

Neonatal Rectal Duplication Cyst: A Rare Presentation

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Abstract

Case Report

Rectal duplication cysts are a rare clinical entity accounting for 4% of all intestinal duplications. The presenting symptoms depend upon their size, location and age of the patient. We report a rare neonatal presentation of rectal duplication cyst which was present as a swelling in the infracoccygeal region in a female child since birth. MRI suggested a cystic swelling adjacent to the rectum. Excision was done by posterior sagittal approach at 1 month of age. Histopathology confirmed it to be a rectal duplication cyst.

Keywords: Rectal, duplication, neonate, cyst.

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INTRODUCTION

Rectal duplication cysts are a rare clinical entity accounting for 4% of all intestinal duplications [1]. These cysts, like other intestinal duplication cysts, are lined by intestinal mucosa and may or may not have submucosal and smooth muscle layers. The presenting symptoms depend upon their size, location and age of the patient. Many of the rectal duplication cysts remain undetected till adult and are found on routine imaging done for some other reason. If the cyst compresses the rectum, it can cause constipation, obstruction or rectal bleeding [2]. If the cyst compresses bladder neck or the distal ureter, it can result in urinary tract obstruction causing urinary infections and back pressure changes [3].

Neonatal presentations are rare. Our patient presented with an external swelling mimicking a sacrococcygeal teratoma or a meningocoele, which has not been documented before as per our literature search.

CASE REPORT

One day old female child delivered at our hospital having a swelling in the infracoccygeal region, just above the natal cleft. The swelling was cystic in nature, 3cm x 3cm in size and more towards the right buttock (Figure 1). Patient passed stools and urine normally after birth.

An ultrasound of local region and abdomen-pelvis revealed a trilobular cystic lesion of size 3x3x2 cm in size, extending into the pre coccygeal region, adjacent to the rectum. There was no communication seen with the rectum.

A magnetic resonance imaging (MRI) of pelvis and local region was done on day of life 5. The report suggested a 3.2cm x 3cm x 2.8cm trilobular cyst in the retro-rectal and pre-coccygeal region (Figures 2 & 3). Differential diagnoses given were:

- Retro-rectal hamartoma/tail gut cyst.
- Lymphangioma.
- Rectal duplication cyst.

All routine haematological investigations were normal. It was decided to follow up the patient on a weekly basis to monitor any sudden increase in size and excise the lesion electively at one month of age.

Patient was operated in prone jack-knife position via muscle complex saving posterior sagittal approach (MCS- PSA) with a red rubber catheter in the rectum. All 3 loculi of the cyst were delineated. 2 of the loculi were excised intact while the third locule got punctured during dissection, hence its wall was completely excised (Figure 4 & 5). The largest locule was adherent to the rectal wall but could be separated without any damage to the rectum. There was no communication between the cyst and the rectum.

Post-operatively the rectal catheter was kept for 3 days and patient discharged after 5 days of intravenous antibiotics. Patient was followed up on post-operative day 10 for suture removal and there was no wound infection.

Histopathological diagnosis was rectal duplication cyst with a fibrocollagenous wall and lining of low cuboidal and columnar cells in monolayer with few short isolated papillary proliferations.

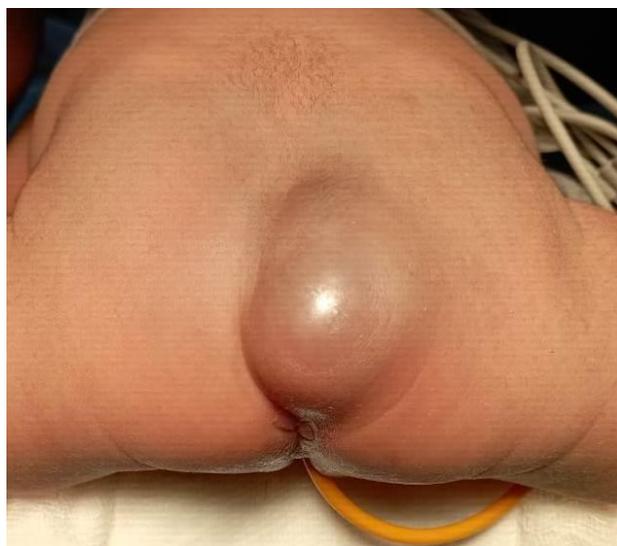


Figure 1

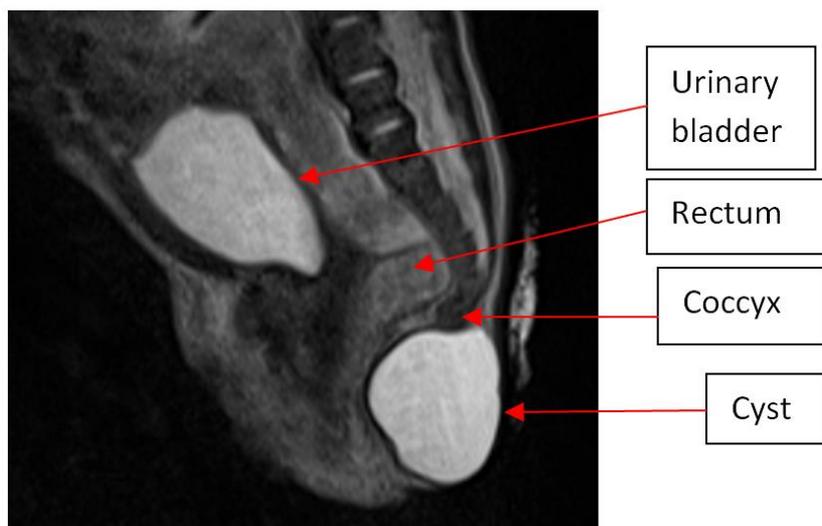


Figure 2

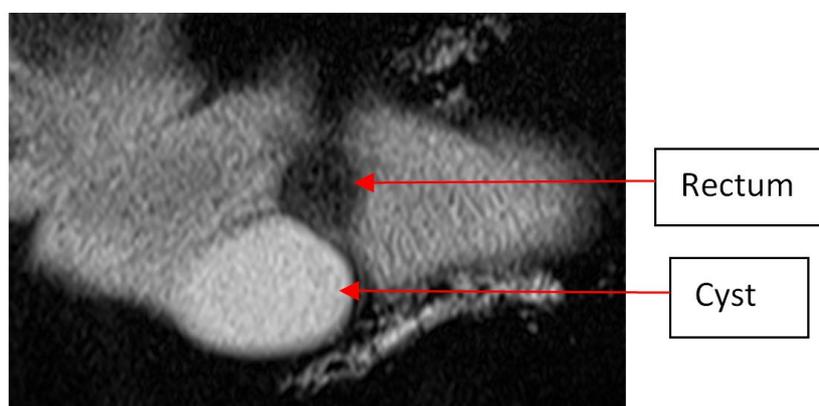


Figure 3



Figure 4



Figure 5

DISCUSSION

Ladd and Gross first described intestinal duplication cysts in 1941 as cysts lined with various types of gastrointestinal mucosa and smooth muscle wall while rectal duplication cyst was described by Middeldorf in 1885 [4]. Approximately 100 cases of rectal duplication have been reported till now [5]. Many presentations of rectal duplication have been documented like rectal bleeding, infection, fistulisation and mass effects like constipation and urinary

symptoms [6]. Our patient presented with a swelling in the infra-coccygeal region just above the natal cleft extending into the right buttock.

MRI is the preferred investigation as it gives detailed account of the relations of the cyst and communication of the cyst with rectum, if any.

Surgical excision is curative. Many surgical approaches have been described like transanal [6], transcoccygeal [1], abdominal [2] and posterior sagittal

[7]. Our patient was feasible for posterior sagittal approach. Care must be taken to excise the cyst in totality without leaving any remnant and taking care not to damage the anal sphincter complex.

We are presenting this case as a rare presentation of rare disease. Although rectal duplication cysts are a known entity, they are extremely rare with only about 100 cases reported. And even rarer is the presentation of swelling in the buttock region mimicking a meningocele or a sacrococcygeal teratoma. Other differential diagnoses should also be kept in mind like lymphangioma and tail gut cyst as their presentation can be similar.

REFERENCES

1. Stringer, M. D., Spitz, L., Abel, R., Kiely, E., Drake, D. P., Agrawal, M., ... & Brereton, R. J. (1995). Management of alimentary tract duplication in children. *British journal of surgery*, 82(1), 74-78.
2. Mboyo, A., Monek, O., Massicot, R., Martin, L., Destuynder, O., Lemouel, A., & Aubert, D. (1997). Cystic rectal duplication: a rare cause of neonatal intestinal obstruction. *Pediatric surgery international*, 12(5), 452-454.
3. Park, W. H., Choi, S. O., & Park, K. K. (2001). Cystic rectal duplication: a rare cause of neonatal bladder-outlet obstruction and hydronephrosis. *Pediatric surgery international*, 17(2), 221-223.
4. Kraft, R. O. (1962). Duplication anomalies of the rectum. *Ann Surg*, 155, 230-2.
5. Knudtson, J., Jackson, R., & Grewal, H. (2003). Rectal duplication. *J Pediatr Surg*, 38, 1119-20.
6. La Quaglia, M. P., Feins, N., Eraklis, A., & Hendren, W. H. (1990). Rectal duplications. *Journal of pediatric surgery*, 25(9), 980-984.
7. Gupta, V., & Sharma, S. B. (2006). Cystic rectal duplication in infants: A case report and review of literature. *Journal of Indian Association of Pediatric Surgeons*, 11(3), 153-154.