RARE APPENDICEAL ESCAPADES IN CHILDHOOD: THE GRANDE EXPERIENCE!

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Abstract
Acute appendicitis in children is known to present in two broad forms: (1) uncomplicated and (2) complicated. Apart from this, a variety of atypical presentations can occur that may pose difficulty in diagnosis or treatment approach. We hereby present a series of such rare experiences namely appendiceal oxyuriasis, sub-hepatic appendicitis and appendiceal mucocele that were encountered and managed accordingly.

INTRODUCTION
The lifetime risk of developing acute appendicitis (AA) throughout the age groups is 7% [1]. The inability to predict formidable complications like perforation and peritonitis influences early operative management [2]. It is important to have an accurate preoperative diagnosis to avoid a negative exploration, for example, mesenteric lymphadenitis; a very close mimic of AA may pose a significant diagnostic challenge in this regard [3]. Similarly, avoidance of operative treatment for an appendiceal oxyuriasis (AO) would be ideally recommended if diagnosed prospectively.

Also the modality of treatment is equally crucial for the best outcome. For instance, sudden right upper abdominal pain would normally be perceived as an upper abdominal pathology in most situations, until guided by sonological findings diagnosing sub-hepatic appendicitis (SHA), which dictates a modification in the surgical approach. Likewise, an appendiceal mucocele (AM) on preoperative sonology would be dealt with open approach to avoid peritoneal contamination when chances of malignancy are predictably high.

CASE SERIES
Case 1
A 10-year-old boy presented to the emergency room with 2 days of abdominal pain initially over the peri-umbilical region and later localizing to right iliac fossa (RIF) with an episode of non-bilious vomiting. Physical examination revealed low-grade temperature (100°F) and tenderness with guarding over RIF. Hematological tests showed polymorph nuclear leukocytosis with left shift without eosinophilia. Biochemical tests and urinalyses were normal. Abdominal radiographs were unremarkable. Ultrasonogram (USG) abdomen could not visualize the appendix, but reported significant probe tenderness in RIF.

With clinical impression of AA, he underwent diagnostic laparoscopy under general anesthesia.

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Intraoperatively, a retro-cecally located appendix was found that was mildly inflamed with surface congestion more towards the tip (with visible leash of prominent vessels) and a healthy base as shown in Fig. 1a. There was no perforation, peritoneal reactive fluid or pus and no omental reaction. Aware of his clinical presentation, a possibility of other pathologies like Meckel diverticulum, mesenteric lymphadenitis and any other small bowel lesions were thought of. On walking the bowel beyond 2 feet proximal to Ileo-Caecal Junction (ICJ), no other obvious lesions were found. Laparoscopic appendectomy was performed. Specimen was retrieved in a plastic bag in an attempt to avoid contamination.

Histopathologically, gross examination confirmed the operative findings showing a 5.5 × 0.7 cm appendix with venous congestion over the serosal surface. Microscopically, the section showed mild congestion and a luminal parasite with features compatible with Enterobius vermicularis as shown in Fig. 1b–d. The specimen was reported to be an AO. He had an uneventful postoperative period and was discharged the next day. At follow up a week later, he had recovered well.

The histopathological finding came as a diagnostic surprise, since historically he never complained of appendiceal colic (AC), anal pruritus or worm defecation in the past. Moreover, eosinophilia was also not observed in the peripheral smear.Attributing his symptom complex to manifestation of AO, he was treated with a single dose of albendazole to be repeated after 2 weeks. His family members were also advised a similar treatment course. At 1 year follow up, he remained symptom free.

Case 2
A 10-year-old boy presented with history of peri-umbilical pain later migrating to right upper abdomen for 3 days and associated three episodes of non-bilious vomiting. He did not have fever, nausea or anorexia. He was moving bowel and bladder normally. Physical examination revealed tachycardia (110 beats per minute) and tenderness over entire right upper quadrant associated with rebound tenderness. Hematological tests were unremarkable. Biochemical tests were normal. Abdominal radiographs were unremarkable. USG abdomen showed a blind ending tubular, non-compressible, non-peristaltic structure measuring 9.5 mm, suggesting an enlarged appendix with its tip located in the sub-hepatic region as shown in Fig. 2a.

With clinical impression of SHA, he underwent an emergency diagnostic laparoscopy under general anesthesia. Intraoperatively, findings were confirmed, an inflamed turgid appendix with its tip just beside the gall bladder in the sub-hepatic region was noted with a healthy base as shown in Fig. 2b.

Laparoscopic appendectomy was performed as shown in Fig. 2c.

Histopathologically, gross findings included an 8 cm long appendix with cut section showing lumen filled with fecal material. Microscopically, the section showed suppuration, edema and congestion along with peri-appendiceal inflammation and focal areas of destruction of the muscular layer suggestive of gangrenous AA.

He had an uneventful postoperative period and was discharged the next day. At follow up a week later and 1 year later, he was doing well.

Case 3
A 13-year-old boy presented with sudden onset RIF pain for a day associated with vomiting, nausea and anorexia without fever. He was moving bowel and bladder normally. Physical examination revealed guarding and rebound tenderness over RIF. Hematological tests showed polymorph nuclear leukocytosis. Biochemical tests and urinalyses were normal. Abdominal radiographs were unremarkable. USG abdomen showed a blind ending tubular, fluid-filled, non-compressible, non-peristaltic structure measuring 13 mm, suggesting an enlarged appendix with mucocoele formation as shown in Fig. 3a.

In view of known associations of mucocoele with malignancy, he underwent open appendectomy as shown in Fig. 3b and c. No visible or palpable mass lesion was found intraoperatively. Histopathologically, gross findings included an 8 cm long appendix with cut section showing lumen filled with purulent material and thinning of wall (2 mm) without an obvious mass lesion as shown in Fig. 3d. Microscopically, the section showed suppuration, edema and congestion along with mucosal erosions. The lumen was filled with neutrophils and fibrinoid material. Peri-appendiceal inflammation was noted along with focal areas of destruction of the muscular layer suggestive of gangrenous AA.

Figure 1: (a) Appendectomy specimen. (b–d) Microscopic appearance of AO at ×10 (b), ×20 (c) and ×40 (d) magnifications, respectively, showing mild congestion and luminal parasite.

Figure 2: (a) SHA diagnosed and located on abdominal ultrasound. (b) Laparoscopic visualization of SHA. (c) Appendectomy specimen of the SHA.
Rarer positions include sub-hepatic, lateral pouch, mesocoeliac, pre- and post-ileal locations in the decreasing order of frequency. Position in 65% cases and less commonly in the pelvis, subcecal, mesenteric abscesses, AO is still not a common cause of AA [8–14]. Operative treatment in such patients is invasive inflammation even when a theoretical possibility of AC exists [15]. Laparoscopic approach was used to localize the infection and subsequent appendectomy could be achieved using a standard three port technique. This was of dual benefit as it avoided a conventional Lanz incision and also a right sub-costal muscle cutting incision that is known for its morbidity, besides the usual advantages of laparoscopy [19].

Lastly, the surgical approach to AA also depends on preoperative predictability of sinister findings as with an appendicular mucocele, which was diagnosed on ultrasound in Case 3. Mucocele of the appendix is an exceedingly rare presentation of AA in children with scarce descriptions and sporadic reports [20]. It implies an appendix that is cystic and dilated with mucinous material as a result of an obstructive lesion. This could be a fecolith, endometriosis, extrinsic compression, inflammation or even a neoplasm. Appendicular malignancies, with an incidence of <0.5%, are a rare occurrence in children with predominant ones being neuro-endocrine variety [21].

With a clinical presentation often mimicking AA, mucinous neoplasms of appendix are difficult to distinguish clinically. Since, tactile examination to confirm tumor in the adjacent large bowel is desirable, an open approach was favored in Case 3 to deal with this potential pathology. However, with normal findings, the procedure could be concluded with a simple appendectomy.

CONCLUSION

1. When on table findings are incongruent with the clinical picture, rarer cause of AA like AO needs to be considered.
2. When symptom complex of AA does not follow a clinic-anatomical correlation, a developmental malformation like abnormal appendico-cecal location must be suspected.
3. When symptom complex of AA is accompanied by an uncommon radiological finding, the surgical approach should be guided by the foreseeable intraoperative findings.

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CONFLICT OF INTEREST STATEMENT

None declared.
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