CASE REPORT

A Functional Ectopic Vaginal Anus: A Rare Clinical Entity

Mukhtar Mehboob[1], Samina Naz[2], Shahina Tabassum[3] and Jamil Ahmed Khan[4]

ABSTRACT

A case of functional ectopic vaginal anus is presented in a 20 years old girl. Patient complained of passage of stool through her vagina. She was continent and had no complaint of constipation. Her examination revealed imperforated anus with functional ectopic vaginal anus. Her surgery was performed in two stages. In first stage, anoplasty was performed at midpoint between the vulval and anal opening. The posterior vaginal wall was repaired in two layers and protective loop colostomy was made. In the second stage, after a period of 3 months loop colostomy was closed. Patient's recovery was smooth, she is not constipated and continent with Wexner score of 3. She was advised pelvic floor rehabilitation exercises. She improved within a month with Wexner score of zero.

Key words: *Vaginal anus. Imperforated anus. Anoplasty.*

INTRODUCTION

Various kinds of anorectal malformation (ARM) have been reported in the literature. These include isolated imperforated anus (low and high level) and imperforated anus associated with rectourethral, rectovaginal, recto-perineal and rectovestibular fistulas. Other associations include pseudotail, anal agenesis, anal atresia with Klein Waardenburg syndrome, anocutaneous fistula with Bardet-Biedl syndrome, rectal duplication, perineal ectopic sinus, anterior ectopic anus and vaginal anus.[1,2]

The term vaginal anus is described as congenital ARM with imperforated anus and intestinal opening in the vulva with normal continence. This ectopic anorectum is situated below the puborectalis muscle, while the opening of rectovaginal fistula is situated above the puborectalis muscle. Ectopic anus was functionally assessed by electromanometer and electromyographic techniques. These methods safely and adequately distinguish a functional ectopic anus from a fistula.[3]

Functional vaginal anus presentation in a young girl is a rare anomaly. The delay in seeking treatment in our setup is due to lack of awareness, illiteracy, social constraint, strong religious belief and residence in a far remote area. It is interesting to note this clinical variant in a teenage group and the case report highlights the treatment.

[1] *Department of Surgery, Shaheed Benazir Bhutto General Hospital, Quetta.*
[2] *Department of Gynaecology, Murshid Hospital and Health Care Centre, Karachi.*
[3] *Department of Radiology / Surgery[4], Sandeman Provincial Teaching Civil Hospital, Quetta.*

Correspondence: Dr. Mukhtar Mehboob, House No. 5-11-84 (777), Gulshan-e-Patel, Quary Road, Quetta.
E-mail: mehboob87300@yahoo.com

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CASE REPORT

A girl of 20 years old came from a far remote area of a neighbouring country. She was complaining of abnormal passage of stool from the vagina. She stated that she had no anal opening since birth. General physical examination revealed a healthy, well-oriented young girl weighing 55 kg. Systemic abdominal examinations was normal. Perineal examination revealed absence of anal opening. There was only a dark dimple on the skin midway between the vaginal and anal opening. The posterior vaginal wall had anal opening.

Her haematological investigations revealed that she had Hb of 13.2 gm/dl, TLC 12500/cu mm, blood glucose of 85 mg/dl and urea of 40 mg/dl; HBsAg and HCV were non-reactive. Urinary analysis revealed 1-2 RBCs, 2-4 WBCs and 2-4 epithelial cells HPF. Her pelvic sonographic examination was without any finding. Barium enema was performed to exclude any hind gut abnormality. The findings of barium enema revealed nothing abnormal.

The diagnosis of the patient was discussed with the patient. Patient was informed about the procedure. The colon was prepared by allowing a low residue diet and mechanical cleaning. Patient's surgery was planned in different stages.

First of all the perineum was pricked with a pin, the dark area of skin puckered, indicating the presence of underlying sphincter muscle. The future site of anal opening was marked midway between the vulval opening and tip of the coccyx. A circular incision was given around the anal opening. The lower anal canal was dissected. It was pulled to the selected site of anus and stitched there. The vulval gap was closed in two layers. Protective loop colostomy was made.

After 3 months, anus and vagina were examined. The wound was healed. The anal opening was snug and had a good sphincteric reflex. After preliminary

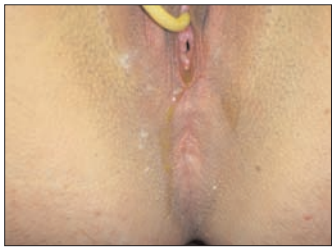

Figure 1: Absence of anus at normal site

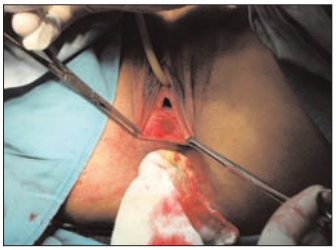

Figure 2: Ectopic anus in posterior vaginal wall.

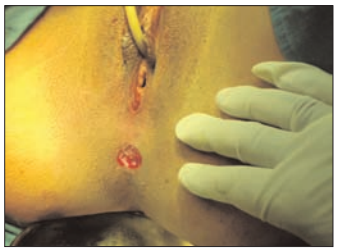

Figure 3: Proposed site of future anus

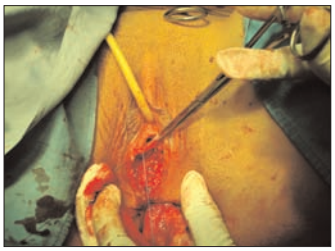

Figure 4: Repair of posterior vaginal wall.

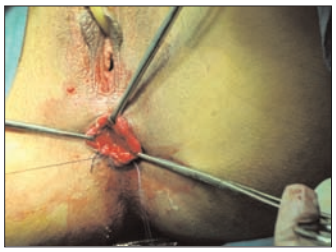

Figure 5: Formation of anus.

preparation of the colon, loop colostomy was closed.

Postoperative recovery of the patient was smooth. Patient was continent without constipation. Later on, patient realized that on some occasions she has no control over flatus. Patient was assessed on the Wexner score. Her score was 3 (3/20). She was offered pelvic exercises. After one month she was assessed again and her score was zero.

DISCUSSION

The prevalence of ARM according to German diagnosis related groups system was about 3.6% (95% CI: 3.4-3.8) per 10,000 or 1:2784 for all different types of anal atresia requiring surgery.[4] Leape and Ramenofsky in 1978 stated that the usual anal position is midway between the vaginal fourchette in female or base of scrotum in male and coccyx. In 1984 Reisner *et al*. presented a simple method, the anal position index (API) to define the normal position of anus in the neonates. When anus was placed more anteriorly, it is termed as anterior ectopic anus.[5] The incidence of anterior displacement of anus (ADA) or anterior ectopic anus and its relation to constipation in children was determined by anal position index (API). It was 43.4% in females and 24.6% in males ($p < 0.01$).[6] Ruiz Moreno *et al*. reported two cases of vaginal anus in teenage group. He described the technique to treat the vaginal anus. In our study, we reported a case of vaginal anus in a 20 years old girl.[7]

The diagnosis of ARM in infants can be made by inverted lateral radiography, distal colostography (loopography), transperitoneal ultrasonography, magnetic resonance imaging, contrast computed tomography and infracoccygeal ultrasonography.[8] However, in this case, the transperitoneal ultrasonography and barium enema revealed no abnormality.

Moreno *et al*. in his two cases of vaginal anus made a new canal. The wound healed promptly with little postoperative pain and reduced risk of postoperative anal stenosis.[7] In this patient, the surgery was planned in two phases. In phase one, the anal opening in the posterior vaginal wall was mobilized and pulled down to midway between the vaginal opening and coccyx, Anoplasty was done. Posterior vaginal wall repaired. Protective loop colostomy was formed. In second phase after a period of 3 months colostomy was closed. Patient's recovery was smooth with good functional outcome.

There are various scoring systems for the severity of fecal incontinence like St. Mark's, Wexner, Pescatori and Rockwood. Patient having a certain score is also scored for their quality of life like 36 items short form health survey (SF-36), Faecal incontinence quality of life scale (FIQLS), Gastro-intestinal quality of life index (GIQLI) and Rockwood's specific fecal incontinence quality of life. In this patient, she was assessed by the Wexner score due to its simplicity. She had a score of 3. She was offered pelvic floor rehabilitation muscle coordinating training. She became perfectly alright.

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