Vestibular anus presenting as a case of primary infertility: An unusual adult presentation

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ABSTRACT

Imperforated anus and ectopic anal orifices are almost always detected in infancy or early childhood. The oldest reported case of imperforated anus in modern literature was in a 7 year old girl. We report a case of vestibular anus in a 23 year old Saudi lady who presented with primary infertility. She was discovered to have an imperforated anus and was managed surgically.

Keywords: Ectopic anal orifice, vestibular anus, infertility.


Imperforated anus and ectopic anal orifices are almost always detected and treated in infancy or early childhood. There is little in the medical literature about this problem in post adolescent patients and when a rare reference is found, it usually describes a patient who underwent less than satisfactory treatment as a child. Most of the cases of imperforated ani and ectopic orifices who presented for the first time as adults were reported over 2 decades ago. In the modern literature, the oldest case of imperforated anus was in a 7 year old girl in Jipijapa, Ecuador. The rare condition reflects the problems associated with rural health care resulting from super-imposed cultural mores which preclude early diagnosis of a significant anomaly. We report this rare case of vestibular anus discovered in a 23 year old Saudi lady, when she presented as a case of primary infertility in our unit.

Case Report. A 23 year old Saudi lady, married for 2 years, presented to the Gynecology Clinic at King Khalid University Hospital, (KKUH), Riyadh, as a case of Primary infertility. She was born at home in a small village in the Southern region of Saudi Arabia and her delivery was assisted by the traditional midwife. She had no significant past medical or surgical history and was married about 2 years ago. There were no marital problems, however, physical examination showed a healthy looking female with no obvious abnormality. At pelvic examination, the vagina was found to be full of stool, which was coming through a huge posterior vaginal wall opening. Upon further examination of the perineum, there was no anus found, and only a small skin dimple was observed at that site (Figure 1). The rest of her examination was of a normal woman.

On further enquiries, the patient was unaware of any unusual problems related to her bowel habits. Similarly, as far as one was aware she had a satisfactory sexual life, inspite of the fact that the vestibular anal orifice was being used for sexual intercourse (Figure 1) and she claimed to have good control of her bowel functions.

Investigations carried out included Barium enema which was performed by inserting a rectal catheter in the ectopic vestibular orifice in the vagina. This showed a 5 cm tract posteriorly between the dye and the anal dimple (Figure 2). The rectum and sigmoid colon were consequently distended due to chronic

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constipation. The findings on Intravenous Pyelogram (IVP) and pelvic ultrasound were normal. Hysterosalpingography (HSG) was performed and this showed patent tubes with normal shape of the uterus. X-ray of sacrum and spine were normal.

The patient was admitted into the hospital for 48 hour bowel preparation before surgery, by bisacodyl, magnesium sulphate, neomycin sulphate, and cleansing enema. Right transverse loop colostomy and posterior sagittal anorectoplasty was carried out as a one-step procedure. After mapping the external sphincter by a nerve, stimulator, an incision was made around the vestibular anus. An extension of the incision was carried out posteriorly in the midline to the tip of coccyx. The vestibular anus was carefully dissected, especially anteriorly where there was a common wall-sharing between vagina and rectum. The rectum was mobilized to the center of the sphincter. The anterior perineal part was repaired, followed by the posterior part, thus fixing the rectum into the posterior rent of levator ani. She recovered satisfactorily after her first surgery. She was re-admitted 3 months later for examination under anesthesia (EUA) and closure of the transverse colostomy. The newly created anus was in good position and there was a good size bridge between the anus and vagina (Figure 3). Anal dilatation was carried out to size no. 17 dilator. The patient was followed up for one year post-operatively, and she was able to have a good control of her bowel habits.

**Discussion.** The incidence of congenital malformations differs, not only from country to country, but even between the various geographical areas of the same country. Ano-rectal malformations were reported to be the most common congenital anomalies of the gastrointestinal tract (GIT) in the new born, with an incidence of 1:2000 - 5000 live births. These differences are due to differences in reporting, rather than in actual incidence, as sometimes, the diagnosis may be missed at birth, especially the low type, as compared with high type anomalies.

In Saudi Arabia, the incidence of anorectal malformations were reported to be 1:1818 in Al-Khobar, and 1:2747 in Holy Makkah and 1:1200 in Qatif. These differences reflect the wide geographical areas that Saudi Arabia cover, and probably in some rural areas, where home deliveries are still carried out, such malformations are often overlooked at birth.

Our patient did not have any associated anomalies, although it is known that ano-rectal malformations could be associated with conditions such as congenital heart disease, mongolism, urological, skeletal, reproductive organ and GIT anomalies.
suspicion of any abnormality was detected by her mother inspite of this not having been her only daughter, and after marriage, she had a satisfactory sexual life, inspite of her condition, until she presented as a case of primary infertility. It was during the routine examination of the pelvis and perianal region that her condition was first discovered.

The recent advances in the management of anorectal malformation have yielded good results, inspite of the combination of transverse colostomy and post agittal anorectoplasty in one stage operations. Since the description of the posterior sagittal anorectoplasty, this procedure has become the standard technique for operative correction of high anorectal malformations. The one stage operation was undertaken here because there was no infection in the perineum as this was a congenital problem. In addition, this has helped to shorten her hospital stay. This procedure has also been successfully reported in infants, as well as a primary repair for adult patients with anorectal fistulas. Although this has produced good results, chronic constipation, which may need enema for relief, may be a long term problem.

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References