Indian Journal of Surgery
https://doi.org/10.1007/s12262-024-04075-0

CASE REPORT

Recurrent Painless Haematuria in a Well Child—A Case Report

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Received: 1 January 2024 / Accepted: 6 April 2024

Abstract
We report a case of appendico-vesical fistula (AVF) in a 12-year-old boy presenting with a 2-month history of solely recurrent painless macroscopic haematuria. Ultrasound and MRI scans were suggestive of an urachus remnant with a calculus in the bladder dome. Cystoscopy showed a bladder diverticulum with mucosal inflammation. Open laparotomy eventually revealed the AVF, as a complication of a clinically 'silent' acute appendicitis. AVF is a rare complication of acute appendicitis. A literature review identified 17 further paediatric cases. Whilst faecaluria and pneumaturia are pathognomonic for AVF, these were present in only 24% of patients. Most patients presented with recurrent urinary tract infections or urinary symptoms, and most had a history of abdominal pain and vomiting. Imaging studies and cystoscopy were often inconclusive, and most diagnoses were made at surgery. A high index of clinical suspicion helps to guide diagnosis and treatment.

Keywords Appendicitis · Appendico-vesical fistula · Urinary infection · Case report

Case Report

A 12-year-old Afro-Caribbean boy presented to his local hospital emergency department with intermittent episodes of painless, macroscopic haematuria for 2 months. Colour varied from bright red blood to orange/tea coloured. He reported occasional clots in the morning and passage of frothy urine. He had no dysuria or frequency, was systemically well with no fever or abdominal pain, and had no history of recent foreign travel. He had a normal CRP and ESR, a hypochromic microcytosis with normal haemoglobin, and was known to have alpha thalassemia trait. Schistosomiasis was ruled out by urine and serology testing. He was treated for a presumed UTI (culture subsequently negative) and discharged with a follow-up ultrasound. The ultrasound showed thickening of the bladder wall at the dome and the presence of calculus, faecolith, or urachal remnant was suspected. With episodic gross haematuria persisting, he was referred to a paediatric urologist where he had a Magnetic Resonance Imaging (MRI) scan which was suggestive of an urachal remnant manifesting as a small tubular urachal diverticulum/cyst with a calculus (Fig. 1).

He underwent a cystoscopy which showed a bladder diverticulum in the upper right aspect of the dome of the bladder with mucosal inflammation around it (Fig. 2). Surgical exploration was done via a Pfannenstiel incision which confirmed an appendico-vesical fistula (AVF). It was resected, appendicectomy was done, and the bladder was repaired, without opening, in two layers. A urethral catheter was placed and removed on day 4. The boy made an uneventful recovery. Histopathology of the specimen showed features of acute appendicitis with perforation. Granulomas were present, and it was suggested to exclude Crohn's disease. He had a normal faecal calprotectin with no other features of Crohn's disease.

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Published online: 17 April 2024

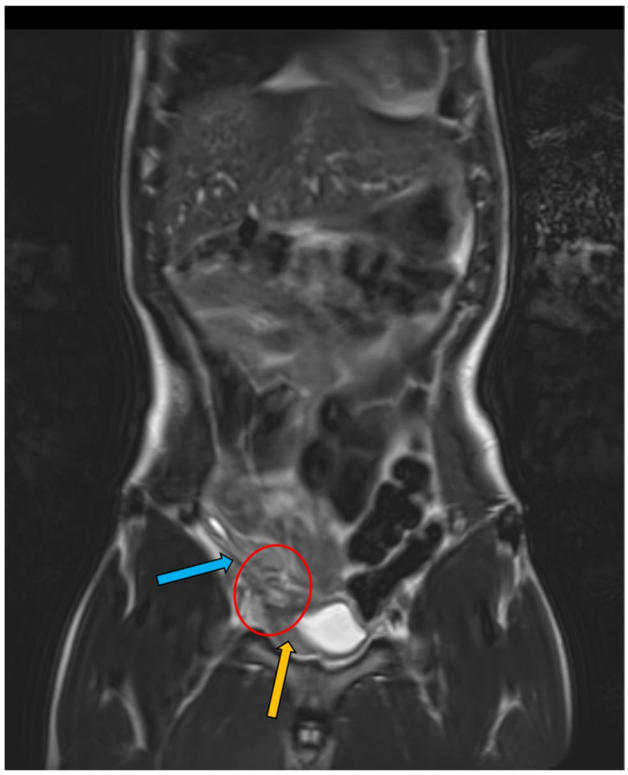

Fig. 1 Preoperative coronal T2 SPACE MRI image of the patient's abdomen showing the inflammatory mass containing what was pre-operatively thought to be a urachus remnant but was confirmed intra-operatively to be an appendico-vesicular fistula (red circle); the appendix (blue arrow) adjacent to the inflammatory mass and bladder, which displays a thickened wall (yellow arrow)

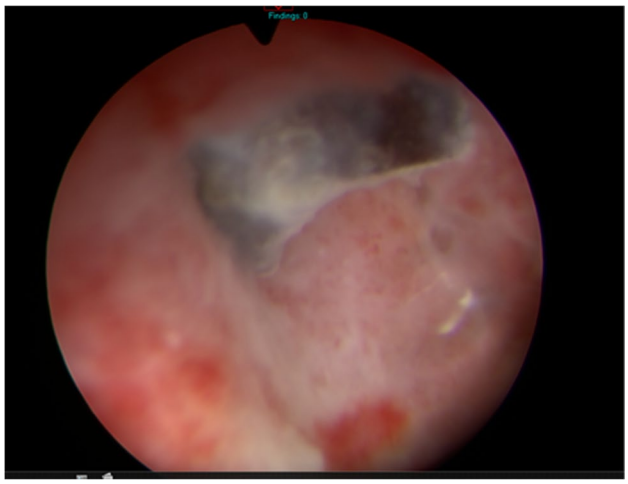

Fig. 2 Preoperative cystoscopy—bladder 'diverticulum' with inflamed mucosa

Discussion

Appendico-vesicular fistula (AVF) is an extremely rare complication of acute appendicitis in children. To our knowledge, this is the first case reported that presented with solely a painless gross haematuria, following a clinically 'silent' acute appendicitis.

AVF is believed to form when an inflamed appendix attaches to the bladder wall, and the ensuing abscess perforates into the bladder [1]. The passing of faecal matter (faecaluria) and intestinal gas (pneumaturia) are seen as pathognomonic for diagnosis [2]. AVF has been long known; a review from 1909 [3] cites 14 cases in children from the nineteenth century, reporting acute appendicitis, penetrating trauma, preceding bladder stone surgery, tuberculosis, malignancy, and anal atresia as underlying causes. A search of more recent literature (Table 1) identified 17 further paediatric cases aged 1–16 years (mean 10 years, median 11 years), 11 of them boys. Males are thought to be more susceptible for anatomical reasons (in females, the uterus prevents contact of the appendix with the bladder). In 12 cases (70%), the cause was acute appendicitis, and single cases each had cystic fibrosis, Hirschsprung's disease, Crohn's disease, previous bladder surgery, and blunt trauma (handlebar injury) as the underlying cause. Most children do have symptoms of acute appendicitis at some point of their illness, often identified in retrospect, with symptom duration between 1 week and 10 years (mean 14 months, median 3 months). Most frequent symptoms of AVF in the cases reviewed (Table 2) include those of recurrent urinary tract infection, abdominal pain, and vomiting (Table 2). Faecaluria and pneumaturia are uncommon (25%), in particular, if the AVF is blocked by a faecalith.

Conclusions

A high index of suspicion and a careful clinical history with a timeline of symptoms help to guide imaging and make the diagnosis. Imaging studies are often inconclusive. A urinary tract ultrasound may show abnormalities of the bladder wall or a faecolith; renal or intestinal contrast studies are often normal; occasionally, a late image in an intestinal contrast study may demonstrate contrast in the bladder. Abdominal CT or MRI studies may give better anatomical information. Cystoscopy may identify mucosal abnormalities which are commonly mistaken for an urachus remnant or polyp. Most definitive diagnoses are made at laparoscopy or laparotomy.

Table 1 Cases identified from the literature review

Reference	Age/sex	Duration of symptoms	Underlying cause
Nesbit (1934)	13 years, girl	10 years	Previous bladder stone operation
Cabot (1956)	5 years, boy	5 months	Acute appendicitis
Hyman 1959)	12 years, boy	8 months	Acute appendicitis
Fitzpatrick (1961)	15 years, boy	2 weeks	Acute appendicitis
Zvara (1964)	5 years, girl	6 weeks	Acute appendicitis
Zvara (1964)	2.5 years, girl	2 months	Acute appendicitis
Khan (1974)	15 years, boy	18 months	Acute appendicitis
Holmlund (1975)	11 years, boy	3 years	Acute appendicitis
Rizen (1976)	9 years, girl	3 months	Acute appendicitis
Dalessandri (1983)	15 years, boy	1 week	Handlebar injury
Yamamoto (1997)	21 months, boy	17 days	Acute appendicitis
Cakmak (1997)	16 years, girl	1 year	Cystic fibrosis
Izawa (1998)	15 years, boy	1 week	Acute appendicitis
Steinberg (1999)	16 years, boy	6 months	Crohn's disease
Roic (2003)	8 years, girl	2 weeks	Acute appendicitis
Lombay (2003)	12 years, boy	1 year	Acute appendicitis
Abubakar (2006)	1 year, boy	1 year	Hirschsprung's disease

References are supplied in supplementary file 1

Table 2 Frequency of symptoms from the literature review on paediatric cases of AVF

Symptom	Frequency *n* = /%
Dysuria/frequency/pyuria	14 (82%)
Abdominal pain	10 (59%)
Vomiting	5 (29%)
Gross haematuria	4 (24%)
Faecaluria	4 (24%)
Pneumaturia	3 (18%)
Fever	3 (18%)
Constipation	2 (12%)
Urinary retention	1 (6%)
Weight loss	1 (6%)

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s12262-024-04075-0.

Author Contribution All authors contributed equally to the concept, writing, and reviewing of this paper.

Data Availability As this is a case report no systematic data collection was undertaken. All data from the literature review are presented in tables 1 and 2 and were directly extracted from the references supplied in appendix 1. There are no other available data.

Declarations

Ethics Approval and Consent to Participate As written informed consent was obtained from the parents of the patient, no formal ethical approval was required by our institution (Newcastle upon Tyne Hospitals NHS Foundation Trust). This case report complies with the CARE guidelines.

Conflict of Interest The authors declare no competing interests.

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