

A Unique Case of Appendicular Leiomyoma: Usual Lesion in an Unusual Site

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Abstract

Appendicular leiomyoma is a rare entity described in 1977. Herein, we present a case of leiomyoma of the appendix in a 45-year-old female who presented with pain in the lower abdomen of 2 months duration. She was diagnosed as recurrent appendicitis based on the physical examination and ultrasonography. The pathological findings showed uni-lobular proliferation of fusiform cells, arranged in net-like sheets or swirls, in a hyaline background with edematous stroma. To our knowledge, there are only 3 cases of appendicular leiomyomas reported in the literature. Although it is a rare tumor, surgeons and pathologists should be aware of it.

Key words: Appendix, Benign appendicular, Leiomyoma

INTRODUCTION

Most appendicular neoplasm in adults are carcinoid tumors.¹ Benign tumors including leiomyoma are extremely rare. Ultrasonography is a useful aid in the diagnosis of appendicitis.² but the sonographic appearance of appendicular leiomyoma is poorly documented and, therefore, it is impossible to diagnose it preoperatively. This is the reason most of the diagnosis is only established after the surgery. Growth and proliferation of smooth muscle tissue in women coincide with estrogenic stimulation. Children rarely develop these tumors.^{3,4} Here, we present a rare case of typical appendicular leiomyoma in a 45-year-old female patient with hemangioma in the liver.

CASE REPORT

A 45-year-old female with the unremarkable past history presented in the surgery clinic for evaluation of the abdominal pain since 2 months. There was no history of trauma

or accident. Underwent tubectomy 8 years back, with a menstrual history suggestive of perimenopause. On surgical examination, tender mass in the right iliac fossa was identified.

The sonography revealed hemangioma in segment vii of liver measuring 1.7 cm diameter and non-compressible appendix, surrounded by a hypoechoic thickened wall more than 2 mm in diameter the clinical and radiological diagnosis of recurrent appendicitis with hemangioma of liver was made and the patient underwent appendicectomy.

The gross pathologic examination revealed a 1.5 cm circumscribed lobulated gray tan mucoid mass arising within the body wall of the appendix, not involving the base or tip (Figure 1). Microscopically, the tumor was composed of fusiform cells with long processes and nuclei with blunted ends, interdigitate with those of the lamina muscularis propria (Figure 2). The mucosa had apparently suffered pressure atrophy to the extent shown in the illustration (Figure 3). On the basis of the characteristic morphologic findings, the diagnosis of appendicular leiomyoma was rendered. At the last follow-up, the patient was well.

DISCUSSION

Leiomyomas of the uterus are the most common benign tumors of smooth muscle origin, but they arise in any

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Figure 1: Appendicectomy specimen cut section showing well-circumscribed mucoid measuring 1.5 cm in diameter

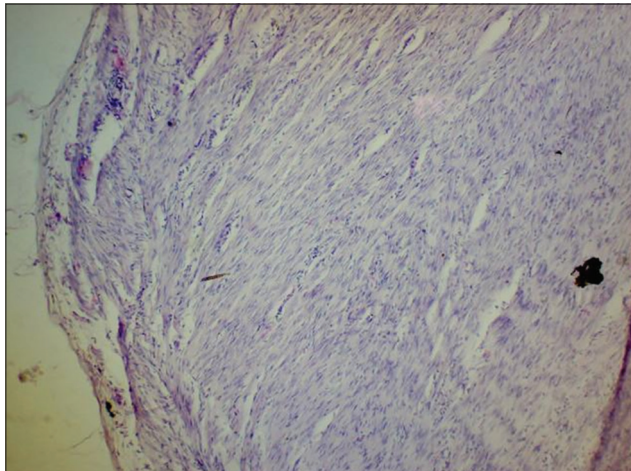


Figure 2: Microscopy hematoxylin and eosin stain showing proliferating spindle cells extending up to the serosa

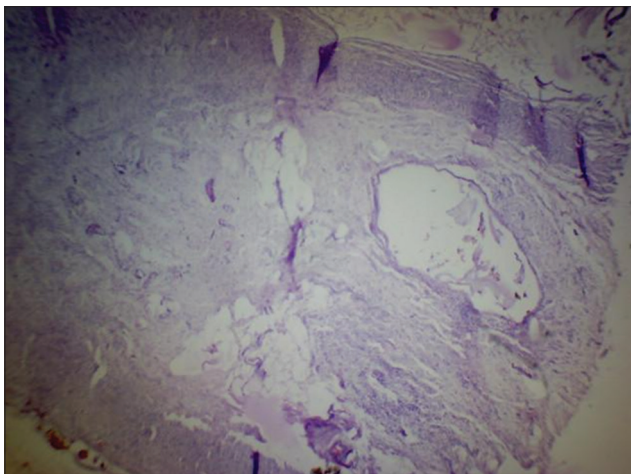


Figure 3: Microscopy hematoxylin and eosin stain showing obliterated mucosa (x100)

portion of the alimentary tract from the esophagus to the rectum and are most common in the stomach.⁵ However, appendicular leiomyomas are quite rare in the literature.^{6,7}

Patient presents with pain abdomen and vomiting with tenderness in the lower abdomen. The reported age range includes 2 years child to those older than 60 years. In our case, patient had appendicular mass as well hemangioma of the liver which was noticed accidentally while scanning. Estrogens have known growth stimulatory effects on the vasculature and most hemangiomas demonstrate estrogen receptors.⁸ In view of this, hyperestrinism in perimenopausal women could be the causes for the development of hemangioma and leiomyoma. In our case, after appendicectomy, the patient had an uneventful post-operative phase and was discharged 8 days after the operation.

Histo-pathology

On gross examination, body of the appendix was bulging. Histology showed a small leiomyoma in the submucosa. There were no inflammatory exudates in the lumen. The adjacent appendicular parenchyma is otherwise unremarkable.

The term “gastrointestinal stromal tumor” has become increasingly popular for variety of mesenchymal tumors, including smooth muscle tumors, spindle cell tumors with possible neural differentiation.⁹ Diagnostic criteria to distinguish leiomyoma from leiomyosarcoma which is highly aggressive depend on patient gender and anatomic site. Most leiomyoma arise in females, bland looking lesions with fewer than 10 mitosis per 50 hpf, located in the uterus, retroperitoneum, and extremities appear to be benign. Such lesions are commonly positive for estrogen and progesterone receptors and appear to have considerable similarities to gastrointestinal leiomyomas. As yet, there are insufficient data concerning comparable retroperitoneal lesions in males, and a diagnosis of leiomyoma in these setting should be made only with extreme caution.^{10,11}

CONCLUSION

Leiomyoma of the appendix is a very rare benign tumor. Clinical appearance and diagnostic exams are usually not sufficient for the definite diagnosis and require a histopathological examination. However, if the surgeons and pathologists are aware of it; conservative surgical treatment would protect the patient from dangers like complete obliteration of the appendicular lumen and tumor progression.

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