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Case Series

Case series: transverse vaginal septum with vaginal atresia

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ABSTRACT

This study includes case series of three cases of transverse vaginal septum with vaginal atresia. Cases presented with either primary amenorrhea or hypomenorrhea or secondary amenorrhea with complaints of cyclical abdominal pain. On examination, all cases had well developed secondary sexual characters, blind vaginal pouch with enlarged uterus felt on per rectal examination. Hormonal profile of all three cases was within normal limit. Karyotyping was done for all three cases, results were 46 XX. Radiological findings were that all cases had hematocolpos and one case had OHVIRA syndrome i.e.; absent right kidney with didelphys uterus with right hemivagina transverse septum. Management of all three cases included resection of transverse vaginal septum with vaginoplasty. Transverse vaginal septum was dissected and was used to create neovagina. Post-operatively, patients were advised to use phantom dilator, for 3-4 times in a day for atleast 3 months, to avoid vaginal stenosis. Cases were followed in OPD every monthly for 3 months, dysmenorrhea was relieved and patients were menstruating regularly with moderate flow for 3-4 days.

Keywords: Amenorrhea, Hypomenorrhea, Hematocolpos, Vaginoplasty

INTRODUCTION

Transverse vaginal septum is a rare developmental disorder of vagina.[1] Transverse vaginal septum occurs due to defect in vertical fusion of Mullerian ducts during embryogenesis. Vaginal atresia occurs due to failure of canalization of sinovaginal bulbs. Transverse vaginal septum has been shown to occur at different depths within the vagina. The incidence of septum in upper one-third is 46%, middle 35%, lower 19%.[2]

Transverse vaginal septum can be either complete (imperforate) or incomplete (perforate).[3] With complete septum, the onset of symptoms usually occurs after puberty. Affected females may present with primary amenorrhea or cyclical pelvic pain or midline suprapubic tender mass because of accumulation of blood in vagina (hematocolpos) or uterus (hematometra).[4] Imperforate transverse vaginal septum may present during either the neonatal period as hydrometrocolpos or at onset of puberty as amenorrhea, dysmenorrhea, and pelvic pain. Perforate transverse vaginal septum presents as hypomenorrhea, dysmenorrhea, dyspareunia, infertility.[5] A pelvic ultrasound would confirm the presence of a uterus and/or ovaries and will reveal fluid collection with internal echoes within endometrial cavity.[4] Magnetic resonance imaging has an indispensable role in distinguishing transverse vaginal septum from the more common imperforate hymen and agenesis or severe hypoplasia of the cervix. MRI is useful before the surgery to determine the thickness and depth of the septum.[4]

We presented three rare cases of transverse vaginal septum with vaginal atresia with different clinical presentation and different MRI findings and management of such cases.

CASE SERIES

Case 1

The patient was a 15 years old girl, presented to clinic with complaints of dysmenorrhea since 4 months and

hypomenorrhea since menarche. On examination, tanner stage 4 for breast and pubic hair development, and vaginal opening not seen and external genitalia normal on local examination. On per rectal examination, 6×6 cm collection felt anteriorly.

Hormonal profile done is within normal limit. USG findings suggestive of right kidney is not present in right renal fossa- agenesis or contracted ectopic kidney, uterus shows hematometrocolpos measuring 4×3 cm. MRI findings suggestive of absent right kidney, complete non-fusion at Mullerian duct, class III Mullerian duct anomaly with obstructed right Mullerian moiety due to transverse vaginal septum Herlyn Werner Wunderlich syndrome (OHVIRA syndrome).

Examination under anesthesia (EUA) left cervix seen, bulge on right side of vaginal septum, 6×6 cm collection felt on per rectal examination. Left cervix visualized. Right vaginal septum resection with hematocolpos drainage was done. A transverse incision was made over the bulge on right side and hematocolpos was drained. Right vaginal septum resection done, margins sutured to vaginal wall with intermittent sutures using vicrly number 2-0. Right cervix felt patent. Patient was discharged in stable condition with advice of daily phantom dilator insertion with lubricant application. Patient was followed up, on follow up visits, patient had no complaints, and had regular menses, with moderate flow for 4-5 days with soaking 3-4 pads per day

Case 2

17 years old girl, presented with complaints of cyclical pain in lower abdomen since 5-6 months and primary amenorrhea. Patient came with USG (a+p) findings s/o-hematometra due to imperforate hymen or transverse vaginal septum. On examination, breast- tanner stage 3, local examination reveals- hymenal tag present, no bulge seen, vaginal opening not seen, external genitalia normal. Per rectal examination- 6×8 cm collection felt anteriorly. Hormonal profile done- within normal limits. USG (A+P)- s/o- hematometra with hematocolpus with distal vaginal atresia. MRI (A+P)- s/o- obstruction at the level of external os or high vaginal level with resultant hematometra. Unicornuate uterus with a possible right rudimentary horn. Findings of examination under anesthesia revealed a blind vaginal pouch of 1 cm. Foley's catheter inserted, myodisection done over the outer vaginal walls of blind vaginal pouch.

A small nick given and blunt dissection done and lower vaginal wall visualized, sims speculum inserted. On per speculum examination, bulging hemorrhage and high vaginal septum seen. A transverse incision taken over the transverse vaginal septum and hematocolpos drained out, flaps of remaining septum sutured along with raw vaginal wall with intermittent circumfrential sutures using vicryl 2-0. Cervix can be felt. Patient was discharged in stable condition with advise of daily phantom dilator insertion with lubricant application. Patient was followed up, patient is menstruating regularly and with moderate flow for 4-5 days.

Case 3

14 years old girl, presented to OPD with complaints of amenorrhea since 8 months. Patient's mother complains of absent vaginal opening. Patient attained menarche 9months back, had spotting and then followed by amenorrhea for 8 months. UPT- negative.

On examination, breast tanner stage 3, local examination reveals- hymenal tag present, vaginal opening not seen, external genitalia normal. Per rectal examination- 6×6 cm collection felt anteriorly. Sr FSH, LH, PRL and TFT-within normal limits. USG (A+P)- s/o- hematocolpus, transverse vaginal septum with atretic vagina. MRI (A+P)- s/o- uterus anteverted, large collection in proximal vagina, cyst located in posterolateral wall, gartner cyst. Examination under anesthesia revealed a blind vagina. Foley's catheter inserted, myodisection done over the outer vaginal walls of blind vaginal pouch. A small nick given and blunt dissection done and lower vaginal wall visualized, sims speculum inserted.

On per speculum examination, bulging hemorrhage and high vaginal septum seen. A transverse incision taken over the transverse vaginal septum and hematocolpos drained out, flaps of remaining septum sutured along with raw vaginal wall with intermittent circumferential sutures using vicryl 2-0. Cervix can be felt.

Patient was discharged in stable condition with advice of daily phantom dilator insertion with lubricant application. Patient was followed up, patient is menstruating regularly and with moderate flow for 4-5 days.

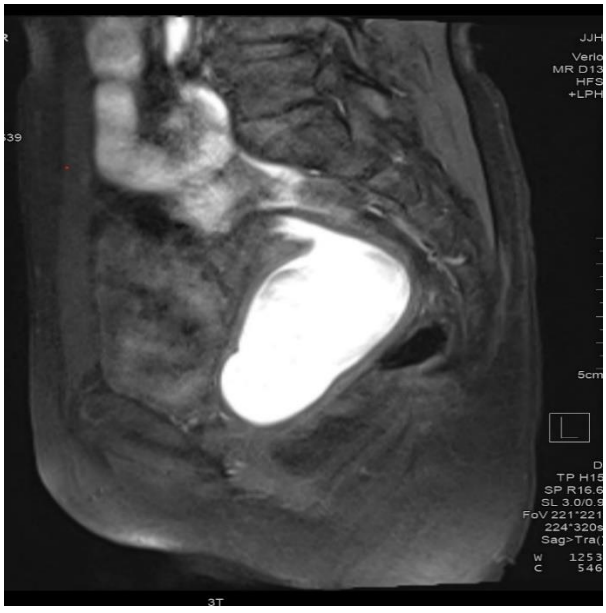

Figure 1: MRI (A+P) lateral view with hematocolpos.

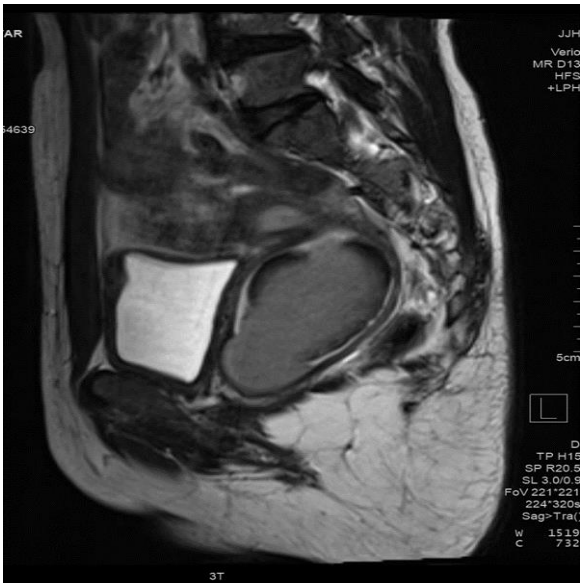

Figure 2: MRI (A+P) lateral view with hematometra.

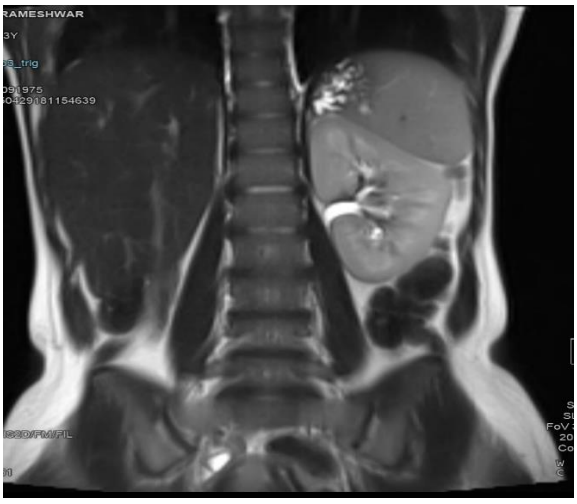

Figure 3: MRI (A+P) with absent right kidney.

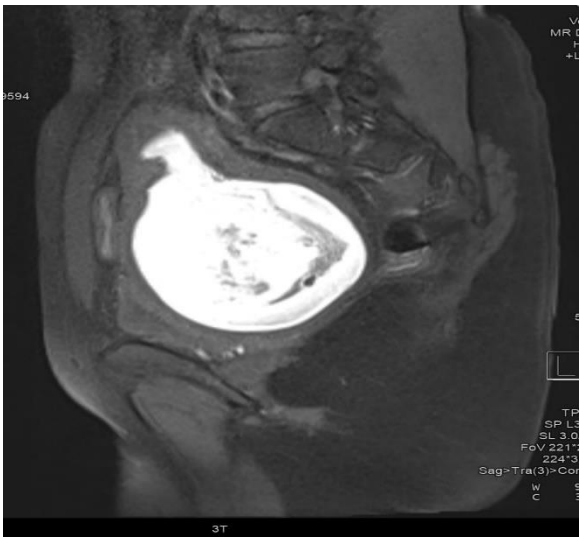

Figure 4: MRI (A+P) of case 2 with hematometra.

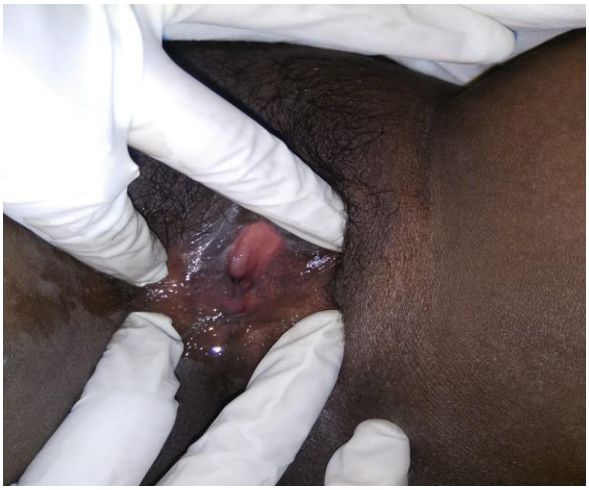

Figure 4: Post operative view of vaginal opening.

DISCUSSION

Development of female genital tract is a complex process, it is dependent on the events involving cellular differentiation, migration, fusion, canalization.[6] Upper portion of vagina is derived from Mullerian duct and lower portion from sinovaginal bulbs (urogenital sinus).[7] Canalization of uterovaginal canal occurs from the caudal to cephalic aspect.[6] Transverse vaginal septum may result from failure of complete canalization of primordial vaginal plate or from lack of union, which represents the junction of urogenital sinus and Mullerian duct contribution.[8] Transverse vaginal septum may be associated with genitourinary, gastrointestinal, musculoskeletal and cardiac anomalies. Genitourinary and gastrointestinal anomalies include imperforate anus, malformation of gut, ectopic ureter with hypoplastic kidney, hydronephrosis.[9] As in our case1, transverse vaginal septum is associated with absent right kidney. The most common location of the transverse septum is the upper vagina (46%), followed by the middle (35%), with least common location being the lower third (13%).[8] The transverse vaginal septum may be either perforate (incomplete) or imperforate (complete). In our case 1 and 2 it was upper vaginal septum while case 3 had lower third vaginal septum.

The onset of symptoms usually in case of complete septum occurs after puberty, with onset of menstruation. Patient usually presents with primary amenorrhea, cyclical pain in abdomen, hypomenorrhea. Patients with low vaginal septum present late than the high septum. In our cases, patients presenting with normogonadotropic normogonadism primary amenorrhoea with findings of blind vaginal pouch aids in diagnosis.

Diagnosis is confirmed by ultrasonography or MRI. MRI in our case not only helped in diagnosis but also to locate position of septum and also any other associated anomaly, as in case 1 OHVIRA syndrome i.e. (obstructed hemivagina and ipsilateral renal anomaly syndrome) was diagnosed on MRI.

According to Dennie's et al, the septum has to be removed when the girl reaches the menarche age, and the operative intervention is easier if the patient presents with hematocolpos before its drainage.[10] Williams et al treated patients affected by TVS in 3 different ways: abdominoperineal vaginoplasty via laparotomy, simple excision using the vaginal approach, and laparoscopic resection of the vaginal septum.[11] Tug et al reported the case of a young patient affected by obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome. In this case, the vaginal septum is completely incised by CO2 laser with hysteroscopy without hymenotomy, and the distal part of the septum is simply cut and sutured.[12]

Wierrani et al used Grünberger method, which consists of a cross-shaped incision on the caudal part of the septum, a cruciate incision on the cranial part, and transverse closure.[13,14] Bijsterveldt et al proposed two novel techniques for the treatment of the vaginal septum: the push through and pull through techniques. The push through technique requires a combined abdominal-vaginal approach, and it is used in patients presenting higher restenosis risk after surgery. The pull through technique is reserved for patients with a simple vaginal obstruction.[15]

Layman et al performed a modification of pull through technique, with a pull through of a proximal distended vagina using an Olbert balloon catheter to facilitate the surgical management and to limit the postoperative narrowing of the vagina.[16]

Sardesai et al described double cross plasty/Z plasty for the management of TVS as a better technique compared with the other surgical methods.[17] Kamal et al used surgical technique based on a transverse incision of the vaginal septum with tarring of edges by a vicrly 2.0.[18] In our cases, myodisection done over the outer vaginal walls of blind vaginal pouch. A small nick given and blunt dissection done and lower vaginal wall visualized, sims speculum inserted. On per speculum examination, bulging hemorrhage and high vaginal septum seen. A transverse incision taken over the transverse vaginal septum and hematocolpos drained out, flaps of remaining septum sutured along with raw vaginal wall with intermittent circumfrential sutures using vicryl 2-0. Flaps of transverse vaginal septum were used create neovagina. Daily phantom dilator insertion with lubricant application was advised, to avoid vaginal stenosis.

CONCLUSION

Transverse vaginal septum is usually undetected until puberty, when patient presents with primary amenorrhea or cyclical abdominal pain for evaluation. USG and MRI are used for confirmation of diagnosis. Transverse vaginal septum with vaginal atresia is treated by using flaps of transverse vaginal septum to create neovagina followed by use of phantom dilator. All three cases responded very well to the above line of management.

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