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Congenital Rectal Atresia with Rectovestibular Fistula: A Case Report

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Abstract
Within the spectrum of anorectal malformations, congenital rectal atresia associated with rectovestibular fistulas is an uncommon and curable entity. It has been reported to have a prevalence of less than 1%. In this study, we report a 9-month-old female presented with the passing of feces andgas through the small orifice of the vaginal wall. The diagnosis of congenital rectal atresia associated with rectovestibular fistula was confirmed by radiological investigations. The patient was successfully treated with transfistula anorectoplasty. Here we present the case, management approach, and a review of the previous literatures.

Keywords: Anorectal malformation, Rectovestibular fistula, Case Report, Rectal Atresia, Transfistula Anorectoplasty.

Introduction

Imperforate anus is amongst the most prevalent birth defects with a global incidence of 1 per 5000 live births.(Kisra, Alkadi, Zerhoni, Ettayebi, & Benhammou, 2005) It is characterized by the absence of anal orifice and the anus being connected with the genitourinary apparatus via a small fistula or ending blind. Atresia ani in a female neonate combined with rectovestibular fistula (RVF) is an exceptionally uncommon disorder.(Aslam, Ashworth, & Spicer, 1996; Khan & Chana, 2013)

Poverty and ignorance have been the causes of a considerable percentage of imperforate anus cases in children who presented to tertiary hospitals throughout the neonatal or preschool periods in poorer regions, such as Yemen, where a major proportion of births are home deliveries.(Pandey, Gupta, Singh, & Verma, 2015) Therefore, we discuss this case due to the rare incidence of this abnormality.

Case Report

The parents of a 9-month-old female with a low-income background presented her to the hospital with the claims of nonexistent anal orifice and complaining that flatus and feces passed through the vestibular opening. The infant was born full-term by vaginal domestic delivery and was solely breastfed since delivery. Her mother did not take any medication through her pregnancy. She was attentive, well-nourished, and weighed 10 kg. Her social skills were typical for her age. On clinical examination, she had soft abdominal distension with redness in the perineum. The stool was observed coming from the small fistulous of about 1 cm in the vaginal wall (**Figure 1**). The ultrasonographic evaluation was normal with no further congenital anomalies discovered. Magnetic resonance imaging

(MRI) of the pelvis was performed, which revealed the anatomical details of RVF and absences of the anal canal.

Figure 1. show the rectovestibular fistula (black arrow) and blind anal canal opening (blue arrow).

Surgical Procedure

Preoperative bowel preparation with saline was carried out with clear fluids administered orally for 24 h until the effluent was clear. Moreover, she received a preoperative intravenous antibiotic. The surgery was performed in lithotomy position with general anesthesia and after foley insertion to the bladder. The anal dimple confirmed the suggested place of the anus. Furthermore, it was validated using a muscular stimulator, and the landmarks to the neonatal place were obtained by a simple stitch. To help the fistula separate from the rectovaginal wall and to reduce the bleeding, a diluted epinephrine solution (0.001%) was inserted beneath the mucosa of the fistulous area. To enhance the retraction during dissection, peri-fistula traction stitches were applied around the fistula wall. A circular incision was created on the mucosa of the fistula, and a submucosal plane was accessed. Next, dissection was continued upward meticulously dividing the shared rectovaginal wall up to the cervix anteriorly, and posteriorly up to the sacral promontory. There was no incision made across the perineum, and the perineum was preserved. The complete width of the distal rectum was achieved at this level. Afterwards, an approximately 1 cm U shape incision was made at the site of the external sphincter complex muscle, which had been marked previously with a plain suture. A hole was produced in the external sphincter complex through which the mobilized rectum was dragged. The main fistula site was repaired in layers with 5-0 vicryl interrupted sutures. Anoplasty was performed with 12 sutures and 5-0 vicryl. Foley catheter was kept in situ for 7 days for better perineal care (**Figure 2**). Oral feeding was started 6–9 h postop and the patient was given fluids and broad-spectrum antibiotics intravenously until the third day post-operation. Afterwards, oral cephalosporin was administered for 7 days. Currently, the patient is passing stools from the normal anal orifice without complication. Informed consent was obtained from the patient's guardians to publish their case details and any accompanying images.

Figure 2. show the postoperative neoanus site.

Discussion

Solo anorectal agenesis in a female newborn is a very uncommon disorder, and its connection with fistulous, such as RVF is much more exceptional with a report of less than 1%.(Khan & Chana, 2013) Although the exact cause of atresia ani is unclear, intravascular thrombosis could be suggested as the main cause.(Aslam, et al., 1996) Careful perineal examination, vaginoscopy, and anal sphincter electrical stimulation are essential to make an accurate diagnosis. A small probe should be used gently to locate any fistulous connection.(Upadhyaya, et al., 2008) In our patient, the fistula orifice was located in the vestibular area of the vaginal wall, which is consistent with the diagnosis of RVF.

Radiological assessment is significant importance in identifying these abnormalities through various modalities. A plain X-ray helps reveal the characteristics of bone anomalies. Uranography and MRI are used to examine the remaining related abnormalities. Conventional investigations with contrast will precisely show the fistulous information that will determine the management route.(Sharma, Sharma, Bhardwaj, Dewan, & Aziz, 2017)

The age of patients presenting with anorectal agenesis with RVF ranged from birth to one year, with a mean of three months.(Choudhury, et al., 2017) Our patient was diagnosed 9 months after delivery.

The anatomic locations of the fistula might vary in neonate females. Chatterjee et al. classified these cases into two categories with Group 1 including the cases in which the fistula tract arises in the anus just overhead of levator ani and matches the typical kind of RVF. Group 2 entails the patients in whom the tract is positioned under the levator ani known as the "perineal canal".(Chatterjee, 1980)

Different approaches were authorized for treating this malformation, such as a transanal end-to-end rectorectal anastomosis, transfistula anorectoplasty approach, anterior sagittal anorectoplasty technique, and posterior sagittal technique(Festen, Severijnen, van der Staak, & Rien, 1996).

Our patient underwent successful takedown of RVF and pull-through of the rectum with transfistula anorectoplasty approach (TFARP). The advantages of TFARP are an intact perineal skin bridge and reducing the possibility of postoperative complications. Furthermore, the levator muscle is recognized but not split. The TFARP is accepted by many surgeons because of the surgical outcome and good aesthetic appearance as leaves no visible scar with suitable strength of neoanus sphincter and accepted continence. In contrast, in the corrective surgery of RVF, anterior sagittal anorectoplasty is accompanied by substantial postoperative complications and a negative cosmetic effect on the anal region.(Patankar, Vidyadhar, Prabhakaran, Bo, & Lsk Loh, 2004)

Many surgeons performed colostomy due to the worry of the healing process and loss of anal continence. However, the accompanying complications of colostomy, such as peristomal scratch, prolapse, hernia in parastomal area, fecal leakage, intra-abdominal adhesions, and obstruction with a range of 28%-74% in previous reports. Recently, colostomy has been only recommended for high-risk patients with other medical or surgical problems or for patients with variable anatomic features who need future surgeries.(Priyatini & Roziana, 2020) Khan et al. carried out the posterior sagittal technique without any colostomy diversion in a young female patient due to the normal development of sphincter muscles.(Khan & Chana, 2013) Priyatini et al. reported atresia ani with RVF in two young female cases which were repaired by TFARP technique without postoperative complications.(Priyatini & Roziana, 2020)

Conclusions

Rectal atresia with RVF is uncommon and necessitates extensive evaluation to rule out any related abnormalities. Furthermore, transfistula anorectoplasty may be performed efficiently without a need for covering colostomy with a proper esthetic appearance, anal continence, and minimal postoperative complications.

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