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Retrorectal Presacral Tailgut Cystic Hamartoma: A Case Report

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Abstract
Tailgut cystic tumours are congenital anomalies which may develop when the embryonic hindgut fails to involute completely. Herewith, we describe a case of tailgut cyst. The patient was a female aged 24 years. She developed a cyst in presacral space. Cyst measured 9x7x7 cms. Anteriorly, cyst displaced rectum and colon. Cyst contained thick mucoid yellowish fluid. Histologically, cyst wall showed thick band of fibrous tissue. Luminal surface was partly lined by squamous epithelium and partly by columnar epithelial cells. The epithelium was ulcerated and showed granulation tissue formation along with lymphocytic infiltration. Cyst was finally diagnosed as inflammed retrorectal presacral cyst.
Keywords: *Cyst, congenital, benign tailgut.*

Introduction

Retrorectal tailgut cysts are hamartomas[1] and rare congenital lesions situated in the Retrorectal space. Anteriorly, it is bounded by the rectum, posteriorly by the sacrum, superiorly by the peritoneal reflection, inferiorly by the levator ani and coccygeal muscles and laterally by the ureters and iliac blood vessels[2]. Tailgut cysts arise from embryonic vestigial postnatal hindgut. The tailgut involutes around the sixth week of gestation. However, if this process fails, a tailgut cyst develops[3].

Case Report

A 24 year old female complained of pain in lower abdomen. Per rectal examination suggested a soft presacral swelling. It measured 9x7x7 cms. Few calcified specks were seen in the wall of the cystic lesion. Specks of adipose tissue were also seen within the lesion. Anteriorly, the lesion was seen displacing the rectum and sigmoid colon with preserved fat planes. A small tract with opening was seen in the overlying skin. Through this opening, radio-opaque contrast dye was injected. However, no obvious internal communication was seen with the cyst. No obvious pelvic and retroperitoneal lymph nodes were seen. No other pathology was seen. Laparoscopic resection of cyst was done. Cyst contained thick mucoid yellowish fluid. Histopathological examination of the cyst wall showed thick fibrous connective tissue along with fibromuscular tissue (Figure 1g). Luminal inner surface of cyst wall was partly lined by stratified squamous epithelium (Figure 1a) and partly by tall columnar or cuboidal epithelium. It was thrown into papillary

projections to form micropapillae. Cyst wall showed organized layer of smooth muscle. Cyst wall also enclosed glands, lined by columnar mucin-secreting epithelium. Few scattered lymphoid aggregates were seen. At places, epithelial lining was ulcerated and granulation tissue consisting of newly formed capillaries and fibrous tissue was seen. Surrounding fibrous tissue also enclosed newly formed capillaries (Figure 1).

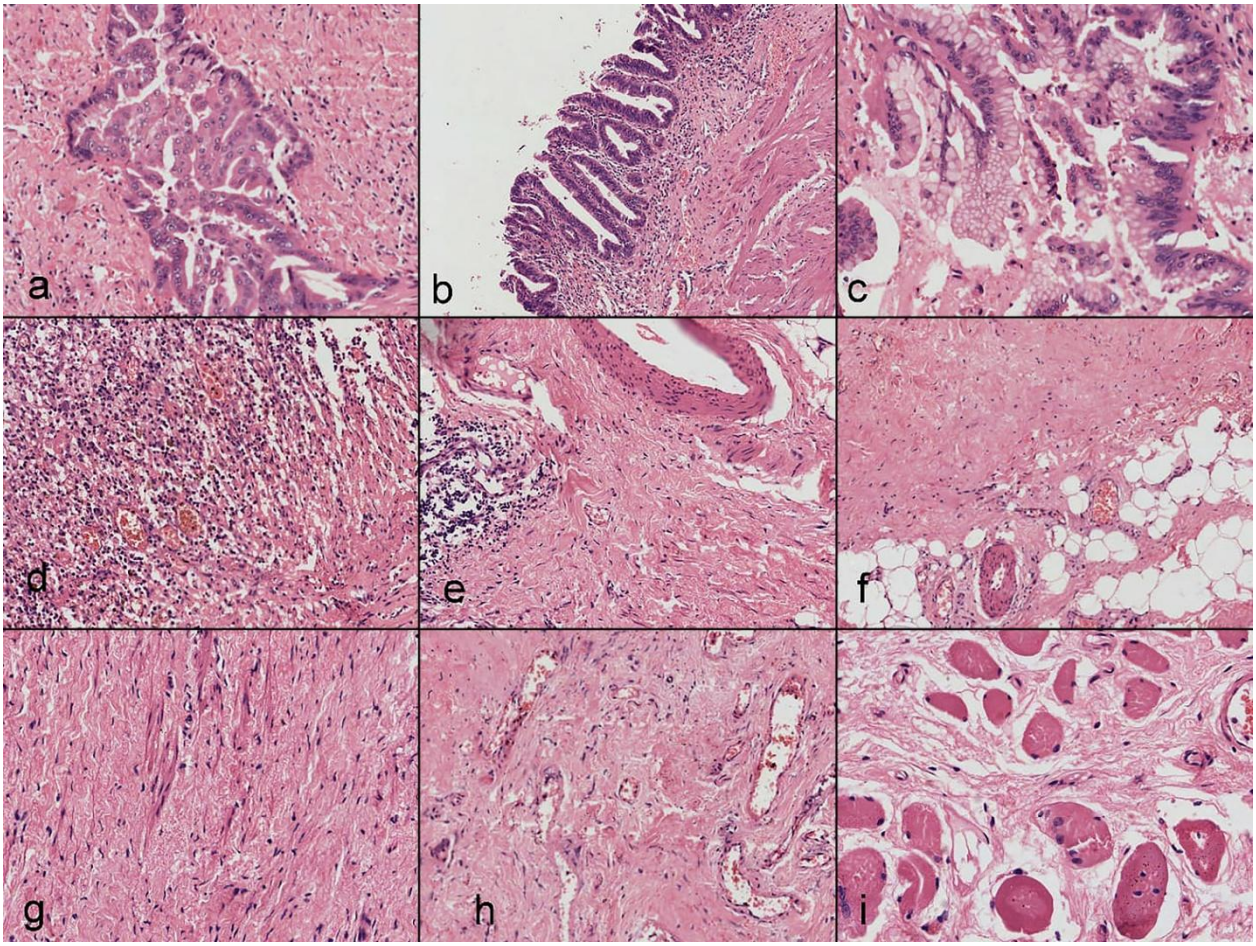

Figure 1: (a) Photomicrograph shows metaplastic squamous epithelium lining the wall of cyst (HEx 400). (b) Photomicrograph shows columnar epithelial cells lining inner luminal surface of cyst (HEx100). (c) Photomicrograph lining the inner surface of cyst wall. Photomicrograph shows inner lining by columnar cells and mucin-secreting epithelial cells (HEx 400). (d) Photomicrograph shows granulation tissue formation in the wall of cyst (HEx 100). (e) Photomicrograph shows focal collection of lymphocytes in the wall of the cyst (HEx 100). (f) Photomicrograph shows fibrofatty tissue and a arteriole in section from wall of cyst (HEx 100). (g) Photomicrograph shows thick band of fibrosis in the wall of cyst (HEx 400). (h) Photomicrograph shows angiogenesis, suggesting new capillary formation(HEx 100). (i) Photomicrograph shows fibromuscular tissue of levater ani (HEx 100).

Discussion

Tailgut cyst is an embryonic vestigial structure which may increase in size and may gradually become symptomatic due to its mass effect. Clinically, the patient may develop urinary obstructive symptoms due to occlusion of ureteric opening. Patient may develop urinary frequency or urinary retention. Further, the patient may also develop constipation or other symptoms like diarrhea. Rarely, malignant tumours e.g. adenocarcinoma[4], carcinoid tumour[5] and/or squamous cell carcinoma may develop. Morever, rectal or anal fistula may also

develop as a result of communication between cyst and wall of rectum or anus. Tailgut cysts are the embryonic remnants of the postnatal component of the hindgut[3]. As the embryo folds during the fourth week of gestation, the cloacal membrane progresses ventrally and encloses a portion of the future gut that is distal to the eventual hindgut-a region known as the tailgut. Majority of tailgut cysts are asymptomatic and may be missed on per rectal examination[3,6]. Rarely, severe sciatica-like pain may develop[3]. Overall rate of malignant transformation may be as high as 26%[7]. Furthermore, tailgut cyts may be associated with elevated carbohydrate antigen- Ca19-9[8].

Conclusion

A rare case of tailgut cyst without a malignant change is described. She was operated by laparoscopy and the cyst was sent to us for histological examination. Cyst wall was lined partly by squamous cells and partly by columnar or cuboidal cells. At places, cyst was lined by mucin-secreting cells. Cyst contained thick mucoid fluid. The patient was operated and was cured post-operatively.

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