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Successful conservative management of a superficial pediatric pseudoaneurysm

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ABSTRACT

Purpose: (1) To report a case of successful non-operative management of a pediatric pseudoaneurysm in a patient with severe factor V deficiency. (2) To review the literature on pediatric pseudoaneurysms managed with a trial of conservative therapy.

Methods: A review of the literature was conducted on OVID-Medline for case reports or series of pediatric pseudoaneurysms managed conservatively (observation or external compression). Only superficial, radiologically confirmed pseudoaneurysms managed conservatively were included. Demographic data, etiology, treatment modalities, success rates, and definitive management for cases where conservative management failed were examined.

Results: Twelve case reports of 13 pseudoaneurysms met inclusion criteria. Including our case, the mean age was 7 years and the most common pseudoaneurysm etiology was iatrogenic (57%). Seventy-nine per cent (11/14) of pseudoaneurysms resolved with conservative management, and 21% (3/14) proceeded to surgery due to increasing size, bleeding, or pain. Of those pseudoaneurysms successfully treated conservatively, 18% (2/11) developed a complication. Five pseudoaneurysms occurred in patients with coagulopathies, all of which resolved conservatively.

Conclusion: In stable, asymptomatic superficial pediatric pseudoaneurysms, a trial of conservative management and close follow-up is a reasonable option, even in patients with coagulopathies.

1. Introduction

Pseudoaneurysms and true aneurysms are both dilatations of vessel lumens. They differ, however, by gross morphology and histology. A pseudoaneurysm is an arterial out-pouching with a narrow neck and a histologically fibrous and disorganized wall, whereas true aneurysms are fusiform and contain all three layers of vessel musculature [1]. Pseudoaneurysms are ubiquitously a complication of arterial trauma. Pathophysiologically, sharp or blunt trauma injures the vessel wall causing hemorrhage which is tamponaded by the surrounding soft tissue, forming a hematoma adjacent to the arterial defect. As the hematoma resolves, the interior is enzymatically resorbed while the exterior surface scars and forms a fibrous capsule. The most common presentation is a compressible soft tissue swelling at a site of previous trauma, either on an acute or subacute basis. There may be pain with overlying erythema, and this may be mistaken for an abscess. A pseudoaneurysm, however, will exhibit a palpable pulse and audible bruit on auscultation [1]. The most economic diagnostic test is Doppler ultrasound. An angiogram can be performed if there is remaining uncertainty.

Surgery has been the traditional management of pseudoaneurysms in order to avoid rupture or thrombosis, causing uncontrollable hemorrhage or distal ischemia. However, there has been a paradigm shift towards minimally invasive options, such as ultrasound guided thrombin injection [2]. There are also reports of spontaneously resolving pseudoaneurysms in adults and children [3,4]. However in the setting of patients with hemophilia or coagulopathy, the likelihood of spontaneous resolution is lower than for those without and there are very few reports of successful conservative management in these patients [5].

We present the first documented case of successful non-operative management of pediatric brachial artery pseudoaneurysm in a patient with severe factor V deficiency (factor V < 1%). There are an increasing number of reports advocating for the use of conservative management of pediatric pseudoaneurysms [4,6]. Thus, our second objective is to review the literature on pediatric pseudoaneurysms managed initially with a trial of conservative management and examine the success and complication rates.

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2. Methodology

A literature search of OVID-Medline was performed in April 2017 using the following medical subject headings and Boolean terms: "pediatrics or neonatology or perinatology or adolescent or child or infant" and "pseudoaneurysm or false aneurysm." Inclusion criteria were: (1) patient age 17 or younger, (2) superficial pseudoaneurysm in the extremities, torso, or extracranial head, and (3) patient underwent a trial of conservative management (defined as medical, observational, or compression bandage), and (4) radiographic evidence of pseudoaneurysm existed before treatment plan made. Exclusion criteria were: (1) patient age 18 or older, (2) pseudoaneurysm located in the neck, cranium, or viscera, (3) article written in a non-English language, (4) article unavailable for download, (5) patient lost to follow up or (6) invasive management (ultrasound guided compression, thrombin injection, interventional coiling, or surgery) was initiated at the time of radiographic diagnosis.

With regards to pseudoaneurysm outcome, documented clinical resolution was required to be defined as successful involution. When a patient proceeded to a more invasive treatment modality (eg. thrombin injection or surgery), this was considered a failed conservative trial. If the authors did not explicitly state a failed conservative trial but a period of 1 week or more elapsed from the time of diagnosis to invasive treatment (without having planned for the intervention electively), this was considered a failed conservative trial. Descriptive statistics were calculated. Demographics, pseudoaneurysm etiology, success rate and complications of conservative management, and definitive treatment in failed cases were examined.

3. Case presentation

A 1-month old female with a history of severe factor V deficiency (factor V < 1%) developed swelling within the right antecubital fossa after multiple attempts at venipuncture. Initial investigation with Doppler ultrasound revealed a hematoma within the biceps muscle. Repeat ultrasound four days later showed a stable hematoma but also revealed a 4.3 mm saccular bulge with arterial pulsations adjacent to the brachial artery, consistent with a pseudoaneurysm (Fig. 1). Repeat ultrasound one week later documented involution of the hematoma, but the pseudoaneurysm remained stable in size (3.8 × 2.2 × 2.3 mm).

Referral was made to the plastic surgery service for consideration of surgical management. On examination, the swelling in the right upper extremity had resolved, and patient was asymptomatic. The digits were pink with adequate capillary refill, and the brachial pulse was palpable. There were no joint restrictions, and median nerve function was normal. A discussion between the parents, medical, and surgical teams occurred to weigh the advantages and disadvantages of surgical intervention. Since the patient was clinically well and showed radiographic evidence of stability, the decision was made to manage conservatively. The parents were educated to monitor for signs of perfusion changes and rapid swelling in the area.

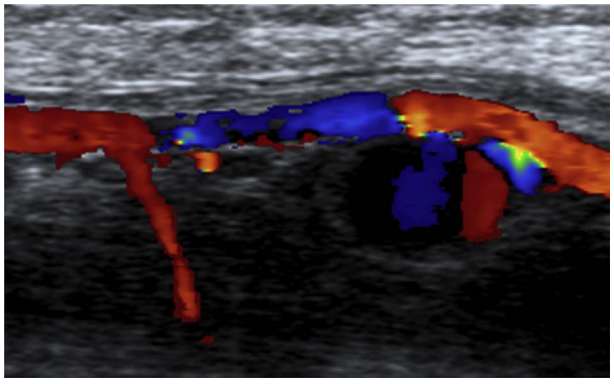

Fig. 1. Ultrasonographic view of the distal brachial artery showing an outpouching with classical "ying-yang" pseudoaneurysm appearance on Doppler view due to turbulent blood flow.

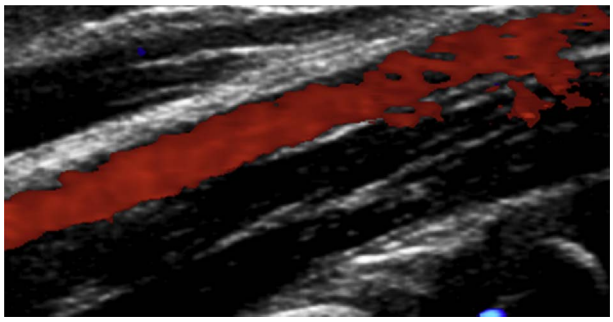

Fig. 2. Complete spontaneous resolution of the pseudoaneurysm two and a half years post-diagnosis.

The patient was initially followed biweekly, then every three months, and eventually every 6 months with serial ultrasound and clinical examination. Throughout this time, the patient remained clinically well with no evidence of skin changes, mass, pain, or vascular compromise. The size of the pseudoaneurysm remained constant at each ultrasound up until the 2 years and 6 month follow up after the initial visit, by which point it had clinically and radiologically completely resolved (Fig. 2).

Throughout the course of this pseudoaneurysm, the patient was followed concurrently by the pediatric plastic surgery and pediatric hematology service. She did not require medical therapy (such as factor replacement).

4. Results

Twelve case reports of 13 superficial pseudoaneurysms met the inclusion criteria [4,6–16] (Fig. 3, Tables 1 and 2). We included our case in the calculation of descriptive statistics for a total of 14 cases (n = 14, 7 males, 5 females, and two patients with unspecified gender). The mean age was 7 years (SD ± 6 years), ranging from 7 days to 16 years old. Twenty-one per cent (3/14) of cases initially managed conservatively required escalation of treatment due to increasing size, pain, or bleeding. One case of a patient with hemophilia A was initially managed conservatively, then went on to ultrasound-guided external compression (ie. transient compression with the ultrasound probe) due to persistent size [10]. Ultrasound guided compression failed but the lesion subsequently resolved spontaneously. This case was classified as successful conservative management. Seventy-nine per cent (11/14) of cases resolved with conservative management. Five of 14 cases (including our own) had concomitant coagulopathy [4,10,11] (n = 4 hemophilia A (1 severe, 1 mild, 2 unspecified severity), n = 1 severe factor V deficiency), and all resolved with conservative management.

Two of eleven patients (18%) treated successfully suffered a complication as a result of conservative management (excluding those patients who proceeded to invasive treatment, Table 1). The first patient [4] had a radial artery pseudoaneurysm and developed skin necrosis due to the compression bandage, which then resolved with dressing changes. The second had a pseudoaneurysm of the cavernosal artery [13] and sustained painless priapism for one month, which did not require drainage and resolved spontaneously.

The most commonly involved artery was radial (n = 4), followed by brachial (n = 2), popliteal (n = 2), femoral (n = 1), superficial palmar arch (n = 1), and cavernosal (n = 1). Three cases had unspecified arterial source. The most common etiology was iatrogenic (57%, 8/14). There was inconsistent reporting of pseudoaneurysm size, time to resolution, and duration of follow up (Tables 1 and 2). For example, some articles reported size in two or three dimensions, others reported size of the aneurysmal neck, and others did not report any size.