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# Appendicular Mucocele: About a Case Observed in Bamako

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## **Abstract**

It is a globally rare condition that can present in a variety of clinical syndromes or can occur as an incidental surgical finding. It poses the double problem of its possible malignancy and the risk of gelatinous disease of the peritoneum in the event of perforation. We report a case treated in the "A" Surgery Department of the Point-G University Hospital Center (CHU) in Mali in 2022. It was a 62-year-old woman, a housewife who presented to the department. "A" surgery at the Point G University Hospital Center for pain in the iliac fossa. As a medical history, she was hypertensive on atenolol and a known diabetic on diet and metformin-based treatment, as well as symptomatic sickle cell disease (AS) and an undocumented history of peptic ulcer disease. The biological assessments revealed hyperleukocyte with granulocyte predominance. C-reactive protein was positive at 32 mg/l. Ultrasound revealed a 27 mm cystic dilation of the appendicitis in favor of appendicular mucocele. We proceeded with the appendix. The surgical specimen containing gelatinous fluid was removed and histological examination was in favor of a mucinous adenocarcinoma of the appendix.

# **Keywords**

Appendicular Mucocele, Appendicular Tumor, Bamako

## 1. Introduction

Appendicular mucocele or appendicular mucus-secreting tumor is defined as fluid distension of the appendicular lumen by accumulation of mucus [1]. It is a globally rare condition that can present in a variety of clinical syndromes or can occur as an incidental surgical finding [2]. Appendicular mucocele has been reported to have an incidence of 0.2% - 0.6% in all appendicitis specimens [1] [3]

[4]. And 8% to 10% of all appendicular tumors [5] [6] [7].

Appendicular mucocele is asymptomatic in 25% to 30% of cases [1] and is manifested by chronic pain in the right iliac fossa in 70% to 75% of cases [8]. The anatomic location of appendicular mucocele includes it in the differential diagnosis of masses in the right lower quadrant of the abdomen [2].

A good diagnosis is necessary to better adapt the surgical procedure. Imaging plays an important role in this diagnosis which is confirmed by histological study, however it is difficult on imaging studies [1] [9]. Appendicular mucocele can be difficult to differentiate from an adnexal mass even on good imaging. Abdominal CT is a better diagnostic tool [10] [11].

The correct management depends on the size and location of the lesion. Its treatment ranges from simple appendectomy in benign forms to right hemicolectomy for cancer in malignant mucoceles [1] [8]. Appendicular mucocele poses the double problem of its possible malignancy and the risk of gelatinous disease of the peritoneum in the event of perforation. We report a case treated in the "A" Surgery Department of the University Hospital Center (CHU) of Point-G in Mali in 2022.

#### 2. Case Presentation

We present the case of a 62-year-old woman, a housewife who presented to the "A" surgery department of the Point G university hospital center for pain in the iliac fossa. She had a history of nausea and vomiting, fever, constipation and tingling sensation. His general condition was altered with weight loss and asthenia. As a medical history, she was hypertensive on atenolol and a known diabetic on diet and metformin-based treatment, as well as symptomatic sickle cell disease (AS) and an undocumented history of peptic ulcer disease.

On admission, the biological examinations, in particular the blood count, there was hyperleukocyte (white blood cell =  $11.8 \times 10^3 / \text{cm}^3$ ) with granulocyte predominance (82.3%). The hemoglobin level was normal at (12.5 g/dl). Liver (ASAT = 30.0 and ALAT = 19.5) and kidney (Urea = 5.66 mmol, creatinine = 109.35  $\mu$ m/L) balance sheets were all normal. C-reactive protein was positive at 32 mg/l. The results of the biological parameters are represented in **Table 1**.

Table 1	<ul> <li>Results</li> </ul>	of biological	parameters.

Biological parameters	Value
White globule	$11.8 \times 10^{3}$ /mm <sup>3</sup>
Red blood cell	$4.28\times10^6/mm^3$
Hemoglobin	12.5 g/dl
Hematocrit	36.5%
Platelets	$346\times10^3/\text{mm}^3$
Granulocytes	82.3%
Lymphocytes	11.8%
Monocytes	5.9%

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VGM	85		
TGMH	29.2		
MCHC	34.2		
AST	30.0		
ALT	19.5		
Urea	5.66 mmol		
Creatinine	109.35 μm/L		
C-reactive protein positive	32 mg/l		

Ultrasound revealed, at the level of the right iliac fossa, a cystic dilation of appendicitis of 27 mm, giving an ultrasound appearance in favor of appendicular mucocele.



The patient received surgery under general anesthesia plus orotrachial intubation. Part in dorsal decubitus we made an incision passing through McBurney's point at the celiotomy, the exploration found a bulky appendix, dilated and hard in favor of an appendicular tumour. We proceeded with the appendix. The surgical specimen containing gelatinous fluid was extracted (**Figure 1**) and pathological examination was done.

After resections of the parts and re-readings, we noted a tumor focus. It consists of tubes and clusters of columnar cells showing cytonular atypia and containing mucus. The stroma is myxoid and inflammatory. Histological appearance compatible with a mucinous adenocarcinoma of the appendix (Figure 2). As part of the patient's follow-up, a CT-TAP was performed postoperatively, which did not reveal any secondary localization.



Figure 1. Appendectomy specimen containing gelatinous fluid.

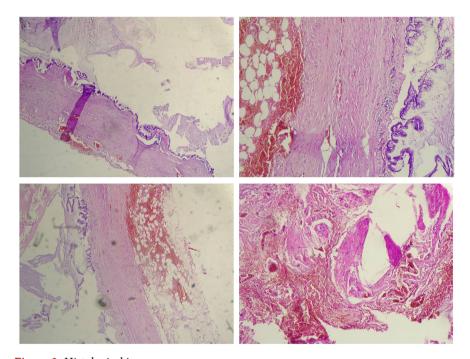


Figure 2. Histological images.

# 3. Discussion

An appendicular mucocele was first described by Rokitansky in 1842 and characterized in 1973 by Aho *et al.* [12]. Appendix mucocele refers to dilation of the appendix with accumulation of mucin [13]. It preferentially affects adults with an average age between 50 and 60 years [2] [9] [14] as we noted in our observation. Previous studies have reported the male to female ratio to be 3 - 4 with an average age of 55 at diagnosis [15].

Regarding sex, it was a woman in our observation. According to the literature, the pathology is four times more frequent in women and generally detected in patients aged < 50 years [16]. The sex ratio is variable from one series to another and would rather be in favor of a female predominance in the latest studies [9] [17]. In a retrospective study of 135 surgically resected patients, 55% were women [18]. On the other hand, a male predominance was noted by Souei-Mhiri [19], in his study planned between 1991 and 1998 in Sousse, which found 6 men against 4 women in a series of 10 cases.

The clinic may be non-specific and according to the literature up to 50% of patients with mucocele may be asymptomatic [20]. The clinical presentation of mucoceles is usually vague, but most often presents with pain in the right lower quadrant [13]. In our study the patient suffered from pain in the iliac fossa. In their study by Wakunga E *et al.* [1] reported a female case suffering from right iliac fossa pain of moderate intensity, in the form of torsion, without timing, not relieved by average analgesics, radiating to the iliac fossa left and right leg moving.

Therefore, radiological imaging is most often sensitive. Ultrasound and CT are the most used, generally documenting a well-encapsulated cystic mass in the right lower quadrant of the abdomen, often associated with mural calcifications [20] [21].

Our case only performed the abdominal ultrasound which objectified a cystic mass of 27 mm in the right iliac fossa. The differential diagnosis could be very important, some patients may have an erroneous diagnosis of appendicitis. According to the study by Lien W et al. [22] in 2006, an appendicular diameter greater than 15 mm documented by ultrasound is correlated with a mucocele with a sensitivity of 83% and a specificity of 92%. Consistent with these results, Saylam et al. confirmed that an appendicitis outer diameter greater than 10 mm on ultrasound and a mean red blood cell count less than 11,000/mm³ were associated with appendicular mucocele. To allow a significant differential diagnosis, the radiologist must give the morphological characteristics and the content of the lesion, but also evaluate its relationship with the main anatomical landmarks of the abdomen and the pelvis in order to determine its origin [23]. CT imaging with contrast is the most commonly used modality for preoperative diagnosis. It clarifies the choice of procedure and avoids complications [7].

On CT scan, it appears as a mass with a cecal base, rounded and well limited, with a thin wall, with fine parietal calcifications. Its wall can be thickened, irregular, with nodules taking the contrast, directing towards a cystadenocarcinoma; however, there are no radiological signs to confirm or exclude with certainty the malignancy of the underlying appendicular tumor [24] [25].

Due to a lack of financial means, we were unable to do a scan. Hence the importance of medical imaging and patented operators to properly guide the clinician in order to arrive at an accurate preoperative diagnosis, thus determining the management, because the rupture of an appendicular mucocele, whatever the stage, in the peritoneal cavity leads to a peritoneal pseudomyxoma also called "gelatinous disease of the peritoneum".

The confirmatory diagnosis is the anatomopathologic examination. Histologically there are four causal pathological conditions according to literature such as: retention cyst, mucosal hyperplasia, cystadenoma and cystadenocarnoma [2] [26]. More than half of mucinous tumors (52% to 58%) are considered malignant tumors of the appendix [7] [27] [28]. In simple mucocele (18%) (inflammatory, obstructive or retention cyst)—degenerative epithelial changes and results in obstruction and distension of the appendix. There is no evidence of mu-

cosal hyperplasia or atypia. In hyperplastic mucocele (20%), dilation of the appendix occurs due to hyperplastic growth of the appendix or cecal mucosa, as do hyperplastic polyps of the colon. Mucinous cystadenoma (52%) is an appendicular tumor with dysplastic epithelium similar to adenomatous polyps of the colon, and mucinous cystadenocarcinoma (10%) has high-grade cellular dysplasia and stromal invasion, in addition to mucosal muscle.

Several strategies have been recommended in the management of appendicular mucocele in previous case reports, and the choice of surgery has varied from simple appendectomy to right hemicolectomy. We proceeded to a simple appendectomy in the patient associated with an exploration which revealed a bulky appendix, dilated and hard in favor of an appendicular tumor containing gelatinous liquid. Dhage-Ivatury and Sugarbaker [20] suggested that laparotomy for thorough exploration would be necessary with surgical treatment of appendicular mucocele to decrease the risk of laparoscopic rupture. However, Morano *et al.* [29] suggested laparoscopic surgery as a safe and effective technique for removal of appendicular mucocele.

Even if some controversies remain as to the clinical and histopathological definition of pseudomixoma peritoneal syndrome, this condition requires a multidisciplinary approach in a reference center, because these treatments are concerned by high morbidity; Five-year survival rates have been reported ranging from 50% to 96% [20]. In our case, no secondary localization was found on the control CT scan.

### 4. Conclusion

Appendicular mucoceles are rare conditions and often pose a diagnostic challenge, in particular they can be confused with a cystic lesion of the right ovary. The clinical presentation is often nonspecific and the clinician should be aware of appendicular mucocele in patients presenting with chronic right lower quadrant pain, adnexal masses, and a picture of acute appendicitis. Diagnosis is more often incidental during imaging or surgery. Surgical resection is potentially curative and rupture of the mucocele should be avoided as it can lead to pseudomyxoma peritonei, a condition with high morbidity and mortality. Exercise due diligence when choosing the surgical approach and procedure.

## **Conflicts of Interest**

The authors declare no conflicts of interest regarding the publication of this paper.

# References

- [1] Wakunga, E., Mukuku, O., Bugeme, M., Tshiband, M., Kipili, A., Mobambo, P., *et al.* (2014) Mucocèle appendiculaire: à propos d'un cas observé à Lubumbashi. *Pan African Medical Journal*, **18**, 36.
- [2] Lakatos, P.L., Gyori, G., Halasz, J., Fuszek, P., Papp, J., Jaray, B., *et al.* (2005) Mucocele of the Appendix: An Unusual Cause of Lower Abdominal Pain in a Patient with

- Ulcerative Colitis: A Case Report and Review of Literature. *World Journal of Gastroenterology*, **11**, 457-459. <a href="https://doi.org/10.3748/wjg.v11.i3.457">https://doi.org/10.3748/wjg.v11.i3.457</a>
- [3] Lorenzon, L., De Dominicis, C., Virgilio, E. and Balducci, G. (2015) The Appropriate Management of an Appendix Mucocele. *BMJ Case Reports*, 2015, bcr2014209045. https://doi.org/10.1136/bcr-2014-209045
- [4] Yakan, S., Caliskan, C., Uguz, A., Korkut, M.A. and Çoker, A. (2011) A Retrospective Study on Mucocele of the Appendix Presented with Acute Abdomen or Acute Appendicitis. *Hong Kong Journal of Emergency Medicine*, 18, 144-149. https://doi.org/10.1177/102490791101800303
- [5] Abuoğlu, H., Yıldız, M.K., Kaya, B. and Odabaşı, M. (2017) Clinicopathological Analysis of Patients Operated for Appendix Mucocele. *Ulusal Travma ve Acil Cerrahi Dergisi-Turkish Journal of Trauma & Emergency Surgery*, **23**, 230-234.
- [6] Salemis, N.S., Nakos, G., Katikaridis, I. and Zografidis, A. (2016) Synchronous Occurrence of Appendix Mucinous Cystadenoma, with Colon Adenocarcinoma and Tubulovillous Rectal Adenoma: Management and Review of the Literature. *Journal of Natural Science, Biology and Medicine*, 7, 173-175. https://doi.org/10.4103/0976-9668.184705
- [7] Motsumi, M.J., Motlaleselelo, P., Ayane, G., Sesay, S.O. and Valdes, J.R. (2017) A Case Report of a Giant Appendix Mucocele and Literature Review. *Pan African Medical Journal*, 28, Article 106. <a href="https://doi.org/10.11604/pamj.2017.28.106.13832">https://doi.org/10.11604/pamj.2017.28.106.13832</a>
- [8] Moujahid, M., Ait, A.A., Achour, A. and Janati, M.I. (2010) Mucocèle appendiculaire: à propos de dix cas. *African Journal of Cancer*, **2**, 107-111. https://doi.org/10.1007/s12558-010-0087-z
- [9] Caspi, B., Cassif, E., Auslender, R., Herman, A., Hagay, Z. and Appelman, Z. (2004) The Onion Skin Sign: A Specific Sonographic Marker of Appendix Mucocele. *Journal of Ultrasound in Medicine*, 23, 117-121. https://doi.org/10.7863/jum.2004.23.1.117
- [10] Lynch, K., Cho, S., Andres, R., Knight, J. and Con, J. (2016) Pre-Operative Identification and Surgical Management of the Appendiceal Mucocele: A Case Report. World Medical Journal, 112, 28-30.
- [11] Panagopoulos, P., Tsokaki, T., Misiakos, E., Domi, V., Christodoulaki, C., Sioutis, D., et al. (2017) Low-Grade Appendiceal Mucinous Neoplasm Presenting as an Adnexal Mass. Case Reports in Obstetrics and Gynecology, 2017, Article ID: 7165321. https://doi.org/10.1155/2017/7165321
- [12] Aho, A.J., Heinonen, R. and Laurén, P. (1973) Benign and Malignant Mucocele of the Appendix. Histological Types and Prognosis. *Acta Chirurgica Scandinavica*, **139**, 392-400.
- [13] Rabie, M.E., Al Shraim, M., Al Skaini, M.S., Alqahtani, S., El Hakeem, I., Al Qahtani, A.S., et al. (2015) Mucus Containing Cystic Lesions "Mucocele" of the Appendix: The Unresolved Issues. *International Journal of Surgical Oncology*, 2015, Article ID: 139461. <a href="https://doi.org/10.1155/2015/139461">https://doi.org/10.1155/2015/139461</a>
- [14] Abdelouafi, A., Essodegui, F., Ousehal, A. and Kadiri, R. (1996) Appendix Mucocele. Apropos 6 Cases. *Annales de Radiologie* (*Paris*), **39**, 119-125.
- Pitiakoudis, M., Argyropoulou, P.I., Tsaroucha, A.K., Prassopoulos, P. and Simopoulos, C. (2003) Cystadenocarcinoma of the Appendix: An Incidental Imaging Finding in a Patient with Adenocarcinomas of the Ascending and the Sigmoid Colon. *BMC Gastroenterology*, 3, Article No. 30. <a href="https://doi.org/10.1186/1471-230X-3-30">https://doi.org/10.1186/1471-230X-3-30</a>
- [16] Wang, H., Chen, Y.Q., Wei, R., Wang, Q.B., Song, B., Wang, C.Y., et al. (2013) Ap-

- pendix Mucocele: A Diagnostic Dilemma in Differentiating Malignant from Benign Lesions with CT. *American Journal of Roentgenology*, **201**, W590-W595. https://doi.org/10.2214/AJR.12.9260
- [17] Lopez, J.P., Kandil, E., Schwartzman, A. and Zenilman, M.E. (2006) Appendiceal Mucocele: Benign or Malignant? *Surgical Rounds*, **29**, 540.
- [18] Stocchi, L., Wolff, B.G., Larson, D.R. and Harrington, J.R. (2003) Surgical Treatment of Appendix Mucocele. *Archives of Surgery*, 138, 585-589. https://doi.org/10.1001/archsurg.138.6.585
- [19] Soueï-Mhiri, M., Tlili-Graies, K., Ben Cherifa, L., Derbel, F., Hmissa, S., Dahmen Y, et al. (2001) Mucocele of the Appendix. Retrospective Study of 10 Cases. *Journal of Radiology*, **82**, 463-468.
- [20] Dhage-Ivatury, S. and Sugarbaker, P.H. (2006) Update on the Surgical Approach to Mucocele of the Appendix. *Journal of the American College of Surgeons*, 202, 680-684. https://doi.org/10.1016/j.jamcollsurg.2005.12.003
- [21] Misdraji, J., Yantiss, R.K., Graeme-Cook, F.M., Balis, U.J. and Young, R.H. (2003) Appendix Mucinous Neoplasms: A Clinicopathologic Analysis of 107 Cases. *American Journal of Surgical Pathology*, 27, 1089-1103. https://doi.org/10.1097/00000478-200308000-00006
- [22] Lien, W.C., Huang, S.P., Chi, C.L., Liu, K.L., Lin, M.T., Lai, T.I., et al. (2006) Appendix Outer Diameter as an Indicator for Differentiating Appendix Mucocele from Appendicitis. American Journal of Emergency Medicine, 24, 801-805. https://doi.org/10.1016/j.ajem.2006.04.003
- [23] Balci, O., Ozdemir, S. and Mahmoud, A.S. (2009) Appendix Mucocele Mimicking a Cystic Right Adnexal Mass. *Taiwanese Journal of Obstetrics & Gynecology*, **48**, 412-414. https://doi.org/10.1016/S1028-4559(09)60333-8
- [24] Fairise, A., Barbary, C., Derelle, A.L., Tissier, S., Granger, P., Marchal, F., et al. (2008) Mucocele of the Appendix and Pseudomyxoma Peritonei. *Journal of Radiology*, **89**, 751-762. <a href="https://doi.org/10.1016/S0221-0363(08)73781-8">https://doi.org/10.1016/S0221-0363(08)73781-8</a>
- [25] Zanati, F. (2007) Appendix Mucocele. *Journal de Chirurgie* (*Paris*), **144**, Article 146. https://doi.org/10.1016/S0021-7697(07)89491-9
- [26] Sasaki, K., Ishida, H., Komatsuda, T., Suzuki, T., Konno, K., Ohtaka, M., *et al.* (2003) Appendix Mucocele: Sonographic Findings. *Abdominal Imaging*, **28**, 15-18. https://doi.org/10.1007/s00261-001-0175-8
- [27] Aggarwal, N. and Bhargava, A. (2019) Appendiceal Mucocele Secondary to Torsion in an Asymptomatic Patient. *Journal of Surgical Case Reports*, **2019**, rjz241. https://doi.org/10.1093/jscr/rjz241
- [28] B.B., S.K. and Jasuja, P. (2019) Appendiceal Mucocele: A Rare Case Report. *International Journal Surgical Case Reports*, 58, 21-25. https://doi.org/10.1016/j.ijscr.2019.04.008
- [29] Morano, W.F., Gleeson, E.M., Sullivan, S.H., Padmanaban, V., Mapow, B.L., Shewokis, P.A., et al. (2018) Clinicopathological Features and Management of Appendiceal Mucoceles: A Systematic Review. The American Surgeon, 84, 273-281. https://doi.org/10.1177/000313481808400237