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Previously perforated transverse vaginal septum becomes obstructed: A case report

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Abstract
Objectives: To present a case of an apparently perforated transverse vaginal septum (rare anomaly of Mullerian duct) which later showed evidence of obstruction during the course of management.
Case report: 30 year old primigravida, with apparently perforated septum, however; post-operatively of her caesarean section, she had complaints consistent with imperforated septum. She was managed by dilatation and insertion of Foley's catheter that successfully maintained the opening of the septum on follow up.
Conclusion: Our case is rare since the pre-operative history indicated a perforated septum that became occluded post-operatively. There are very few reports of such conditions, and no clear explanation based on evidence. The proposed hypothesis is obstruction of micro-perforation by blood and tissue debris.

Keywords: Transverse vaginal septum, perforated, obstructed, congenital, anomaly

Introduction

Transverse vaginal septum (TVS) is one of the rare congenital anomalies of the development of the Mullerian duct [1], it was first reported in 1872 by Delaunay [2], then reappeared in literature again 72 years later in a report by Dannreuther [3]. Most of the publications about TVS seem to agree on an incidence that varies between 1:40,000 and 1:84,000 [1, 4, 5]. Unlike the etiology which remains a matter of debate, although recently it is believed to be multifactorial [6]. TVSs are broadly classified as perforated or imperforated, based on the presence or absence of communication between the vagina and the uterus [1], accordingly, clinical symptoms vary. An imperforated TVS usually presents early, as early as the neonatal period due to obstruction of flow of vaginal secretions (hydrometrocolpos), or in the early puberty period, with features of obstruction of menstrual flow, such as primary amenorrhea, hematocolpos, and hematometra, which are usually associated with the complaint of cyclic lower abdominal pain [1, 5]. On the contrary, perforated TVS may be asymptomatic and an incidental finding upon vaginal examination due to complains such as dysmenorrhea, dyspareunia, or infertility [7]. Regardless of perforation, there is a consensus on the frequency of TVS with regards to position in the vagina, with 46% located in the upper, 35% in the middle, and 19% in the lower vagina [4, 5] based on the findings of the largest available to date case series of female genitourinary congenital anomalies [8].

Here we present a rare case report of a female with presumably perforated TVS based on history that later on became obstructed during the course of management. We followed the CARE checklist of information to include when writing a case report. Both approval of the local institutional review board, and the patient's consent were formally obtained.

Case Report

A 30 years old primigravida Ethiopian lady, not previously following with us in the antenatal period presented to the ER of Maternity hospital with the complaint of labor pain. She gave no history of previous medical illness, surgical interventions, or gynecological complaints. She confirmed having regular menstrual cycles every 28 days, lasting for 5-6 days, with average amount of blood. Interestingly, she gave history of recent leaking vaginally. She was hemodynamically stable and afebrile. Abdominal ultrasound (U/S) revealed a viable single fetus with cephalic presentation of 32 weeks gestational age, and a normally situated placenta. Upon PV examination the cervix could not be identified, instead a blind vaginal pouch about 5 cm deep was felt.

We initially planned to admit her as a case of preterm labor and to do formal U/S, however; due to a non-reassuring cardiotocography (CTG) she was taken to the theater for emergency caesarian section (CS). At that time all her lab investigations were within normal ranges. Intra-operatively the CS was eventless with minimal blood loss, apart from the finding of anhydramnios and thick old meconium, we manually explored the lower uterine segment, however; we couldn't feel a definite cervical canal, and palpated a blind pouch without any definite perforations. Since the patient was consented only for CS, and based on the recommendations of senior consultants we decided to close the abdomen and counsel the patient post-operatively for examination under anesthesia and further surgical management of her TVS, which she initially refused due to socio-economic reasons. In the first 3 post-operative days she was doing well, hemodynamically stable, afebrile, with a clean surgical wound. However; there was absolutely no lochia or any vaginal discharge. On the fourth day she started to complain of lower abdominal pain and distension, U/S (figure 1) and computerized tomography (CT) scan (figure 2) both reported a distended lower uterine segment and dilated cervix with a collection of blood measuring 65 by 58 mm, magnetic resonance imaging (MRI) (figure 3) confirmed those findings with the addition of loss of clear communication between the uterine cavity and the vagina. In view of her aggravating lower abdominal pain she eventually consented to EUA and further management. In the second surgery, vaginal examination from the lithotomy position revealed a short blind vaginal pouch of about 5 cm that was completely occluded by a fibro-vascular septum, there were no visible nor palpable perforations in the septum. Under U/S guidance, we introduced a needle in the middle of septum which withdrew old blood, so with continuous U/S guidance we introduced dilators in the point of needle puncture, up to size 8, followed by a Foley's catheter that drained about 300 ml of dark old blood, the septum was about 1.5 cm thick. The balloon of the catheter was inflated, and catheter fixed in place by adhesive tape after confirming its position by U/S. Post-operatively, the patient was covered by Cefazolin 500 mg intravenously (IV) every 6 hours for 48 hours, Metronidazol 500 mg IV infusion every 8 hours for 48 hours, Clexane 40 mg subcutaneously daily for 5 days, and analgesia. The uterine Foley's catheter was kept for 14 days, then removed and the patient was discharged home, she followed in our outpatient clinic 3 months later, and there was no stenosis or narrowing of the opening in the septum, and the patient reported no complaints.

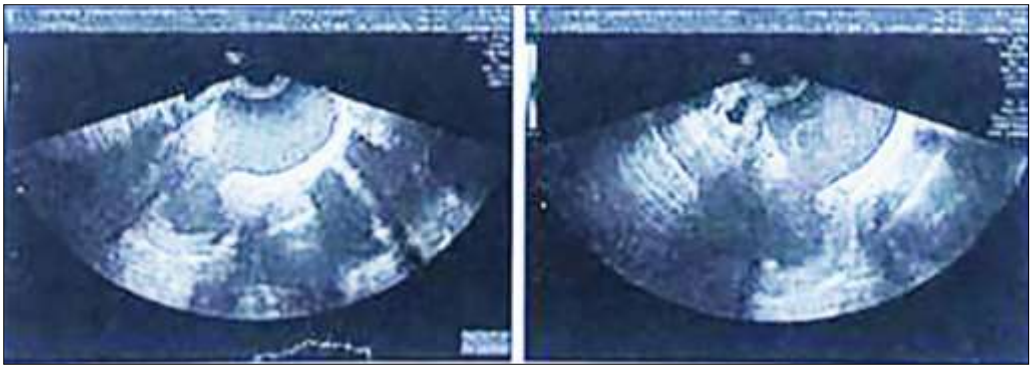
Discussion

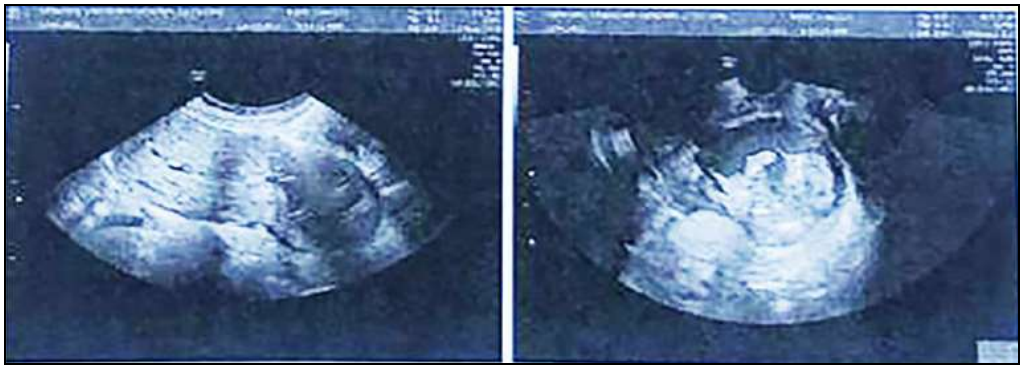
The etiology of TVS remains a debatable issue, between the theories of failure of fusion of the Mullerian system which forms the upper vaginal part and the sinovaginal bulb which forms the lower part, the theory of failure of canalization of the vaginal plate, or a combination of both [1, 4]. The first theory fusion failure is support by histopathological examination of TVSs, in which the cranial side is lined by columnar epithelium of Mullerian origin, whereas, the caudal side is lined by squamous epithelium of the urogenital sinus. The theory of failed canalization is supported by the presence of TVSs at various levels in the vagina [1, 9]. According to the consensus of the European Society of Human Reproduction and Embryology (HSHRE) and European Society of Gynecological Endoscopy (ESGE) [10], our patient would be classified as U0C0V3, since she had no anomalies of either the uterus or the cervix with a TVS.

We used almost all the diagnostic tools reported to be beneficial in this case with our patient, we performed transvaginal and transabdominal U/S which could identify the pathological condition and vaginal structure, we also performed CT scan and MRI both of which are also reported to be used in the diagnosis of the condition, with MRI having the advantage of ability to distinguish between highly situated TVS and congenitally absent cervix by visualization of the cervix [6].

Our initial expectant plan of management has also been advocated by others [11], however; it was replaced by CS delivery due to non-reassuring CTG. Afterwards, we opted for a non-surgical management of the patient's TVS using dilators and Foley's catheter which was also reported to be successful by others [4], although the dilatation method is advised by some reports for small septa, or as an adjunct to surgery to improve outcomes [6]. Several surgical techniques have been proposed, particularly for cases with low TVS such as transverse incision over the vault of the vagina followed by anastomosis [12], or Z plasty especially with septa of less than 1 cm thickness, while larger septa (> 1 cm) may be managed by longitudinal Z plasty [6], or Y plasty [13], although none of the surgical management techniques has been without complications, the most outstanding among which post-operative stenosis is the most common [6, 12].

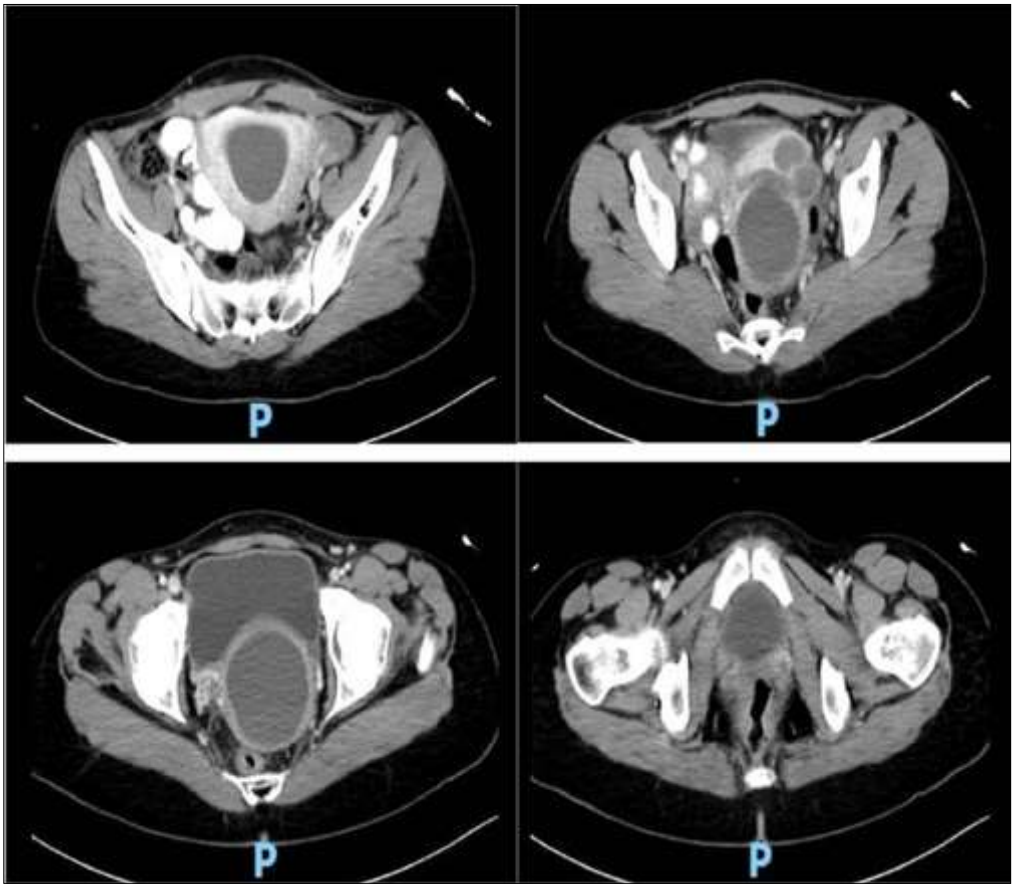
The rarity and singularity of our case arise from several points, first, the history given by the patient indicates a perforated TVS, yet, we could not identify the perforation both by examination of the lower uterine segment during her CS, and by vaginal examination under anesthesia. Second, her post-operative course indicates that the septum became obstructed, resulting in accumulation of blood within the uterine cavity, which was confirmed by CT and MRI. We were able to find only two similar reports of obstruction of a presumably perforated TVS [1, 13], both reports couldn't fully explain this occlusion, and proposed a hypothesis of obstruction of a micro-perforation by blood and tissue debris or septal scarring. Possible other causes of obstruction may include infection during pregnancy, or spontaneous transformation.





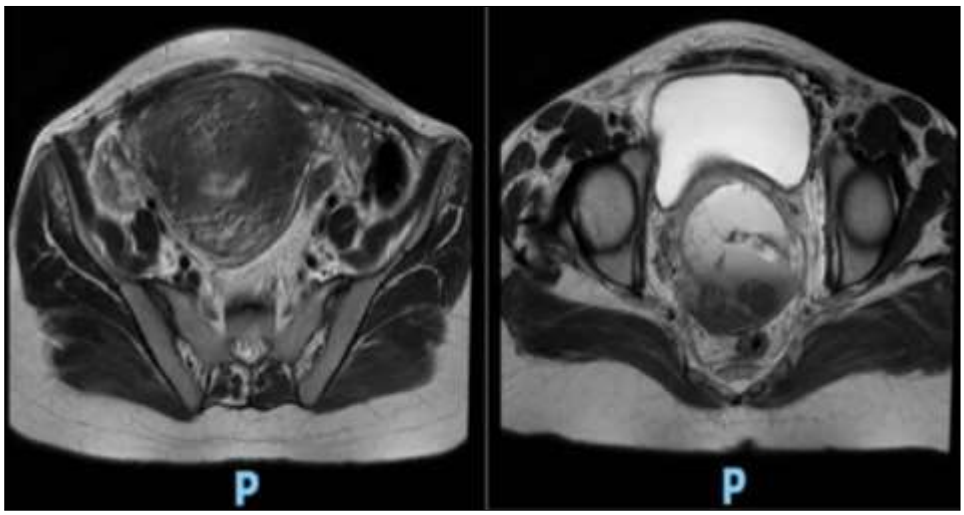
Dilated cervix, and distended lower uterine segment with blood collection

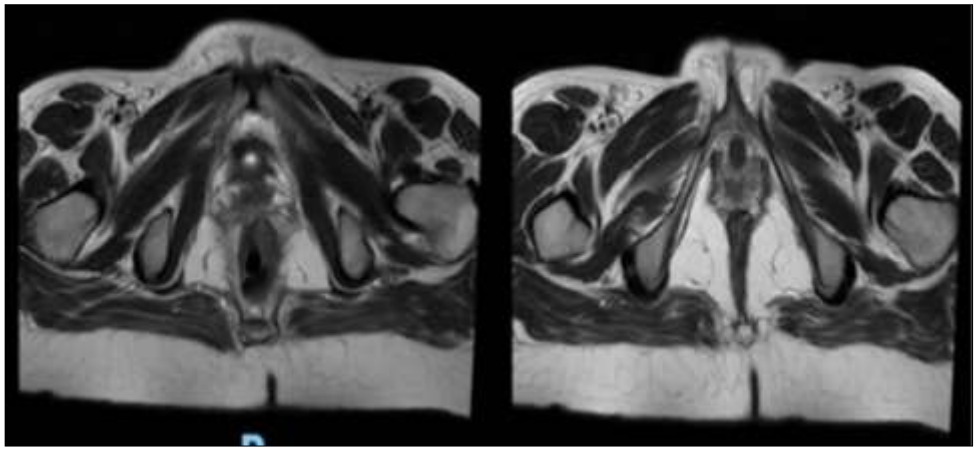
Fig 1: Trans-abdominal and Transvaginal ultrasound



Fluid filled dilated endometrial and endo-cervical canal, with no evidence of extra-uterine free collection.

Fig 2: Computerized Tomography





Heterogeneous collection in cervical canal, with loss of clear communication with vagina.

Fig 3: Magnetic resonance imaging

Conclusion

Perforated TVS may present as an incidental finding during labor, however, identification of the perforation site may not be possible in a micro-perforated TVS, and such TVS may become obstructed later on. Further studies and investigations are required to ascertain the causes of transformation.

Financial and conflict of interests' declaration

No personal or institutional financial support was obtained for this article. All authors declare no conflict of interests.

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