Uncommon incidental pseudoaneurysm

Diagnostic and management challenges

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ABSTRACT

الأهداف: دراسة المشاهدات السريريه و الشعاعيه لمرضى يعانون من أم الدم الكاذبة العرضية غير الشائعة و التي لم تحدث نتيجة القسطرة، ولملاحظة هذه المشاهدات بالإضافة إلى طرق العلاج مع الإشارة جزئياً إلى أم الدم الكاذبة نتيجة القسطرة.

الطريقة: تم إجراء الدراسة بطريقة إسترجاعيه على 11 مريض، 8 ذكور و 3 أنثى – مستشفى جامعة الأردن – عمان – الأردن ما بين الفترة 2002 وحتى 2008. تم إجراء دراسة للصور الشعاعيه والتي التي تحتوي على التصوير الطبقي المحوري (DS)، والتصوير المقطعي (CT)، والتصوير العادي، والتصويربالرنين المغناطيسي (MRI) و التصوير الملون للأوعية الدموية (MRA).

النتائج: تحدث أم الدم الكاذبة أكثر عند الذكور اليافعين (63.6%) و بالأخص في الأطراف السفلى (36%) أما الأعراض السريريه فكانت ذات قيمة تشخيصيه في (27%) من الحالات. أدى التصوير الطبقي المحوري (DS) إلى التشخيص الفوري لأربعة مرضى من أصل ثمانية تم إجراء الفحص لهم. بينما فشل تشخيص الآفة المرضية في مريض من أصل خمسه مرضى بواسطة (CT). وفشل تخطيط الأوعية الدموية كذلك في مريض من أصل خمسة مرضى. أظهر MRA و MRI تشخيص الأم الكاذبه لمريضين فقط.

خاتمة: يجب التفكير في تشخيص أم الدم الكاذبة العرضية بعد أي مداخلات طبية أو أذيات نافذة أو غير نافذة وخاصة بعد مرور وقت على حدوثها. و إن الذكور الشباب هم أكثر عرضه من الإناث. وكما أن التصوير الطبقي المحوري أقل حساسية في التشخيص بينما CTذو دقة عالية عندما تكون الآفة في الشرايين الصغيرة و المتوسطة. أن فحص MRI و MRA ذو قيمة عالية في تشخيص أم الدم الكاذبة العرضية ويضاهي التصوير العادي.

Objectives: To analyze patients with uncommon incidental pseudoaneurysms, secondary to non-catheterization causes, and to discuss the peculiar clinical spectrum, and focus on some aspects of difference from post-catheterization pseudoaneurysms.

Methods: Eleven patients, 8 males and 3 females, were studied retrospectively in Jordan University Hospital, Amman, Jordan, between 2002-2008. Radiological studies performed included duplex sonography (DS), computed tomography (CT), conventional angiography, magnetic resonance imaging (MRI), and magnetic resonance angiography (MRA).

Results: Pseudoaneurysms were most commonly encountered in young males (63.6%), especially in the lower limb vessels (36%). Clinical findings were suggestive of pseudoaneurysms in 27% of our cases. Four out of the 8 DS scans showed the neck of pseudoaneurysms, and the "to and fro" waveform, the strongest indicators for pseudoaneurysms. Both CT with intravenous contrast and angiography failed to establish the diagnosis in one out of 5 cases. The MRI with MRA showed the pseudoaneurysms in 2 patients that underwent the scan.

Conclusion: Incidental pseudoaneurysms are considered following iatrogenic procedures, penetrating, or blunt traumas with variable delay time. Young healthy males are at increased risks, as opposed to elderly females with calcified vessels in post-catheterization cases. Duplex sonography is less sensitive in incidental than post-catheterization pseudoaneurysms. The CT scan with intravenous contrast has high accuracy in establishing the diagnosis in small, or medium sized pseudoaneurysms. The MRI and MRA are accurate valuable studies and comparable to conventional angiography.

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Pseudoaneurysm (PSA) or false aneurysm is a focal enlargement of the vascular lumen resulting from the disruption of one or more arterial wall layers. It ranges from a vascular space contained by media and adventitia, to a blood space contained only by surrounding soft tissues. Clinical presentation vary, from silent to symptomatic patients mostly due to possible complications, which include rupture, distal embolization, local pain, neuropathy, or local skin ischemia, with rupture being the most serious cause of morbidity, and carrying the risk of life threatening shock.[1,2] The PSA can affect virtually any artery, while some vessels are very common, and typical sites for PSA formation such as femoral artery, other vessels are involved only very rarely. Several factors play a role in the frequency of affection of vessels with PSA, that includes anatomical aspects in terms of vessels that lack protection from surrounding soft tissues and underlying bony structures,[3] vessels that are frequently used as access sites for common diagnostic and interventional procedures, and the use of antiplatelets and anticoagulants.[1] The introduction of modern imaging techniques increase rates of PSA detection, although a large number are asymptomatic, and discovered incidentally, with regard to the cause, large main group includes PSA's in the setting of planned arterial punctures or catheterization, whether diagnostic, therapeutic, or related to hemodialysis. Bearing in mind the large number of diagnostic catheterizations carried out world wide, and the massive increase in the number of interventional procedures, this group is very common nowadays, and represents a significant burden. The other much less common group includes PSA's secondary to incidental arterial injury, either due to iatrogenic procedures such as biopsy, drainage catheter, or orthopedic interventions, penetrating injuries, infections, or the least common, blunt trauma. Due to the rare nature of the second group, only a limited number of studies regarding its diagnosis and management were conducted, and when available, a limited number of patients were studied each time. History and physical examination play only a minor role in the diagnosis and confirmation of the presence of incidental uncommon PSA,[3,4] the diagnosis remains largely the task of imaging studies. Angiography was the mode of diagnosis until 1987 when Mitchell et al[5] reported the diagnosis of PSA with color Doppler ultrasound, and today, this is the diagnostic procedure of choice in post catheterization PSA's.[4] However, other radiological investigations are mandatory in many instances, especially in the uncommon incidental PSA group to confirm the diagnosis, define anatomical relations, or to determine the cause of PSA particularly when ultrasound fails to give accurate diagnosis. This retrospective study was conducted on 11 patients with PSA's secondary to incidental arterial trauma (not related to interventional arterial punctures) aiming at determining the role of various imaging techniques in their diagnosis and management. This study concentrated on the uncommon incidental PSA group with unusual location, the role of various imaging techniques in the diagnosis and management with a comparison between these, and the post catheterization group.

Methods. After obtaining the Institutional Review Board (IRB) approval from Jordan University Hospital, Amman, Jordan, the medical records and imaging studies of all patients referred to our institution between May 2002 and July 2008, who had final diagnosis of PSA, and underwent treatment for PSA formation were retrospectively reviewed. Imaging studies were evaluated by 3 experienced radiologists, and they excluded all post catheterization PSA's whether diagnostic, therapeutic, or related to hemodialysis access. The study group comprised 11 patients, 8 males and 3 females. Their ages ranged between 16 and 58 years with a mean age of 32 years. The site of PSA formation, relevant clinical data, and physical examination, cause of PSA, and time lag between insult and presentation are presented in **Table 1**. Clinical criteria included: history of swelling or pain in the anatomical region of PSA formation, signs of localized tender mass, audible bruit, palpable thrill or ecchymosis, in addition to any symptoms or signs suggestive of mass effect upon surrounding structures, such as arterial or venous obstruction or narrowing, and airway compromise, or neuropathy. Finally, a review was made for the presence or absence of potential complications of PSA's like; rupture, deteriorating organ function, or distal embolization. As for the imaging studies that the patients underwent, and which lead to the diagnosis of PSA, 8 of the patients underwent duplex sonography (DUS), whether as the first, or following other radiological investigations. Of these 8 cases, 3 patients does not require any further investigation, and were managed according to DUS results. As for the other 5, 2 of them proceeded into conventional angiography, one patient underwent angiography, and computed tomography (CT) scan as well, one underwent CT scan only, and the last patient underwent CT scan, magnetic resonance imaging (MRI), magnetic resonance angiography (MRA) and magnetic resonance venography (MRV) studies. Two patients were investigated directly, and only by contrast enhanced CT, while MRA and MRI were the sole diagnostic investigations carried out for the last patient. Ultrasound (US) reports and images were reviewed

for any presence of anechoic, or hypoechoic mass adjacent to the parent vessels, presence of a channel or neck communicating the mass with the parent vessel, swirling internal echoes on DUS yin-yang sign,[6] and by the presence of "to-and-fro" wave form,[7] and finally any other pertinent findings were seen. Visualization of the neck, and the "to-and-fro" wave form are the most specific US findings for PSA. The CT, MRI, and MRA scans were all evaluated for presence of fluid signal structures arising from a donor artery, after a contrast injection filling of the sac with contrast material and anatomical evaluation. Nine of the patients were managed surgically, as for the other 2 patients, one was managed using US guided compression, while the other using minimally invasive technique (stent graft insertion).

Results. The patient's clinical data are summarized in **Table 1**. Young males were most frequently diagnosed to have PSAs (63.6%), particularly in the lower limb vessels (36%). Vessels involved in PSA formation were: common carotid artery (CCA), subscapular artery, intramammary artery, intrarenal artery, splenic artery, intratesticular artery, superficial femoral artery, popliteal artery, posterior tibial artery, peroneal artery, and occipital artery. Swelling in the anatomical region of PSA was the most common clinical finding seen in 9 of the 11 patients (81.8%). Pain, ranging from dull pain to severe and unrelenting, was seen in 8 (72.7%) of the patients. Clinical data was strongly suggestive of PSA in only 3 (27.2%) patients in terms of palpable thrill and audible bruit. The time interval between the insult and presentation for PSA formation ranged between 12 hours in the case of intratesticular PSA, and 8 months in the case of occipital artery PSA. The etiology of PSA formation varied between iatrogenic in 3 patients (patients number 6, 8, & 9). Penetrating trauma in 3 patients (patients number 3, 4, & 10). Secondary to blunt trauma 3 patients (patients number 1, 5, & 11). Pancreatic disease (acute on top of chronic pancreatitis with pseudocyst [patient number 7]), and the last one was due to enlarging femoral osteochondroma, while self-induced PSA was not encountered in any of our patients. The DUS was performed in 8 of our patients: in 4 of them (50% [breast, testicular, popliteal and posterior tibial arteries]) (**Figure 1**), all ultrasound criteria for diagnosing PSA were present, and no further work up was carried out for 3 of those patients. For the fourth (patient number 2) further investigation was carried out to reveal the reason of PSA formation. As for the fifth patient (patient number 11), the sac was very large, and DUS did not succeed in showing either the neck of the PSA, or the "to-and-fro" wave form, therefore an angiography was conducted, and revealed the diagnosis. In CCA-PSA, duplex showed a cystic soft tissue neck mass engulfing CCA with a swirling flow, thereafter, CT scan showed the connecting neck between the cystic mass and CCA by filling the sac with

Table 1 - Patients' characteristics, site of pseudoaneurysm (PSA) formation, delay time, and pertinent signs and symptoms (N=11).

Patients number	Age	Gender	Site of PSA	Cause of PSA	Delay time*	Pertinent signs and symptoms	Systemic disease
1	58	F	Left breast	Falling from her bed	72 hours	Pain, ecchymosis, lump	Hypertension
2	16	M	Left popliteal artery	Osteochondroma	6 months	Progressive swelling, pain, mass, thrill, bruit	Diaphyseal aclasis
3	21	M	Right peroneal artery	Stab wound	4 months	Pain, swelling, redness	-
4	26	M	Left posterior tibial artery	Gunshot wound	2 weeks	Pain, swelling	-
5	24	M	Right intratesticular artery	Blunt trauma	12 hours	Pain, swelling, ecchymosis	-
6	40	M	Intrarenal artery	Transplanted kidney biopsy	24 hours	Dull pain, hematuria, high creatinine	End stage renal disease
7	35	M	Splenic artery	Acute on top of chronic pancreatitis	72 hours	Back pain, vomiting, hematemesis	Chronic pancreatitis
8	35	F	Right supscapular artery	Traumatic chest tube insertion	3 months	Painless mass	End stage renal disease
9	42	F	Right common carotid artery	Core needle biopsy	1 month	Change in voice, paralysis of left vocal cord, left 9^{th}, 10^{th}, 11^{th} cranial nerves	Tuberculosis
10	20	M	Left occipital artery	Stab wound	8 months	Pulsatile mass, thrill, bruit	-
11	48	M	Right SFA	Trauma	3 months	Pulsatile mass, lower limb ischemia	Burger's disease

*time between the start of swelling and diagnosis, PSA - pseudoaneurysm, SFA - superficial femoral artery

contrast media. As for intrarenal PSA, due to its small size it was seen as a small cystic lesion with transmitted pulsations only. Occipital artery PSA was masked on DUS and angiography by the presence of arteriovenous fistula (AVF), and was only seen during surgery. The DUS findings are analyzed in **Table 2**. A total of 5 patients underwent CT scanning, and in 3 of them CT was diagnostic (CCA, subscapular, and splenic artery), while the fourth patient with popliteal artery PSA, CT scan images revealed a large soft tissue mass with peripheral calcification adjacent to the lower femoral shaft known as osteochondroma, raising the suspicion of malignant transformation. For the fifth patient with the large distal superficial femoral artery PSA, contrast opacification was faint due to dilution and high flow volume giving the appearance of soft tissue mass lesion. The MRI and MRA were carried out in 2 patients (patients number 2, & 3) giving an accurate diagnosis. Five patients underwent angiography, in 4 of them; renal artery, subscapular artery (**Figure 2**), popliteal artery, and superficial femoral artery PSA were noted. The diagnosis of PSA was confirmed. The last patient with occipital artery PSA, the diagnosis of PSA was missed, due to large AVF and rapid venous shunting, and the diagnosis was confirmed only surgically. Nine patients were managed surgically, and the surgical options used included; venous grafts, venous patch, ligation, and repair. Superficial femoral artery PSA was managed surgically because angiography showed wide neck artery PSA, and one artery in distal run indicating a high risk patient, which is not suitable for endovascular treatment. Breast PSA was managed successfully by US guided compression, while renal PSA was managed using stent graft insertion. Follow up periods ranged

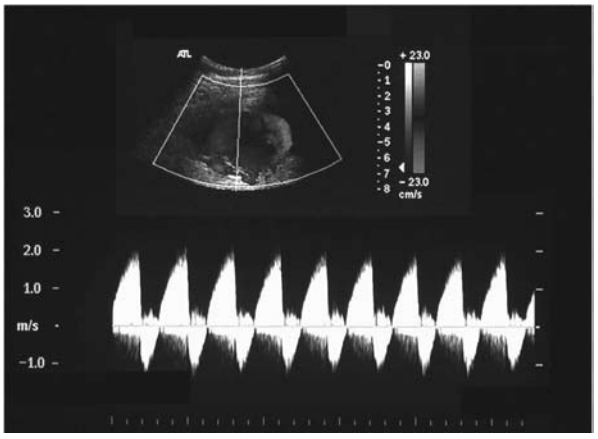

Figure 1 - Duplex ultrasound scan of left posterior tibial artery showing a cystic mass with "to-and-fro" waveform highly suggestive of pseudoaneurysm.

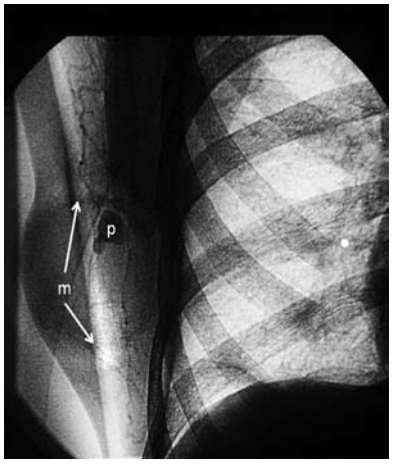

Figure 2 - Selective angiogram of right subscapular artery demonstrates pseudoaneurysmal sac (p) with the surrounding hematoma (m).

Table 2 - Duplex sonography findings in 8 patients.

Patient number	Site of PSA	Cystic mass	Neck of PSA	Swirling flow	To-and-fro wave form	Others	Other radiological studies
1	Breast	√	√	√	√	Surrounding hematoma	No further W/U
2	Popliteal artery	√	√	√	√		CT, MRI, MRA, MRV
5	Testicular artery	√	√	√	√	Enlarging hematoma	No further W/U
4	Posterior tibial artery	√	√	√	√		No further W/U
6	Renal artery	√	X	Pulsation	X	Ultrasound PSA	Angiography
9	Common carotid artery	√ irregular cystic mass	X	√	X	Soft tissue neck mass engulfing CCA	CT
10	Occipital artery	X	X	X	X	US suggestive of AVF	Angiography
11	Superficial femoral artery	√	X	√	X		Angiography

√ - present, X - absent, W/U - work up, PSA - pseudoaneurysm, CT - computed tomography, MRI - magnetic resonance imaging, MRV - magnetic resonance venography, CCA - common carotid artery, US - ultrasound, AVF - arteriovenous fistula

between one month and 3 years, and all patients were doing fine with no complications, or recurrence.

Discussion. Pseudoaneurysms result from disruption of the arterial wall continuity, whether due to planned or incidental punctures, with resultant formation of a blood filled space that is connected to the parent vessel by a neck, and is either contained by remnants of the vessel wall, or by the surrounding soft tissues. Femoral artery is the entry site in most catheterization and interventional procedures, and so it is by all means the most common vessel to develop PSA with different reported incidences varying between 1.1-7%.[8] In this study, uncommon PSA's discovered incidentally with unusual location and etiology were chosen to be analyzed. Although far less common than the planned arterial puncture group, there are still several vessels that are more frequently affected than others, either due to anatomical factors, or certain interventions.

Common carotid artery PSA's have been reported after carotid endarterectomies complicating approximately 0.3-0.47% of cases,[9] and after ear, nose, and throat infections, particularly in children.[10] Renal PSA's are reported to complicate 2-3% of biopsied kidneys.[11] Abdominal trauma, nephrostomy, nephroureterolithotomy, and nephron sparing surgeries are less common causes.[12] Seventy cases of popliteal artery PSA's were reported in the civilian settings, 40 of which are secondary to osteochondroma.[13-15] The peroneal artery is injured less frequently than popliteal and tibial arteries, due to its deep lying position in the axis of the lower extremity with PSA formation reported as a very rare complication.[16] Breast PSA's have been reported after core biopsies,[17] and the case presented in this study was the first reported after blunt trauma.[18] Subscapular, occipital, and testicular PSA's are very rare with only few case reports published worldwide.[19,20] The incidence of PSA in pancreatitis has been estimated at up to 10%, and with the splenic artery as the most common site.[21] Our patient was admitted with hemosuccus pancreaticus, which progressed to upper gastrointestinal bleeding, due to PSA bleed in the pseudocyst that ruptured into the stomach posteriorly, necessitating immediate surgical intervention. The diagnosis of PSA remains largely the role of imaging studies, such as angiography (noninvasive [CTA, MRA], or conventional] is required not only for confirmative diagnosis, but also for planning either operative, or percutaneous intervention.[22] Although conventional angiography is the gold standard for diagnosing PSA,[23,24] however, due to its invasive nature, it is reserved only for cases where other imaging modalities fail to establish the diagnosis, or when further details regarding the anatomical relations are needed. Recent reports suggest that angiography may fail to show PSA in the case of total thrombosis, or proximal arterial occlusion.[25] In this study, angiography failed to demonstrate a small PSA originating from occipital artery, which was masked by the presence of AVF, and the small PSA was discovered intraoperatively.

The DUS is the diagnostic procedure of choice with reported sensitivity of 94%, and specificity of 97% in post catheterization PSA's.[2,4] However, in non-post catheterization cases, and due to technical limitation in terms of deep vessels, small arterial branches, bony coverage, and gaseous reflection, especially in visceral organs, US has limited sensitivity.[26] In this study, it was found that US was less sensitive in this group than in the post catheterization group. Computed tomography is now used increasingly, especially with the advent of multislice scanners, and the ability to perform CT angiography, and post processing with 3D imaging. Reported sensitivity and specificity of CT angiography in detecting arterial injuries in the extremities ranged between 90-95.1% (sensitivity) and 98.7-100% (specificity).[27]

The MRI and MRA (**Figures 3a & 3b**) have been used successfully in diagnosing PSA, but due to long procedure time, high cost, and limited availability, these render it as not being a first line of investigation in PSA cases. However, MR studies are commonly encountered in cases where presentation lags far beyond the causative incidence, thus the clinician index of suspicion for PSA may be low, and other differentials like tumor, may be on top of the list. Several treatment options for PSA's

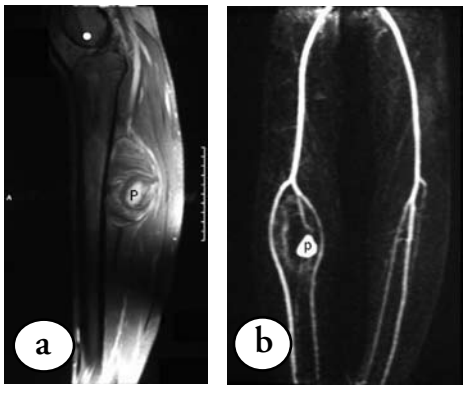

Figure 3 - Magnetic resonance (MR) imaging of the right leg of patient 3 showing **a)** Sagittal T2 fat saturation MR scan shows a pseudoaneurysm (p) arising from the right peroneal artery with surrounding concentric thrombosis. MR angiogram **b)** pseudoaneurysm (p) arising from the right peroneal artery.

including non-invasive; US guided compression, or US guided thrombin injection, and minimally invasive; covered stent insertion or coil, particles or liquid embolization, and conventional surgeries have been used, however, small PSA may resolve spontaneously.[22] The US guided thrombin injection, is now the treatment modality of choice in post catheterization PSA's with over 95% of patients having successful thrombosis of PSA after one, or 2 injections.[1,2,8,28] In this study, breast PSA was successfully occluded using this method.[18] In renal artery PSA, segmental arteries are most commonly involved though interlobar arteries, and rarely major division of the renal artery maybe involved.[22] Morrissey[29] suggested that renal artery lesions remain better suited to open, rather than endovascular repair, while Huppert et al[11] and Cohenpour et al[12] reported high success rates in treating renal PSA using endovascular techniques. Rich et al[30] suggested that blunt trauma leads to a late presentation of symptoms compared to penetrating, or iatrogenic traumas. However, in our study blunt trauma resulted in PSA's presenting earlier than PSA's secondary to penetrating, or iatrogenic causes. Post catheterization PSA's are known to be most common in females, other risk factors include anticoagulation, larger sheath size, therapeutic rather than diagnostic procedures, manual compression versus closure device, obesity, or calcified vessels.[8,26] In our study, we found that males are more frequently affected than females. A suggested explanation is that males are more frequently involved in violent acts and quarrels than females, and in war settings, the male gender preference rises dramatically. Most of our patients were healthy young adults, only 2 of our patients had renal failure due to adult polycystic kidney disease, and chronic membranous glomerulonephritis, one had hypertension, and one had multiple hereditary exostosis, chronic pancreatitis, and Burger's disease. This suggests that no significant risk factors could increase the possibility, and thus, direct us to the diagnosis of incidental PSA.

In conclusion, although this is a retrospective study with small number of patients, we conclude that young healthy males are the group most frequently affected, with the lower limb vessels most commonly involved. Pain and swelling were the most frequent presenting symptoms, and clinical examination proved to be of little value in suggesting the diagnosis, especially when there is a long time lag between the causative incidence and presentation, making the clinical index of suspicion towards PSA very low. Duplex sonography is less sensitive in incidental compared to post catheterization PSA, particularly in very large or very small PSA's, and in deep organs. Multi-slice CT with intravenous contrast has high accuracy in establishing the diagnosis in small or medium sized PSA's, but in large PSA's, contrast dilution may render the study non-informative. The MRI and MRA are very valuable studies with high accuracy comparable to conventional angiography. Future studies must include a larger sample size, and deal with pathology with a more specific non-invasive radiological criteria.

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