CASE REPORT

Female anorectal malformation in a woman

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Accepted 9 July 2015

SUMMARY

Delayed presentation of patients with anorectal malformation is not uncommon, especially in developing countries. However, presentation beyond teenage years is not commonplace. We describe a case of a 28-year-old woman who presented for treatment due to marital issues.

BACKGROUND

Anorectal malformations (ARMs) are one of the most common congenital anomalies faced by paediatric surgeons, with reported incidence of approximately 1 in 5000 live births.¹ Vestibular fistula represents the most common anorectal defect seen in girls with ARM. Adult presentation of this malformation is rare.² In a developing country such as India, illiteracy and poverty can lead to delayed medical advice, particularly in female patients who, besides having an abnormal anal opening, may remain asymptomatic.³

We treated one such patient. Being an uncommon entity, it is being presented with a brief review of the relevant literature.

CASE PRESENTATION

A 28-year-old woman presented in the outpatient clinic with an abnormal anal opening since birth. She did not have any specific problem. After marriage, her husband had noticed the perineal abnormality, which disturbed him greatly; the patient subsequently sought medical advice. There was no incontinence for flatus or faeces. On examination, there was an anterior ectopic anus. The sacrum was normal. There was no associated problem.

INVESTIGATIONS

Investigations included a haemogram, serum electrolytes, kidney function test and chest X-ray, all of which were normal.

TREATMENT

After administering anaesthesia, a diverting sigmoid loop colostomy was made, as the first stage of surgery. After about 2 months, anterior sagittal anorectoplasty (ASARP) was performed (figures 1–3). The patient was placed in lithotomy position. The anus was mobilised from anterior and lateral sides. Since the tissues were matured, dissection was easier, but more force was needed for mobilisation. Next, dissection proceeded between the rectum and vagina. Again, as the patient was an adult, the tissues were robust, compared with those of young children; hence the dissection between the rectum and vagina was clearer. Also, as compared to young children, there was less chances of

injury to the vagina or the rectum, but more force was needed for dissection. The rectum was mobilised up to the posterior fornix anteriorly. Overall, the rectum was mobilised all around to such an extent that anoplasty could be performed without any tension. After ASARP, the postoperative period was uneventful.

Anal dilation was performed as per the protocol given by Levitt and Pena.⁴ This includes anal dilations 2 weeks after the surgery. A dilator that fits snugly into the anus is passed through twice. Every week, the size of the dilator is increased, until the rectum reaches the desired size, which depends on the patient's age. In our patient, it was Hegar dilator size 18. Once this desired size is reached, the colostomy may be closed. The frequency of dilation may be reduced once the dilator enters easily with no resistance according to the following protocol: once a day for 1 month, every third day for 1 month, twice a week for 1 month, once a week for 1 month and once a month for 3 months.

After about 3 months of ASARP, colostomy closure was performed. The postoperative period was uneventful. The patient was able to pass stools normally. Continence assessment by Kelly score was 5.

OUTCOME AND FOLLOW-UP

After the colostomy closure, the patient was in follow-up for 1 year. There have been no subsequent problems.

DISCUSSION

ARMs are correctable congenital malformations. They can achieve reasonably good prognosis. Most of these anomalies can be easily detected on a clinical examination in newborns. This report highlights some important issues. Delayed presentation is common in developing countries; however, delaying the treatment up to the age of 28 years is uncommon. Delayed diagnosis may be attributed to lack of knowledge, awareness, poverty and poor

Figure 1 Picture showing anterior ectopic anus. The normal anus is absent.

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To cite: Pandey A, Gupta V, Singh SP, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-211456

Figure 2 Intraoperative photo showing mobilised rectum.

care offered to girl children in some parts of this country.^{5 6} Besides, sometimes, wrong advice regarding the correct age of treatment also leads to delay in treatment. In our case, the problem of a failing marriage compelled the patient to seek medical advice. The scenario is different in developed countries where constipation and an abnormally positioned anal opening are detected by parents much earlier.⁷

Delayed presentation may affect the surgical management, and contribute to functional and psychological problems for adult patients.¹ Surgically, tissue planes are better delineated and dissected in adults, with less risk of tissue damage as compared to that in children. In this operation, we had to arrange instruments from an adult surgical set and had to use thicker sutures for proper outcome. As the patient presented late, we chose not

Figure 3 Postoperative picture after the anterior sagittal anorectoplasty procedure.

to perform a primary ASARP. The rectum may be dilated in these patients, and there may be a problem of placement of the rectum into the muscle complex. Besides, there may be a chance of sutural dehiscence. Hence, a three-stage operation was undertaken, without any complication. As compared to children, this patient, being a woman in her late 20s, needed more privacy for adequate examination and advice during follow-up.

This type of presentation has not been mentioned in a patient beyond 25 years of age. There have been reports of late teenage presentations in the literature.^{3 7} There are reports of patients with diagnosis of vestibular fistula at the age of 23 years.^{2 8}

Learning points

- ▶ Delayed presentation of anorectal malformation, especially in females, is common in developing countries.
- ▶ Presentation beyond the teenage years is less common.
- ▶ Surgical correction at any age of presentation is possible, with good results.

Contributors AP and RV operated on the patient. AP, SPS and VG reviewed the literature. All the authors read and approved the final draft.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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