

Unusual traumatic uterine injury: first reported cervicouterine transection

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Cervical agenesis is one of the Müllerian developmental anomalies that can occur and is usually associated with vaginal atresia rarely isolated. Here we are reporting a case that has been referred as cervical agenesis and found to be a cervicouterine transection, so far not reported in literature. We report a case of traumatic cervicouterine transection in teenager patient who presented with amenorrhea and hematometra. She was primarily investigated and found to have intact full length cervical canal, normal uterus, and urinary system. Operative management confirmed our diagnosis of transection rather than agenesis with her history of trauma at the age of 3 years. We report our case with medical diagnostic approach and

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Introduction

Cervical developmental pathology is a very rare condition, with atresia being so far reported in rare cases and often associated with vaginal atresia. Clinical diagnosis is usually difficult before surgery. Transverse vaginal septum or vaginal agenesis is also a rare condition that results from incomplete fusion between the vaginal components of the Müllerian ducts and the Phallic part of the urogenital sinus. Clinical presentation depends on whether it is partial or complete. Complete agenesis leads to the accumulation of menstrual blood in the uterus and the structures above it [1–6]. In this paper, the management of the first case of traumatic cervicouterine transection is reported.

Case report

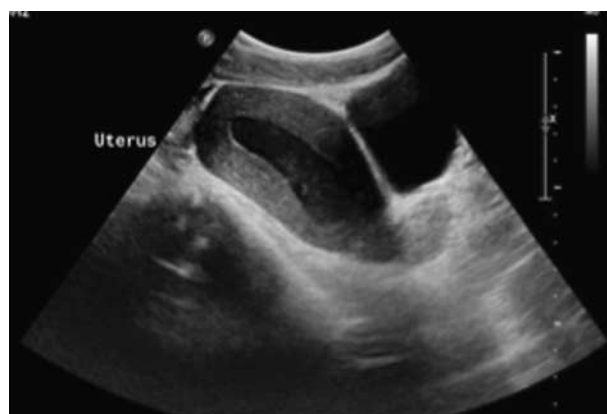
A 14-year-old Saudi girl was referred to King Abdulaziz University in Jeddah from Taif as a case of primary amenorrhea with cyclic abdominal pain for 1 year, with the transabdominal ultrasound (US) report from local hospital showing hematometra and hematosalpinx. Patient consent was acquired from the patient and the parents. On examination, she had normal secondary sexual characteristics: breast Tanner stage 3; normal axillary and pubic hair; and normal external genitalia, hymen, and vaginal opening. According to her medical history, she had a traumatic car accident at the age of 3.5 years. A car drove over her pelvis, causing multiple pelvic fractures, leading to admission to ICU and mechanical ventilation with hematuria and per vaginal bleeding that continued for 1 week; she received conservative treatment only for 2 months in the hospital.

Transabdominal US and MRI of the pelvis indicated an enlarged uterus with hematometra and hematosalpinx, normal ovaries (Fig. 1), normal vagina, and a cervical canal with an area of stenosis or agenesis at the cervicouterine junction ~1 cm in length. The rest of the study was normal in terms of other organs and lymph nodes (Fig. 2). Hormonal profile and chromosomal analysis were normal as well.

The examination of the patient under general anesthesia with hysteroscopy showed a normal vaginal shape and size, with a normal cervical canal three centimeters in length; thus, using intraoperative US, cervical dilatation was attempted with a Hegar dilator. During the procedure, posterior perforation was diagnosed by US and immediate laparoscopy was performed, showing a bulky uterus with bilateral distended tubes and fused fimbrial ends and multiple endometriotic spots in the uterovesical pouch and minimal pelvic adhesions. The Hegar dilator was placed posteriorly just at the level of the uterosacral ligament, which was removed under direct vision. No bleeding could be observed. During manipulation of the left fallopian tube, some of the old collected blood came out. Suction irrigation was performed, and again no bleeding was observed at the site of the perforation. A suction drain was inserted and the procedure was completed with a plan for laparotomy and uterocervical reconstruction later.

Three weeks later, she was admitted for cervicouterine junction reconstruction. An abdominovaginal approach

Fig. 1



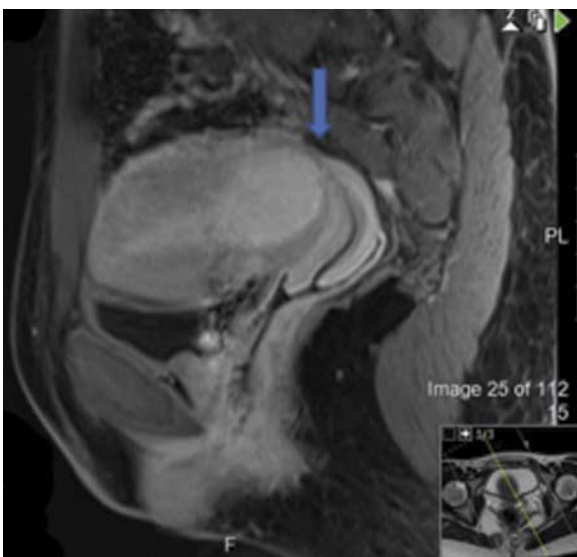
Dilated uterus full of blood.

was planned. After laparotomy, both the uterus and the fallopian tubes were distended with old menstrual blood, the peritoneum was dissected, and bladder was pushed down to expose the anterior uterine wall and well-developed lower uterine part and full cervix with normal thickness and length with a small area of fibrosis at the junction between the cervix and the uterus about 1-cm in length. This finding indicated the transection of the uterus at the level of the internal cervical ostium, which means that the uterus was transected at the time of the car accident that she had at the age of 3 (Fig. 3).

A longitudinal incision on the lower anterior uterine wall was performed, old blood was drained out, and a uterine

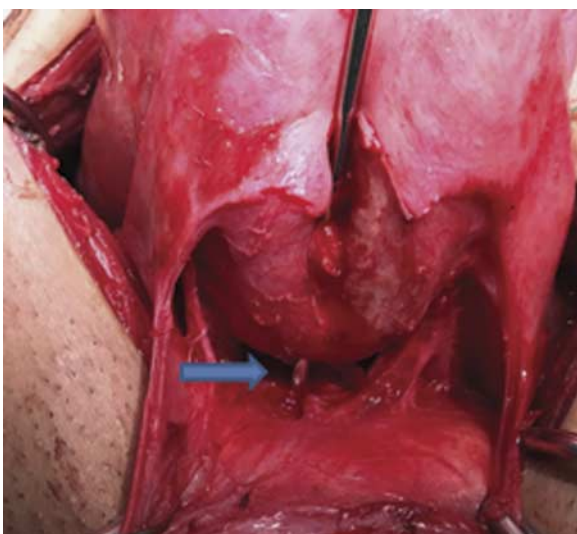
probe was inserted through the incision and directed toward the cervix to dilate the fibrous area. At the same time, a transvaginal small dilator was inserted through the cervical canal and forced through the fibrous transected end, and then gradual dilatation of both fibrous ends till a size 16 F, foley catheter was passed through the cervix to the uterine cavity (Fig. 4). The uterine incision was closed and then the cervicouterine junction was sutured with multiple interrupted stitches (Fig. 5). Medium suction soft drain was inserted into the pelvic cavity. The procedure was completed and the uterine catheter was kept inside for 1 month with antibiotics coverage.

Fig. 2



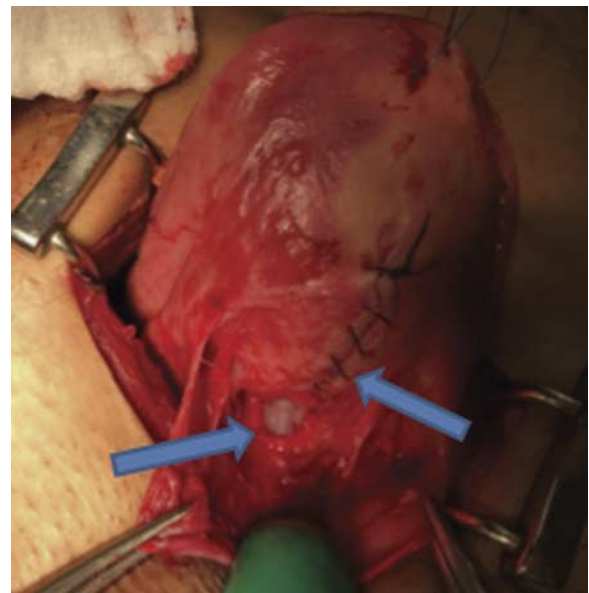
MRI pelvis showing the dilated uterus with normal cervical length and a small area of fibrosis in between (arrow).

Fig. 3



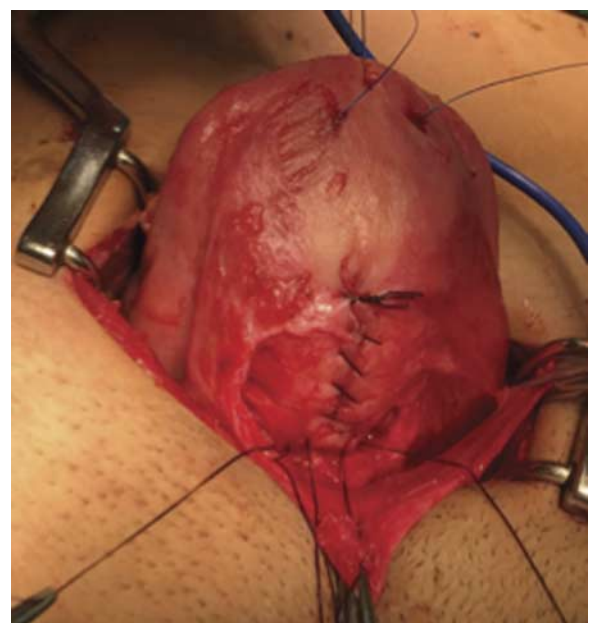
Hegar dilator passing through the cervix at the level of the internal OS (arrow) to the uterine cavity.

Fig. 4



Uterus and cervix with the catheter (arrows) passing and connecting both.

Fig. 5



Anastomosis of the uterus with the cervix.

Fig. 6



Catheter passing through the cervix to the uterine cavity with the dye showing normal uterine cavity.

During her postoperative period, she was stable, afebrile with no pain, and there was visibility of menstrual blood through the uterocervical stent. After 1 month, the patient was admitted for removal of the catheter, with an examination under anesthesia. The intraoperative catheter could be removed easily with minimal bloody discharge, and washing with saline was performed, followed by hysterosalpingogram, which showed a normal cervical canal in continuity with the uterine cavity (Figs 6 and 7).

Discussion

We report this case as an unusual presentation of primary amenorrhea with hematometra likely because of cervical agenesis. On review of the literature, cervical agenesis or aplasia is a rare congenital variation of Müllerian anomalies and commonly associated with partial or complete vaginal atresia, agenesis, or septum [7]. These patients usually present after puberty with cyclic abdominal pain with different-size pelviabdominal masses depending on the time of presentation after puberty. US is a less reliable imaging tool, but still remains the first method for diagnosis. However, MRI is known to be the best method for diagnosis in these cases; still, it is difficult to differentiate between complete agenesis and dysgenesis of the cervix, and the diagnosis needs to be confirmed intraoperatively [8]. In our patient, the MRI (Fig. 2), with her clinical history of trauma with documented different pelvic organ injuries at the age of 3 years with bleeding per vagina for a period of time, and considering that only conservative management had been followed, misdiagnosis of uterine transection was expected. This diagnosis was supported by our intraoperative findings of adhesions between the base of the bladder and the cervicouterine area and cervix, and also normal cervical length and patent canal with a normal uterus and other pelvic organs.

Fig. 7



The dye is passing out through the vagina.

Although different surgical techniques are reported for the management of cervical agenesis, the method of choice remains controversial [9]. Hysterectomy was the management for these cases before because of the complication of cervical recanalization and the difficulty of having normal viable pregnancy [10]. Improved surgical techniques and preservation of the uterus for future fertility are considered to be the first line of treatment at present [11]. In this case, we report a traumatic cervicouterine transection that was repaired with anastomosis of the transected part over the Foley's catheter, which kept in as a stent to promote epithelization of the cervicouterine junction and prevent stenosis or obstruction. Follow-up of these patients is recommended as restenosis or obstruction can occur again as reported by others [10–12].

Conclusion

Cervical agenesis isolated or with different degrees of vaginal aplasia is rare and always reported as the congenital cause of primary amenorrhea and hematometra in adolescent patients. In our patient, traumatic misdiagnosed uterocervical transection during her childhood was the diagnosis on the basis of her history and intraoperative findings. To our knowledge, this is the first reported case of cervicouterine obstruction because of traumatic cervicouterine transection injury.

Conflicts of interest

There are no conflicts of interest.

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