

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

A case report and comprehensive review of the literature

Hajrunisa Cubro, MD, MSc^a, Vesna Cengic, MD, MSc^b, Nina Burina, MD^c, Zlatko Kravic, MD^d, Esad Beciragic, MD^c, Semir Vranic, MD, PhD^{e,*}

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 tubo-ovarian 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]

Editor: N/A.

The publication of this article was funded by the Qatar National Library.

The authors have no conflicts of interest to disclose.

^a Division of Nephrology and Hypertension, Mayo Clinic, Rochester, Minnesota, ^b Department of Anesthesiology and Critical Care, ^c Department of Obstetrics and Gynecology, ^d Department of General Surgery, General Hospital "Prim.dr. Abdulah Nakas", Sarajevo, Bosnia and Herzegovina, ^e College of Medicine, Qatar University Health, Qatar University, Doha, Qatar.

* Correspondence: Semir Vranic, College of Medicine, Qatar University Health, Qatar University, PO Box 2713, Doha, Qatar (e-mail: semir.vranic@gmail.com, svranic@qu.edu.qa).

Copyright © 2019 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial License 4.0 (CCBY-NC), where it is permissible to download, share, remix, transform, and buildup the work provided it is properly cited. The work cannot be used commercially without permission from the journal.

How to cite this article: Cubro H, Cengic V, Burina N, Kravic Z, Beciragic E, Vranic S. Mucocele of the appendix presenting as an exacerbated chronic tubo-ovarian abscess. *Medicine* 2019;98:39(e17149).

Received: 11 February 2019 / Received in final form: 17 July 2019 / Accepted: 20 August 2019

<http://dx.doi.org/10.1097/MD.00000000000017149>

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

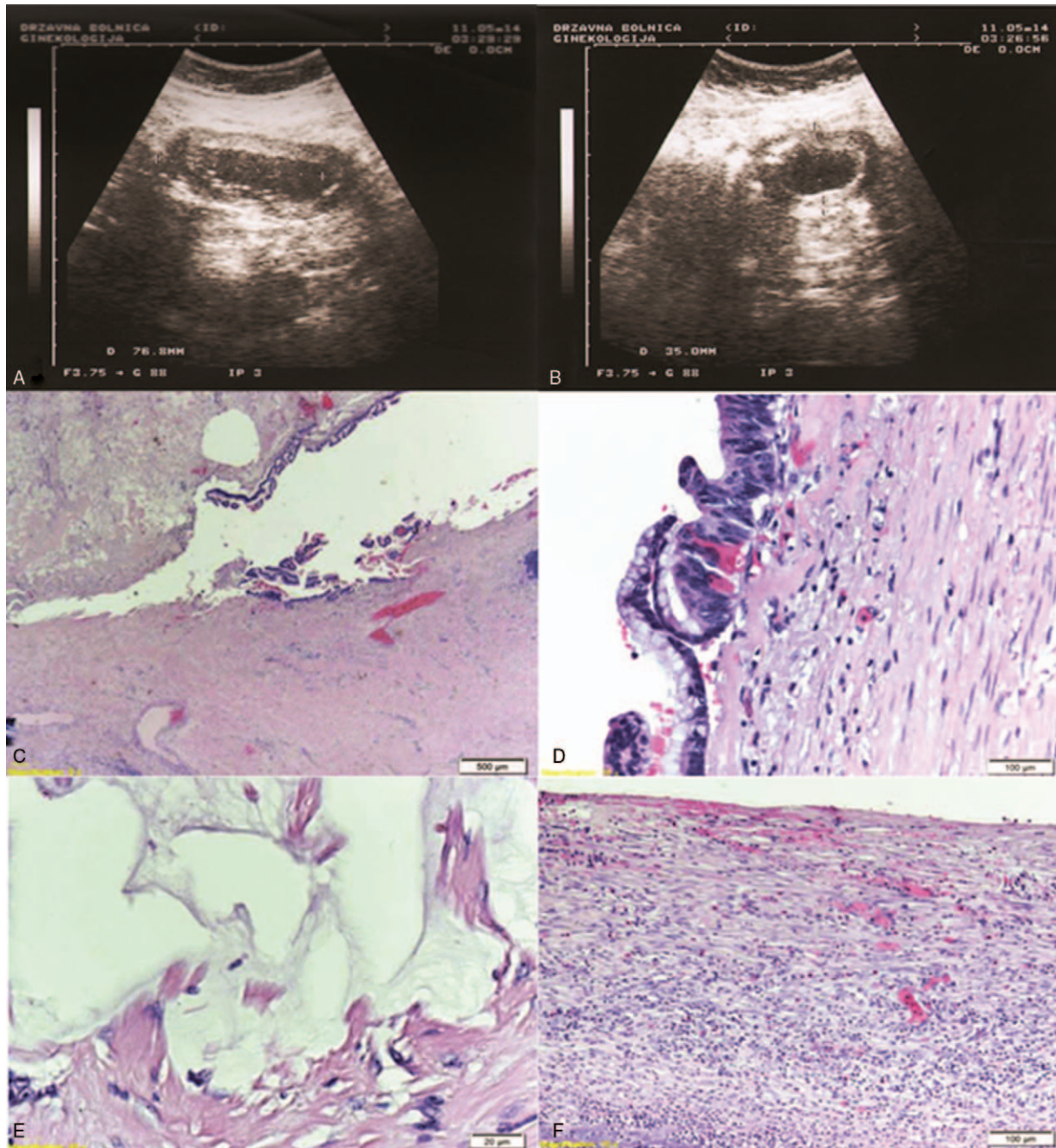


Figure 1. A–B: Transvaginal sonography of the appendiceal mucocele showing ovoid formation with thickened wall, filled with anechogenic material, and no intraluminal proliferations. C: Lumen of the appendix was cystically dilated and contained mucus. The wall of appendix was partly necrotic with dysplastic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

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 $^{\circ}$ 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 112g/L, Hematocrit 0.33), with all other standard lab test parameters within the reference ranges.

Table 1
Review of 22 cases of AM mimicking adnexal mass from the literature.

Authors	Patients characteristics	Imaging method	Preop. Dg.	Treatment option	Timing of the diagnosis	Mucocele type
Abu Zidan et al. ^[13]	-	US	38 weeks pregnant. Torqued ovarian cyst.	LPT. Caesarean section and appendectomy.	Intra/postoperative	Simple AM
Akman et al. ^[9]	81 F	US, MRI	Right adnexa mass.	Appendectomy, R hemicolectomy with ileo-transverse anastomosis TAH.	Intra/postoperative	Mucinous cystadenoma
Arrington et al. ^[14]	15 F	TVS	Ovarian torsion	LPSC. Appendectomy and partial cecotomy.	Intra/postoperative.	Simple AM
Bahia and Wilson ^[15]	46 F	US, Barium enema.	Adnexal mass.	Appendectomy	Intra/postoperative	AM
Balci et al. ^[10]	71 F	TVS, MRI	Ovarian cyst.	LPT. Appendectomy. TAH	Intra/postoperative	AM
Bartlet et al. ^[11]	80 F	US, CT	Ovarian cyst.	LPT. Appendectomy.	Intra/postoperative	Mucinous cystadenoma
Cristian et al. ^[16]	61 F	US, CT	Hydrosalpinx or ovarian cyst.	LPSC. Appendectomy.	Intra/postoperative.	LAMN
Dragoumis et al. ^[17]	70 F	TVS, CT.	Ovarian cystic tumor.	TAH with bilateral salpingo-oophorectomy. Appendectomy.	Intra/postoperative	Mucinous cystadenoma
Driman et al. ^[18]	34 F.	X-ray colonoscopy, CT.	Adnexal mass	R hemicolectomy, ovarian cystectomy, and removal of the mucinous cysts from the cul-de-sac.	Intra/postoperative	Foci of endometriosis in the proximal appendix and an AM
	31 F	Explorative LPSC	Endometriosis.	Laser excision of the endometrioid deposits and appendectomy.		Pelvic endometriosis. AM
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 adenocarcinoma
Hutchinson et al. ^[21]	47 F.	US, CT, MRI.	Hydrosalpinx, AM, Tumor.	Reoperation. Hemicolectomy.	Intra/postoperative.	Carcinoma in situ
Kalu and Croucher. ^[22]	42 F	TVS	Cystic adnexal mass	Surgical. Approach unclear. Appendectomy	Intra/postoperative	AM
O'Sullivan et al. ^[23]	31 F	IWU, US, and CT.	Hydronephrosis, hydroureter.	Excision of the caecum and terminal ileum with an ileocolic anastomosis, limited ureterolysis and a ureteric stent inserted through an ureterotomy.	Intra/postoperative.	Endometriosis of the appendix resulting in AM and endometriosis of the terminal ileum.
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 tumor.	Appendectomy. TAH.	Intra/postoperative	Mucinous cystadenoma
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 tumor.	Open appendectomy.	Intra/postoperative	Mucinous cystadenoma
Yildiz and Abassoglu ^[30]	54 F	TAS, CT, colonoscopy.	Ovarian mass.	Appendectomy	Intra/postoperative	AM

MRI = magnetic resonance imaging.

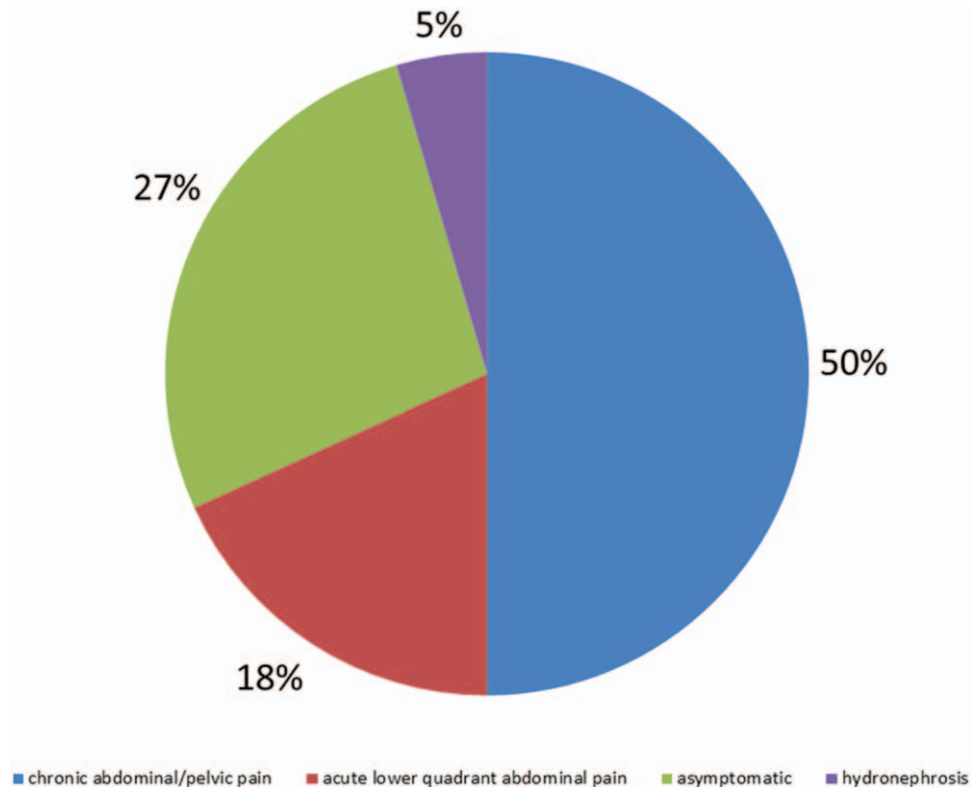


Figure 2. Most common symptoms of AM mimicking adnexal mass described in literature. AM=appendiceal mucocele.

Trans-vaginal ultrasonography (TVS) showed a well-defined, encapsulated, oval, unilocular mass just above the uterus with a heteroechoic 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 × 7.5 × 6 cm, was diffusely dilated, filled with fecal contents, entailing 2 concerns 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 intra-operatively 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.^{17]}

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**Cases of AM presenting with the acute abdomen.**

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	US	AA	LPT. Appendectomy	Intra/postoperative	Mucinous cystadenoma
	31 F	US	AA	LPT. Appendectomy	Intra/postoperative	Neuroendocrine tumor and retention cyst
	39 F	US, CT	AA	LPT. Appendectomy	Intra/postoperative	Retention cyst
	42 M	CT	AA	LPT. R hemicolectomy	Intra/postoperative	Mucinous cystadenoma, pseudomyxoma peritonei.
Bestman et al ^[33]	49 F	US, CT	AA	LPT. Appendectomy	Intra/postoperative	Mucinous cystadenoma
	61 F.	US, CT	AA	LPT. Appendectomy	Intra/postoperative	Mucinous cystadenoma
	62 M	US, CT	AA	LPT. Appendectomy	Intra/postoperative	Mucinous cystadenoma
	76 M	US	AA.	LPT. Appendectomy	Intra/postoperative	Mucinous cystadenoma
	35 F	Abdominal x-ray.	AA	LPSC. Appendectomy	Intra/postoperative	Retention AM
Caliskan et al ^[34]	11 patients (2 F/6 M), median age 70.	CT, US	6 AA. 2 intestinal obstruction.	Emergency surgery. Appendectomy 5/8. Small bowel/sigmoid colon resection/hemicolectomy 3/8.	Intra/postoperative	AM
Casey et al ^[35]	36 F	US	AA, 21 weeks gestation	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]	54 M	US	AA	LPT. Appendectomy.	Intra/postoperative	Mucinous cystadenoma
Fatima Ezzahra et al ^[38]	55 M	-	AA	Unclear. Appendectomy.	Intra/postoperative	Mucinous cystadenoma
Hamada et al ^[39]	79 M	US	Torqued AM	LPT. Appendectomy.	Preoperative	Mucinous cystadenoma
Hebert and Pickhardt ^[40]	59 M	CT	Torqued AM	LPT. Appendectomy.	Preoperative	Mucinous cystadenoma
Idris et al ^[41]	35 F. Pregnant	US	AA	LPT. Appendectomy.	Intra/postoperative	AM
Karakaya et al ^[42]	82 M	US, CT.	AA.	LPT. Appendectomy.	Intra/postoperative	Mucinous cystadenoma.
	65 F	Abdominal x-ray, US, CT	Ileus	LPT. Ileal and cecal mesenteric ischemia.	Intra/postoperative	Simple mucocele.
Kehagias et al ^[43]	72 F	CT	Acute abdomen	LPT. R Hemicolectomy.	Intra/postoperative	LAMN
Kelemouridou et al ^[44]	71 M	Imaging	AA	Open appendectomy.	Intra/postoperative	Mucinous cystadenoma
Kilic et al. ^[45]	52 F	Unclear	AA	LPSC. Appendectomy.	Intra/postoperative	LAMN
Kimura et al ^[46]	41 F	Ba enema	Acute abdomen	LPT. Appendectomy.	Intra/postoperative	Mucosal hyperplasia in AM
Kitagawa et al ^[47]	34 M	CT	Inflamed appendiceal tumor	LPT. Appendectomy.	Intra/postoperative	Mucinous cystadenoma
Komo et al ^[48]	79 F	CT	Mechanical small intestinal obstruction.	LPT. Appendectomy.	Intra/postoperative	LAMN
Lee et al ^[49]	78 F	CT	AM with periappendicitis.	LPT. Appendectomy	Preoperative	AM
Malasi et al ^[50]	58 M	CT	Ruptured AM.	LPT. Appendectomy.	Preoperative	AM
Malya et al ^[51]	30 M	US, CT	AA	LPT. Appendectomy.	Intra/postoperative	AM
	56M	US, CT.	AA	LPT. Appendectomy.	Intra/postoperative	AM
	39 M	US, CT.	AA	LPT. Appendectomy.	Intra/postoperative	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. Intussusception.	Laparoscopic appendectomy	Intra/postoperative	AM
Sertkaya et al ^[56]	61 F	US, CT	Acute abdomen.	LPT. Appendectomy	Preoperative suspected	Mucinous cystadenoma
Singal et al ^[57]	23 F	US	AA	LPT. Appendectomy	Intra/postoperative	LAMN
	46 F	US	AA	LPT. Appendectomy	Intra/postoperative	LAMN
	57 M	US	AA	LPT. Appendectomy	Intra/postoperative	LAMN
	35 M	US, CT	AA	LPT. Appendectomy	Intra/postoperative	LAMN
Stark et al ^[58]	34 F	US, MRI	Torsion of the appendix	LPSC. Appendectomy	Intra/postoperative	AM
Tarcoveanu et al ^[59]	60 M	US	AA	LPSC. Appendectomy	Intra/postoperative	Mucinous cystadenoma
Wani et al ^[60]	76 M.	Abdominal x-ray	AA	Open appendectomy	Intra/postoperative	Retention AM
Zaharie et al ^[61]	39 F	X-ray	Intestinal obstruction	LPT. Appendectomy	Intra/postoperative	Mucinous cystadenoma
Xu et al ^[62]	76 M	CT	Intestinal obstruction	LPT. Appendectomy.	Intra/postoperative	Mucinous cystadenoma

MRI=magnetic resonance imaging.

Table 3
The most frequent symptoms of the appendiceal mucocele presenting as the acute abdomen reported in the literature.

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 postmenopausal 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%),^[13] 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.

Acknowledgments

The authors thank Allan Ackerman for the proofreading of the manuscript and suggestions about improvement of the style of writing.

Author contributions

Conceptualization: Hajrunisa Cubro, Vesna Cengic, Semir Vranic.

Data curation: Nina Burina, Zlatko Kravic, Esad Beciragic.

Formal analysis: Hajrunisa Cubro, Vesna Cengic, Nina Burina, Semir Vranic.

Investigation: Nina Burina, Esad Beciragic.

Writing – original draft: Hajrunisa Cubro, Vesna Cengic, Nina Burina, Semir Vranic.

Writing – review & editing: Semir Vranic.

Semir Vranic orcid: 0000-0001-9743-7265.

References

- O'Brien T. Top 3 Differentials in Radiology: A Case Review. Stuttgart, Germany: Thieme; 2011.
- Athanassiou E, Spyridakis M, Karasavvidou F, et al. Low-grade appendiceal mucinous neoplasm presenting as a surgical emergency: a case report. *Case Rep Oncol* 2009;2:7–11.
- Ruiz-Tovar J, Teruel DG, Castiñeiras VM, et al. Mucocele of the appendix. *World J Surg* 2007;31:542–8.
- Aho AJ, Heinonen R, Lauren P. Benign and malignant mucocele of the appendix. Histological types and prognosis. *Acta Chir Scand* 1973;139:392–400.
- Pai RK, Longacre TA. Appendiceal mucinous tumors and pseudomyxoma peritonei: histologic features, diagnostic problems, and proposed classification. *Adv Anat Pathol* 2005;12:291–311.
- Carr NJ, Cecil TD, Mohamed F, et al. A Consensus for classification and pathologic reporting of pseudomyxoma peritonei and associated appendiceal neoplasia: the results of the Peritoneal Surface Oncology Group International (PSOGI) Modified Delphi Process. *Am J Surg Pathol* 2016;40:14–26.
- Kruszka PS, Kruszka SJ. Evaluation of acute pelvic pain in women. *Am Fam Physician* 2010;82:141–7.
- Protopapas AG, Diakomanolis ES, Milingos SD, et al. Tubo-ovarian abscesses in postmenopausal women: gynecological malignancy until proven otherwise? *Eur J Obstet Gynecol Reprod Biol* 2004;114:203–9.
- Akman L, Hursitoglu BS, Hortu I, et al. Large mucinous neoplasm of the appendix mimicking adnexal mass in a postmenopausal woman. *Int J Surg Case Rep* 2014;5:1265–7.
- Balci O, Ozdemir S, Mahmoud AS. Appendiceal mucocele mimicking a cystic right adnexal mass. *Taiwan J Obstet Gynecol* 2009;48:412–4.
- Bartlett C, Manoharan M, Jackson A. Mucocele of the appendix - a diagnostic dilemma: a case report. *J Med Case Rep* 2007;1:183.
- Chen J, Zhu L, Wu B. [Appendiceal mucocele mimicking right adnexal mass: a report of 25 cases]. *Zhonghua Yi Xue Za Zhi* 2011;91:1637–9.
- Abu Zidan FM, al-Hilaly MA, al-Atrabi N. Torsion of a mucocele of the appendix in a pregnant woman. *Acta Obstet Gynecol Scand* 1992; 71:140–2.
- Arrington D, Jewett B, Sterner S, et al. Incidental mucocele of the appendix in a 15-year-old girl. *Pediatr Emerg Care* 2014;30:555–7.
- Bahia JO, Wilson MH. Mucocele of the appendix presenting as an adnexal mass. *J Clin Ultrasound* 1989;17:62–6.
- Cristian DA, Grama FA, Becheanu G, et al. Low-grade appendiceal mucinous neoplasm mimicking an adnexal mass. *Rom J Morphol Embryol* 2015;56(2 suppl):837–42.
- Dragoumis K, Mikos T, Zafrakas M, et al. Mucocele of the vermiform appendix with sonographic appearance of an adnexal mass. *Gynecol Obstet Invest* 2005;59:162–4.
- Driman DK, Melega DE, Vilos GA, et al. Mucocele of the appendix secondary to endometriosis. Report of two cases, one with localized pseudomyxoma peritonei. *Am J Clin Pathol* 2000;113:860–4.
- Gortchev G, Tomov S, Dimitrov D, et al. Appendiceal mucocele presenting as a right adnexal mass: a case report. *Obstet Gynecol Int* 2010;2010:pii: 281053.
- Hajiran A, Baker K, Jain P, et al. Case of an appendiceal mucinous adenocarcinoma presenting as a left adnexal mass. *Int J Surg Case Rep* 2014;5:172–4.
- Hutchinson C, Lyske J, Patel V, et al. Mucinous neoplasm of the appendix as a mimic of cystic adnexal pathology. *J Clin Imaging Sci* 2018;8:32.
- Kalu E, Croucher C. Appendiceal mucocele: a rare differential diagnosis of a cystic right adnexal mass. *Arch Gynecol Obstet* 2005;271:86–8.
- O'Sullivan MJ, Kumar U, Kiely EA. Ureteric obstruction with mucocele of the appendix due to endometriosis. *BJOG* 2001;108:124–5.
- Ortiz-Mendoza CM, Dircio AC. A rare tumor resembling ovarian cysts: mucocele of the appendix. *Int J Gynecol Obstet Res* 2013;1:1–3.
- Paladino E, Bellantone M, Conway F, et al. Large mucocele of the appendix at laparoscopy presenting as an adnexal mass in a postmenopausal woman: a case report. *Case Rep Obstet Gynecol* 2014;2014:486078.
- Papoutsis D, Protopappas A, Belitsos P, et al. Mucocele of the vermiform appendix misdiagnosed as an adnexal mass on transvaginal sonography. *J Clin Ultrasound* 2012;40:522–5.
- Rudloff U, Malhotra S. Volvulus of an appendiceal mucocele: report of a case. *Surg Today* 2007;37:514–7.
- Scaffa C, Di Bella O, Tartaglia E, et al. Surgical approach to appendiceal mucocele mimicking an adnexal complex mass: case report. *Eur J Gynaecol Oncol* 2007;28:503–5.
- Shimizu T, Shimizu M, Kawaguchi K, et al. Mucinous cystadenoma of the appendix with raised serum carcinoembryonic antigen concentration: clinical and pathological features. *J Clin Pathol* 1997; 50:613–4.
- Yildiz BD, Abbasoglu O. Two unusual presentations of appendiceal masses. *Int J Surg* 2008;6:e9–11.
- Abuoglu H, Yildiz MK, Kaya B, et al. Clinicopathological analysis of patients operated for appendiceal mucocele. *Ulus Travma Acil Cerrahi Derg* 2017;23:230–4.
- Basak F, Hasbahceci M, Yucel M, et al. Does it matter if it is appendix mucocele instead of appendicitis? Case series and brief review of literature. *J Cancer Res Ther* 2018;14:1355–60.
- Bestman TJ, van Cleemput M, Detournay G. Torsion of the vermiform appendix: a case report. *Acta Chir Belg* 2006;106:228–9.
- Caliskan K, Yildirim S, Bal N, et al. Mucinous cystadenoma of the appendix: a rare cause of acute abdomen. *Ulus Travma Acil Cerrahi Derg* 2008;14:303–7.
- Casey RG, Tan M, Salman R, et al. Acute abdomen in pregnancy due to mucinous cystadenoma of the appendix. *J Obstet Gynaecol* 2003;23: 566–7.
- Costa V, Demuro JP. Low-grade appendiceal neoplasm presenting as a volvulus of the cecum. *Gastroenterol Rep (Oxf)* 2013;1:207–10.
- Demetashvili Z, Chkhaidze M, Khutsishvili K, et al. Mucocele of the appendix: case report and review of literature. *Int Surg* 2012;97: 266–9.
- Fatima Ezzahra Z, Ibn Majdoub K, Hicham A, et al. Uncommon mimics of appendicitis: giant mucocele. *Pan Afr Med J* 2010;7:7.
- Hamada T, Kosaka K, Shigeoka N, et al. Torsion of the appendix secondary to appendiceal mucocele: gray scale and contrast-enhanced sonographic findings. *J Ultrasound Med* 2007;26:111–5.
- Hebert JJ, Pickhardt PJ. MDCT diagnosis of an appendiceal mucocele with acute torsion. *AJR Am J Roentgenol* 2007;189:W4–6.
- Idris LO, Olaofe OO, Adejumbi OM, et al. Giant mucocele of the appendix in pregnancy: a case report and review of literature. *Int J Surg Case Rep* 2015;9:95–7.
- Karakaya K, Barut F, Emre AU, et al. Appendiceal mucocele: case reports and review of current literature. *World J Gastroenterol* 2008; 14:2280–3.
- Kehagias I, Zygomalas A, Markopoulos G, et al. Diagnosis and treatment of mucinous appendiceal neoplasm presented as acute appendicitis. *Case Rep Oncol Med* 2016;2016:3612014.
- Kelemouridou E, Mogrampi SA, Tsavis G, et al. Mucinous cystadenoma of the appendix. A diagnostic dilemma? *Chirurgia (Bucur)* 2011;106: 251–4.
- Kilic MO, Inan A, Bozer M. Four mucinous cystadenoma of the appendix treated by different approaches. *Ulus Cerrahi Derg* 2014;30: 97–9.
- Kimura H, Konishi K, Yabushita K, et al. Intussusception of a mucocele of the appendix secondary to an obstruction by endometriosis: report of a case. *Surg Today* 1999;29:629–32.
- Kitagawa M, Kotani T, Yamano T, et al. Secondary torsion of vermiform appendix with mucinous cystadenoma. *Case Rep Gastroenterol* 2007;1:32–7.
- Komo T, Kohashi T, Hihara J, et al. Intestinal obstruction caused by low-grade appendiceal mucinous neoplasm: a case report and review of the literature. *Int J Surg Case Rep* 2018;51:37–40.

- [49] Lee CH, Lee MR, Kim JC, et al. Torsion of a mucocele of the vermiform appendix: a case report and review of the literature. *J Korean Surg Soc* 2011;81(suppl):S47–50.
- [50] Malasi S, Kadi R, Haven F, et al. An acute abdomen mimicking appendicitis. *J Belg Soc Radiol* 2016;100:11.
- [51] Malya FU, Hasbahceci M, Serter A, et al. Appendiceal mucocele: clinical and imaging features of 14 cases. *Chirurgia (Bucur)* 2014;109:788–93.
- [52] Mishin I, Ghidirim G, Zastavitsky G, et al. Torsion of an appendiceal mucinous cystadenoma. Report of a case and review of literature. *Ann Ital Chir* 2012;83:75–8.
- [53] Nopajaroonsri C, Mreyoud N. Retention mucocele of appendix due to endometriosis. *South Med J* 1994;87:833–5.
- [54] Opreanu RC, Sobinsky J, Basson MD. Appendicitis and benign appendiceal mucocele presenting as large bowel obstruction. *J Gastrointest Surg* 2013;17:609–10.
- [55] Park JK, Kwon TH, Kim HK, et al. Adult intussusception caused by an appendiceal mucocele and reduced by colonoscopy. *Clin Endosc* 2011;44:133–6.
- [56] Sertkaya M, Emre A, Pircanoglu EM, et al. Giant appendicular mucocele due to mucinous cystadenoma. *Euroasian J Hepatogastroenterol* 2016; 6:186–9.
- [57] Singal R, Zaman M, Sharma BP. Unusual entities of appendix mimicking appendicitis clinically - emphasis on diagnosis and treatment. *Maedica (Buchar)* 2017;12:23–9.
- [58] Stark C, Jousi M, Enholm B. Preoperative assessment and treatment of appendiceal mucocele complicated by acute torsion: a case report. *BMC Res Notes* 2014;7:1.
- [59] Tarcoveanu E, Vasilescu A, Hee RV, et al. Appendicular mucocele: possibilities and limits of laparoscopy. brief series and review of the literature. *Chirurgia (Bucur)* 2015;110:530–7.
- [60] Wani I, Kitagawa M, Rather M, et al. Torsion of vermiform appendix with fecalith: a case report. *Cases J* 2008;1:20.
- [61] Zaharie F, Tomus C, Mocan L, et al. Intestinal obstruction secondary to appendiceal mucocele. *Chirurgia (Bucur)* 2012;107:802–4.
- [62] Xu ZS, Xu W, Ying JQ, et al. Mechanical intestinal obstruction secondary to appendiceal mucinous cystadenoma: A case report and brief review. *Medicine (Baltimore)* 2017;96:e6016.
- [63] Caspi B, Cassif E, Auslender R, et al. The onion skin sign: a specific sonographic marker of appendiceal mucocele. *J Ultrasound Med* 2004; 23:117–21. quiz 122-113.
- [64] Federle MPAV. Mucocele of the appendix. In: *Diagnostic Imaging Abdomen*. Salt Lake City: Amirsys; 2004.