Case Report

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Modified transanal repair of congenital H-type rectovestibular fistula: A technique to avoid recurrence

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ABSTRACT

Congenital H-type rectovestibular fistulas are rare in the spectrum of anorectal malformations. Repair is associated with recurrence rates of up to 30%, using perineal repair, vestibuloanal pull-through or anterior anorectoplasty. The rarity of the malformation has limited experience with the surgical approach; hence, the rate of wound breakdown. We describe the use of a modification of the transanal approach for the repair in a 7-month-old female patient.

Key words: Anorectal malformation, H-type fistula, transanal

INTRODUCTION

H-type rectovestibular fistula or rectovaginal fistula with normally sited anal openings are rare types of defects seen in the spectrum of anorectal malformations. Since the first description by Bryndorf and Madsen in 1960,[1] very few cases have been documented in the literature. There are two variants of these anomalies – a type seen in Asia, which tends to follow an inflammatory process,[2] and the type seen elsewhere in the absence of a preceding inflammation, that is, the congenital type.[3] The congenital type is extremely rare in sub-Saharan Africa, also, and only two cases have ever been documented, in the literature, from Africa – in a series of 1410 patients with ARM from six countries.[4]

H-type rectovestibular or rectovaginal fistulas have been treated by perineal repair, vestibuloanal pull-through, anterior anorectoplasty and posterior sagittal anorectoplasty.[3,5] The rarity of H-type fistulas, however, has not allowed significant experience in its management by any one paediatric surgeon and recurrence rates, following perineal wound breakdown after repair, have ranged from 5% to 30%.[2,5-7]

We recently treated a 7-month-old female who presented to our hospital with a congenital H-type rectovestibular fistula and herein describe the operative technique used to avoid recurrence.

CASE REPORT

A 7-month-old female presented with passage of faeces from the external genitalia since birth. The child was noticed to be passing stool from both the external genitalia and the anus. However, the quantity from the anus was more than that from the external genitalia. This was noticeably appreciated anytime the child had diarrhoea. There was no abdominal distension but the child strained to pass stools.

On examination, at presentation, she was neither pale nor febrile. The chest and cardiovascular system examination were essentially normal. Her external genitalia was stained with faecal matter and there was a fistulous opening at the vestibule of the vagina. The anus was patent and stained with faeces. A diagnosis of H-type rectovestibular fistula was made.

She had a fistulogram done, which showed a fistulous tract connecting the rectum to the external genitalia. Preceding scout film reported as showing what appeared to be a calcified mass at the centre of the fistula. She was then worked up for surgery.

Under general anaesthesia with inhalational technique, using a non-kinkable endotracheal tube, the patient was placed in the prone position with sand bags placed appropriately to allow for chest movements and prevent undue pressure against the abdominal cavity. The fistula was demonstrated under direct vision with a probe inserted in it [Figure 1a] at 1.5 cm above the anal verge.

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There was no presacral mass and no mass was found in the fistula.

An incision was made over the perineal body extending from the anterior anal circumference to the vestibule and deepened to the fistula. A modified transanal technique was used. Stay sutures (silk) were placed at the anocutaneous junction [Figure 1a] to facilitate dissection and mobilization of the rectum. An incision was made in the anterior part of the circumference of the anorectal canal, and adjoining the anal canal and the rectum mobilized from the level of the incision up to 1 cm above the fistula [Figure 1b].

Throughout the surgery, the posterior part of the anocutaneous circumference and adjacent anal and rectal walls were left intact to preserve faecal continence mechanisms [Figure 2a].

The fistula was resected with the distal part of the anterior anal canal and rectal wall. The vestibular end of the fistula was repaired in a single layer. The distal end of the normal rectal wall (anteriorly) was then pulled down and sutured to the anal canal [Figure 2b] to ensure that the repaired vestibular wall does not lie against a repaired rectal wall.[5] The perineal body was reconstructed using 4-0 polyglycolic sutures [Figure 2b].

The patient had a satisfactory post-operative recovery, was discharged home after 7 days stay without untoward incident and has been followed-up for 9 months. There is no recurrence and she no longer strains to pass stools.

DISCUSSION

Congenital H-type rectovestibular or rectovaginal fistulas are extremely rare entities in the spectrum of anorectal malformations constituting 0.7% of the malformations seen in females.[5] The fistula can be located in the vestibule, vagina or labia.[3,5] In addition to passage of stool through the vagina, H-type fistulas may be associated with anal stenosis or presacral mass and these should be excluded.[5] In the patient presented, neither of these associated anomalies was present.

In Asia, H-type rectovestibular or rectovaginal fistulas are much more common, with proportions as high as 3% to 7% of all types of anorectal malformations.[2,7,8] Furthermore, the variant of H-type fistula seen in Asia tend to follow an inflammatory process,[2,8,9] and are, often, not congenital as occurs in Europe[3] and North America.[5] This condition is also rare in Africa[4] and from the case reported here, the pattern of occurrence may be similar to the congenital variant seen in Europe and America, further confirming geographical variation in the etiology.

The diagnosis of H-type fistulas is usually made clinically. A fistulogram has been employed to outline the tract,[10] but the yield is often disappointing.[5] In our patient, the erroneous interpretation of the fistulogram made us to incise the perineal body during the surgery, looking for the 'calcified mass' that was reported. Examination under anaesthesia appears to be the best way of outlining the fistula, and a probe can be inserted to demonstrate this.

Earlier, children with H-type fistulas were usually operated after a diverting colostomy has been constructed.[1,7,8]

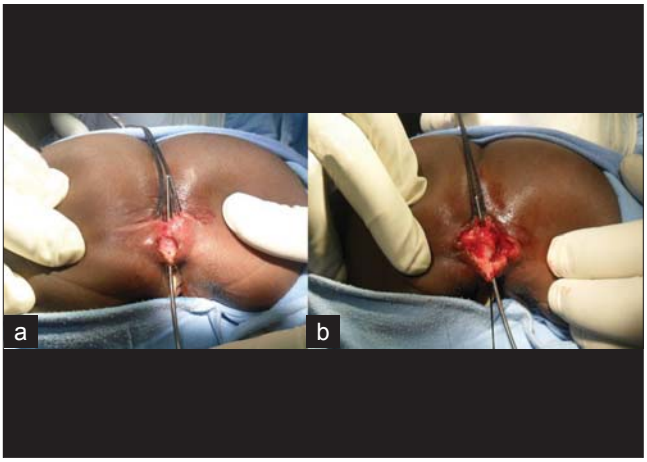


Figure 1: (a) Probe inserted into the fistula from the vestibular end and stay sutures placed at the anocutaneous junction to facilitate dissection. (b) Transanal mobilization of the anterior lip of the anorectal circumference and adjacent anal and rectal canal

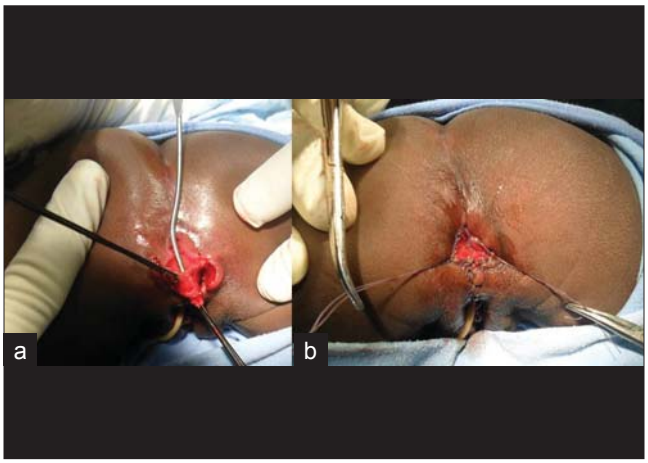


Figure 2: (a) The posterior part of the circumference of the anocutaneous junction with adjacent anorectal canal was left undisturbed. (b) Repair completed and perineal body reconstituted

More recently, however, most paediatric surgeons are comfortable with a primary definitive surgery, without this approach affecting the rate of wound dehiscence.[5] In the case reported here, the child was operated upon without a prior diverting colostomy and the outcome was satisfactory.

Although repair of these malformations in females has been described for over 50 years, there is still no consensus on the optimal treatment. The various surgical approaches that have been utilized include simple fistulectomy, fistulotomy, perineal repair, vestibuloanal pull-through, ASARP, ASARVP, PSARP and limited PSARP among others.[2,3,6,7,10] The option of treatment has been varied because of the occurrence of perineal wound dehiscence and fistula recurrence that have been reported to vary from 5% to 30%.[2,3,7] A transanal approach leaving the perineal body intact has recently been described as a good option for repair.[5] In this case, we employed a modification of the technique, because of the attempt to explore the fistula for the 'calcified mass'. The essential step in preventing wound breakdown is to prevent repaired surfaces of the vagina and rectum, after fistula excision, from being in contact. The fistula was relatively low, so we succeeded in mobilizing the rectum and pulling healthy rectum down, via transanal mobilization of the anterior and lateral rectal walls only, without undue tension on the recto-anal repair. In patients with the fistula located higher to prevent undue tension on the repair, a full circumferential mobilization of the rectum is advised, ensuring that the recto-anal anastomosis is made proximal to the dentate line, in order to preserve the anal continence mechanism.

We therefore recommend the transanal approach, similar to that used for pull-through in patients with the Hirschsprung disease for the treatment of the rarely encountered H-type rectovestibular or rectovaginal fistula, to prevent wound dehiscence or fistula recurrence.

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