

External cystic rectal duplication: an unusual presentation of rectal duplication cyst

Karaman I, Karaman A, Arda N, Çakmak O

ABSTRACT

Duplications of gastrointestinal tract are rare anomalies, and rectal duplications account for five percent of the alimentary tract duplications. We present an unusual case of rectal duplication, which was located externally in a newborn female, and discuss the types of distal hindgut duplications.

Keywords: duplication cyst, external cystic rectal duplication, gastrointestinal duplication, rectal duplication

Singapore Med J 2007; 48(11):e287–e288

INTRODUCTION

Gastrointestinal tract duplications are rare congenital anomalies. They may occur throughout the alimentary tract and only 5% of them occur in the rectum.⁽¹⁾ Most of the rectal duplications are located in the retrorectal space.^(2,3) We present a case of cystic rectal duplication that was derived from the dentate line, and which had prolapsed through the anus and was located externally. To our knowledge, this form of rectal duplication has not been previously described in the English language literature.

CASE REPORT

A newborn female, delivered vaginally at 36 weeks gestation to a 24-year-old mother, was referred to our hospital for the problem of a large mass prolapsing through her anus. Prenatal ultrasonography was not performed. On perineal examination, a cystic mass measuring 6 cm × 7 cm, derived from the dentate line, was detected (Fig. 1). The mass was protruding posterior to the dentate line in a bundle that was of 1.5 cm width and 3 mm thickness. Radiographs of the lumbosacral spine were normal. The mass was excised totally via the transanal route and the defect in its place was repaired primarily. Upon sectioning the mass, it was found to be filled with mucous fluid. Pathological examination revealed that the wall of the cystic mass consisted of nonstriated muscle layers and rectal mucosa that is pathognomonic to rectal

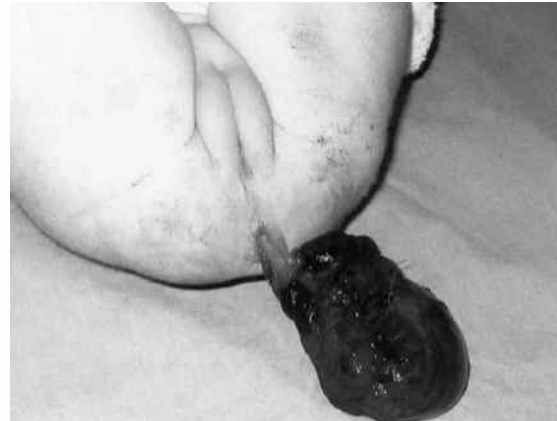


Fig. 1 Photograph shows the cystic mass derived from the dentate line.

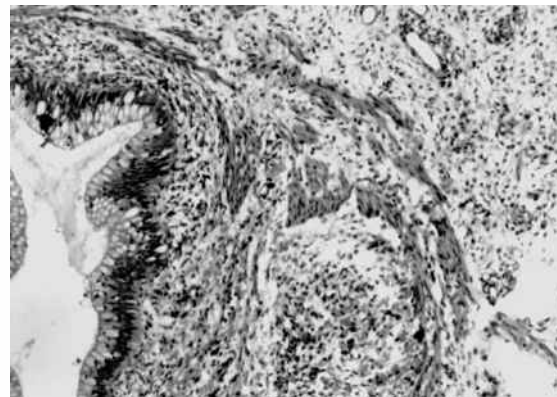


Fig. 2 Photomicrograph shows that the wall of cystic lesion consists of rectal mucosa and smooth muscle layers (Haematoxylin & eosin, × 200).

duplication (Fig. 2). At one year follow-up, no clinical recurrence was observed and the defaecation function of the child was normal.

DISCUSSION

Distal hindgut duplications are rare forms of intestinal duplications. Duplications in this region are classified as anal canal duplications (ACD) and rectal duplications (RD). ACDs are located at the posterior side of anal canal with an external

Department of
Paediatric Surgery,
Dr Sami Ulus
Childrens Hospital,
Babür Cad.,
Altındag 06080,
Ankara,
Turkey

Karaman I, MD
Paediatric Surgeon

Karaman A, MD
Paediatric Surgeon

Çakmak O, MD
Associate Professor
and Director

Department of
Pathology

Arda N, MD
Pathologist

Correspondence to:
Dr Ibrahim Karaman
Köylüler sok. 15/2,
Cebeci 06590,
Ankara,
Turkey
Tel: (90) 312 305 6208
Fax: (90) 312 317 0353
Email: ibrahimkaraman
@hotmail.com

opening just behind the normal anus, at six o'clock at lithotomy position, without any communication with the anorectum. This type of duplication is usually asymptomatic, and diagnosis can be made by perineal examination. The wall of ACD consists of transitional or squamous epithelium, smooth-muscle fibres and apocrine glands.⁽⁴⁾

RDs are generally cystic and they may be located at the posterior, lateral or anterior side of the rectum.⁽⁴⁾ 20%–45% of RDs have fistulous communication with the perineum or anus at the posterior⁽⁵⁾ or anterolateral⁽²⁾ position. Presenting symptoms include intestinal obstruction, bladder outlet obstruction, dysuria, pelvic pain, mucous or purulent drainage from the rectum or perianal fistula, rectal bleeding from the presence of heterotopic gastric mucosa, constipation and rectal prolapse. The wall of RD consists of colonic or anorectal mucosa, which sometimes includes squamous or urothelial cells and smooth muscle layers.^(2,5)

The prolapse of the intussusceptum, rectal polyp or rectal prolapsus can be considered in the differential diagnosis, but in this case, all these could be ruled out clinically. In our case, the mucous-filled cystic lesion, which was derived from the dentate line, was completely externally located. The wall of the cystic lesion consisted of rectal mucosa and smooth muscle layers. To our knowledge, this form of RD has not been published previously.

REFERENCES

1. Park WH, Choi SO, Park KK. Cystic rectal duplication: a rare cause of neonatal bladder-outlet obstruction and hydronephrosis. *Pediatr Surg Int* 2001; 17:221-3.
2. Knudtson J, Jackson R, Grewal H. Rectal duplication. *J Pediatr Surg* 2003; 38:1119-20.
3. Kızılcın F, Tanyel FC, Kale G, Hiçsönmez A. Duplication of the rectum resembling a juvenile polyp. *Turk J Pediatr* 1992; 34:193-5.
4. Jacquier C, Dobremez E, Piolat C, Dyon JF, Nugues F. Anal canal duplication in infants and children – a series of 6 cases. *Eur J Pediatr Surg* 2001; 11:186-91.
5. La Quaglia MP, Feins N, Eraklis A, Hendren WH. Rectal duplications. *J Pediatr Surg* 1990; 25:980-4.