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Female ARM Left to Loiter: Attempt for Delayed Primary PSARP with Meticulous Outcome

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Abstract: ***Delayed presentation of anorectal malformation (ARM) at adolescence is rare but poses a challenge to the surgeon in terms of poor long term outcome. To lessen the stress further, early diagnosis and definitive surgery, thereafter, becomes necessary. We present a case of female anovestibular fistula seeking advice at 13 years of age and treated with primary posterior sagittal anorectoplasty (PSARP).***

Keywords: Adolescent; Anorectal malformation; Constipation; Fecal incontinence; Posterior sagittal anorectoplasty

1. Introduction

Anorectal malformations include a diversity of congenital conditions involving rectum and anal canal. Other common associated anomalies include cardiac, renal, vertebral and spinal. The reported incidence is 1 in 5000 live births with a slighter male preponderance [1]. Although commonly diagnosed in infancy, few cases may be reported at a much later age due to poor socio- economic condition and fear of social isolation [2]. Presentations among female patients are usually sub acute but hospital delivery and proper examination of the newborn helps in early detection and counseling for early treatment. Anorectal malformations detected in older age group have an unfortunate female inclination. The posterior sagittal anorectoplasty is considered ideal for treatment of anorectal malformations [3].With sub acute presentation in females, it can be performed as early as 1 – 2 months of age. For older age group, staged repair is generally recommended [4]. Following is a presentation of ARM in a 13 year old female child treated with a successful attempted primary PSARP.

2. Case Report

A 13 year old female patient was brought to us with complaints of abnormal position of anus inside the vestibule and constipation since birth. Due to social inhibitions and poor economic status, they never consulted a clinician before. Now that the child reached the age of puberty, her post marital concerns led her mother to consult our OPD. Detailed clinical examination was done in the presence of a gynecologist. General condition of the patient appeared fair. There were no thoracic, abdominal or cardiac anomalies. On local examination of perineum, the anal opening was placed inside the vestibule and stenotic [Figure 1]. The vaginal and urethral openings were normally positioned. The labia and clitoris were looking normal. Diagnosis of ano-vestibular fistula was made and investigated further. Ultrasonography detected normal mullerian structures and kidneys. Echocardiography and X- ray pelvis also revealed normal study. Surgery was thence planned and different surgical options like 3- stage anorectoplasty and primary posterior sagittal anorectoplasty were discussed with the parents. Due to financial constraints, they opted for primary PSARP. Pre-operatively bowel was cleansed by total gut irrigation. Surgery was done under general anesthesia in prone jack knife position with adequate cotton padding underneath the groin area. After induction and positioning of the patient, per urethral catheterization was done. Posterior sagittal incision grouped with circumferential incision around fistulous opening was given to reach and mobilize the ano-rectum. Position of neo anus was determined with the help of a muscle stimulator. After delineating muscle complex and mobilizing ano-rectum, adequate neo anus created and perineal body repaired. There was no iatrogenic vaginal injury. Post surgery, patient was kept nil per oral for 5 days. Post operative period was uneventful and patient was discharged in fair condition on day 7. Child's mother seemed happy with the aesthetic appearance [Figure 2] and also the relief in symptoms.

3. Discussion

In view of sub acute presentation, poor socio economic condition and lack of access to advanced medical care, many cases of female ARM present at a later age group [5]. Fecal incontinence is reported in almost 30- 50 % of the adolescent patients after surgery [6]. The long term outcome relates with ARM sub types, age at presentation and associated anomalies [7]. To prevent delayed presentation and improved outcome by early surgery, it is necessary to create awareness about the entity among poor and illiterate population with the help of health care professionals. Regardless of the age of presentation, immediate surgical intervention is mostly advisable [8].

4. Conclusion

Anorectal malformation is a surgically treatable entity. Early restorative surgery leads to good outcome. Time being the

essence, awareness about the condition, hospital delivery and detailed neonatal examination is advisable. In developing countries, where delayed presentation of the condition is a frequent encounter, early surgery is always commendable in terms of social acceptance and a sound psyche.

5. Conflict of interest

None

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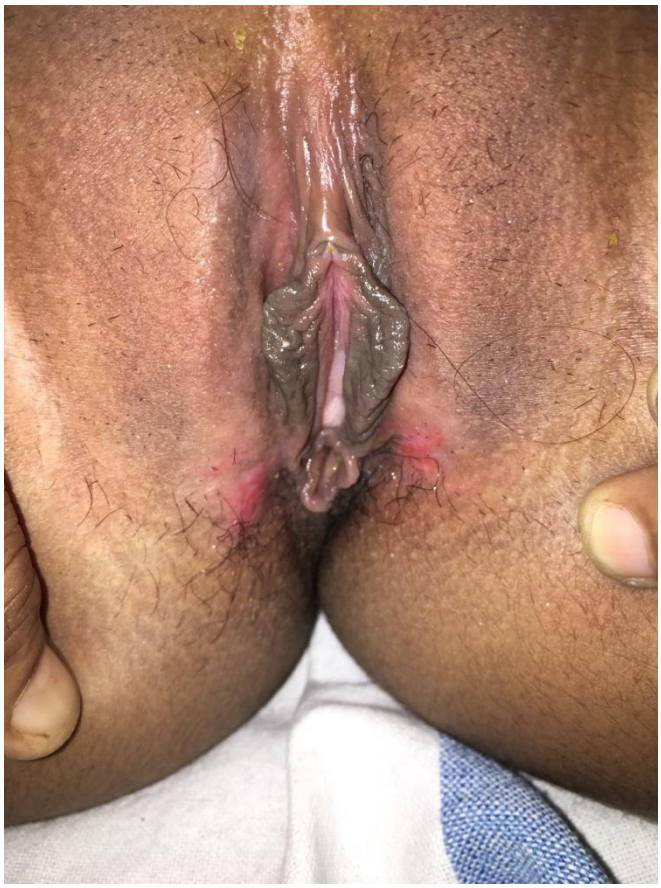

Figure 1: Anorectal malformation in a 13 year old female

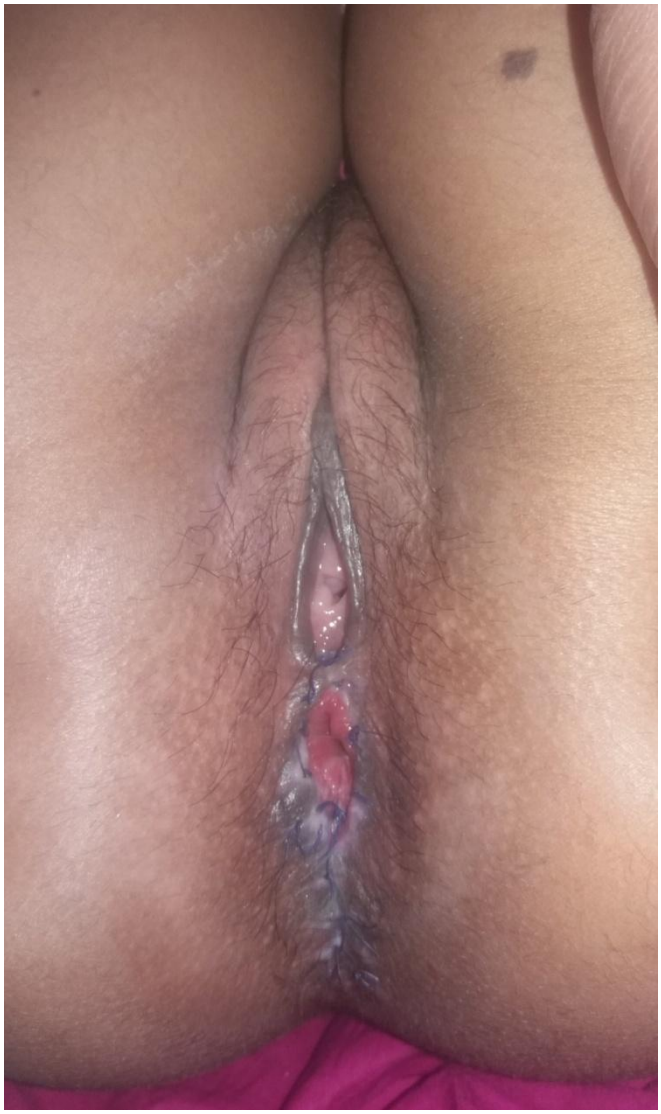

Figure 2: post operative aesthetic appearance