Lymphangioma of the Anal Canal

Amina Ismaeel, MSc, DPhil* Fayek Alhilli, Ph.D**

ABSTRACT

Lymphangiomas of the large gut are rare vascular malformation presents with nonspecific symptoms. We report herein a new case of incidental solitary pedunculated polypoid superficial lymphangioma of the anal canal in a 32-year-old man in which the diagnosis was made on histological examination of the excised lesion.

Keywords: Anal canal, lymphangioma, gastrointestinal tract, vascular malformation, anorectal pathology.

INTRODUCTION

Lymphangiomas are rare benign lesions of the gastrointestinal tract, and the anal canal is the least involved site.¹⁻⁴ Most of the entries of these lesions in the literatures appeared in standard textbooks as a passing term to complete a listing, or in large review series of anal and perianal lesions to record a finding.^{1,2,4} We found only six publications adequately describing this lesion.^{1,5-9}. We report herein a new case of incidental solitary pedunculated polypoid superficial lymphangioma of the anal canal in a 32-year-old man in which the diagnosis was made on histological examination of the excised lesion.

CASE REPORT

A 32-year-old man, not known to have any illness, presented with 4 months history of left side abdominal pain associated with heartburn, constipation, flatulence unrelated to food intake and occasional rectal bleeding. Clinical examination was unremarkable apart from epigastric tenderness. Laboratory investigations, abdominal ultrasound examination and esophagogastroduodenoscopy were unremarkable. Gastric and duodenal mucosal biopsies showed features of mild chronic gastritis and duodenitis, respectively. Colonoscopy showed solitary soft pedunculated polyp paler than the surrounding mucosa measuring 1.3 cm in length and 1 cm in width arising from the dentate line of the left lateral wall of the anal canal (Figure 1A). The polyp was snared and submitted for histological examination. No other anorectal or colonic pathology noted. The patient experienced partial improvement of his symptoms after polypectomy. He was prescribed Chlorodiazepoxide 5 mg and Omeprazol 20 mg. He was lost for follow up since then.

Grossly, the polyp was firm grayish white lesion with smooth surface measuring $10\times8\times4$ mm. The entire specimen was processed into paraffin sectioning. Microscopy showed polypoid superficial lymphangioma composed of anal squamous epithelium covering fibrous

core containing numerous thin-walled dilated multicystic cavernous vascular channels of different sizes arising from the papillary zone of the anoderm (Figure 1 and Figure 2A). These channels were filled with faint eosinophilic homogenous substance and lined by flattened Factor VIII+, CD31+, CD34- endothelial cells (Figure 2B, 2C and 2D). The overlying epithelium showed hyperkeratosis, acanthosis and elongation of the rete ridge which partially surrounded the channels forming collarettes. The channels appeared in the superficial parts of the polyp in close proximity to the anoderm and extended into the fibrous core and in close proximity to the pedicle of the polyp. The core also showed scattered lymphocytes. No mitosis, dysplasia or malignancy noted.

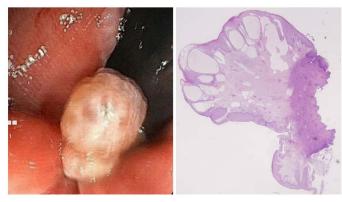


Figure 1. (A) Endoscopy image showing 1.3 cm polyp arising from the dentate line of the left lateral wall of the anal canal. (B) H&E-stained section of the entire polyp showing multicystic cavernous lymphatic channels in the papillary y zone of the anoderm extending into the core and pedicle of the polyp.

- Department of Pathology
 College of Medicine and Medical Sciences
 Arabian Gulf University, Manama, Kingdom of Bahrain.
 - Email: aminaaya@agu.edu.bh
- ** Department of Pathology, King Hamad University Hospital, Busaiteen, Kingdom of Bahrain. Email: falhilli@gmail.com

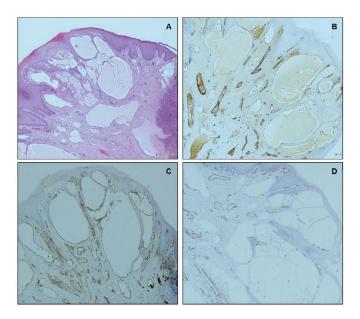


Figure 2. (A) Histological details of the lymphangiomatous polyp showing the thin-walled dilated multicystic vascular channels filled with faint eosinophilic homogenous substance and lined by flattened Factor VIII+ (B), CD31+ (C), and CD34- (D) endothelial cells.

DISCUSSION

Anal lymphangiomas are developmental vascular malformation rather than truly neoplastic lesions arising from progressive dilatation of primitive mal-developed sequestered lymphatic vessels due to failure of communication with local lymphatic system or the presence of deep cistern in the same region.5 The lesion is similar to other vascular malformations notably cutaneous superficial lymphangiomas (lymphangioma circumscriptum) and angiokeratomas except that in the latter the vascular channels are filled with blood and lined by hematogenous endothelium. However, it has been suggested that anal lymphangiomas may also develop secondary to acquired obstructive anorectal pathology (e.g. previous surgery, irradiation, infection) which results in failure of communication with underlying deep lymphatics.⁵ This does not seem a likely explanation because any such "acquired" obstruction would involve the hematogenous and lymphatic channels and not selectively the latter. The channels formed may best be regarded as secondary lymphangectasis rather than developmental lymphangiomas. It is also worth noting here, that in the present case report, no associated (acquired) anorectal lesion was found. In the other six cases reported in the literature, three were not associated with anorectal pathology, one had an anal fistula, one had ulcerative colitis with hemorrhoids and one had hemorrhoids only. 1,5-9.

In the present case, the diagnosis of superficial variant of anal cavernous lymphangioma was made in view of the pattern and distribution of multicystic dilated channels of various sizes filled with faint eosinophilic homogenous lymph located primarily in the papillary zone of the anoderm. ^{1,2,5-8} The endothelium of these channels is Factor VIII+, CD31+ and CD34-. The latter, stains only the hemangiomatous endothelium and as such is a good marker to distinguish between these two endothelial cells. Another useful marker is D2-40.⁵

The histological appearance of the superficial variant of these lesions characteristically forms multicystic cavernous channels of various

sizes seen in the papillary zone of the anoderm filled with faint eosinophilic homogenous lymph.^{1,5-8} However, in the present case report, the extension of some channels into the core and pedicle of the polyp not only indicated incomplete excision but also the possible presence of either a local obstruction or the existence of a connection with a deep cistern. As such, recurrence of such lesions must always be considered. Regrettably, our patient was lost for follow up soon after polypectomy and further investigations to rule out the presence of any vascular pathology was not possible. We recommend therefore that the pedicle of such polypoid lesions should be included in the polypectomy specimen so as to assess the level of excision.

CONCLUSION

The present case has several common attributes with the other six previously reported cases. ^{1,5-9}. In all, the symptoms were vague and nonspecific, and the lesion was an incidental pedunculated solitary polyp. There was also no characteristic gross or endoscopic appearance of the polyps, and the diagnosis was made only after the histological examination of the polypectomy specimens.

Authorship Contribution: All authors share equal effort contribution towards (1) substantial contributions to conception and design, acquisition, analysis and interpretation of data; (2) drafting the article and revising it critically for important intellectual content; and (3) final approval of the manuscript version to be published. Yes.

Potential Conflict of Interest: None

Competing Interest: None

Acceptance Date: 21-11-2024

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