Torsion of the Vermiform Appendix: A Case Report and Review of Literature

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Patient: Male, 30
Final Diagnosis: Torsion of appendix
Symptoms: Abdominal pain • anorexia • nausea
Medication: —
Clinical Procedure: Laparoscopic appendicectomy
Specialty: Surgery

Objective: Rare disease
Background: Torsion of the vermiform appendix is a rare condition that presents with symptoms analogous to those of common acute appendicitis; therefore, it is often diagnosed during surgery. It was first described by Payne et al. in 1918. Since then, there has been wide recognition of a primary and a secondary form of the condition, affecting both the pediatric and adult populations. We present a case of an adult patient and conducted a literature review in the adult demographic.

Case Report: We report the case of a 30-year-old man who presented with clinically acute appendicitis. Laparoscopically, we diagnosed a torsion of the vermiform appendix secondary to a mucocele process. Histology confirmed a low-grade mucinous cystoadenoma, with a hemorrhagic necrosis of the wall, in keeping with torsion.

Conclusions: Torsion of the vermiform appendix is a rare condition that presents similar to acute appendicitis, and therefore is often diagnosed intraoperatively. Since first described, 33 cases in adults were identified in the English literature, and recognition of a primary or secondary form has emerged. Preoperative radiological imaging is rarely useful in diagnosis. To the best of our knowledge, this is the eighth reported case in the English literature of a torsion of the vermiform appendix secondary to a mucinous cystoadenoma.

MeSH Keywords: Appendix • Cystadenoma, Mucinous • Torsion Abnormality

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Background

Torsion of the vermiform appendix is a rare condition that presents with symptoms analogous to those of common acute appendicitis; thus, it is often diagnosed during surgery. It was first described by Payne et al. in 1918 [1]. Since then, there has been wide recognition of a primary and a secondary form of the condition, affecting both the pediatric and adult populations. We present a case of torsion of the vermiform appendix secondary to mucinous cystoadenoma, with a review of the literature in the adult demographic.

Case Report

A 30-year-old man presented with a 24-h history of progressive onset of abdominal pain associated with nausea and anorexia. The pain was localized centrally and migrated to the right iliac fossa. He reports there was no radiation of pain or any febrile symptoms. His bedside observations were within normal limits. Abdominal examination revealed significant tenderness with localized peritonism on the right iliac fossa. Rovsing’s sign was also positive. Laboratory findings were unremarkable except for leukocytosis of 15.1×10^9/L (normal range 4.0–11.0). Radiological imaging was not performed, as he was clinically diagnosed with acute appendicitis.

Upon laparoscopy, the appendix was located in the right iliac fossa but was grossly distended and gangrenous in appearance (Figure 1). It was rotated 720 degrees anticlockwise at its base. A standard laparoscopic appendicectomy was performed with two 5-mm ports inserted at the left iliac fossa and suprapubic area. The appendix was assessed carefully and deemed appropriate to proceed laparoscopically, as it did not appear to be necrotic or at high risk of rupture with manipulation. Its position was paracecal, which did not require significant laparoscopic manipulation to define the anatomy. The appendix was untwisted completely, and the mesoappendix was dissected with the appendiceal artery clipped and transected. The base of appendix was ligated using 2 loops of polydioxanone suture (PDS) and the appendix was retrieved using a specimen pouch plastic bag. The umbilical port site wound had to be extended inferiorly to deliver the specimen. The patient recovered post-operatively without any complications and was discharged home the next day. The specimen measured 120 mm in length, with a maximum diameter of 30 mm (Figure 2). Histology finding was a low-grade mucinous cystoadenoma, with a hemorrhagic necrosis of the wall, in keeping with torsion.

Discussion

Torsion of the vermiform appendix is a rare disorder that presents with a clinical picture similar to acute appendicitis; therefore, it is often diagnosed intraoperatively, to the surgeon’s surprise. It was first reported in the English literature by Payne et al. in 1918 [1]. Since then, there has been widespread recognition of a primary and secondary form of appendiceal torsion.

In our review of the English literature, 33 cases of torsion of the vermiform appendix in adults were identified, including the present case [1–32] (Table 1). The mean age is 42 years old, with a range of 18 to 79 years old, and a 19: 14 female-to-male sex ratio. The rotation of torsion varies from 180 to 1800 degrees, and although anticlockwise rotation is often reported as the most common rotation, our review of the literature in the adult population shows that clockwise rotation is most common (12 clockwise vs. 8 anticlockwise).

Primary torsion has been associated with anatomical variation in which the mesoappendix is fan-shaped, with a narrow base, and the absence of the azygotic fold that normally attaches the appendix laterally, or a long appendix [33,34]. It

Figure 1. Laparoscopic view of the appendix mucocele. Arrow pointing to torsion at the base of the appendix.

Figure 2. Appendix specimen.
Table 1. Appendiceal torsion in an adult.

| Author            | Ref  | Year | Age/Sex | Presenting complain | Degree/direction of torsion | Length, cm | Preoperative USS/CT/MRI | Secondary cause            |
|-------------------|------|------|---------|---------------------|-----------------------------|------------|-------------------------|---------------------------|
| Payne JE          | [1]  | 1918 | 37 F    | RIF pain NV         | 1080                        | NA         | 7                       | No                        | Fecalith                  |
| Bevers EC         | [2]  | 1920 | 35 F    | RIF pain NV Fever   | 720                         | NA         | 7.6                     | No                        |                           |
| Flatley G         | [3]  | 1936 | 22 F    | Periumbilical pain  | 900                         | NA         | 9.5                     | No                        |                           |
| Chan KP           | [4]  | 1965 | 18 F    | RIF pain            | 1260                        | NA         | 10                      | No                        | Simple mucocele            |
| Ghent WR          | [5]  | 1966 | 21 M    | RIF pain NV Fever   | 450                         | AC         | NA                      | No                        |                           |
| De Bruin Al       | [6]  | 1969 | 28 F    | Lower Abd Pain Fever| 360                         | AC         | 10.5                    | No                        |                           |
| Killam AR         | [7]  | 1969 | 47 M    | RIF pain NV Fever   | NA                          | NA         | 7                       | No                        | Mesoappendiceal lipoma     |
| Legg NGM          | [8]  | 1973 | 29 M    | RIF pain            | 360                         | NA         | 10                      | No                        |                           |
| Finch DRA         | [9]  | 1974 | 38 F    | Suprapubic pain NV  | 540                         | NA         | 10                      | No                        |                           |
| Cassie GF         | [10] | 1974 | 25 M    | RIF pain            | 720                         | AC         | 11                      | No                        | Carcinoid tumor            |
| Petersen KR       | [11] | 1982 | 59 F    | Suprapubic pain NV  | 540                         | NA         | 10                      | No                        |                           |
| Dickson DR        | [12] | 1982 | 60 F    | RIF pain            | 720                         | NA         | 15                      | No                        | Simple mucocele            |
| Won OH            | [13] | 1982 | 35 M    | RIF pain            | NA                          | NA         | 12                      | No                        |                           |
| Abu Zidan FM      | [14] | 1992 | 37 F    | RIF pain            | NA                          | C          | 11                      | No                        | Mucinous cystoadenoma      |
| Moten AL          | [15] | 2002 | 44 F    | Abd pain            | 360                         | NA         | 10                      | No                        | Mucinous cystoadenoma      |
| Tzilinis A        | [16] | 2002 | 44 M    | Suprapubic pain NV  | 540                         | C          | 5.5                     | No                        |                           |
| Bowling CB        | [17] | 2006 | Middle age F| Abd pain NV      | NA                          | C          | 12                      | CT                        | Mucinous cystoadenoma      |
| Bestman TJR       | [18] | 2006 | 35 F    | Abd pain            | NA                          | NA         | 7.5                     | USS                       |                           |
| Rajendran N       | [19] | 2006 | 29 F    | RIF pain            | 360                         | NA         | 11                      | No                        | Simple mucocele            |
| Rudloff U         | [20] | 2007 | 28 F    | RLQ pain            | 900                         | C          | 5                       | CT                        |                           |
| Kitagawa M        | [21] | 2007 | 34 M    | Periumbilical pain  | 180                         | C          | 10                      | CT                        | Mucinous cystoadenoma      |
| Hebert JJ         | [22] | 2007 | 59 M    | RLQ pain            | NA                          | NA         | 12                      | CT                        | Mucinous cystoadenoma      |
| Hamada T          | [23] | 2007 | 79 M    | Abd pain            | 180                         | AC         | NA                      | USS                       | Mucinous cystoadenoma      |
| Wani I            | [24] | 2008 | 76 M    | RIF pain            | 540                         | AC         | 10                      | No                        | Fecalith                  |
| Wani I            | [25] | 2010 | 38 F    | RIF pain NV fever   | 180                         | C          | 20                      | CT                        | Adhesion from ovarian mucocele torsion |
| Lee CH            | [26] | 2011 | 78 F    | RIF pain NV         | 900                         | AC         | 10.5                    | CT                        | Simple mucocele            |
| Misurati FI       | [27] | 2011 | 48 M    | RIF pain            | 360                         | C          | NA                      | No                        | Simple mucocele            |
| Dimitriadis PA    | [28] | 2012 | 52 F    | RUQ pain            | 540                         | AC         | 9                       | USS                       | Caecal malposition         |
| Mishin I          | [29] | 2012 | 30 M    | RIF pain, N        | 360                         | C          | 11                      | No                        | Mucinous cystoadenoma      |
| Stark C           | [30] | 2014 | 34 F    | RIF pain NV         | 720                         | C          | 8                       | US and MRI                | Mucinous cystoadenoma      |
| Grebic D          | [31] | 2015 | 70 M    | RIF pain            | 1800                        | C          | 9                       | USS                       | Mesoappendiceal lipoma     |
| Dubhashi SP       | [32] | 2016 | 52 F    | RIF pain fever      | 180                         | C          | 8                       | USS                       |                           |
| Current           |      | 2017 | 30 M    | RIF pain            | 720                         | C          | 12                      | No                        | Mucinous cystoadenoma      |

F – Female; M – Male; RIF – right iliac fossa; NV – nausea vomiting; NA – not available; AC – anticlockwise; C – clockwise; USS – ultrasound scan; CT – computed tomography scan; MRI – magnetic resonance imaging; Ref – reference. Secondary causes, where clearly mentioned, are listed. Otherwise, it was left blank and presumed as primary torsion or undefined.
has also been described as a secondary ischemic or necrotic change with luminal distension distally to the torsion site in the absence of any primary lesion.

Secondary torsion is associated with an identifiable patholo-
gy such as a fecalith, mucocele, carcinoid tumor, or cystoad-
enoma. Theoretically this would cause the appendix to first
be engorged and distended, before rendering it unstable and
more likely to twist.

Preoperative imaging is often unhelpful in diagnosis. Out of the 33 reported cases in adults, 13 had preoperative imaging that is
not a plain film, and of those, only 3 reported cases had a radi-
ologically diagnosed twisted appendix [22,23,30]. Hamada et al.
described a target-like appearance at the base of the appendix
on ultrasound akin to that seen in cases of ovarian and testicular
torsion, whereas in the case reported by Herbert et al., a whirl
of mesenteric fat and vessels around the appendiceal axis was
seen on CT. On MRI, Stark et al. considered mesenteric edema and
abrupt tapering of the base of appendix to be signs of torsion.

Conclusions

Torsion of the vermiform appendix is a rare condition that
presents similar to acute appendicitis and is therefore often
diagnosed intraoperatively. Since first described, 33 cases in
adults were identified in the English literature, and recogni-
tion of a primary or secondary form has emerged. Preoperative
radiological imaging is rarely useful in diagnosis. To the best
of our knowledge, the present case is the eighth report in the
English literature of a torsion of the vermiform appendix, sec-
ondary to a mucinous cystadenoma. Whether surgery is car-
rried out in open or laparoscopic approach, great care should
be taken to avoid iatrogenic rupture of the appendiceal mu-
occele. We would not hesitate to convert to open to prevent
iatrogenic rupture and spillage of content/cystadenoma in a
more difficult situation.

Conflict of interest

None.

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