

Mucocele of the appendix presenting as an exacerbated chronic tubo-ovarian abscess

A case report and comprehensive review of the literature

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Abstract

Rationale: Appendiceal mucocele is a rare entity of mucinous cystic dilatation of the appendix. It has no typical clinical presentation and is considered a potentially premalignant condition.

Patient concerns: We present a case of accidental intraoperative finding of an appendiceal mucocele in a 54-year old woman that clinically presented with an exacerbated chronic tubo-ovarian abscess.

Diagnoses: Trans-vaginal ultrasonography showed an encapsulated, oval, unilocular mass above the uterus with a heteroechogenic structure, homogeneous fluid content, and smooth regular walls without inner proliferation. The histopathologic diagnosis was consistent with an appendiceal cystadenoma.

Interventions: The patient underwent a simple appendectomy.

Outcomes: There were no clinical, biochemical or imaging signs of the disease recurrence at 6 months follow up.

Lessons: To our knowledge, this is the only well-documented case of appendiceal mucocele mimicking exacerbated chronic tuboovarian abscess reported in the literature. Awareness of a rare entity such as an appendiceal mucocele, which is frequently misdiagnosed as a potential cause of acute abdomen, is necessary for the appropriate management strategy in order to prevent complications.

Abbreviations: AM = appendiceal mucocele, HAMN = high-grade appendiceal mucinous neoplasm, LAMN = low-grade appendiceal mucinous neoplasm.

Keywords: acute abdomen, adnexal mass, appendiceal mucocele, high-grade appendiceal mucinous neoplasm, low-grade appendiceal mucinous neoplasm, mucinous cystadenocarcinoma, mucinous cystadenoma, tubo-ovarian abscess

1. Introduction

Common conditions that cause acute lower abdominal pain in a woman include: appendicitis/periappendicular abscess, adnexal mass, cecal/appendiceal carcinoma, mucocele of the appendix and lymphoma.^[1]

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Mucocele of the appendix (appendiceal mucocele [AM]) is a clinical descriptive term for obstructive dilatation of the appendix caused by intraluminal mucinous accumulation.^[2] The AMs were previously classified into 4 pathologic entities according to the characteristics of the epithelium: simple or retention mucocele, mucocele with local or diffuse villous hyperplastic epithelium (5–25%), mucinous adenoma/cystadenoma (63–84%), and mucinous cystadenocarcinoma (11–20%).^[3–5] The recently established nomenclature suggests that the AM term should be used only as a clinical term and that histologic diagnosis should classify AMs into either low-grade appendiceal mucinous neoplasms (LAMN) or high-grade appendiceal mucinous neoplasms (HAMN). The term mucinous cystadenoma should no longer be used.^[6]

The clinical significance of AM is reflected in the fact that it is often misdiagnosed and therefore a variety of complications can arise due to improper detection/management. Our objective was therefore to emphasize the broad array of differential diagnoses associated with acute lower abdominal pain in women to which appendiceal mucocele is a member. Furthermore, we stress the need for adequate preparation to effectively manage this condition.

In this report we present a case of appendiceal mucocele in a postmenopausal woman that was treated under the clinical diagnosis of an exacerbated chronic tubo-ovarian abscess (TOA). In addition to this specific case, a comprehensive literature review was performed.

2. Case report

A 54-year-old Caucasian woman presented with severe right lower abdominal and pelvic pain, vomiting, fever, and chills. The

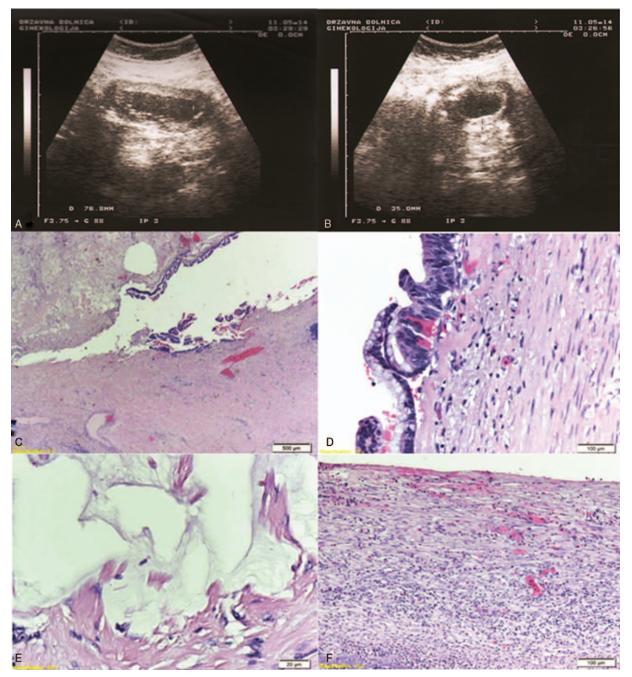


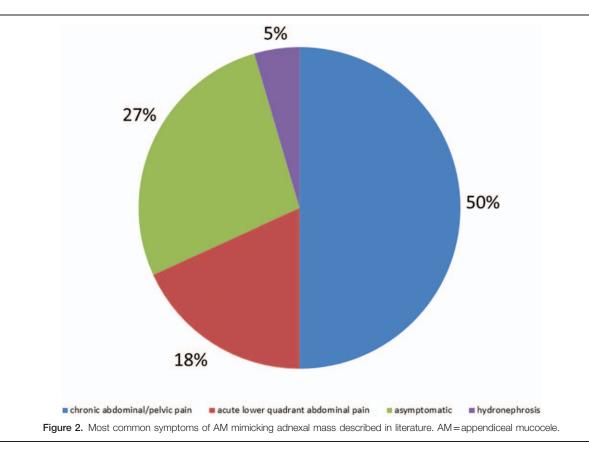
Figure 1. A–B: Transvaginal sonography of the appendiceal mucocele showing ovoid formation with thickened wall, filled with anechogenic material, and no intraluminal proliferations. C: Lumen of the appendix was cystically dilated and contained mucus. The wall of appendix was partly necrotic with dysplastic epithelium on its surface (Hematoxylin and eosin staining, [H&E], 5×). D: Surface epithelium showing moderate to severe dysplastic changes (H&E, 40×). E: Mucus in the lumen of appendix (H&E, ×100). F: Periappendicitis (H&E, 40×).

vague symptoms started 7 days prior to, gradually increased and worsened 2 days before the hospital admission. She delayed consulting her physician until the pain became severe. She had no history of irregular bowel movements or abnormal changes in the stool. The patient denied having any prior episodes of chronic abdominal pain, as well as any gastrointestinal, gynecologic, or other symptoms, including any recent weight loss. Her last menstrual period was approximately 3 years ago.

Previous medical and surgical histories including a family history for malignancy were unremarkable. On physical examination, the patient presented with pain, fever (39°C), and tachycardia. All other vital signs were within the normal range. Abdominal examination showed right lower abdominal quadrant tenderness, with moderate guarding, muscle rigidity, and rebound tenderness. There was a right adnexal tenderness along with cervical motion tenderness on pelvic examination. A scant amount of cervical discharge was also observed.

Laboratory tests showed mild anemia (RBC 3.84, hemoglobin 112 g/L, Hematocrit 0.33), with all other standard lab test parameters within the reference ranges.

Table 1 Review of 22 cases of	i AM mimicking adr	Table 1 Review of 22 cases of AM mimicking adnexal mass from the literature.	ature.			
Authors	Patients characteristics	Imaging method	Preop. Dg.	Treatment option	Timing of the diagnosis	Mucocele type
Abu Zidan et al ^[13]	I	SU	38 weeks pregnant. Torqued	LPT. Caesarean section and	Intra/postoperative	Simple AM
Akman et al. ^[9]	81 F	US, MRI	Right adnexa mass.	Appendectomy, R hemicolectomy	Intra/postoperative	Mucinous cystadenoma
Arrington et al, ^[14]	15 F	SVT	Ovarian torsion	with ileo-transverse anastomosis TAH. LPSC. Appendectomy and partial	Intra/postoperative.	Simple AM
12				cecotomy.		
Bahia and Wilson ^[15]	46 F	US, Barium enema.	Aadnexal mass.	Appendectomy	Intra/postoperative	AM
Balci et al, ^[10]	71 F	TVS, MRI	Ovarian cyst.	LPT. Appendectomy. TAH	Intra/postoperative	AM
Bartlet et al ¹¹¹	80 F	US, CT	Ovarian cyst.	LPT. Appendectomy.	Intra/postoperative	Mucinous cystadenoma
Cristian et al ^{t roj}	61 F 30 r	US, CT	Hydrosalpinx or ovarian cyst.	LPSC. Appendectomy.	Intra/postoperative.	LAMN
Dragoumis et al,	7.0 F	IV5, UI.	Uvarian cystic tumor.	IAH With Dilateral salpingo- oonhorectomy Annendectomy	Intra/postoperative	Mucinous cystadenoma
Driman et al. ^[18]	34 F.	X-rav colonoscopy. CT.	Adnexal mass	R hemicolectomy. ovarian	Intra/postoperative	Foci of endometriosis in the
				cystectomy, and removal of	-	proximal appendix and an AM
				the mucinous cysts from the cul-de-sac.		
	31 F	Evulorative LPSC	Endometrioeie	l sear avricion of the		Palvic andomatriosis AM
	- -)			endometrioid deposits and		
				appendectomy.		
Gortchev et al, ^[19]	68 F	TVS	Cystic adnexal mass	LPSC. Appendectomy	Intra/postoperative.	AM
Hajiran et al, ^[20]	50 F	TVS, MRI	Adnexal mass	Appendectomy.	Intra/postoperative.	Low-grade mucinous adenocarci-
				Reoperation. Hemicolectomy.		noma
Hutchinson et al, ^[21]	47 F.	US, CT, MRI.	Hydrosalpinx, AM, Tumor.	Surgical. Approach unclear.	Intra/postoperative.	Carcinoma in situ
Kalu and Croucher, [22]	42 F	TVS	Cystic adnexal mass	Appendectomy	Intra/postoperative	AM
0'Sullivan et al, ^[23]	31 F	IVU, US, and CT.	Hydronephrosis, hydroureter.	Excision of the caecum and	Intra/postoperative.	Endometriosis of the appendix
				terminal ileum with an		resulting in AM and endometrio-
				ileocolic anastomosis, limited		sis of the terminal ileum.
				ureterolysis and a ureteric		
				stent inserted through an		
				ureterotomy.		
Ortiz-Mendoza ^[24]	46 F	TAS	Ovarian cyst.	LPT. Right hemicolectomy.	Intra/postoperative	AM
Paladino et al, ^[25]	79 F	MRI, TVS	Adnexal mass.	LPSC. Appendectomy.	Intra/postoperative	AM
Papoutsis et al, ^[26]	78 F	TVS, TAS, MRI	Potentially malignant right ovarian	Appendectomy. TAH.	Intra/postoperative	Mucinous cystadenoma
			tumor.			
Rudloff and Malhotra ^[27]	28 F	CT	Ruptured ovarian cyst.	LPSC. Appendectomy.	Intra/postoperative	AM
Scaffa et al, ^[28]	36 F.	TVS and CD	Adnexal complex mass.	Appendectomy	Intra/postoperative	Mucinous cystadenoma
Shimizu et al, ^[29]	75 F	US, CT, Barium enema	Potentially malignant ovarian	Open appendectomy.	Intra/postoperative	Mucinous cystadenoma
Vildiz and Abassoclut ^[30]	54 F	TAS CT colonosconv	tumor. Ovarian mass	Annendectomy	Intra/nostonerative	AM
MRI = magnetic resonance imaging.	Jg.					



Trans-vaginal ultrasonography (TVS) showed a well-defined, encapsulated, oval, unilocular mass just above the uterus with a heteroechogenic structure, homogeneous fluid content, and smooth regular walls without inner proliferation. Doppler sonography did not detect flow within the structure. The left ovary appeared normal, whereas it was not possible to localize the parenchyma of the right ovary (Fig. 1A–B).

The history, physical, laboratory, and imaging finding led to the preliminary diagnosis of exacerbated chronic TOA and an open surgery was planned.

Intraoperative finding showed a normal sized uterus, ovaries, and fallopian tubes, and a cystic mass of the appendix; there was no evidence of ascites in the peritoneal cavity. The mass measured 11.5 cm in diameter and was loosely adherent to the surrounding tissue. Signs of periappendiceal inflammation were negative. The mass was suspected to be a mucocele based on the surgeon's experience. Given this presentation, and the apparently uninvolved appendiceal base, a decision was made to perform a simple appendectomy, with special attention towards preserving the integrity of the mass. The peritoneal cavity was subsequently thoroughly irrigated.

On gross examination the appendix measured $11.5 \times 7.5 \times 6$ cm, was diffusely dilated, filled with fecal contents, entailing 2 concernments along with some amount of mucus. Focal wall thickening up to 5 mm, and intramural calcifications were also noted. Microscopic examination of the tissue showed LAMN with moderate to severe focal dysplastic changes in the epithelium and chronic inflammation of the surrounding structures (periappendicitis) (Fig. 1C–F).

The patient was discharged from the hospital on the fourth postoperative day. Patient's further clinical course remained uneventful with no signs of disease progression at a follow up examination 6 months after surgery. The patient was counseled about association of mucinous neoplasms of the appendix with cancer in other organs, such as ovaries, colon, endometrium. Follow up transvaginal and transabdominal ultrasound were normal.

This study was conducted in accordance with the ethical standards laid down in 1964 Declaration of Helsinki. The case report was shared with the local ethical committee but the policy of the committee is not to review case reports. Informed written consent was obtained from the patient for publication of this case report and accompanying images.

3. Discussion

In this article, we present a case of appendiceal mucocele (AM) mimicking a right adnexal mass, which was diagnosed intraoperatively and treated with a simple appendectomy and uneventful clinical follow-up.

The differential diagnosis of the acute lower quadrant abdominal pain in a postmenopausal women is broad and may involve gynecologic (pelvic inflammatory disease, adenomyosis, degenerating uterine fibroid, ruptured ovarian cyst, TOA), gastrointestinal (appendicitis, diverticulitis, bowel obstruction, inguinal hernia, mesenteric venous thrombosis, perirectal abscess, complication of inflammatory bowel disease), and urinary (ureterolithiasis, cystitis, pyelonephritis) etiologies. The most common causes, however, include: pelvic inflammatory disease, ruptured ovarian cyst, and appendicitis.^[7]

In the case of our patient, a preliminary diagnosis of exacerbated chronic TOA was based on detection of fluid-filled adnexal mass associated with positive 2 of 4 SIRS criteria (fever

Table 2

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Authors	Patients characteristics	Imaging method	Preop. Dg.	Treatment option	Timing of the diagnosis	Mucocele type
Abuoglu et al ^[31]	16 cases	US, CT	AA	LPT. 14/16 appendectomy. 2/16 R hemicolectomy	Intra/postoperative	AM, Mucinous cystadenoma
Basak et al ^[32]	26 M 31 F	US US	AA AA	LPT. Appendectomy LPT. Appendectomy	Intra/postoperative Intra/postoperative	Mucinous cystadenoma Neuroendocrine tumor and retention cyst
	39 F 42 M	US, CT CT	AA AA	LPT. Appendectomy LPT. R hemicolectomy	Intra/postoperative Intra/postoperative	Retention cyst Retention cyst Mucinous cystadenoma, pseudomyxoma peritonei.
	49 F 61 F. 62 M	US, CT US, CT US, CT	AA AA AA	LPT. Appendectomy LPT. Appendectomy LPT. Appendectomy	Intra/postoperative Intra/postoperative Intra/postoperative	Mucinous cystadenoma Mucinous cystadenoma Mucinous cystadenoma
Bestman et al ⁽³³⁾ Caliskan et al ⁽³⁴⁾	76 M 35 F 11 patients (2 F/ 6 M), median age 70.	US Abdominal x-ray. CT, US	AA. AA 6 AA. 2 intestinal obstruction.	LPT. Appendectomy LPSC. Appendectomy Emergency surgery. Appendectomy 5/8. Small bowel/sigmoid colon resection/	Intra/postoperative Intra/postoperative Intra/postoperative	Mucinous cystadenoma Retention AM AM
Casey et al ^[35]	36 F	US	AA, 21 weeks gestation	hemicolectomy 3/8. Exploratory LPT. Appendectomy.	Intra/postoperative	Mucinous cystadenoma
Costa and Demuro ^[36]	77 M	CT	Cecal volvulus.	Exploratory laparotomy. Right hemicolectomy.	Intra/postoperative	AM
Demetrashvili et al ⁽³⁷⁾ Fatima Ezzahra et al ⁽³⁸⁾ Hamada et al ⁽³⁹⁾ Hebert and Pickhardt ⁽⁴⁰⁾ Idris et al ⁽⁴¹⁾ Karakaya et al ⁽⁴²⁾	54 M 55 M 79 M 59 M 35 F. Pregnant 82 M	US - US CT US US, CT.	AA AA Torqued AM Torqued AM AA AA.	LPT. Appendectomy. Unclear. Appendectomy. LPT. Appendectomy. LPT. Appendectomy. LPT. Appendectomy. LPT. Appendectomy.	Intra/postoperative Intra/postoperative Preoperative Preoperative Intra/postoperative Intra/postoperative	Mucinous cystadenoma Mucinous cystadenoma Mucinous cystadenoma Mucinous cystadenoma AM Mucinous cystadenoma.
Kehagias et al ^[43]	65 F 72 F	Abdominal x-ray, US, CT CT	lleus Acute abdomen	LPT. Ileal and cecal mesenteric ischemia. LPT. R Hemicolectomy.	Intra/postoperative	Simple mucocele.
Kelemouridou et al ^[44] Kilic et al. ^[45] Kimura et al ^[46]	71 M 52 F 41 F	Imaging Unclear Ba enema	AA AA ACute abdomen	Open appendectomy. LPSC. Appendectomy. LPT. Appendectomy.	Intra/postoperative Intra/postoperative Intra/postoperative	Mucinous cystadenoma LAMN Mucosal hyperplasia in
Kitagawa et al ^[47]	34 M	CT	Inflamed appendi- ceal tumor	LPT. Appendectomy.	Intra/postoperative	AM Mucinous cystadenoma
Komo et al ^[48]	79 F	CT	Mechan-ical small intestinal obstruction.	LPT. Appendectomy.	Intra/postoperative	LAMN
Lee et al ^[49]	78 F	CT	AM with periap- pendicitis.	LPT. Appendectomy	Preoperative	AM
Malasi et al ⁽⁵⁰⁾ Malya et al ⁽⁵¹⁾	58 M 30 M 56M 39 M	CT US, CT US, CT. US, CT.	Ruptured AM. AA AA AA	LPT. Appendectomy. LPT. Appendectomy.	Preoperative Intra/postoperative	AM AM
Mishin et al ⁽⁵²⁾	30 M	Diagnostic LPSC.	AA	LPSC. Conversion to LPT. Appendectomy.	Intra/postoperative	Mucinous cystadenoma
Nopajaroonsri and Mrejoud ^[53]	22 F	-	AA	Exploratory LPT. Appendectomy.	Intra/postoperative	Retention cyst
Opreanu et al ^[54]	51 F	CT	Large bowel obstruction.	LPT. Ileocecal resection and primary anastomosis	Intra/postoperative	AM
Park et al ^[55]	69 M	CT	Infectious colitis. Intususception.	Laparoscopic appendectomy	Intra/postoperative	AM
Sertkaya et al ⁽⁵⁶⁾ Singal et al ⁽⁵⁷⁾	61 F 23 F 46 F 57 M 35 M	US, CT US US US US, CT	Acute abdomen. AA AA AA AA AA	LPT. Appendectomy LPT. Appendectomy LPT. Appendectomy LPT. Appendectomy LPT. Appendectomy	Preoperative suspected Intra/postoperative Intra/postoperative Intra/postoperative Intra/postoperative	Mucinous cystadenoma LAMN LAMN LAMN LAMN
Stark et al ^[58]	34 F	US, MRI	Torsion of the appendix	LPSC. Appendectomy	Intra/postoperative	AM
Tarcoveanu et al ^[59] Wani et al ^[60] Zaharie et al ^[61]	60 M 76 M. 39 F	US Abdominal x-ray X-ray	AA AA Intestinal obstruc-	LPSC. Appendectomy Open appendectomy LPT. Appendectomy	Intra/postoperative Intra/postoperative Intra/postoperative	Mucinous cystadenoma Retention AM Mucinous cystadenoma
Xu et al ^[62]	76 M	CT	tion Intestinal obstruc- tion	LPT. Appendectomy.	Intra/postoperative	Mucinous cystadenoma

MRI = magnetic resonance imaging.

The	most	frequent	symptoms	of	the	appendiceal	mucocele
pres	enting	as the ac	ute abdome	n re	eport	ed in the litera	ature.

Symptom	Frequency
Acute abdominal pain	96.6%
Nausea	34.5%
Vomiting	20.7%
Fever	10.3%
Abdominal distension	3.4%
Other	6.9%

and tachycardia); right lower abdominal and adnexal tenderness associated with cervical motion tenderness; absence of involuntary guarding and rebound as well the lack of free peritoneal fluid in a postmenopausal woman.

The most common overlapping features between right-sided TOA and uncomplicated AM include the shared location of pain/ tenderness and fluid filled mass. Of note, uncomplicated AM is mostly asymptomatic, whereas complicated AM (associated with intestinal wall inflammation/necrosis) such as in our patient, is associated with the additional overlapping signs, namely fever, and tachycardia. TOA is less common, but still possible in postmeno-pausal women.^[8] Oppositely, AM is rare, however, more common in postmenopausal than in women of reproductive age.^[9–11]

The diagnosis of AM as opposed to TOA in our patient was limited by the rare occurrence of the disease, multiple overlapping and nonspecific clinical and ultrasonographic features, and lack of preoperative CT/magnetic resonance imaging (MRI) diagnostic imaging results.

An AM case series reported that up to 40% of AMs are misdiagnosed as adnexal masses.^[12] To the best of our knowledge, there are only 22 reports of AM mimicking an adnexal mass in the published literature (Table 1).

Cases occurred in both women of reproductive age and peri-/postmenopausal women. Most of the reported cases were chronic, often presenting with prolonged abdominal/pelvic pain (50%), with or without weight loss (Fig. 2). Acute presentations were reported in 13%, with 2 cases suspected to be ovarian torsion^[9,13,14] and 1 ruptured ovarian cyst.^[27] In contrast to majority of previous reports, our case had an acute presentation. In addition, it presented with fever. All the diagnoses of AM among the 22 reports involving adnexal pathology in the differential diagnosis were presumptive intraoperatively and confirmed by histopathology. The majority of these cases were treated with a simple appendectomy (68%), whereas some extent of bowel resection was employed in 27.2%. The final pathological diagnoses were appendiceal mucocele (50%), followed by LAMN/formerly mucinous cystadenoma (31.8%) with only 2 cases of HAMN/formerly mucinous cystadenocarcinoma or carcinoma in situ.

Furthermore, we conducted a comprehensive literature survey (PubMed/MEDLINE, Google Scholar) on the cases of AM presenting as an acute surgical emergency, these results are summarized in Table 2.

A total of 67 cases were found, mostly presenting with acute abdominal pain (96.6%) (Tables 2 and 3). Among them, only 7.5% were correctly diagnosed prior to surgery. The employed surgical approach for the reported acute AM cases was predominantly laparotomy (91%) and the treatment modality of choice was a simple appendectomy (85.1%). Both, the surgical approach and the treatment of choice, are consistent with our case, including the final

diagnosis of the LAMN, which was also most frequently seen in cases of AM that presented as acute surgical emergency (64.2%). The open surgery proved to be an appropriate therapeutic option not only because of the urgent nature of the condition but also because of the need for careful handling of the appendiceal mass in order to preserve its integrity. Simple appendectomy in our case was appropriate because there was neither appendiceal base nor the lymph node involvement; there was also a lack of free peritoneal fluid. The subsequent pathology report of LAMN reconfirmed the appropriateness of the treatment strategy.

Our case is similar to previously reported ones that lacked specific findings, and in which the diagnosis and management strategy were made intraoperatively. The literature has reported mainly AM cases of chronic abdominal pain as suspected adnexal masses/tumors. However, reports of acute gynecological conditions that resulted in the diagnosis of AM are rare which makes our case valuable.

The limitations of our case report include the failure to perform more frequently used diagnostic imaging methods, such as CT or MRI preoperatively as well as intraoperative frozen tissue section examination. Further diagnostic methods were not applied mainly because of the acuteness of the patient's condition, which required an urgent exploratory laparotomy but also because some of them are were not readily available in our hospital.

In summary, AM in gynecologic and obstetric pathology is a rare occurrence. It is seen more often in chronic than in acute settings. Ours as well as other reported cases raise the awareness of the possibility of existence of an AM instead of adnexal pathology and the need for the implementation of the appropriate treatment strategy to prevent intra- and postoperative complications. The preoperative diagnosis is rare in the literature (15-29%),^[3] and in the acute setting even less (7.5%), because of the lack of a specific clinical presentation.^[11] Imaging methods may be helpful, especially "the onion skin" sign on TVS.^[10,25,63] Even if preoperative diagnosis fails, the frozen tissue section pathology availability to complement the surgical experience and the awareness of the significance of AM can help to manage the patient properly. Careful attention should be employed in order to prevent breakage of the appendiceal contents and to employ more aggressive measures in cases where malignancy is suspected. Furthermore, intraoperative inspection of the adnexa and colon deserves particular scrutiny, since appendiceal mucocele can be related to mucinous tumors of these organs (in 10-20%), especially if associated with pseudomyxoma peritonei.[34] Patients should be referred for postoperative colonoscopic and gynecologic surveillance because of the increased risk of colorectal carcinoma associated with this condition.^[64]

4. Conclusions

Awareness of rare entity such as an AM, which is frequently misdiagnosed as a potential cause of acute abdomen, is necessary. Appropriate diagnosis and management of appendiceal mucocele prevents potential complications including pseudomyxoma peritonei, which has a high mortality rate. Postoperative colonoscopic and gynecologic surveillance of patients is warranted.

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