ORIGINAL RESEARCH PAPER
Volume - 11 | Issue - 02 | February - 2022 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

SESSILE SERRATED APPENDIX ADENOMA

Surgery

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KEYWORDS

INTRODUCTION

The sessile serrated adenoma is a recently described rare condition characterized by a dysplastic sawtooth-like epithelium. Its location in the appendix is rare and the degree of malignancy is still a matter of debate. We describe a rare case of sessile serrated appendix adenoma.

HEADINGS: Sessile serrated adenoma, appendix, adenocarcinoma

CASE REPORT

A 56-year-old woman without history of prior illnesses returned for a 15-day history of right iliac fossa pain. It was associated with nausea without vomiting and she reported having had a fever measured at 38.2°C. Laboratory tests and computed tomography of the abdomen and pelvis with endovenous contrast showed a distended appendix with intraluminal fluid density, and a soft tissue of 2.0 cm x 2.3 cm x 1.7 cm density occluding the appendiceal orifice.

Diagnostic videolaparoscopy was indicated to elucidate the diagnosis, which identified the cause of persistent pain with a picture compatible with grade II acute appendicitis.

A videolaparoscopic appendectomy was performed in accordance with the technique, the surgical specimen was sent for anatomopathological examination and the inventory of the abdominal cavity did not present other alterations. The postoperative period was uneventful, with complete involution of the abdominal pain. Pathological examination of the surgical specimen revealed an uncommon appendicular lesion: tubulovillous adenoma. Since the appendicular resection was complete, no additional procedure on the cecum or right colon was proposed. The search for personal or family history of gastrointestinal tumor was negative.

Histology of endoscopic biopsy of the overlying mucosa revealed a sessile serrated adenomatous tissue with no evidence of lymphadenopathy or adjacent focal lymphadenopathy inflammatory changes were not seen.

DISCUSSION

Serrated adenoma has the characteristic histology of dysplastic epithelium similar to a sawtooth, which is found in more than 50% of the basal area, with branching crypts, dilation of its base and growth of crypts parallel to the mucosal muscle(1). Sessile serrated adenomas are a recently described category, occurring most commonly in the right colon, being part of the adenocarcinoma sequence of colon malignancy, with 20 to 30% of colorectal cancer cases arising in the serrated polyp pathway. Literature review revealed that only one case of serrated appendix adenoma was published until Rubio's first report on serrated appendix adenomas. In his report, Rubio stated that villi of the serrated appendix adenomas appeared to be highly aggressive lesions rather than colon rectal adenomas (1–3).

Its location in the appendix is rare and its association with a malignancy is still a matter for discussion (4). A study of the serrated neoplastic pathways between the colorectal part and the appendix showed that, in the last decade, the molecular classification of colorectal polyps has evolved to include multiple carcinogenesis pathways arising from polyp precursors with distinct morphological lesions. Appendicular polyps often show morphological similarities with their colorectal counterparts, due to the anatomical uniqueness of the organ itself and the polyps that originate from its mucous lining (3,5). However, there is limited and often conflicting literature on changes in the serrated appendix lesions to support the adoption of colorectal diagnostic terminology in the appendix. Thus, the results indicated that serrated appendix lesions that resemble hyperplastic polyps (HPs) and sessile serrated adenoma (SSA/Ps) often harbor KRAS mutations and rarely exhibit BRAF mutations. These results suggest that the serrated pathway in the appendix is different from the serrated pathway in the colon and rectum and, therefore, colorectal terminology may not be applicable to appendicular lesions (1,2,6).

Thus, all appendix precursor lesions were classified using a simplified scheme into the categories based on the presence of serrated epithelium and dysplastic cytology: non-dysplastic serrated, dysplastic serrated, and dysplastic non-serrated. Epithelial serrations were defined as epithelial crypts demonstrating luminal folding in a prominent sawtooth. Appendicular lesions exhibiting an epithelial growth pattern undulation without prominent sawtooth and luminous folds were classified as non-serrated. Therefore, serrated appendix lesions were grouped using standard colorectal diagnostic terminology as described by the World Health Organization (WHO) [10] for the following categories: hyperplastic polyp, sessile serrated adenoma with cytological dysplasia, and traditional serrated adenoma (TSA). Then, the serrated lesions of the dysplastic appendix were divided into mucinous and non-mucinous types defined by the presence of gross appendicular luminal mucin or histological evidence of abundant cytoplasmic mucin within the neoplastic epithelium (6).

Due to the lack of information, the incidence of serrated appendix adenoma has not been fully clarified (1,2,7), but it is estimated that appendix tumors are the rarest among gastrointestinal tumors, making up about 0.5 % of the total. Its diagnosis usually occurs incidentally in appendectomy or necropsy (1). It can still be rarely diagnosed in colonoscopy when masses appear in the appendicular orifice, as

described in two case reports (1,2,7). Diagnosis is based primarily on architectural features, including branching of the crypts, dilation of the base of the crypts, and a growth pattern in which the crypts appear to grow parallel to the mucosal muscle, often creating a T or L shape in the inverted crypt. This growth pattern is often accompanied by the presence of mature cells with goblet cells or foveolar gastric type at the base of the crypt (8,9).

Sessile serrated polyps have a predilection for the proximal colon and are associated with female gender and smoking, but no consistent effect of other factors on their formation has been reported. In Contrast, Wallace et al. 13 found that obesity, smoking, dietary fat intake, caloric intake, and red meat consumption were associated with an increased risk of distal serrated polyps, including hyperplastic polyps, sessile serrated polyps, and traditional serrated adenomas (8). Furthermore, these associated risk factors may present as ulcerative colitis, familial adenomatous polyps and association with other tumors.

Mucocele

Mucocele is a rare entity characterized by dilation of the appendicular lumen and excessive mucus collection, which can develop in different ways (5,9). A cyst is formed by obstruction of the appendicular lumen, with normal mucus secretion, and may be caused by fecalitis, gallstones, endometriosis, postoperative adhesions, appendix volvulus, or intestinal tumors. Appendix mucocele is often misdiagnosed, leading to a variety of complications due to improper detection / management (5,9).

It results from an intraluminal accumulation of mucoid fluid, estimated to be found in 0.2-0.3% of all appendectomies (10). It predominates in females, with a mean age at diagnosis of 50-55 years (5.9).

Appendix mucoceles were previously classified into 4 pathological entities according to epithelial characteristics: simple or retention mucocele, mucocele with local or diffuse hyperplastic villous epithelium (5–25%), mucinous adenoma/cystadenoma (63-84%) and mucinous cystadenocarcinoma (11–20%).

It does not have a typical clinical presentation and is considered a potentially premalignant condition (5,9,11). In most cases these tumors are asymptomatic or mildly symptomatic. Although clinical symptoms are not specific, usually a palpable mass is found in the right lower quadrant, in addition to stiffness and pain in the same location (9,11,12). It can sometimes accompany urinary tract infection or hematuria and, very rarely, lower gastrointestinal bleeding caused by intussusception (9).

Some complications can occur when the mucin-producing cells form abscesses and rupture or when there is tubular obstruction of the adenoma, with an increase in intraluminal pressure and its rupture (2). An important complication is peritoneal pseudomyxoma (PMP), which consists of large amounts of mucinous ascites associated with mucinous peritoneal and omental implants due to perforation of the organ, which has its pressure increased by the accumulation of mucin. The organ that is more likely to develop PMP is the appendix, but it can also originate from other organs(13,15). Furthermore, it can be categorized, according to the WHO, as low or high (15). It is a rare, low-incidence complication that affects the population with a mean age of 50 years, with no difference in prevalence between genders. However, PMP is potentially lethal, as with the perforation of the organ there is dissemination of cancer cells within the peritoneal cavity (9,15). Rarely, PMP spreads through the bloodstream or lymphatic vessels, but there are reports of cases where there was intrathoracic involvement, presenting a worse prognosis at high mortality rates (16).

The manifestation occurs progressively (over months or even years) by the slow accumulation of mucus inside the abdominal cavity and is characterized by gradual abdominal distension. The chronicity of the condition has a poor evolution, as there is formation of fibrosis and adhesions in the peritoneum (9). The preoperative diagnosis of PMP is very important, as this allows the surgeon to prepare and take extra care in order to avoid the leakage of cells or mucin into the peritoneum during surgery and thus prevent peritoneal dissemination. However, peritoneal pseudomyxoma can develop during surgery, if the cystic fluid leaks during surgery, or even postoperatively, in some cases, and therefore, patients at risk need to be monitored (11,13) . When present, PMP may have surgical debulking and appendectomy as initial therapy (15). This complication can occur in both benign and malignant neoplasms, but the prognosis is even worse when it develops from a malignant lesion, with a low survival rate (9).

CONCLUSION

The finding of sessile serrated adenoma located in the appendix is rare and, in general, its diagnosis is made incidentally. This report demonstrates one of the cases, where the clinical presentation was suggestive of acute appendicitis and only through the anatomopathological examination was it possible to clarify the probable etiology. There is still a lot to be understood, but in this case, the appendectomy was resolute, with no need for other interventions.

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