CLINICAL CASE

Tubulovillous adenoma of the appendix: A case report and review of the literature

Adénome tubulovilleux appendiculaire, mise au point à propos d’un cas

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Summary  We report the case of a young woman hospitalized for a chronic appendicular syndrome. The histological examination of the resected specimen revealed a rare tumor: tubulovillous adenoma, discovered in 0.02% of all appendectomy procedures. Treatment is most often limited to appendectomy, but in the event of incomplete excision or associated adenocarcinoma, right hemicolectomy may be required. After surgery, a follow-up colonoscopy is recommended due to the higher risk of second gastrointestinal neoplasms in patients with appendicular tumors.

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Résumé Nous rapportons le cas d’une jeune femme hospitalisée pour un syndrome appendiculaire chronique. L'analyse de la pièce d'appendicectomie a révélé une tumeur peu fréquente: un adénome tubulovilleux. Elle est en effet mise en évidence sur 0,02 % des pièces d'appendicectomie. La prise en charge se limite le plus souvent à une appendicectomie mais elle peut nécessiter une colectomie droite en cas d'exérèse incomplète ou d'adénocarcinome associé. La coloscopie est également recommandée en postopératoire car la fréquence des tumeurs digestives augmente chez les patients atteints par ces lésions.

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Case report

A 19-year-old woman with an uneventful history sought care for chronic pain in the right iliac fossa. Routine laboratory tests were normal and the computed tomography of the abdomen and pelvis were noncontributive. Exploratory laparoscopy was proposed to identify the cause of the persistent pain and revealed an inflammatory appendix. Appendectomy was performed.

The postoperative period was uneventful, with total involution of the abdominal pain. Pathology examination of the surgical specimen disclosed an uncommon appendicular lesion: tubulovillous adenoma. Since the appendicular resection had been complete, no complementary procedure on the cecum or right colon was proposed. Search for personal or family history of gastrointestinal tumor was negative.
The colonoscopy performed 1 month after appendectomy failed to identify any suspect lesion.

Discussion

Epidemiology

Pathology reports show that five out of 100 appendectomy specimens harbor a tumor [1,2]. Tubulovillous adenoma is amongst the rarest, identified in 0.02 to 0.06% of appendectomy specimens [3,4]. About 50 cases of appendicular tubulovillous adenoma have been described in the literature, almost exclusively in case reports with a review of the literature. One large series of six cases has been published [5].

Clinical presentation

In the report by Hameed [3], mean patient age at diagnosis was 45 years. In the most recent series, it was 65 years [5]. The clinical presentation of appendicular adenoma is nonspecific. Generally, the patient presents a more or less acute appendicular syndrome, but as in our patient, without signs of inflammation. Fortuitous discovery during colonoscopy [6–8] or abdominal surgery performed for a reason other than appendicular disease [9,10] have been reported, as have been intestinal invagination and intussusception [11,12]. In adults, these manifestations suggest an underlying surgical anomaly. Rarely, diarrhea and melena have led to diagnosis [9].

Pathology

Adenomas of the colon and the appendix are dysplastic polyloid formations. Three types are described:

- tubulous adenoma related to the proliferation of Lieberkühn glands in the chorion;
- villous adenoma formed by epithelium-coated gland-free finger-like expansions;
- tubulovillous adenoma where the two continents are associated [13].

The potential for malignant degeneration depends on tumor size (less than 5% for lesions measuring less than 1 cm and more than 50% for those measuring more than 2 cm [14]) and pathology type (5% for tubulous adenomas, 40 for villous adenomas and 20% for tubulovillous adenomas [15]). Suggestive features include irregular glandular formations bordered with a nonsecreting pluristratified epithelium with nuclear anomalies and mitosis configurations.

Treatment

Tubulovillous adenomas of the appendix are rare tumors. The diagnosis is often established at the pathology examination of the surgical specimen. A complementary procedure should be discussed, depending on the pathology findings, but appropriate management has been widely debated. For some authors, appendectomy or cecal resection is sufficient for benign lesions or low-grade adenocarcinoma [9,16,17]. For others, complementary resection of the ascending colon is necessary considering the risk of malignancy and recurrence as well as the cecal origin of the lesions [18]. At the present time, it is generally proposed to perform right hemicolectomy for:

- tumor size greater than 2 cm;
- mesoappendicular invasion;
- nodal extension;
- contaminated margin;
- lymphatic or vascular emboli [5].

In the event of an endoscopic diagnosis, biopsy or resection should be followed by surgical treatment since endoscopic resection may be incomplete or introduce a risk of intraluminal dissemination of malignant cells [19,20] (Figs. 1 and 2).

Follow-up

The course of these appendicular adenomas or adenocarcinomas is similar to that observed for the colonic or
rectal localizations [16]. Once the diagnosis has been established, a complete control colonoscopy should be scheduled at 1 month. This control is designed essentially to search for an associated carcinomatous or adenomatous syndrome because of the higher incidence of malignant gastrointestinal tumors in these patients [21].

**Conclusion**

Appendicular adenoma is a rare benign tumor for which appendectomy can be considered sufficient treatment if the tumor measures less than 2 cm, there is no mesoappendicular or nodal spread and the resection margins are healthy. Otherwise, right hemicolectomy with nodal dissection is recommended. Blood tests and imaging results may be misleading, as was the case in this young woman. In the event of an overt discordance between a highly suggestive clinical presentation of a chronic appendicular syndrome and negative complementary tests, adequate diagnostic management should include exploratory laparoscopy with appendectomy.

**Conflict of interest statement**

None.

**References**