Rare Presentation of Isolated Pseudoaneurysms of the External Iliac Artery with Acute Appendicitis in a Type 1 Diabetic Patient - A Case Report

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INTRODUCTION

A 16-year-old female presented with pain in the right iliac fossa (RIF) and fever in diabetic ketoacidosis (DKA) with no significant findings consistent with a differential of co-existing aneurysms. Ultrasound of her abdomen was suggestive of likely sealed off perforation of the appendix with collection in the RIF. After resolution of her DKA, she underwent appendicectomy laparoscopically which was converted to open in view of dense adhesions. She developed per rectal bleeding post appendicectomy evaluation of which with computed tomography (CT) angiography was suggestive of two isolated right external iliac artery (EIA) aneurysms which were then treated with covered metallic stents.

While isolated aneurysm of the external iliac artery itself are extremely rare, the association of appendicitis with pseudoaneurysm of the EIA reinstates one of the mechanisms of formation of such aneurysms via direct extension of infection. Herein, we discuss the successful management of ruptured pseudoaneurysms of the right EIA in an operated case of acute appendicitis with type 1 diabetes mellitus.

First described by William Osler in 1885, mycotic pseudoaneurysm were termed "mycotic" for he found that in patients of infective endocarditis, the aortic arch aneurysms resembled fleshy fungal vegetations.[1] Pseudoaneurysm as a complication of appendicitis, is an extremely rare condition and delay in diagnosis results in sepsis, arterial rupture and death.[2-8] We present a similar case of right EIA pseudoaneurysm associated with acute appendicitis in a type 1 diabetic young female treated successfully with an endovascular covered stent.

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PRESENTATION OF CASE

A 16-year-old female presented to the emergency department with complaints of pain in the RIF and fever for a duration of 3 days. She had no history of abdominal intervention or surgery, or any history of trauma or arterial catheterization. On admission, she was febrile, tachycardic and tender in the right iliac fossa (RIF) but no signs of hypotension or pulsatile mass were elicited. No stigmata of any connective tissue or infective endocarditis disease were present.

Blood investigations were suggestive of elevated white blood cells (WBC) counts of 26000/mm cu and low potassium of 2.4 mEq/L. Her blood sugar levels were 449 mg/dL with ketones 3+ and HbA1c of 17.3 and she was diagnosed as a type 1 diabetic presenting with diabetic ketoacidosis. A provisional diagnosis of acute appendicitis in DKA was made clinically which was confirmed with ultrasound of the abdomen suggestive of possibility of a sealed of appendicular perforation.

After consultation with a paediatrician and paediatric surgeon, she was managed for the diabetic ketoacidosis in paediatric ICU and taken up for laparoscopic appendectomy. In view of dense adhesions, it was converted to open appendectomy. Intra-operatively, after separating the omentum from the appendix and the caecum, appendicular base and the anterior wall of the caecum showed partial thickness sloughing with whitish creamy wall. Appendectomy was done with drain placement in the RIF.

On post-operative day 5, she showed signs of surgical site infection which was culture positive for *Enterococcus faecium* and *rhizopus*. Injectable liposomal amphotericin B was started and rigorous irrigation of the wound was done.

On post-operative day 20, she developed an episode of large quantity per rectal bleeding and hypotension while straining for passing stools. She was evaluated with a CT angiography which was suggested of a collection of approximately 6 X 4.8 cm in the RIF involving and communicating with the caecum (Fig. 1). The right EIA showed two out pouchings which were approximately 5.8 cm and 7.1 cm from the bifurcation of the common iliac artery on its lateral aspect measuring 1.5 X 1 cm and 1.6 X 1 cm, consistent with a diagnosis of mycotic pseudoaneurysms with increasing size and active extravasations of contrast from the caudal one.

An interventional radiologist was consulted, and pelvis angiography stent grafting was done via an 8F left femoral artery access. Two stents were deployed (Covera plus 7 x 60 mm and Fluency 6 x 60 mm) in the right EIA across the diseased segment with exclusion of the aneurysm from the circulation with no residual extravasation. Post procedure patient was stable and had no further episodes of bleeding per rectally.

Blood cultures showed no growth of organism, bacterial or fungal. The patient received 4 weeks of liposomal amphotericin B for the treatment of surgical site infections (SSI).

Further recovery was uneventful. Histological specimen of the appendix was suggestive of inflammation without any discontinuity in the wall to confirm an absence of perforation or malignancy.

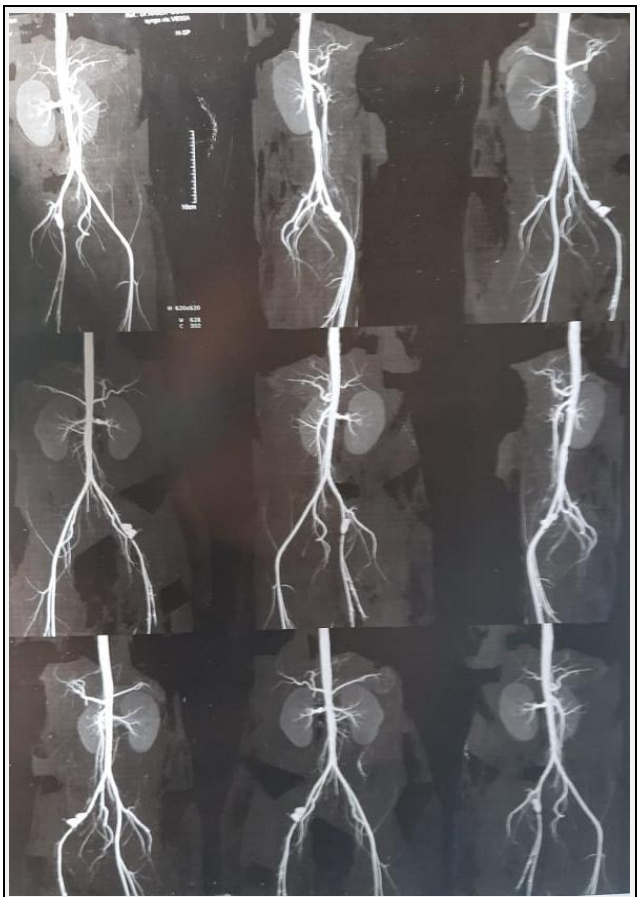

Figure 1. CT Angiography showing Right EIA with Two Outpouchings which are Approximately 5.8 cm and 7.1 cm from the Bifurcation of the Common Iliac Artery on its Lateral Aspect Measuring 1.5 X 1 cm and 1.6 X 1 cm

CLINICAL DIAGNOSIS

From the above sequence of events a likely diagnosis of mycotic aneurysms of EIA as a complication of acute appendicitis resulting from direct extension can be made.

DIFFERENTIAL DIAGNOSIS

The timing of the aneurysm becoming symptomatic after the event of acute appendicitis, in addition to the fact that they are isolated aneurysms of the EIA which is in close proximity to the inflamed appendix, likely suggests a provisional diagnosis of mycotic aneurysms as a complication of acute appendicitis.

Apart from above, the possibility of a collagen vascular disease being unmasked during an episode of acute appendicitis resulting in aneurysms of the EIA in the setting of an unknown type 1 diabetes, correlates with the genetic malfunction at play. Therefore, it is recommended to screen for the same in this case.

Another plausibility is pre-existing EIA aneurysms that became symptomatic in the advent of diabetic ketoacidosis with superadded subclinical bacteraemia in the setting of an acute episode of appendicitis.

PATHOLOGICAL DISCUSSION

Our case is unusual because multiple pseudoaneurysms arising from the EIA alone are extremely rare and are usually associated with trauma or non-degenerative causes but once they begin, they are extremely resistant to aneurysmal degeneration.[9,10,11]

Because most studied routes of inoculation of organisms include septic embolus (like from infective endocarditis) implanting bacteria over the intimal surface and a pre-existing vessel injury and super-added bacteraemia like in old individuals with atherosclerotic plaques, the extension of infection from local pathology such as appendicitis is rare but known.[12] The sudden development of an aneurysm in a toxic patient points towards the development of a mycotic aneurysm.

Albeit, the exact aetiology of these multiple aneurysms remains unclear. It seems highly unlikely that an occult source of systemic fungemia/bacteraemia would seed a localised part of right EIA without being detected in an earlier examination. Since the family and personal history did not reveal any similar history and given the patient's young age, infection in a pre-existing aneurysm seems unlikely as well. However, it seems plausible to say that an episode of acute appendicitis may have seeded the right EIA through direct extension from the collection in the RIF due to their close proximity to each other at the pelvic brim, given the immunosuppressed status of the patient.[13] Moreover, the probability of a missed external injury to the right EIA during appendicectomy should be considered in with the presence of intra-operative extensive adhesions.[14,15,16] Therefore, the aneurysm may be classified as Type I or Type IX based on the clinic-pathological classification proposed by Sarkar, et al.[17] Needless to say, the possibility of a pre-existing collagen vascular disease might have played a role as well.[18]

Isolated iliac artery aneurysms are frequently associated with gastro-intestinal symptoms, specially pre rectal bleeding and perforation.[19] However, in our case, the possibility of a stump blow-out with collection in the RIF with concurrent aneurysmal rupture into the RIF collection might explain the per rectal bleeding.

DISCUSSION OF MANAGEMENT

The primary goal of an aneurysm repair is to exclude the flow and remove wall tension to prevent expansion which might lead to rupture.[20] Endovascular repair (EVR) has overtaken open repair as they can be performed through an easily accessible site (femoral artery), eliminates the need for deep pelvis resection and achieve a faster control of bleeding in patient with hemodynamic compromise especially cases similar to ours.[15] But however, they do require long term follow ups and potential need for second operation.[21,22,23]

Since the size of the aneurysm in our case was small and the patient was haemodynamically compromised, EVR with covered stent was the appropriate choice of treatment.[24,25,26] The major disadvantage in a young age patient is the potential of future growth requiring a second revascularization procedure in adulthood. Therefore, annual surveillance would be recommended to look for endograft infection, subsequent pseudoaneurysms and blow out.

A major limitation of this case is that the nature of the aneurysm formation cannot be explained unless tissue analysis of the same is done, which might have been possible with open repair/ post-mortem autopsy. Therefore, the diagnosis remains probable based on clinical, biological and imaging parameter.

FINAL DIAGNOSIS

Pseudoaneurysms as a complication of acute appendicitis of the iliac arteries should be borne in mind while evaluating patients of appendicitis given the common occurrence of the latter in young individuals, moreover in patients who are type 1 diabetics and/or immunocompromised due to other ailments.

CONCLUSIONS

The discussed case might throw light on one of the modes of formation of a mycotic aneurysm, that is, via direct extension through local pathology.

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