Chronic perianal fistula: beware of rectal duplication

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Summary: A child with cystic duplication of the rectum containing ectopic duodenal mucosa and aberrant pancreatic tissue presenting as a chronic perianal fistula is reported. The duplication was removed by subtotal excision of the cyst along with mucosal sleeve resection from the common septum with the rectal wall.

Introduction

Rectal duplications are rare and usually present as a retrorectal mass lesion. We report a child who presented with a chronic perianal fistula secondary to rectal duplication. We are aware of only one earlier report of duplication of rectum with such an unusual presentation in the paediatric age group.¹

Case report

A 9 year old girl was admitted with a perianal fistula and pruritus ani since the age of one year. The child was normal at birth but developed a painful and warm swelling in the left perianal region at one year of age, incisional drainage of which resulted in a nonhealing perianal fistula.

The anus was normal and there was cutaneous excoriation surrounding the fistulous opening situated about 2 cm from the left side of the anal verge (Figure 1). Thin purulent fluid was being discharged from the fistula. Digital rectal examination revealed a well defined oblong cystic mass of about 5 × 3 cm size in the retrorectal area. Compression of this mass resulted in pus discharge from the perianal fistula. Other physical examination was normal. An injection of radiopaque dye into the fistulous tract demonstrated a smooth walled cyst in the retrorectal region (Figure 2).

After controlling the infection with antibiotics and saline irrigations, the cyst was excised through a perineal approach. A para-anal curved incision was made, the sinus tract was dissected off the ischiorectal

Figure 1 Clinical photograph of the child with chronic perianal fistula.

fossa and the cyst was excised from all aspects except anteriorly where it was found to have a common wall with the rectum. The mucosa of the cyst was cored out at this region leaving the common wall with the rectum intact. The resultant cavity was allowed to heal secondarily.

The postoperative recovery was uneventful. Duodenal mucosa and islands of aberrant pancreatic tissue were demonstrated on histology of the cyst wall. No ecto- or mesodermal elements were detected in any of the sections.

Discussion

Cystic duplications of rectum usually present as a retrorectal mass lesion, prolapse through the rectum,

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Figure 2 Lateral view of the pelvis with contrast injection into the fistula showing a well defined cystic lesion in the retrorectal area. Air is seen in the rectum.

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bleeding per rectum, abnormal opening into the vagina or urinary tract or they may be discovered incidentally from infancy to old age with or without associated anomalies.^{2,3,4} They are known to undergo carcinomatous change.

The differential diagnosis includes anterior meningocele, benign cystic teratoma, dermoids or pilonidal cyst. The cutaneous excoriation and dermatitis may indicate secretions from ectopic gastric mucosa and/or pancreatic tissue.

Whenever heterotopic mucosa or tissue is suspected, total excision of the cyst mucosa is mandatory and septostomy with internal drainage of the cyst at its distal end (as practised sometimes in duplications of other areas)⁵ into the normal rectum should be avoided as it does not remove the mucosa of the cyst. Excision of the cyst with resection of the part of the rectal wall and its primary closure is difficult and is not free of complications. Subtotal excision of the cyst, from the posterior and lateral aspects combined with mucosal sleeve resection of the anterior cyst wall, offers a logical and effective alternative in the surgical treatment of these rare duplications of rectum containing heterotopic tissue.