International Journal of Science and Research (IJSR)
ISSN: 2319-7064
ResearchGate Impact Factor (2018): 0.28 | SJIF (2019): 7.583

A Rare Case of Vestibular Anus - Presentation during Pregnancy

Dr Kavita Ghanwat, Dr Jyotsna Deshmukh

Abstract: ***This is the rare case of vestibular anus (ARM- Anorectal Malformation) case presenting during pregnancy and labour. So far very few cases of vestibular anus are reported. ARM usually presents during paediatric age group, but occasionally can be presented in adults too. 20 year old primigravida presented in latent labour to hospital who diagnosed to have vestibular anus with imperforate anal pit. However, there was no fischulous connection between vagina and rectum. Patient was continent for faeces. Delivery was conducted by LSCS (lower segment caesarean section). Conventionally, ARM are treated surgically. But we have opted for conservative management as patient was continent for faeces and new formation of anal opening can have doubtful sphincteric integrity.***

Keywords: Vestibular anus, Anorectal malformation, fecal incontinence, Pregnancy

1. Introduction

ARM are rare birth defects concerning anus and rectum. Approximately 1 in 2, 500 to 1 in 5, 000 new born babies are affected [1-3]. Different degrees of severity are distinguished, ranging from mild anal stenosis over anal atresia with or without fistula to persistent cloaca or even cloacal exstrophy [4]. Vestibular fistula represents the most common anorectal defect seen in girls with ARM. Adult presentation of this malformation is rare[5].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[6]

2. Case Presentation

20 year old Primigravida, married for a year, unbooked, residing at rural part of Maharashtra came to labour room as 39 weeks of pregnancy. She was in latent labour on admission. Per vaginal findings were cervical os 2cm dilated, 50% effaced, membranes absent, station of head at -1. While examining her, something unusual was noticed. Anal opening was seen in vestibule and anal pit was imperforate. There was thin mucosa present between vaginal and anal opening. Anal sphincter was thin. However, no fistulous connection was seen in same. After patient was enquired in detail about her complaints regarding defecation. Surprisingly she was asymptomatic. She was absolutely continent for both liquid and solid stools and flatus too. She was not having any sexual symptoms also. However her mother was known about this anatomic abnormality but she failed to seek medical attention due to some reasons.

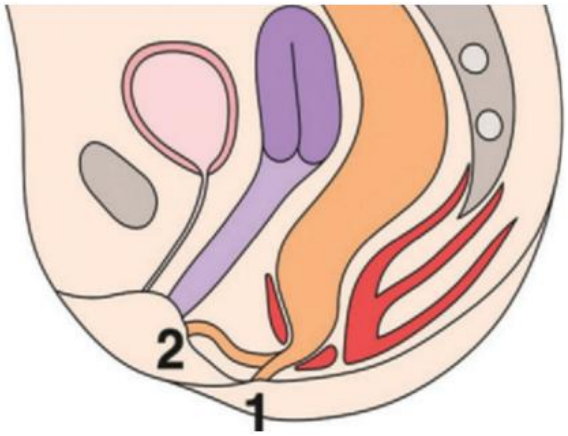
As patient was already in labour, she was taken up for emergency LSCS as to avoid perineal tear. Perineal body was present posterior to anus in this case, as opposed to its normal situation in between vestibule and anus. So patient could not be given trial for vaginal delivery which might haven't been resulted into unforeseen tear into anus and possibly rectovaginal fischula.

As patient was continent for faeces and flatus both, decision for conservative management was taken. Surgical repair and formation of new anal opening may not have given appropriate sphincteric action which was adopted by patient since childhood. [7]

3. Discussion

ARMs are easily diagnosed conditions. Most of the cases are diagnosed in neonatal cases. But late diagnosis is not uncommon in developing countries. In early embryonic life, the terminal portion of the hindgut—the primitive cloaca—is divided into dorsal and ventral parts by a coronal sheet of mesenchyme—the urorectal septum—and separated from the amniotic cavity by the cloacal membrane [9, 10]. Most ARMs result from abnormal development of the urorectal septum.

Possible locations of fistulas in females with ARMs according to the Krickenbeck classification. Low-type ARMs have an external opening in the perineum (1) or vestibular area (2)[11]



Krickenbeck's classification of ARM[12, 13]

Type 1	Anal stenosis, imperforate anus without fistula, anal agenesis with rectoperineal or rectovestibular fistula
Type 2	Anal or anorectal agenesis without fistula, rectal atresia, cloacal malformtions with short (<3 cm) or long (>3 cm) common canal

Present case can be mentioned as anal agenesis with rectovestibularfischula which diagrammatically represented as above. Similar case was reported by Anand Pandey and et al (2015) [8].But as their patient was symptomatic, they offered surgical management. They did diverting colostomy followed by anterior parietal anorectoplasty at later date. Results were good. Longterm follow up showed no sequelae and satisfactory outcome.

Same case has been reported by Rahulkumar Chavan, Bhargav Chikkala et al.

In 2015 of rectovestibularfischula.[14]In contrast to our case, their patient presented in emergency as intestinal obstruction due to collection of hard stools in fischulous tract. Similar to Pandey et al study, they aslodid diverting colostomy followed by formation of new anus at later date.

Thus, anorectal malformations can be presented in adulthood also. These can be successfully treated with PSARP (posterior sagittal anorectoplsty), ASARP (Anterior sagittal anorectoplasty) or TSARP (trans sphincter anorectoplasty). [15] Although surgery offers definitive anatomical correction, it could be associated with postoperative infections, scarring. Formation of new sphincter may not give satisfactory continence for liquid stools. Considering pros and cons of surgery, we offered a choice of treatment to patient and her relatives. As she had already undergone one surgery of lscs and she was completely asymptomatic towards this condition, they opted for conservative management.

4. Follow up

After 6 months, she was again examined and enquired about symptoms. Fortunately she was asymptomatic till date.

References

[1] Forrester MB, Merz RD. Risk of Selected Birth Defects with Prenatal Illicit Drug Use, Hawaii, 1986-2002. J Toxicol Environ Health A. 2007;70:7–18. doi: 10.1080/15287390600748799. [PubMed] [CrossRef] [Google Scholar]
[2] International Clearinghouse Birth Defects Surveillance and Research. http://www.icbdsr.org/
[3] Jenetzky E. Prevalence estimation of anorectal malformations using German diagnosis related groups system. Pediatr Surg Int. 2007;23:1161–5. doi: 10.1007/s00383-007-2023-6. [PubMed] [CrossRef] [Google Scholar]
[4] Cho S, Moore SP, Fangman T. One Hundred Three Consecutive Patients with Anorectal Malformations and Their Associated Anomalies. Arch PediatrAdolesc Med. 2001;155:587–91. [PubMed] [Google Scholar]
[5] Bokhari I, Ali Su, Khan A. Late presentation of a patient with an anorectal malformation (ARM). J Coll Physicians Surg Pak 2010;20:825–7. [PubMed] [Google Scholar]
[6] Chakravartty S, Maity K, Ghosh D et al. Successful management in neglected cases of adult anorectal malformation. Singapore Med J 2009;50:e280. [PubMed] [Google Scholar]
[7] Padubidri VG, Daftary SN. Howkins and Bourne Shaw's Textbook of Gynaecology.16th edition. Elsevier.India;2016.p 135, 136
[8] Pandey A, Gupta V, Verma R. Female Anoractal Malformation in Woman.BMJ case reports.2015 July 22;bcr2015211456
[9] Moore KL, Persaud TVN, eds. The developing human. Philadelphia, Pa: Saunders, 1998.
[10] Nievelstein RA, van der Werff JF, Verbeek FJ, Valk J, Vermeij-Keers C. Normal and abnormal embryonic development of the anorectum in human embryos. Teratology 1998;57(2):70–78.
[11] Alamano L., Meyrat BF., et al. Anorectal Malformations: Finding the pathway out of labyrinth. Radio graphics. Vol 33 no 2.2013;33;491-512.
[12] Holschneider A, Hutson J, Peña A, et al. Preliminary report on the international conference for the development of standards for the treatment of anorectal malformations. J PediatrSurg 2005;40(10): 1521–1526.
[13] Peña A. Anorectal malformations. Semin Pediatr Surg 1995;4(1):35–47.
[14] Chavan R, Chikkala B. Anorectal malformation: paediatric problem presenting in adult. Case Reports in surgery. 2015 Oct 11;vol 2015.http://dx.doi.org/10.1155/2015/625474
[15] J. S. Kamal, “Anal transposition (trans-sphincterican-rectoplasty) for recto-vestibular fistula, ” Saudi Journal forHealth Sciences, vol. 1, no. 2, pp. 89–91, 2012.