCT         | Multi-slice computed tomography |
| CTA          | Computed tomography angiography |
| MRI          | Magnetic resonance imaging |

**Consent of patient**

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

**Availability of data and materials**

The datasets generated during and/or analyzed during the current study are not publicly available because the Data were obtained from the hospital computer-based in-house system. Data are available from the corresponding author upon reasonable request.

**Provenance and peer review**

Not commissioned, externally peer-reviewed.

**Ethical approval**

Institutional review board approval is not required for deidentified single case reports or histories based on institutional policies.

**Funding**

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

**Guarantor**

Omar Al Lham.

**Research registration number**

N/A.

**CRediT authorship contribution statement**

OA: Conceptualization, resources, who wrote, original drafted, edited, visualized, validated, literature review, and reviewed the manuscript.

AS: Resources, supervision, validation, and review of the manuscript.
OY: Vascular Surgery senior 1st assistant during the operation, supervision, project administration, and review of the manuscript.
HH: Vascular Surgery Specialist, who performed and supervised the operation, supervision, project administration, and review of the manuscript.
OA: The corresponding author who submitted the paper for publication.

All authors read and approved the final manuscript.

Declaration of competing interest
The authors declare that they have no competing interests.

Acknowledgements
Pathology Department at the Pediatrics University Hospital, Damascus, Syria.

References
[1] A. Namin, S.E. Starnes, C.M. Plikaitis, Pediatric craniofacial pseudoaneurysm with a variable history of antecedent trauma, J Craniofac Surg. 26 (3) (2015 May) 796–799, https://doi.org/10.1097/sce.0000000000001425.
[2] P.C. Guzzetta, Congenital and acquired aneurysmal disease, Semin. Pediatr. Surg. 3 (2) (1994 May) 97–102. PMID: 8062661.
[3] D. Landau, R. Schreiber, G. Szendro, L. Golcman, A. Klimov, C. Yefim, B. Johnatan, E. Avrahami, & M.R. Nehler, L.M. Taylor Jr., J.M. Porter, Iatrogenic vascular trauma, Semin. Vasc. Surg. 18 (2005) 212–223, https://doi.org/10.1053/j.semvascsurg.2005.09.008.
[4] J.G. O'Sullivan, S.A. Ray, J.L. Lewis, A.J. Lopez, B.W. Powell, A.H. Moss, J. A. Dormandy, A. M. Belli, T. M. Buckenham, A review of alternative approaches in the management of iatrogenic femoral pseudoaneurysms, Ann. R. Coll. Surg. Engl. 81 (1999) 226–234. PMID: 10615187.
[5] J. Woodley-Cook, M. Konieczny, M. Simons, The ulnar artery pseudoaneurysm, BMJ Case Rep. 2015 (2015), bcr2015212791, https://doi.org/10.1136/bcr-2015-212791.
[6] J.A. Spittel, Aneurysms of the hand and wrist, Med. Clin. N. Am. 42 (1958) 1007–1010, https://doi.org/10.1016/s0025-7125(16)34252-3.
[7] R. Sarkar, A.G. Coran, R.E. Cilley, Arterial aneurysms in children: clinicopathologic classification, J. Vasc. Surg. 13 (1991) 47–50. PMID: 1987369.
[8] B.F. Coughlin, et al., Peripheral pseudoaneurysm: evaluation with duplex US, Radiology 168 (1988) 339–342. PMID: 32393107.
[9] K. Winkler, E. Xenos, J. Lynch, Posttraumatic versus mycotic dorsalis pedis pseudoaneurysm, Int. J. Angiol. 22 (2) (2013) 135–136, https://doi.org/10.1016/j.ijanjo.2013.03.006.
[10] B. Ertuk, A. Ates, Post-traumatic dorsalis pedis pseudo-aneurysm caused by crush injury, EJVES Short Rep. 44 (2019) 29–32, https://doi.org/10.1016/j.ejves.2019.07.003.
[11] A. Lennox, M. Griffin, A. Nicolaides, A. Mansfield, Regarding, “Percutaneous ultrasound guided thrombin injection: a new method for treating post catheterization femoral pseudo aneurysm”, J. Vasc. Surg. 27 (1998) 1032–1038, https://doi.org/10.1016/s0741-5214(98)00441-2.
[12] U. Ferreira, J. Aragão, A. Lenik, I. Aragão, F. Aragão, W. Leão, C. Nunes, F. Reis, Aneurisma verdadeiro de arteria dorsal do pé: relato de caso, J. Vasc. Bras. 17 (2) (2018) 152–155, https://doi.org/10.1590/1677-5449.012817.