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CASE REPORT

Mucinous cystoadenoma of cecal appendix, pre-surgical diagnosis and treatment: case report

Cistoadenoma mucinoso de apendice cecal, diagnóstico prequirúrgico y tratamiento: reporte de caso

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ABSTRACT

Introduction: mucinous tumors of the appendix are a condition characterized by obstructive dilation of the cecal appendix caused by the intraluminal accumulation of mucous material. Their incidence is relatively low, accounting for approximately 0,2-0,3 % of all appendectomies performed. Mucinous lesions are divided into two groups: 1. Non-neoplastic lesions (mucocele) 2. Neoplastic lesions (serrated polyps, hyperplastic polyps, low-grade mucinous appendiceal neoplasia, high-grade mucinous appendiceal neoplasia, mucinous adenocarcinoma). There is no typical clinical presentation for this pathology; however, it often presents as acute appendicitis or as a mass in the right lower quadrant. Improper management could lead to pseudomyxoma peritonei, which has a high mortality rate.
Clinic case: a case of a patient diagnosed preoperatively with a large appendiceal mucocele is presented. An exploratory laparotomy, right hemicolectomy, and ileotransverse anastomosis were performed with a good postoperative recovery. The histopathological result reported a mucinous cystadenoma of the appendix.
Conclusion: mucinous cystadenoma is an extremely rare pathology, and its perioperative identification is challenging due to its nonspecific clinical presentation. For this reason, tomography is recommended in the study of neoplasms of the right iliac fossa.

Key words: Appendix; Appendiceal Neoplasms; Mucocele; Colectomy.

RESUMEN

Introducción: los tumores mucinosos del apéndice son una entidad caracterizada por una dilatación obstructiva del apéndice cecal ocasionada por la acumulación intraluminal de material mucoide, su incidencia es relativamente baja y corresponde aproximadamente al 0,2-0,3 % de todas las apendicetomías realizadas. Las lesiones mucinosas se dividen en dos grupos: 1. Lesiones no neoplásicas (mucocele) 2. Lesiones neoplásicas (pólipos dentados, pólipos hiperplásicos, neoplasia mucinosa apendicular de bajo grado, neoplasia mucinosa apendicular de alto grado, adenocarcinoma mucinoso). No existe cuadro clínico típico de esta patología sin embargo suele presentarse como una apendicitis aguda o como la presencia de una tumoración en el cuadrante inferior derecho. El manejo incorrecto podría ocasionar un pseudomixoma peritoneal el mismo que tiene una alta mortalidad.

Caso clínico: se presenta el caso de una paciente a la que se le diagnostica preoperatoriamente de un mucocele apendicular de gran tamaño, se realizo una laparotomia exploratoria, una hemicolectomia derecha y una anastomosis ileotransversa; con una adecuada evolucion post quirurgica. El resultado de el histopatológico reporta un cistoadenoma mucinoso de apendice.
Conclusión: el cistoadenoma mucinoso es una patologia en extremo rara, más aun su identificación perioperatoria por un cuadro clínico inespecifico; por esta razon se recomienda la Tomografia en el estudio de las neoplasias de la fosa iliaca derecha.

Palabras Clave: Apéndice; Neoplasias del apéndice; Mucocele; Colectomía.

INTRODUCTION

The term mucocele was initially described by Rokitansky in 1842 to refer to cystic dilatation of the appendix with an accumulation of mucus inside.[1,2] This definition includes everything from simple retention cysts to mucinous adenocarcinomas. Currently, the term mucocele has become obsolete,[1,3] and it is recommended to use the term mucinous tumors of the appendix (MAT) to refer to these entities.[1]

MAT is a heterogeneous entity characterized by the abnormal accumulation of mucin within the cecal appendix,[4,5,6,7] this term encompasses some causal pathologies, which include benign and malignant pathologies; among the first ones, we can mention epithelial hyperplasia, simple obstruction, and mucinous cystadenoma; meanwhile, malignant lesions can include neoplasms such as mucinous cystadenocarcinoma.[5,6,7]

Epidemiologically, it is a rare entity that corresponds to 0,2-0,3 % of all appendicular pathologies.[2,7,8,9] Simple TMA can be identified in approximately 29 % of cases, while epithelial hyperplasia and mucinous cystadenoma correspond to 31-34 %.[5] Mucinous cystadenocarcinoma is even rarer and causes 5 % of all appendicular mucoceles.[5] It usually occurs more frequently in women than in men and usually appears after the age of 50.[7,8]

They do not have a specific clinical picture, and in 50 % of cases, they present asymptomatically; their finding is usually accidental during radiological or endoscopic examinations or surgery;[4,5] when they present symptoms, they can include pain in the right lower quadrant, mass or tumor, nausea, vomiting, weight loss or gastrointestinal bleeding, sometimes a picture of intestinal invagination can occur.[1,4,5,7,10]

The preoperative diagnosis is unusual; however, it is considered that tomography is the most precise method and will help us to define more specific signs related to mucinous tumors of the appendix.[1,4,5] It is considered that benign tumors are usually smaller than 2 centimeters in size, while neoplastic tumors are usually larger.[6,8] The most frequent finding in the tomography is the dilatation of the appendix (53,8 %) with liquid density in its interior; the visualization of the appendix with a diameter greater than 15mm suggests the possibility of a mucinous tumor of the appendix with a sensitivity of 83 % and a specificity of 92 %. Another criterion for distinguishing between acute appendicitis and appendiceal mucinous tumor is the thickness of the wall; in the latter, it is usually greater than 6mm.[11] With respect to tumor markers, it is known that carcinoembryonic antigen and CA19-9 are associated with mucinous cystadenoma and cystadenocarcinoma,[5] However, the information is scarce and the available data suggest that they are elevated in advanced appendiceal mucinous tumors, at this point they can be useful for post-surgical follow-up; the current recommendation is to perform basal determination of CEA and CA19-9 in a mandatory manner.[11]

Primary tumors of the appendix can be classified according to the WHO 2019 classification into three main groups according to their cell type: epithelial tumors, mesenchymal tumors, and lymphomas; epithelial tumors are subclassified into mucinous, nonmucinous, neuroendocrine, and mixed nodal-endocrine tumors. Neoplastic lesions (serrated polyps, hyperplastic polyps, low-grade appendiceal mucinous neoplasm, high-grade appendiceal mucinous neoplasm, mucinous adenocarcinoma).[9,11] Mucinous cystadenoma is considered to be included in the low-grade appendiceal neoplastic lesions.[9] Treatment is of great importance since the treatment of mucinous cystadenoma is of great importance.

The treatment is of great importance since the surgical objective is to keep the tumor intact and avoid the spilling of its content to the peritoneal cavity; if this is the case, there is the possibility of developing a peritoneal pseudomyxoma.[12] besides, its treatment is not very encouraging and has high mortality; survival at 10 years is 10-32 %.[2,4,6] The surgical technique to be developed will depend on some factors, among them if the mucocele is perforated or not if the appendicular base is compromised, or if there are positive ganglions; patients may require simple appendectomy, which is curative as long as the margins of resection are clear, to right hemicolectomy with or without cytoreductive surgery and/or intraperitoneal chemotherapy.[1,2,4,10]

Wide resection, including cecum and terminal ileum, or even right hemicolectomy, may be required when a broad-based mucocele of the cecal wall or ileum is involved or when malignancy is suspected.[2,6,7,10]

The prognosis of benign mucinous tumors of the appendix after complete resection with negative margins is excellent, with a 5-year survival of 100 %.[7] In benign forms, progression to peritoneal dissemination only occurs

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in approximately 2 %, while in mucinous adenocarcinomas, it is up to 23 %.[11]

CLINICAL CASE

Female patient, 42 years old, primary education, occupation: housewife, catholic religion, marital status: married. No clinical or surgical history of importance. She came to the outpatient surgery department for presenting symptoms of approximately 3 years of evolution characterized by the presence of a tumor in the right iliac fossa that has increased progressively, accompanied by sporadic pain, apparently triggered after suffering a contusive blow in that region.

On initial physical examination, vital signs were within normal parameters, with a body mass index of 28.7. During abdominal palpation, attention was drawn to the presence of a tumor in the right iliac fossa, approximately 10 centimeters in diameter, painful on deep palpation, with no signs of peritoneal irritation. Complementary imaging studies were requested; the ultrasonography reported an oval echo lucent image with fine internal echoes of dense liquid content measuring approximately 11,9x3,9x39 centimeters with an approximate volume of 95 milliliters (figure 1A); the CT scan showed an oval echo lucent image with fine internal echoes. 1A) In the simple and contrasted tomography of the abdomen, it is reported in the right iliac fossa the presence of a tubular mass, depending on the base of the cecum, which ends in a cecum sac, with a liquid density (30UH), which extends to the midline and measures 12x4x4,1 centimeters in its longitudinal, anteroposterior and transverse diameters respectively. The mass could correspond as a first possibility to appendicular mucocele. No abdominal adenopathies were identified (figure 1B). Laboratory tests showed no evidence of leukocytosis or neutrophilia, anemia, or significant malnutrition. Tumor markers within normal parameters.

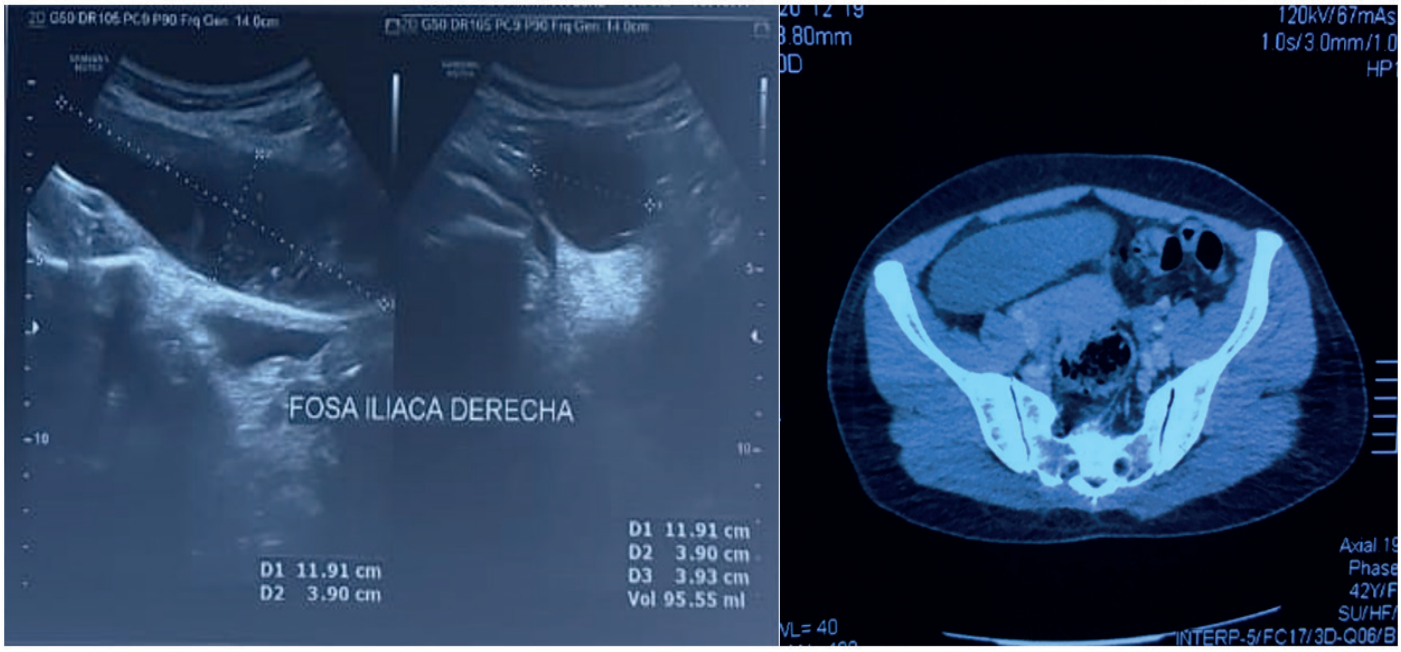

Figure 1. A) Abdominal ultrasound B) CT scan showing an image of a tubular tumor suggestive of an appendiceal mucocele with an adequate cleavage plane; no adenopathy is reported

With the diagnosis of appendicular mucocele, it was decided to perform an exploratory laparotomy in which it was evidenced: Cecal appendix of approximately 15x5 cm, undamaged; the presence of lymphadenopathy in the appendicular base of 1 cm in diameter of woody consistency, retroperitoneal structures identified and preserved (figure 2A). Given these findings, it was decided to perform a right hemicolectomy, resection of the terminal ileum, and laterolateral ileotransverse anastomosis procedure without complications (figure 2B).

The oral diet was started on the fourth day for patients with favorable postoperative evolution. The patient tolerated and presented adequate intestinal transit and no complications, so it was decided to discharge the patient from the hospital. The results of the histopathological study of the sent surgical specimen were subsequently received and reported: mucinous cystadenoma of the cecal appendix, mild chronic inflammation and hyperplasia of lymphoid follicles in the terminal ileum, moderate chronic inflammation in the colon, thus confirming the initial presumptive diagnosis.

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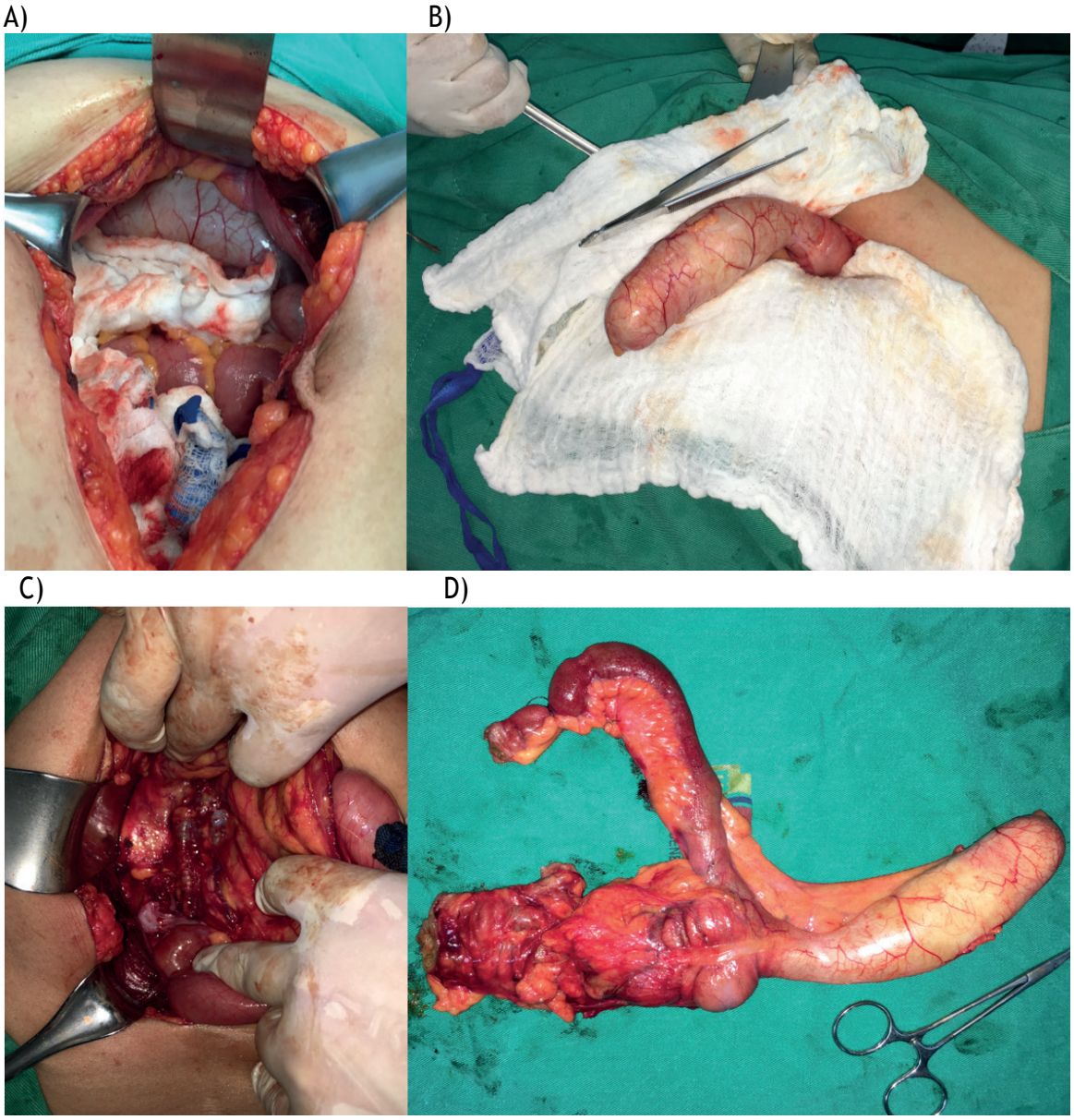

Figure 2. A) Exploratory laparotomy and initial finding. B) eviscerated undamaged appendiceal mucocele. C) Retroperitoneal structures without the presence of macroscopic pathology. D) Hemicolectomy product

DISCUSSION

Appendiceal mucoceles are a rare entity. However, the mucinous cystadenoma lineage is more unusual and generally considered benign. NMA is diagnosed during the surgical procedure. However, we present a case in which the diagnosis is made preoperatively, and a scheduled surgery is performed. In a retrospective study by Nutu et al.[1], it was found that preoperative diagnosis was achieved in 58,3 %; the 30,5 % of synchronous or metachronous tumors of the colon is noteworthy.

The diagnosis was made by computed tomography, which reported a tubular image without local invasion or adenopathies. In their article, Shinohara et al.[2] consider CT and colonoscopy the best tests to reach a pre-surgical diagnosis.

The treatment was a right hemicolectomy because the appendicular base exceeded two centimeters, and during the procedure, a lymph node could be seen in the region of the appendicular base. The surgical conduct is controversial, probably with a pre-surgical diagnosis, a thin base, and the absence of adenopathy; a safe treatment modality would be simple appendectomy, conventional or laparoscopic, as Merino et al.[13] described in their article. The most important thing in managing an NMA is to keep it intact.

In the case of a preoperative rupture or an accidental transoperative rupture with peritoneal dissemination, referral to a specialized center for cytoreduction and hyperthermic intraperitoneal chemotherapy is required. However, Zih et al.[14] report that observation is an alternative in low-grade TMA with limited dissemination

without decreasing survival and disease-free period at 5 years.

CONCLUSIONS

Mucinous cystadenoma is an extremely rare pathology, even more so its perioperative identification due to an unspecific clinical picture; for this reason, CT is recommended in the study of neoplasms of the right iliac fossa. The treatment is still controversial; however, removing the undamaged specimen is mandatory to avoid complications.

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CONSENT

Consent was obtained from the patient for the performance of this work.

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

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