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Case Report

A congenital long rectoscrotal fistula: A rare variant ☆

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ABSTRACT

Congenital rectoscrotal fistula is a rare variant of anorectal malformations (ARM) as per pena classification. Its nomenclature as well as the management, obviously has not been discussed in literature so far. One such rare case and its management in a male neonate have been discussed here. Non or occasionally deflating congenital recto scrotal fistula has to be managed like an intermediate ARM.

Introduction

Anorectal malformations (ARM) have an incidence ranging from 1:1,500 to 1:5,000 live births, with mild variants requiring only minor surgical interventions to more complicated ones needing multi-staged procedures. Though the cause of ARM is unknown, the arrest in the descent of the urorectal septum towards the cloacal membrane between the 4th to 8th weeks of gestation has been suggested.

Rectoperineal fistula, first named by Wilkinson, is an intermediate anomaly wherein a bowel of normal or greater caliber ends above the diaphragmatic part of the levator ani and has a long narrow tract that passes between the limbs of the sling fibers to reach the anterior perineum, the scrotum, or the ventrum of the penis of boys and the vestibule of girls.

Case summary

A day 1male neonate born by full term vaginal delivery, to a nonconsanguineously married gravid 3 mother, with 2.5 kg birth weight noted to have anorectal malformation and hence referred to us for further management. On examination there was an absence of an anal orifice, but a speck of meconium was seen in the middle of the scrotum (Fig. 1A). Prone cross table lateral shoot through radiography was done showing presence of gas shadow below pubococcygeal line. In view of persistent abdominal distension even after 24hours without any deflation of meconium from fistulous orifice; a high sigmoid colostomy was done (Fig. 1B).

Baby used to pass slight mucus from the recto scrotal fistulous site on follow up. Pressure augmented distal loop

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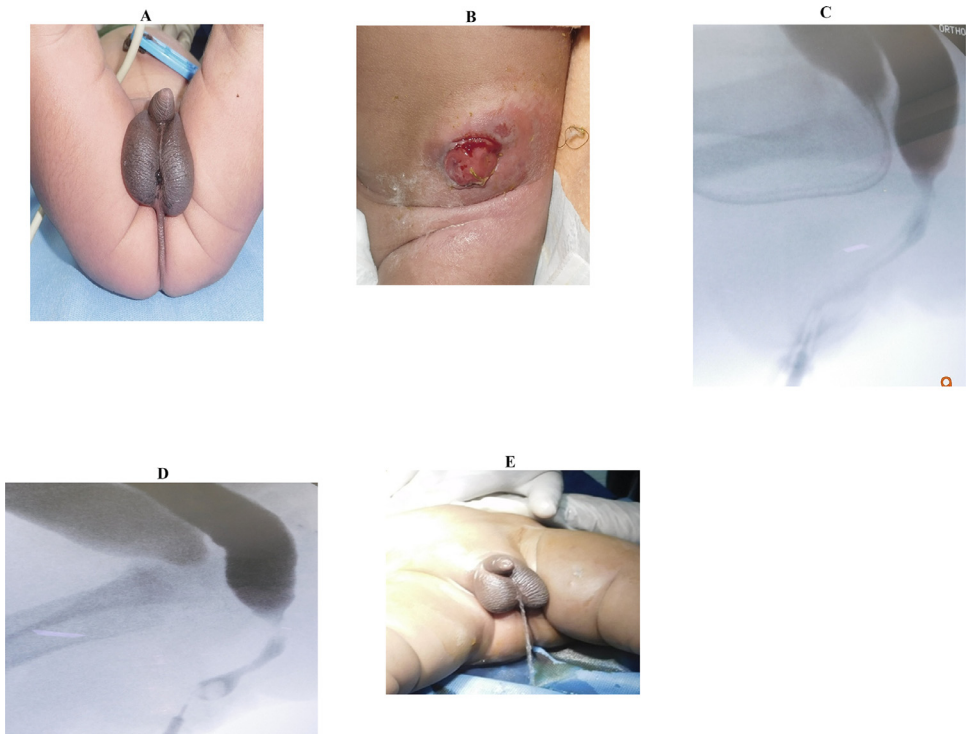

Fig. 1 – (A)—Clinical appearance of rectoscrotal fistula. (B)—high sigmoid colostomy. (C)—augmented distal loopogram showing no connection to the urinary tract. (D)—ante grade dye study showing long recto scrotal fistula without any connection. (E)—dye passed via fistula.

cologram on follow up revealed, distal pouch of bowel getting narrowed to a long fistulous tract curving anteriorly to the middle level of scrotum running parallel to the urethra without having any communication with it (Fig. 1C). Same was noticed via retrograde urograffin dye study (Figs. 1D and E). Subsequently baby underwent posterior sagittal anorectoplasty(PSARP). Rectum with its fistulous tract directed towards scrotum was disconnected and anorectoplasty was done after laying open the fistulous tract in the perineum (Figs. 2F–H). Subsequently colostomy closure was done and baby is now continent of stools and urine (Fig. 2I).

Discussion

As per Peñas classification, the commonest defect in males is the rectourethral fistula followed by the cutaneous perineal fistula [1].

A rectoperineal fistula is traditionally known as a low defect where in, the rectum is located within most of the sphincter complex, only the lowest part of the rectum is anteriorly mislocated . Sometimes the fistula does not open into the perineum, but rather follows a sub epithelial midline tract, opening somewhere along the midline perineal raphe, scrotum, or even at the base of the penis. The diagnosis is established by perineal inspection alone requiring anoplasty [1–4].

In an intermediate level of anorectal malformations (ARM) in males, the hindgut terminates as a fistula into the lower urinary tract, but less commonly without a fistula, or having a fistula terminating externally in the perineum [1–4].

A congenital long rectoscrotal fistula has not been described so far in the literature. To the attending clinician it might look like a low ARM having meconium speck at the tiny fistulous orifice, but however, this tiny orifice is unable to deflate the thick meconium filled distal bowel. Even a prone cross table lateral shoot through radiography can reveal air shadow below the pc line; making the surgical decision challenging. To avoid the morbidity of inappropriate initial perineal surgical procedures injuring the delicate sphincter complex and the urethra, a diverting colostomy is an ideal option, in such doubtful embryological, anatomical and clinically intriguing cases in view of their diagnosis in the neonatal period.

Author, is presenting here a one day old male neonate having a long congenital rectoscrotal fistula without much deflation of the thick meconium loaded distal bowel associated with persistent abdominal distension .A high sigmoid loop colostomy at day 2, followed by posterior sagittal anorectoplasty(PSARP) was done when baby weighed 3kg and subsequently colostomy closure, once baby attained 5kg weight uneventfully. Baby is doing well on follow up.

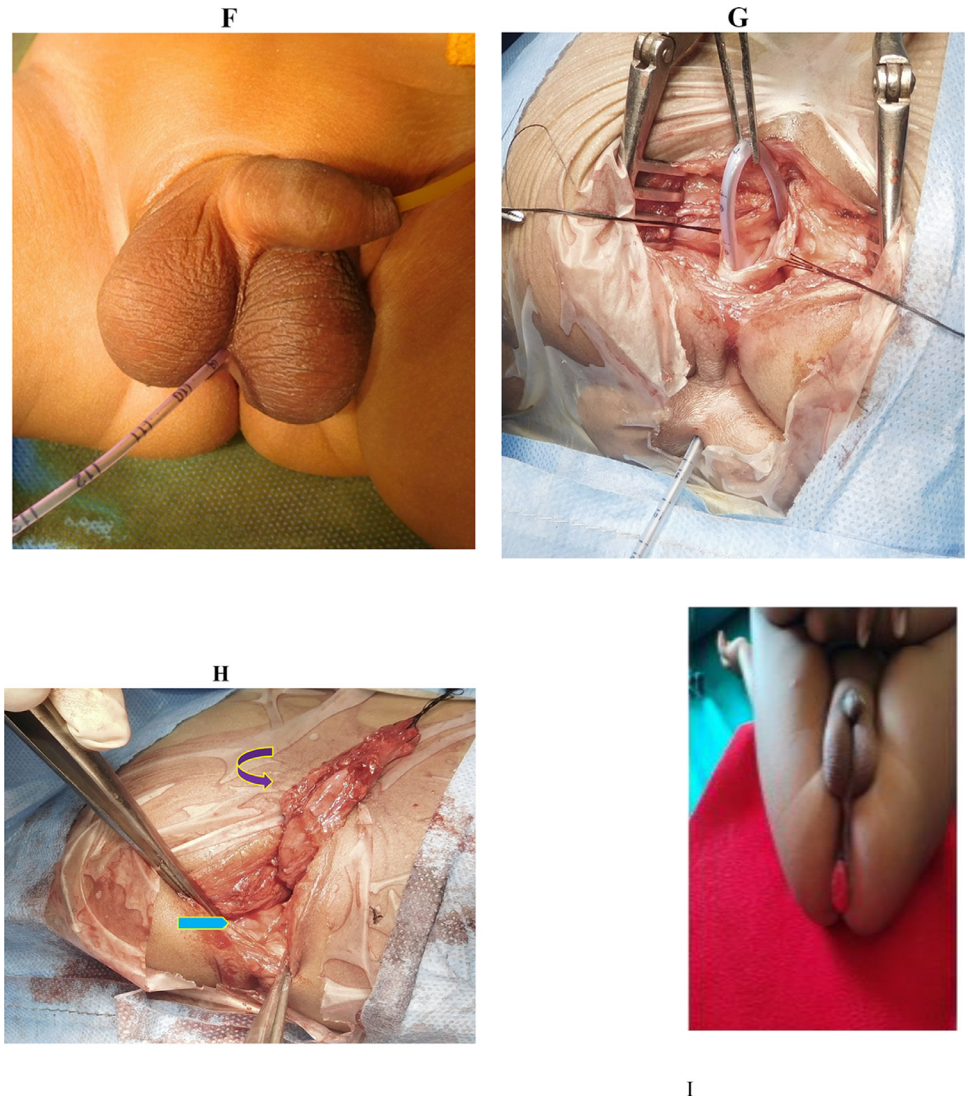

Fig. 2 – (F)—preoperative catheterization of the recto scrotal fistula with infant feeding tube (IFT). (G)—intraoperative delineation of distal rectum with IFT in it. (H)—Blue arrow depicting laid open fistulous tract after its disconcertion with rectum, Violet arrow pointing at distal bowel. (I)—postoperative appearance of PSARP.

Conclusion

Non or occasionally deflating congenital long recto scrotal fistula, has to be managed a like an intermediate ARM with an initial high sigmoid colostomy avoiding the morbidity of, possibility of urinary and fecal incontinence following initial perineal procedure.

The work has been reported in line with the SCARE 2020 Criteria [5].

Provenance and peer review

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Ethical approval

Yes

Author contribution

Author, Dr Jayalaxmi Shripati Aihole, has contributed in concept, design, drafting of manuscript, and approved final version of the manuscript.

Patient Consent

Both verbal and written informed consent has been obtained from parents for the publication of this manuscript

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Patient consent

Author has submitted signed consent form from legal guardian of the patient and available with editorial office.

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