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Anorectal Malformation Presented in Adolescence – Case Report

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Abstract: Case Report

Introduction: Anorectal malformations are one of the common pediatric surgical problems and it is usually treated by pediatric surgeons. Very occasionally, patients with anorectal malformations present to general surgeon in their adolescence. Here, we report a case of young lady with Anorectal malformations which is successfully treated. ***Case report:*** Our reported patient has anorectal malformation and searching surgical repair before her marriage. We also describe about the detail surgical technique. ***Discussion:*** Nowadays, it can be assumed that most of the patients with anorectal malformations are treated in their early infant. But, a few patients miss early reconstruction and presents lately in their adolescence. There are various surgical approaches for adult patients. The reported procedure, anal transposition or trans-sphincter anorectoplasty, can be done in lithotomy position under general or regional anesthesia and it does not need to divide muscle complex. ***Conclusion:*** This case report highlights the satisfactory outcomes of or trans-sphincter anorectoplasty in adult and it is also reminding the existence of adult patients with anorectal malformations in our community.

Keywords: Anorectal Malformation, Trans-Sphincter Anorectoplasty, Adult Patient.

INTRODUCTION

Anorectal malformations (ARM) are one of the common pediatric surgical problems and it is usually treated by pediatric surgeons. Very occasionally, patients with ARM present to general surgeon in their adolescence. Some authors report that delay presentation of ARM may be due to poverty, lack of health education, and negligence (Chakravartty, S *et al*., 2009). A few cases of ARM presented in adolescence are also encountered in Myanmar. The posterior sagittal ano-rectoplasty (PSARP) is a recommended procedure for ARM in pediatric patients. To our knowledge, guidelines and recommendations regarding procedure of choice for repair of ARM in adolescence and adult patients are not too much. Some surgeons recommend trans-sphincter ano-rectoplasty (TSARP) in adult patients with ARM (Kamal, J. S. 2012). Actually, successful management of an adult patient with ARM is a great challenge for general surgeons and here we report a case of young lady with ARM which is successfully treated by TSARP.

Case Report

An 18 - year - old young lady presented to our hospital with an intention for the closure of transverse loop colostomy which has been done in her age of 11 year at a tertiary pediatric hospital for fecal diversion purpose. She was born by traditional birth attendant at a remote area and her parents missed to undergo tertiary pediatric hospital

for repair of her congenital anorectal malformation. Fortunately, her bowel had opened since birth with fecal passage from vaginal opening. But there were significant social issues when she became a teenager. Therefore, she was sent to tertiary pediatric hospital and underwent diverting colostomy as a stage procedure. Again, she missed her follow up and definitive surgery. She encountered social issue when she was a young adult lady, at the time of her marriage. So, she sought surgical consultation urgently and presented to our hospital. We performed complete work up to define the extent of anorectal malformation. Her perineal examination revealed no separate anal opening, and, dry fecal mass was palpable through an opening in vestibule and another small opening was noted on the right side of the vulva as shown in Figure (1).

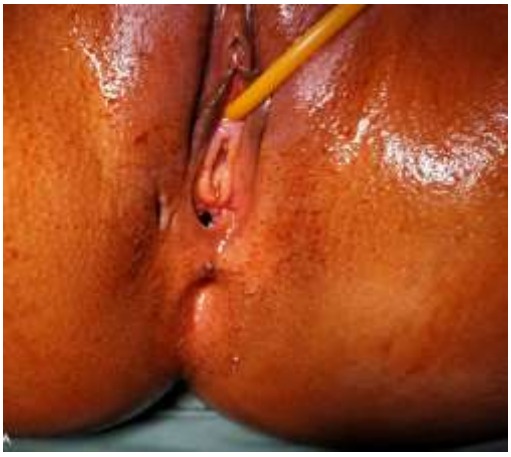

Figure 1: Ano-vestibular fistula with anal dimple and well-formed gluteal cleft

Transverse diameter of the fistula opening was very large and passed examined index finger loosely. A distal loop colonography was done and it failed to show rectum and fistula tract because of the obscure of filling defect resulting from huge fecal impaction. Therefore, fistulogram was carried out and which showed low rectovaginal fistula. Her abdominal ultrasound scan examination was unremarkable. She had a fully developed sacrum, a normal gluteal cleft, and no other congenital abnormalities. Finally, she underwent trans-sphincter ano-rectoplasty.

Under spinal anesthesia, patient was put in lithotomy position, thorough examination was done, and previous clinical findings were confirmed. After that, impacted huge fecal mass was crushed and evacuated. Then, vaginal tract and operating site were cleaned with iodine solution. Traction sutures were applied to expose the operative area. The circumferential incision was made around fistulous opening and posterior wall of vagina was separated from the anterior wall of rectum. Then, anorectum was circumferentially mobilized up to the level of levator ani muscle (Figure-2).

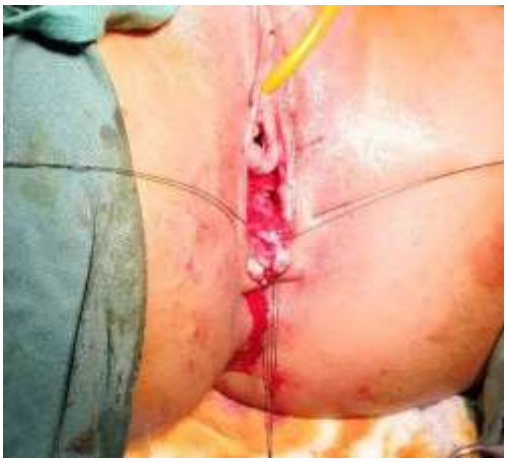

Figure 2: Mobilization of anorectum up to levator ani muscle

The anal sphincter complex was identified in the perineum and an opening was made within it by making a skin incision. At that point, special attention was made to minimize the injury to sphincter complex. Via the created neo-anal opening, a tunnel was created through anal sphincter complex and which was slightly stretched with surgeon's finger up to two fingers width (Figure-3).

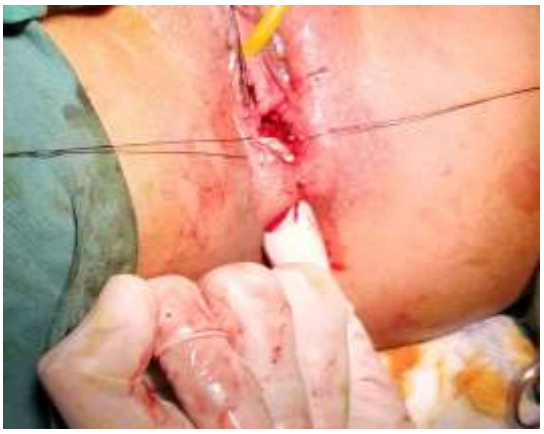

Figure 3: Creating neo anus and tunneling for anal transposition

The mobilized anorectum was pulled down through the tunnel, assessed the tightness of anal canal and mucosal end was fixed at the new anal opening by using absorbable suture (3/0 Polyglactin 910). Finally, vaginal wall was repaired by layer with similar absorbable suture (Figure 4).

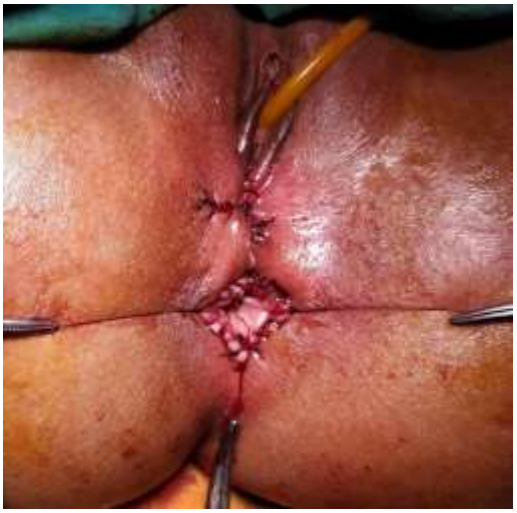

Figure 4: Suturing of neo anus and vaginal repair

The wounds were healed well uneventfully and she was discharged from hospital with an instruction for self- anal dilatation starting two weeks after the operation by using Hegar's dilators. She was also prescribed stool softener and educated to take high fiber diet. Three months later, distal loop colonography was done to identify the patent passage of her bowel. The perineal examination was carried out. Her neo anus was healed well and anal tone was normal. There was no fistula opening in her vestibule and vaginal tract. But there was a palpable large fecal impaction in her lower rectum. So, it was crushed and evacuated before the closure of her temporary colostomy. Finally, her transverse loop colostomy was closed. Postoperatively, she had proper defecation and there were no soiling, incontinence and constipation. She was able to differentiate between feces and flatus very well. On digital rectal examination, she had strong effective squeeze and there was no anal stenosis. The anal continence was assessed by Kelly's score and her overall score was six. The patient and her parent were satisfied with scars of her perianal wounds. On her discharge from hospital, she was educated for dietary modifications.

DISCUSSION

The ARMs are frequently encountered congenital anomalies in neonatal pediatric surgery, but it is undoubtedly rare in daily practice of adult care provider likes general surgeon. Historically, surgeons have attempted to treat infants with ARM for centuries. Since A.D.600, the treatment of ARM has occurred as Paul of Aegina first advocated the blind plunging of a knife into the perineum (Rosenblatt, M. S., & Gustavson, R. G. 1958). Up to 1953, ARM was empirically treated with the rectum pulled down to the perineum using a perineal approach for "low" defects or a combination of abdominoperineal approaches for "high" defects. At that time, the outcomes of bowel control of patients were very poor (CL, S. 1997). During the 1980s, the PSARP was implemented worldwide and encountered better patient outcomes and it became a recommended treatment of ARM (Peña, A., & Devries, P. A. 1982).

At present, accepted surgical options for ARM in pediatrics are neonatal perineoplasty, pull-through operation during infancy following colostomy, PSARP and laparoscopy-assisted surgery (Iwai, N., & Fumino, S. 2013). But, because of extreme rarity of incidence in adult patients, there is no recommended treatment and possibility of PSARP is not certain yet (Chakravartty S, *et al*., 2009). There are various surgical approaches like PSARP, anterior sagittal anorectoplasty (ASARP) and anal transposition or TSARP for adult patients (Kamal, J. S. 2012). Furthermore, some also advocated laparoscopically assisted anorectal pull through for adult patients (Miglani, R. K, *et al*., 2012). However, the goal in treatment of ARM in adulthood should be the normal integrity of sphincter complex.

Nowadays, it can be assumed that most of the patients with ARM are treated in their

early infant. Unfortunately, a few patients miss early reconstruction and presents lately in their adolescence. The possible reasons for persistence of this pediatric problem until adulthood may be lack of health education, improper neonatal care, inappropriate medical facilities, poverty and lack of social support (Bokhari, I, *et al*., 2010). In our patient, home delivery by traditional birth attendance at rural area may be the leading cause of delay diagnosis for her AVF. Furthermore, lack of social support and financial limitations cause missing for reconstruction at tertiary hospital.

As in our report, anovestibular fistula (ARM) or rectovaginal fistula (RVF) is the most common form of anorectal anomaly in female infants and in which the fistula opening is located near the vagina at the posterior fourchette (Javid, P. J, *et al*., 1998; Peña, A., & Levitt, M. A. 2006). To our knowledge, there is very limited evidence of adulthood presentation of ARM. The largest case series reported in one retrospective cohort study included 16 female patients who had unrecognized or inadequately treated RVF and one patient who had rectoperineal fistula. The mean age of patients in this study group was 25 years, ranging from 15 to 55 years of age (de Blaauw, I, *et al*., 2013). The patient in our report recognized RVF only in her age of 11 years and her main presenting complaint was constipation due to fecal impaction. She underwent diverting colostomy for emergency measure and was planned for pull through operation. But she missed again for adequate repair up to 18 years of age and the main reason for coming to our hospital was due to the decision of marriage as in Terzi's case report.

As our report, Terzi *et al*., reported two cases of RVF, in which a 19 years old lady presented with main complaints of constipation and passage of feces via vagina sought for treatment due to the decision of marriage. In his report, another 18-year-old lady had frequent attacks of urinary tract infection which was not encountered in our case (Terzi, A, *et al*., 2008). Most of the reported patients with ARM presented in adulthood were females but one reported a young male patient with ARM and rectourethral fistula which needed meticulous urethral repair (Chakravartty, S, *et al*., 2009). The earliest case review regarding adult patient with ARM was found in 1978 which included 29 adult patients. Among these patients, 19 were treated surgically (Katz, L. D, *et al*., 1978). Furthermore, Hamdy *et al*., reported one adult patient with rectovaginal fistula treated by PSARP in 1993 (Hamdy, H, *et al*., 1994). Another case report described treatment with anal transposition procedure for late presentation of ARM which encountered complication of anal stenosis postoperatively and needed anoplasty (Bokhari, I, *et al*., 2010). This complication was not encountered in our reported case. Similarly, Chavan *et al*., reported a successful anal transposition procedure or TSARP for ARM in an adult female patient (Chavan R. N, *et al*., 2015).

There are isolated and series of case reports about successful repair of ARM in adult with PSARP but this technique has to divide levator ani muscle and muscle complex which play important role in continence. Furthermore, PSARP procedure has to be done in prone position with general anesthesia which may definitely have some challenges to theatre team, anesthetists and patient, him or herself. These limitations can be avoided in TSARP, which is the selected procedure of our case report. The TSARP procedure can be done in lithotomy position under general or regional anesthesia and it does not need to divide muscle complex. Additional benefit of TSARP encountered in our report is feasibility to evacuate huge fecal impaction via the fistula opening which can obscure the dissection from posterior approach.

CONCLUSION

This case report highlights the satisfactory outcomes of TSARP in adult and it is also reminding the existence of adult patients with ARM in community. Furthermore, this report points up the importance of individualized patient care and

common sense in making choice of surgical method. The adult care surgeons should be aware of successful management in adult with ARM for the benefits of these forgotten patients.

CONSENT

Written informed consent was obtained from patient for publication of this case report and corresponding photographs.

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