CASE REPORT

Medley of Gynecological Abnormalities Around Hematocolpos

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Pak Pediatr J 2026; **50(1):** 88-91

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Received for publication: Jan. 3, 2026
Revised received: Feb 14, 2026
Accepted for publication: Feb. 14, 2026

How to cite this: Fathima T, Mahalingam S, Joshi J, Mahale AR

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DOI:
https://doi.org/10.66347/ppj.v50i1.666

ABS TRACT

A 12 year old girl not attained menarche yet, presented with urinary retention and abdominal pain. Clinical examination and imaging confirmed the presence of hematocolpos secondary to both imperforate hymen and transverse vaginal septum. There was evidence of hydroureteronephrosis due to mass effect along with a septate uterus and vaginal aplasia (ESHRE Type U2b, C0, V4). Following examination under anesthesia, excision of the transverse vaginal septum was done along with drainage of menstrual blood and vaginoplasty to correct the vaginal aplasia. This is a very rare and possibly one of the first reported cases of concurrent imperforate hymen, transverse vaginal septum (TVS), septate uterus and vaginal aplasia with hydroureteronephrosis.

Key Words: *Hematocolpos, Transvere vaginal septum, Imperforate hymen, Septate uterus, Vaginal aplasia*

INTRODUCTION

Hematocolpos is characterized by accumulation of menstrual blood in the vagina. One of the causes is imperforate hymen, which after menarche traps menstrual blood behind the hymen and is noticed as a bluish bulge at the introitus. This with cyclical menstruation may lead to complications like hematometra or hematosalpinx. A similar presentation can be caused by transverse vaginal septum, which is believed to arise from failure of fusion of mullerian duct or failed canalization of mullerian plate. Patients usually present with primary amenorrhea, cyclical pain, urinary retention, difficulty in defecation and abdominal pain. Symptoms are present in the presence of normal secondary sexual development and vaginal examination generally reveals absence of the bulging of bluish membrane. Diagnosis is made on the basis of genital and gynecological examination and imaging with ultrasound scans of abdomen and pelvis or MRI.[1] Surgery is the mainstay of treatment in this condition and must not be delayed as management after puberty is associated with high rate of vaginal stenosis.[2] Moreover, retrograde menstruation can lead to development of endometriosis.[3]

CASE REPORT

A 12 year old girl presented to us with a history of recurrent urinary retention, which was relieved the previous time with a urinary catherization. Symptoms recurred again over a span of 1 month and hence child was referred to our centre with Foley's catheter in situ. She also had complaints of constipation and lower abdominal pain. The child had not attained menarche yet. Mother had attained menarche at 16 years of age.

On clinical examination, the abdomen was slightly distended with tenderness noted on deep palpation and a mass in the left iliac fossa. Local examination revealed bluish discoloration at introitus with absent vaginal orifice. Abdomen and pelvic ultrasound revealed hematocolpos secondary to imperforate hymen/transverse vaginal septum and

bilateral mild hydroureteronephrosis likely due to mass effect of the retained blood. MRI pelvis showed - probable septate uterus with hematocolpos and distal vaginal aplasia (ESHRE Type U2b, C0, V4)

Further, examination under anesthesia (EUA) with excision of transverse septum along with vaginal pull through was done under general anesthesia by the pediatric surgeon. A thin membranous septum with dark discoloration was found in the distal vagina and around 500ml of menstrual blood was drained. The post operative period was uneventful and the child is well on follow up.

DISCUSSION

Urinary retention at menarche due to imperforate hymen/TVS along with uterine and renal anomalies is rare and needs evaluation.

One of the causes for hematocolpos is Imperforate hymen i.e. a hymenal defect occurring due to failure of canalization of the inferior end of vaginal plate. Its incidence is approximately 1 in 1000 to 2000 females. It accounts for 90% of the cases of hematocolpos being the most common anomaly of the female genital tract.[4] It is entirely of urogenital origin without associated urogenital anomalies unlike a transverse vaginal septum.[1]

TVS, a type of vertical fusion defect is a rare mullerian duct anomaly, with an incidence of 1 in 70,000 females, resulting from failure of fusion and/or canalization of the urogenital sinus with the mullerian ducts. TVS presents with similar presentation like our child with hematocolpos or hematometra.[5,6] It can occur at any level within the vagina, 50% being in the upper vagina and corresponding to the junction between the vaginal plate and the caudal end of the fused müllerian and 10% of the cases in the lower portion. This can either be obstructive i.e. complete or non-obstructive i.e. incomplete with fenestrations.[7] An obstructive septum accumulates blood (hematocolpos , which was noted in our child) or mucus (mucocolpos) and non-obstructive septum presents with blood and mucus egress[1] and such patients complain of dysmenorrhea, hypomenorrhea, foul-smelling vaginal discharge and dystocia.[8]

Hematocolpos usually presents with a TVS or an imperforate hymen independently. The concurrence of both these anomalies is very rare as evident by the lack of literature on them, which makes our case a very unique.

Diagnosis is made based on the clinical and physical examination along with the radiological findings. In case of an imperforate hymen, a bluish bulge due to accumalated blood, is often seen between the labia which undergoes visible distension on application of suprapubic pressure and is absent in the case of TVS.[7] In our case, the child was noted to have an imperforate hymen, proven by the bluish bulge and confirmed by imaging which also showed evidence of a transverse vaginal septum.

Ultrasonography and MRI are gold standard in diagnosis. However, MRI is needed prior to surgery to confirm the thickness and depth of transverse vaginal septum and helps to identify the presence of cervix, hence differentiating a high vaginal septum from cervical agenesis.[1] In our MRI suggested a septate uterus with a normal cervix.

Another rare cause for hematocolpos is a bicornuate/didelphys uterus that occurs due to incomplete lateral fusion of mullerian ducts. This is often associated with renal agenesis[9] and along with an obstructed hemivagina is called the OHVIRA syndrome or Herlyn- Werner-Wunderlich syndrome. A bicornuate uterus can complicate the hematocolpos causing hematometra or hematosalpinx and is very rarely seen with vaginal aplasia.[10] Vaginal aplasia is a rare congenital anomaly and its commonest cause is Mayer-Rokitansky-Kuster-Hauser syndrome (MRKH syndrome).[11] Although our case does not fit into the OHVIRA syndrome or the MRKH syndrome completely, it is of note that a septate uterus was present along with vaginal aplasia and hydro-ureteronephrosis which has not been reported as yet.

Review of literature discusses cases of hematocolpos due to either TVS/imperforate hymen/hydroureteronephrosis due to mass effect/vaginal aplasia/uterine anomalies but rarely in combination or concurrently with other abnormalities. Our case seems to have a very rare medley of all the above mentioned defects with a possible syndromic association.

Surgical repair is the mode of management in these cases. Here, after performing an

examination under anesthesia, the hematocolpos was drained by making an excision of the imperforate hymen. The transverse vaginal septum was excised and the retained blood drained (500 ml). A vaginoplasty was then performed to correct the distal vaginal atresia in our patient.

A case of OHVIRA syndrome was reported by Jain and Mutyapwar where the TVS and vaginal aplasia were noted as associations. They managed the case with resection of TVS with vaginoplasty.[12] In another literature report another procedure was attempted by using the abdominal approach with laparoscopy for the TVS resection.[13]

Recently a minimally invasive technique was used by Ling et al, where a hysteroscope was inserted through the hymen under general anesthesia and an electric needle was used to divide the septum using ultrasound guidance.[14]

Despite the various types of surgical options, the importance lies in identifying the cause and completing the surgical repair without needing further corrective procedures as was done in this child.

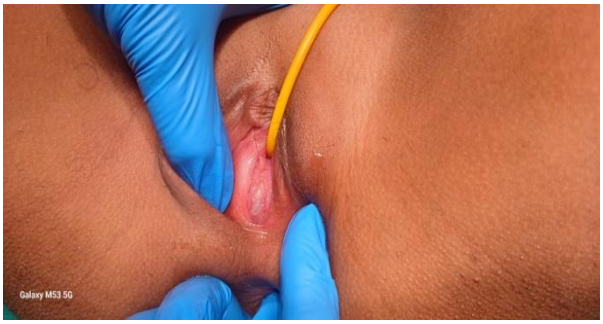

Fig 1: Bluish bulge noted at the introitus

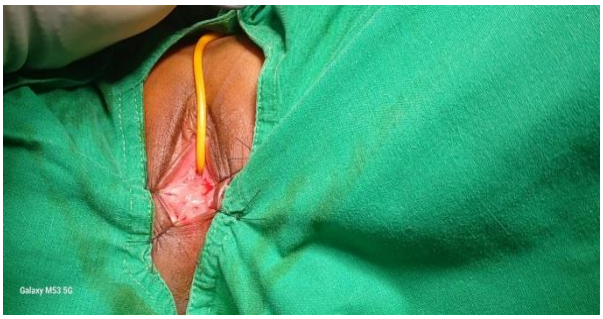

Fig 2: Absent vaginal orifice, urinary catheter in situ

Fig 3: Excision of the TVS

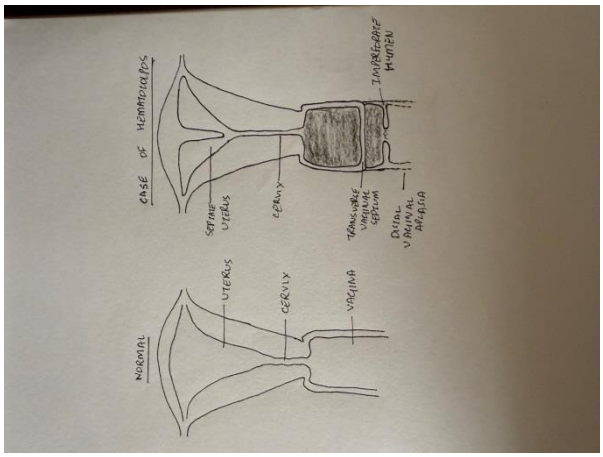

Fig 4: Diaphragmatic representation of Hematocolpos

Source of funding: None
Conflict of interest: None

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Authors' contribution

TF: Original draft writing, literature review and references
SM: Proposed topic, literature review, reviewed and edited the manuscript
JJ: Primary surgeon managing the patient, provided clinical insights and interpretation
ARM: Provided and interpreted radiological images, supervised manuscript writing
All the authors have approved the final manuscript draft and accept the responsibility of research integrity.