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

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
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Conflict of interest: None declared

Patient: Male, 30
Final Diagnosis: Torsion of appendix
Symptoms: Abdominal pain • anorexia • nausea
Medication: —
Clinical Procedure: Laparoscopic appendectomy
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 mucocoele process. Histology confirmed a low-grade mucinous cystadenoma, 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 cystadenoma.

MeSH Keywords: Appendix • Cystadenoma, Mucinous • Torsion Abnormality

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/908725>

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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 cystadenoma, 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 $15.1 \times 10^9/L$ (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

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

Figure 2. Appendix specimen.

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 cystadenoma, 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

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 NV Fever	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 AJ	[6]	1969	28 F	Lower Abd Pain Fever	360	AC	10.5	No	Mesoappendiceal lipoma
Killam AR	[7]	1969	47 M	RIF pain NV Fever	NA	NA	7	No	
Legg NGM	[8]	1973	29 M	RIF pain Fever	360	NA	10	No	
Finch DRA	[9]	1974	38 F	RIF pain NV Fever	360	NA	NA	No	Simple mucocele
Cassie GF	[10]	1953	25 M	RIF pain N	720	AC	11	No	
Petersen KR	[11]	1982	59 F	Suprapubic pain NV	540	NA	10	No	Carcinoid tumor
Dickson DR	[12]	1953	60 F	RIF pain NV	720		15	No	
Won OH	[13]	1977	35 M	RIF pain	NA	NA	12	No	Simple mucocele
Abu Zidan FM	[14]	1992	32 F	RIF pain	NA	C	NA	USS	
Moten AL	[15]	2002	44 F	Abd pain	360	AC	NA	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 N	NA	NA	7.5	USS	
Rajendran N	[19]	2006	29 F	RIF pain N	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	
Hamada T	[23]	2007	79 M	Abd pain	180	AC	NA	USS	Mucinous cystoadenoma
Wani I	[24]	2008	76 M	RIF pain N	540	AC	10	No	
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	
Marsdin EL	[27]	2011	48 M	RIF NV	360	C	NA	No	Simple mucocele
Dimitriadis PA	[28]	2012	52 F	RUQ pain	540	AC	9	USS	
Mishin I	[29]	2012	30 M	RIF pain, N	360	C	11	No	Caecal malposition
Stark C	[30]	2014	34 F	RIF pain NV	720	C	8	US and MRI	
Grebic D	[31]	2015	70 M	RIF pain	1800	C	9	USS	Mucinous cystoadenoma
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 pathology such as a fecalith, mucocele, carcinoid tumor, or cystoadenoma. 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 radiologically 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 whorl 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.

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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 recognition 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, secondary to a mucinous cystadenoma. Whether surgery is carried out in open or laparoscopic approach, great care should be taken to avoid iatrogenic rupture of the appendiceal mucocele. 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.