CASE REPORT

Complicated appendicitis manifesting as bladder tumour in a child: A case report and literature review

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SUMMARY

Cases of complicated appendicitis have a spectrum of different clinical presentation, the array of which vary greatly depending on adjacent structures the inflamed appendix is in contact with. Urological manifestations are not uncommon. A wide array of symptoms; from urinary retention, obstructive uropathy with hydronephrosis, cystitis, appendicovesical fistula, perinephric abscess and even formation of inflammatory mass with contiguous structures bring an added risk of misdiagnosis should the physicians not be sufficiently diligent. We herein report a case of an eight-year-old girl who underwent bladder dome tumour laparotomy, with rhabdomyosarcoma being the suspected cause. Intra-operatively, the bladder dome tumour identified with the tip of the appendix. The omentum was adhering densely to the tip of the appendix with the fused structure forming a mass-like lesion. An en bloc excision of the bladder dome together with the appendix was done. A postoperative pathological examination found no evidence of malignancy and a diagnosis of chronic xanthogranulomatous inflammation sequela of perforated appendicitis was rendered. A later literature review of similar cases with bladder tumour like appearance suggests initial non-operative management with interval appendicectomy have lower complication rates.

INTRODUCTION

The appendix is often found in intimate proximity to the bladder. Appendiceal pathologies thus on frequent occasions present with urinary symptoms which maybe apparent singularly or in any combination of; urinary retention, obstructive uropathy with hydronephrosis, cystitis, appendicovesical fistula, perinephric abscess and formation of inflammatory mass with contiguous structures.[1-7] These urological manifestations quite often confound the examiner / investigator clouding radiological examinations and rendering results, inconclusive. Only subsequent laparoscopic examination and/or exploratory surgery can in the end provide for a confident and definitive diagnosis. We herein document in our report, an exceptionally rare case of appendicular phlegmon masquerading as bladder tumour in a child. It is a new complicated appendicitis case which we would like to document and share with fellow medical practitioners in the public domain.

A literature review of documented cases of complicated appendicitis in combination with radiological appearances consistent with bladder tumour served as a preamble to the writing of this report. Pertinent articles were collected and compiled. Data were sorted with respect to relevant parameters namely; gender, age, presentation duration, intraoperative finding, treatment given and histopathological examination observations were arranged in Table I.

CASE PRESENTATION

An eight-year-old girl with no known comorbidities, and no past surgical history was admitted on suspicion of having acute appendicitis. She presented with lower abdominal pain, dysuria and low grade fever for a duration of 2 weeks prior. Clinically a mass was palpable at the suprapubic region and ultrasound of the abdomen revealed a lobulated heterogeneous pelvic mass at the superior aspect of the bladder dome measuring approximately 5.0 x 6.4 x 4.6cm (AP x W x CC) as shown in Figure 1(a). Minimal vascularity was observed within. There was also focal irregular thickening of the adjacent urinary bladder wall. The appendix was obscured by overlying bowel gas. As such, proper visual examination by ultrasound was not possible. An abdominal computerized tomography (CT) revealed a well-defined, lobulated, heterogeneously enhancing solid lesion with internal hypodensities seen arising from the dome of the bladder as shown in Figure 1(b). It measured approximately 5.0 x 5.5 x 4.5 (AP x W x CC). No internal calcifications or fat density was identified. The appendix could not be clearly delineated. Laboratory biomarkers were non-specific. C-reactive protein (CRP) was raised (69.23mg/L), however white blood cell count was normal (7.5 x 10^9/L). Urine biochemistry showed ketone 1+ and protein 2+. Consequently, a provisional diagnosis of urinary bladder malignancy (likely rhabdomyosarcoma) was rendered. It was decided that a primary excision of what appears to be a tumour to be made intraoperatively. The medical team reasoned that a preoperative cystoscopic biopsy would have subjected the child to general anaesthesia twice in rapid succession and was judged to be unnecessary considering its functional limitations when used to obtain biopsy sample from muscular layer. The bladder dome tumour was identified with the tip of the appendix and the omentum

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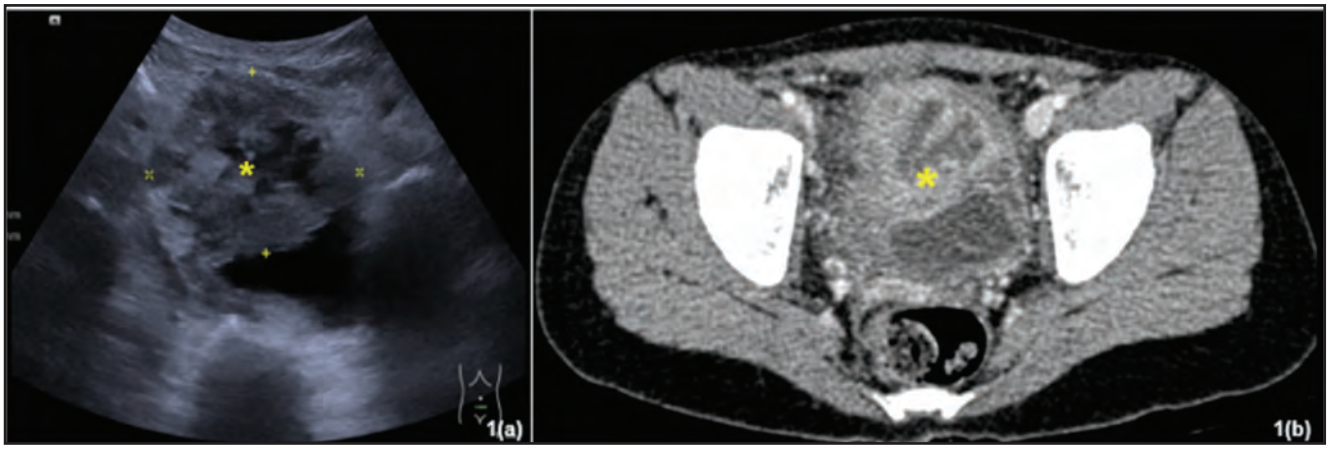

Fig. 1: Preoperative abdominal ultrasonography showing a pelvic mass at the superior aspect of the bladder dome (a) and the solid lesion was confirmed on the contrast enhanced computerized tomography of the abdomen (b)

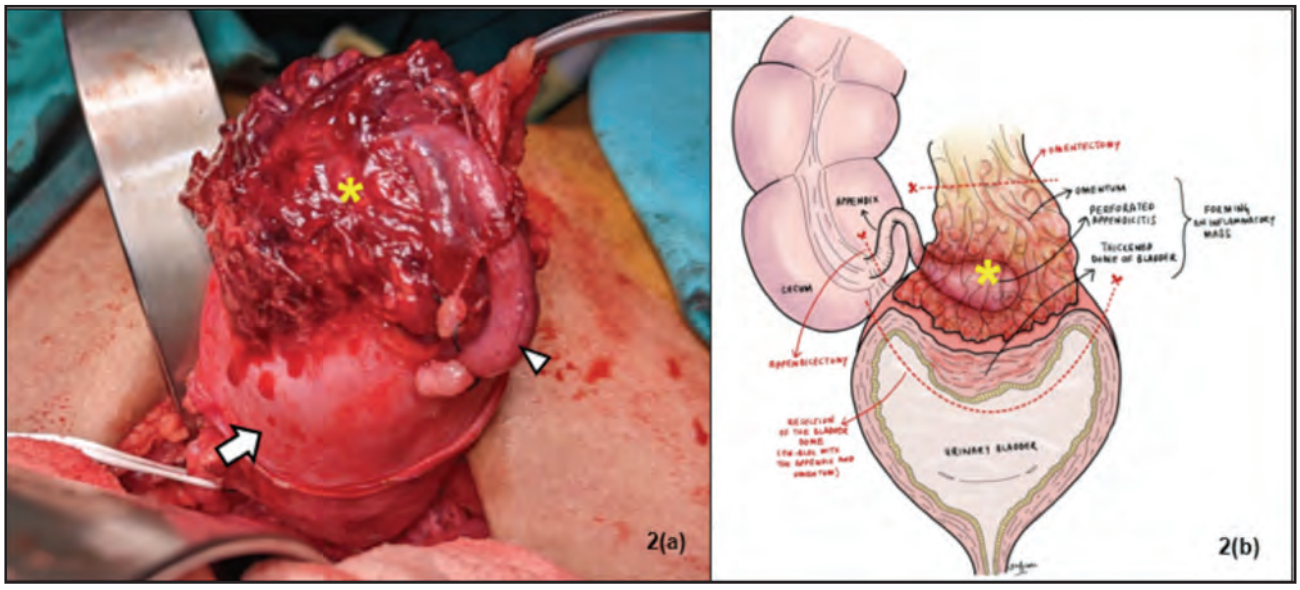

Fig. 2: Intraoperative photo (a) and the illustration (b) demonstrate the mass seen at the bladder dome (asterisk) with tip of appendix adhered within it (arrow head shows the appendiceal body and arrow is pointing to the bladder)

appeared to adhere to it forming a mass-like lesion as shown in Figure 2(a) and Figure 2(b). Otherwise, both ovaries appeared normal. Subsequently, an en bloc excision of the bladder tumour together with the appendix was performed. This was done to better facilitate histopathological examination as opposed to separating the mass from the bladder. Opting to perform the latter would have resulted in a much-reduced degree of confidence in the resulting histopathological interpretation. The bladder dome was excised and primary repaired in two layers. Postoperative recovery was uneventful. Oral feeding was initiated 1 day post-surgery and the patient discharged 1 week post operatively.

Histopathological examination revealed near total effacement and transmural large collections of foamy macrophages and histiocytes with abundant foamy to granular eosinophilic cytoplasm admixed with variable amounts of lymphocytes, plasma cells, eosinophils, and neutrophils representing xanthogranulomatous inflammation as shown in Figure 3(a), Figure 3(b) and Figure 3(c). There was no evidence of malignancy. The overlying bladder mucosa at the resection margin was lined by a benign urothelial epithelium. Focal surface erosion and subepithelial vascular congestion are observed. The adhered part of the appendix shows focal mucosal ulceration, perforation of the muscularis propria, and subjacent transmural collections of xanthogranulomatous inflammation as described above as shown in Figure 3(d). A pathological diagnosis of chronic xanthogranulomatous inflammation, a possible sequelae of perforated appendicitis was rendered.

Table I: A summary of available literature on bladder tumour as a complication of appendicitis

Author (year)	Age / Gender	Presentation	Duration of presentation	Investigation	Appendicitis Suspected	Intraoperative finding	Treatment	Histopathological examination
Our case (2023)	8 / Female	Abdominal pain, fever & dysuria	2 weeks	1) USG: lobulated heterogenous pelvic mass in continuity with the urinary bladder dome, possibly infected urachal cyst 2) CT: urinary bladder malignancy (rhabdomyosarcoma)	No	Bladder tumour arising from the dome of the bladder Tip of the appendix & omentum adhered to the tumour.	Excision of bladder tumour and appendicectomy	Operative specimen: Chronic xanthogranulomatous inflammation as sequelae of perforated appendicitis
Johal et al.[14] (2005)	27 / Female	Intermittent right iliac fossa pain, dysuria, anorexia & weight loss	5 months	1) USG: suggestive a bladder tumour. 2) Cystoscopy: erythematous urothelium with solid mass bulging into the bladder posteriorly 3) CT: mass from the right superior aspect of the bladder	No	Diagnostic laparoscopy: appendiceal phlegmon adherent to the bladder and omentum tethered in the pouch of Douglas	Systemic antimicrobial treatment and a repeat CT scan at 6 months revealed complete resolution of the phlegmon. No surgical intervention planned	Both cystoscopic biopsy and CT biopsy showed inflammation with no malignancy
Lombay et al.[15] (2003)	12 / Male	Gross haematuria & right lower quadrant abdominal pain	4 weeks	1) USG: bladder wall thickening & soft-tissue mass with central calcification 2) IVU: normal pelvicalyceal systems & ureters 3) CT: extravesical mass with central calcification	Yes	Inflammed small-bowel mass involving the bladder wall together with a perforated appendix	Appendicectomy	
Palnaes et al.16 (1991)	35 / Male	Recurrent gross haematuria & frequency	2 years	1) IVU: calcified density adjacent to the right side of the bladder 2) CT: a process located close to the bladder wall but without relation to the urinary tract 3) Cystoscopy: friable polypoid bleeding tumour at the right side on the bladder wall	No	4 X 4 cm large tumour between the cecum and the terminal ileum adherent to the bladder. Dissection revealed an abscess with a 2-cm long appendix stump and a 3 X 2 cm large fecalith	Appendicectomy	1) Cystoscopic biopsy: chronic inflammation and glandular metaplasia with dilated cystic glands of colonic type without signs of malignancy 2) Operative specimen: Chronic inflammatory process in the appendix mucosa without signs of malignancy
Richie et al.[17] (1975) (case 1)	64 / Female	Bilateral lower abdominal pain, watery diarrhea, emesis, dysuria, foul-smelling urine & fever	24 days	1) IVU: medial deviation of the right ureter 2) Cystoscopy: marked edema of the trigone and bladder base 3) Sigmoidoscopy: normal 4) Barium enema: appendicovesical fistula	No	Inflammend appendix & a probe-patent fistulous tract containing an olive pit was found between the appendix and the bladder. Marked edema and cystitis were present in the bladder.	Appendicectomy & partial cystectomy	1) Cystoscopic biopsy: atypical transitional cell hyperplasia with severe acute inflammation 2) Endometrial biopsy: negative for malignancy
Richie et al.[17] (1975) (case 2)	66 / Male	Midabdominal pain, fever, emesis & gross haematuria	36 hours	IVU: delayed emptying from the right kidney and a possible bladder tumour	No	Acutely inflammed appendix	Appendicectomy	Operative specimen: Periappendiceal perforation
Richie et al.[17] (1975) (case 3)	34 / Female	Crampy lower abdominal pain, fever, purulent greenish per vaginal discharge	10 days	1) IVU: right ureteral obstruction at the pelvic brim 2) Cystoscopy: trigonitis	No	Perforated pelvic appendix with abscess and drainage through the vaginal cuff	Appendicectomy, cystostomy & right ureterolysis	Vaginal cuff biopsy: negative

USG: Ultrasonography, CT: Computed Tomography scan, IVU: Intravenous Urography

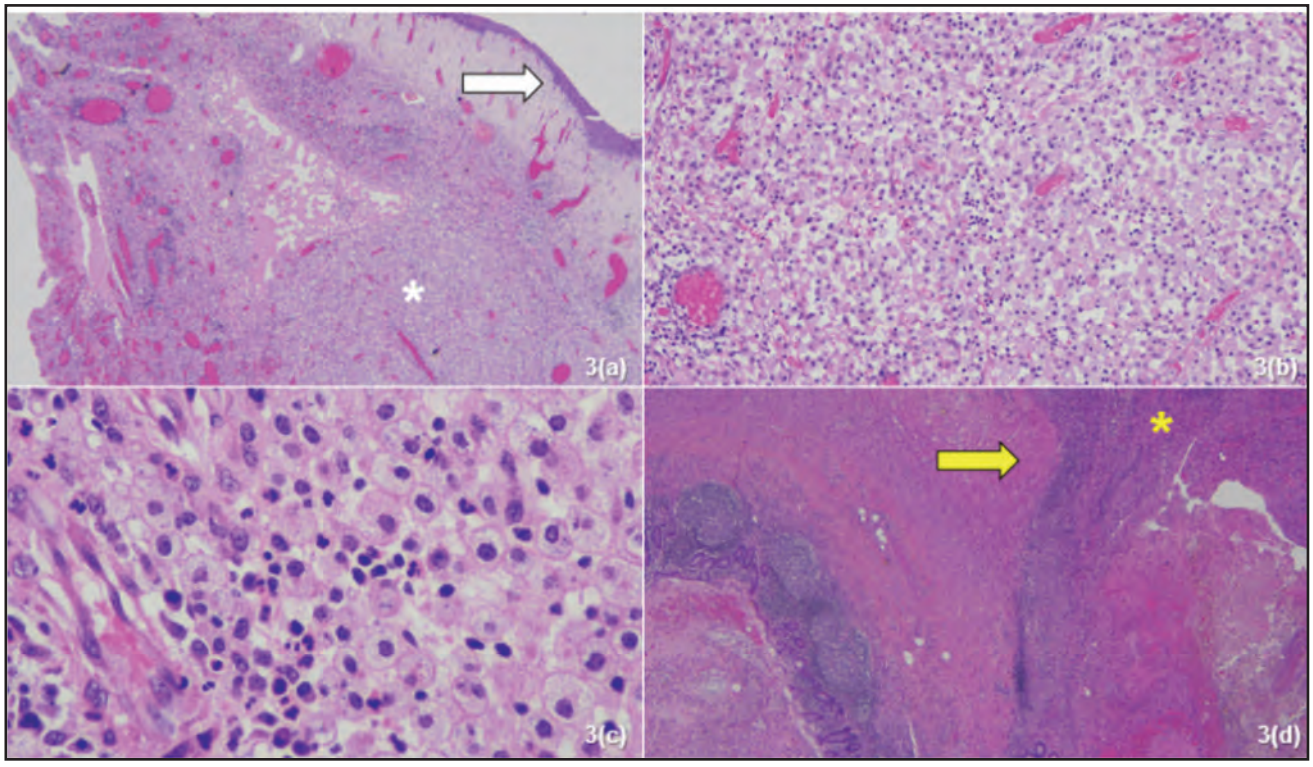

Fig. 3: Histopathological examination of the bladder tumour showed a large collections of foamy macrophages and histiocytes with abundant foamy to granular eosinophilic cytoplasm admixed with variable amounts of lymphocytes, plasma cells, eosinophils, and neutrophils representing the xanthogranulomatous inflammation (white asterisk), and the overlying bladder mucosa at the resection margin is lined by a benign urothelial epithelium. Subepithelial vascular congestion is observed (white arrow) (a) and its magnification at 10x (b) and 40x (c). The adhered part of the appendix shows focal mucosal ulceration, perforation of the muscularis propria (yellow arrow), and subjacent transmural collections of xanthogranulomatous inflammation (yellow asterisk)

DISCUSSION

Complicated appendicitis has a wide range of different clinical manifestations depending on the location of the inflamed appendix in relation to the surrounding structures. Due to close proximity, urological manifestations often accompany appendicitis. These may lead to misdiagnosis and delay in appropriate treatment. Several urological manifestations have previously been reported such as hydronephrosis resulting from compression of the ureter by an undiagnosed appendiceal abscess and can be unilateral or bilateral.[2,3] Appendicitis may also present with perinephric abscess when the appendix is in the retrocaecal position. If an enlarged or ruptured appendix accompanies the perinephric abscess, appendicectomy and abscess drainage is required.[7]

Diana et al. concluded that inflamed or ruptured appendix can affect bladder function, and even more likely when the appendix is located in the pelvic position.[8] Reported complications include cystitis, appendicovesical fistula, irritative bladder, and urinary tract infections. Both Brewster et al and Lemieux et al reported cystitis as symptoms of appendicitis.[4,5] Appendicovesical fistula was reported by Rainuli et al although it is a rare complication of appendicitis typically accompanied by coprosuria, pneumaturia, and recurrent urinary tract infections.[6] In several cases, appendicular abscesses may present as bladder tumours such as in this case. It is hence important to consider differential diagnosis upon identification of bladder tumours either via cystoscopy or CT scan.

Concerning bladder tumour, the most common 'true' bladder tumour in paediatric age group is rhabdomyosarcoma. Its morphology tends to be botryoid and is commonly located at the trigone or bladder neck. Tissue diagnosis (endoscopic or percutaneous) may be necessary when complete excision is not feasible or would result in significant morbidity, such as radical cystoprostatectomy.

Bladder tumour in children is not similar in adults where mucosal biopsy from cystoscopy is feasible without the need for general anaesthesia. Rhabdomyosarcoma require deeper mucosal sampling thus in a well localized tumour as in this case, we think primary resection is feasible for both therapeutic and diagnostic purposes, while minimizing number of general anaesthesia and avoiding treatment delay.

For this case, we were considering rhabdomyosarcoma as our first differential diagnosis despite the unusual location in this case i.e. bladder dome rather than the trigone or bladder neck which is most prevalent. The tumour was resected for

both diagnostic and therapeutic measures as it was feasible with assumptions that other medical therapies would ensue thereafter if deemed necessary. Intraoperatively, we suspected complicated appendicitis with appendicular mass but we could not dismiss the relevance of significant thickened bladder wall from preoperative imaging. Hence, the decision for partial cystectomy together with appendicectomy.

St. Peter et al. published a recent review of operative management of appendicitis specifically those categorized as complicated.[9] The authors described a recent meta-analysis of complicated appendicitis patients published in 2010 encompassing 847 patients who underwent interval appendicectomy and 725 who underwent early appendicectomy. Those who make up the interval appendicectomy group were found to experience less complications overall. Their surgical wounds were also less likely to become infected. It was also noted that likelihood of requiring reoperation was also lower.

When the same sensitivity analysis was deliberated exclusively for pediatric patients, the interval appendicectomy group also had an overall much lower complication rate.[9,10] On the basis of this data, one can conclude that initial non-operative management with interval appendicectomy is well tolerated in selected patients, specifically those who present with an abscess or well-formed phlegmon. Early suspicion of a complicated appendicitis could see initial non-operative management with antibiotics prescribe followed by interval appendicectomy. This should permit avoidance of unnecessary bladder resection.

Table I summarizes the six (6) reported cases of bladder tumour mimicry in association with appendicitis we found in our literature search. We have included our own case in the same table thus making it seven (7) cases in total. Of the six, only one case was reported in children before this to our knowledge. This case is the youngest of age in the series.

In every one of the seven (7) cases, definitive diagnosis was only rendered intraoperatively. Out of all seven (7), only in one case was appendicitis suspected i.e., the one reported by Lombay et al.[11] Based on the aforementioned observation; the likelihood of an accurate preoperative diagnosis is at a low 14%.

CONCLUSION

Rhabdomyosarcoma is the commonest paediatric soft tissue sarcoma and most commonly sited in the trigone or bladder neck. A lesson reinforced is that unusually located bladder tumour should raise one's suspicions of other aetiology such as an inflammatory mass from a complicated appendicitis. A second lesson is that with ever improving quality of imaging, we still need to balance this tool with proper clinical assessment.

Complicated appendicitis patients with an abscess or well-formed phlegmon who receive initial non-operative management with interval appendectomy have lower complication rate compared to early appendicectomy.

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DECLARATION

The authors declare no actual or potential conflict of interest in relation to this article. The patient's legal guardian formally consented to publication of this case report on May 25, 2023 by putting signature to an informed consent form.

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