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Case Report

Rectal duplication cyst with divarication of recti and urinary retention in a neonate

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ARTICLE INFO

Article history:

Received 9 September 2017

Accepted 30 November 2017

Available online 27 December 2017

Keywords:

Rectal cyst

Rectal duplication cyst

Divarication of recti

Neonate

Urinary obstruction

Introduction

Rectal duplication cysts are the rarest of all intestinal duplications. They account for approximately 4% of all intestinal duplications.¹ Worldwide only about 100 cases are reported so far.^{2,3} They usually present in childhood with varying presentation like constipation, tenesmus, urinary retention, bleeding, prolapse, with localized infection, as fistula and even rarely transforming into malignancy.^{1,3–5} We report a neonate with rectal duplication cyst for its rarity, unusual presentation and discuss pertinent review of literature.

Case report

A female new born patient with normal antenatal ultrasound findings, was noticed at birth, to have a midline abdominal swelling on anterior abdominal wall suggestive of divarication of recti (Fig. 1). Per abdomen no mass was palpable and the anal opening and the external genitalia were normal. On day 3 of life neonate developed recurrent urinary retention which did not respond to gentle suprapubic pressure and required repeated bladder catheterisations to drain the urine. Post-urinary drainage a 4 cm × 3 cm lump was felt in the deep pelvis region. Plain X-ray of abdomen and spine were normal.

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<https://doi.org/10.1016/j.mjafi.2017.11.008>

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Fig. 1 – Divarication of racti noticed at birth.

Ultrasound abdomen on day 3 revealed distended urinary bladder and a well-defined cystic hypoechoic lesion (31 mm × 17 mm) in presacral pelvis location compressing both the ureters causing bilateral hydronephrosis. Magnetic Resonance Imaging (MRI) abdomen on day 4 of life revealed 38 mm × 43 mm × 83 mm pelvico abdominal cystic lesion with well-defined fluid level, right anterolateral displacement of uterus and urinary bladder and compression of bilateral distal ureters by the cystic mass, resulting in bilateral hydronephrosis (Fig. 2). Laboratory investigations revealed normal haematological parameters, serum electrolytes and renal function tests. Though the ultrasound raised the possibility of anterior sacral meningocele or sacrococcygeal teratoma, anterior sacral meningocele was ruled out by X-ray of spine which showed no bony defect. Similarly, Currarino triad (a triad of pre sacral mass, sacrococcygeal bone defect and anorectal malformation)⁶ was also ruled out as there was no ano rectal malformation and no sacrococcygeal osseous defect. We also ruled out sacrococcygeal teratoma by the MRI. Bladder diverticulum was another possibility, but the MRI showed the mass not to be communicating with bladder. Meanwhile the baby remained on urinary catheter, and was on feeds awaiting surgery. The abdomen remained distended, though she passed stools normally.

The neonate was subjected to surgery on day 7 of life in view of increasing size of the cyst mass and urinary retention due to compressing of bladder neck. Intra-operatively cystic swelling was approximately 5 cm in diameter located antero-medial to rectum and sharing common wall with rectum. The cyst was communicating with rectum about 2 cm proximal to anal verge (Fig. 3). The cyst contained thick mucoid material and inspissated meconium. The cyst was completely excised leaving a portion of cyst wall common with normal rectum (Fig. 4). The site of communication was closed with interrupted sutures. Proximal right transverse loop colostomy was done to protect the distal repair. Stoma started functioning from 2nd

Fig. 2 – MRI abdomen: T2 weighted sagittal image showing a large well encapsulated pelvico abdominal cystic lesion with air fluid level within the mass, displacing the uterus anteriorly.

post-operative day and baby was started on oral feeds, which she tolerated well. Histopathology of the excised cyst revealed a fibro collagenous tissue with chronic granulation. On follow up, at 1 month of age, the baby was gaining weight normally and had a healthy and functional colostomy.

Discussion

Duplications of alimentary tract are rare anomalies, characterized by an epithelial lining, well developed muscle coat and attachment to any portion of gastrointestinal tract. Rectal

Fig. 3 – Intra op image showing the point of communication of the duplication cyst with rectum.

Fig. 4 – Intra op image showing margins of the rectal duplication cyst.

uplications are a very small subgroup of these intestinal duplications. In rectal duplications, posterior rectal duplications are more common than the anterior ones.³ One of the largest case series involving 11 of rectal duplications had a presentation ranging from new born to 18 years,⁷ though there are instances of presentation even in adults as well.⁸ The clinical presentation is variable, depending on the location and size. The common clinical presentations are with constipation, intestinal prolapse as 'polyp', perianal swelling, presacral mass, perineal fistula, bleeding per rectum, or with urinary obstruction.^{5,7}

Our case presented with large divarication of recti at birth and recurrent urinary retention on day 3 of life. Association of divarication of recti in this patient could be due to antenatal

foetal urinary retention causing increase in intra-abdominal pressure and thus widening of inter recti distance. Pre-op diagnostic challenge was to find out the origin of the cyst. The differential diagnoses of the rectal duplication cyst, were anterior sacral meningocele, sacrococcygeal teratoma, Currarino triad, and bladder diverticulum. In our case the diagnosis was eventually confirmed intra-operatively. The management remains complete excision with preservation of anorectal functions.⁹ Surgical excision is curative, provides relief from the symptoms and prevents the complications. The options for surgical access are transanal, transcoccygeal and posterior sagittal (most preferred approach).¹⁰ Our patient was operated by pfannenstiell incision, as the cyst was anteriorly located.

To conclude, we have presented a case of rectal duplication cyst, the rarest of all intestinal duplications, which has unusual presentation with divarication of recti at birth and as a rare cause of recurrent urinary retention during very early neonatal period.

Conflicts of interest

The authors have none to declare.

REFERENCES

1. Flint R, Strang J, Bissett I, Clark M, Neill M, Parry B. Rectal duplication cyst presenting as perianal sepsis: report of two cases and review of the literature. *Dis Colon Rectum*. 2014;47(12):2208–2210 [Epub 20.01.05].
2. Patil PS, Kothari P, Gupta R, et al. An unusual presentation of rectal atresia with rectal duplication cyst in a neonate: a case report. *Indian J Case Rep*. 2015;1(2):47–49.
3. Gupta V, Sharma SB. Cystic rectal duplication in infants: a case report and review of literature. *J Indian Assoc Pediatr Surg*. 2006;11(3):153–154.
4. Delarue A, Garcia Meric P, Martin C, et al. Antenatal rupture of diverticular rectal duplication with neonatal perineal fistulization. *Pediatr Surg Int*. 1998;13(4):288–289.
5. Park WH, Chai SO, Park KK. Cystic rectal duplication: a rare cause of neonatal bladder outlet obstruction and hydronephrosis. *Pediatr Surg Int*. 2001;17(2–3):221–223.
6. Vinod MS, Shaw SC, Devgan A, Mukherjee S. The currarino triad. *Med J Armed Forces India*. 2017. <http://dx.doi.org/10.1016/j.mjafi.2017.07.006>.
7. La Quaglia MP, Feins N, Eraklis A, Hendren WH. Rectal duplications. *J Pediatr Surg*. 1990;25(9):980–984.
8. Jackson KL, Peché WJ, Rollins MD. An unusual presentation of a rectal duplication cyst. *Int J Surg Case Rep*. 2012;3(7):314–315.
9. Amjadi K, Poenaru D, Saboleski D, Hurlbut D, Kamal I. Anterior rectal duplication: a diagnostic challenge. *J Pediatr Surg*. 2000;35:613–614.
10. Delarue A, Garcia-Meric P, Martin C, et al. Antenatal rupture of a diverticular rectal duplication with neonatal perineal fistulisation. *Pediatr Surg Int*. 1998;13:288–289.