CASE REPORT

Recurrent formation of haematocolpos in a young girl with multiple congenital anomalies of the urogenital tract

Mehreen Yousaf Rana, Syeda Hira Naqvi, Rubina Hussain

Abstract

An 11-year-old girl, a known case of left crossed fused renal ectopia and sacral hypoplasia presented to the gynaecological OPD in Karachi, Pakistan, in February 2019 with complaints of abdominal pain. On examination, she was found to have a septum covering her vaginal orifice. She was subsequently diagnosed with haematocolpos secondary to imperforate hymen. Incision and drainage was done. However, despite surgical management, she continued to have recurrent formation of haematocolpos for the next two months secondary to multiple complete and partial transverse vaginal septa and post-operative formation of adhesions. Definitive management was done with ultrasound guided needle puncture and drainage, followed by post-operative tampon use to maintain patency.

Keywords: Haematocolpos, urogenital abnormalities, imperforate hymen

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Introduction

In approximately one in 2,000 females, the hymen fails to perforate during the development of the genitourinary system leading to formation of haematometrocolpos.[1] Transverse vaginal septum is also a rare condition that results from incomplete fusion between the Mullerian ducts and the urogenital sinus.[2] It is associated with genitourinary and gastrointestinal tract anomalies such as imperforate anus, ectopic ureter with hypoplastic kidney and musculoskeletal defects such as sacral hypoplasia.[3] Presentation may vary depending on whether the septum is complete or partial.[4] After taking due consent from the patient and her parents, we report the case of a young girl with multiple congenital anomalies who despite undergoing incision and drainage, presented with recurrent haematocolpos.

Case Report

In February 2019, an 11-year-old girl, presented to the gynaecology OPD at Dr. Ziauddin Hospital Clifton with a three day history of lower abdominal pain. She was pre-menarche and had no other associated symptoms.

According to her parents, she was born at full term via spontaneous vaginal delivery with no anomalies detected prior to or at the time of birth. However, at the age of 1.5 years, she had anal dilatation surgery as her parents noticed that her anal opening appeared smaller than usual. Around that time, she was also found to have sacral hypoplasia and left crossed fused renal ectopia with both the ureters opening into the vagina causing constant dribbling of urine. Subsequently, she underwent bilateral transureteric reimplantation at the age of two. Thereafter, she suffered from nocturnal enuresis and stress incontinence but her symptoms had improved over time.

When she presented to us, she was vitally stable with no significant abdominal tenderness. Genital examination revealed a thick septum covering the vaginal orifice. The rest of the external genitalia appeared normal.

Initial CBC revealed Hb: 11.6 gm/dl, TLC: 11 x 10^9/L, PLT: 258 x 10^9/L.

On ultrasound, the right kidney appeared smaller and in close proximity to the left kidney with possible fusion at the hilum of the left kidney. These findings were consistent with the already known left crossed fused renal ectopia.

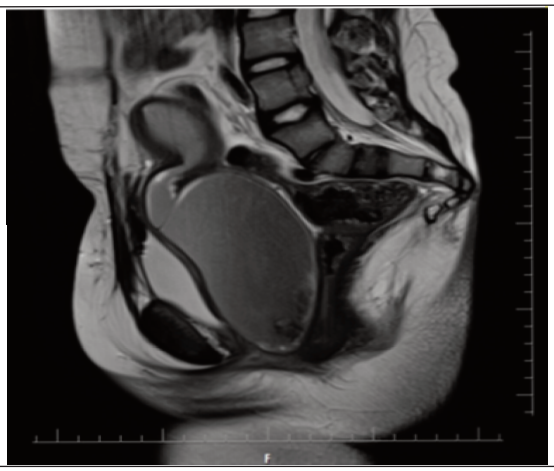

Figure-1: MRI pelvis with contrast showing an elongated and distended vagina with central haemorrhage displacing the uterus and urinary bladder as well haemorrhage in the endometrial canal suggestive of haematometrocolpos.

Department of Obstetrics and Gynaecology, Ziauddin Hospital, Clifton, Karachi, Pakistan.

Correspondence: Syeda Hira Naqvi. e-mail: hiranaqvi95@gmail.com

Dilated vagina was seen which measured approximately 7.5 x 5.7cm. Uterus showed thickened endometrium measuring 1.8 cm. Rest of the scan was unremarkable and overall findings represented haematocolpos. Following the ultrasound findings, an MRI of the pelvis with contrast (Figure-1) was done which was suggestive of haematometrocolpos due to imperforate hymen.

Subsequently, she was admitted for incision and drainage under general anaesthesia. After taking all aseptic measures, the patient was cleaned and draped. The urethra appeared grossly dilated and drops of urine were dribbling out even after insertion of Foleys catheter. Anal opening was separately identified. A thick septum was seen covering the vaginal orifice. A deep cruciate incision was given, extending laterally from 3 to 9 o'clock position and along the midline from 12 to 6 o'clock position. The septum was approximately 5mm thick. 500 ml of blood was drained and stitches were applied at the 2, 4, 8 and 10 o'clock positions. Vaginal patency was assessed with index finger and cervix was palpated. Post operatively she made a good recovery and was discharged the next day. Following the procedure, she menstruated for three days.

A month later, she presented with similar complaints of abdominal pain. On examination, the vaginal orifice was found to be closed again. An ultrasound was repeated and examination under anaesthesia was planned. Hegar's dilator was gently pushed into the vaginal orifice for recanalisation and 5ml of mucus was drained. However, the cervix could not be palpated. Vaginal packing was done and the patient was discharged the same day after removal of the packing. The patient was advised daily dressing with Bactigras for the next few days.

The third time her symptoms reoccurred, the MRI showed left haematosalpinx and haematometrocolpos with distended vagina measuring 11 cm, ending blindly, owing to the presence of septum in the distal vagina measuring 5-10 mm. There were also multiple incomplete thin septa noted in the lower 1/3rd of the vagina.

Ultrasound guided needle puncture of the vaginal orifice was done under general anaesthesia and a wire was passed into the uterine cavity. The length of the vagina measured around 10 cm. A nick was given above the wire and approximately 500ml of dark coloured blood was drained. Index finger was inserted and the septa were broken. Vaginal orifice was created by putting interrupted stitches at 2, 4, 8, 10 o'clock positions. Haemostasis was secured. Intraoperatively, on ultrasound, the uterine cavity appeared empty. All the blood had been drained and patency was confirmed by inserting a finger up to the cervix. The patient was discharged the next day and advised the use of tampons daily until follow up to prevent reformation of adhesions. Two days later, the girl spontaneously started menstruating.

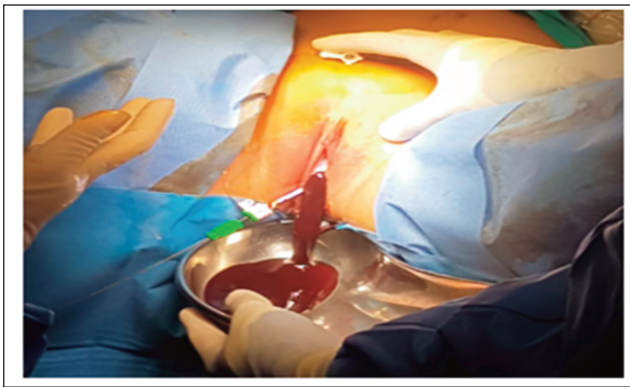

Figure-2: Ultrasound guided needle puncture and drainage of blood.

Discussion

In the case reported above, a young girl with multiple congenital anomalies, presented with haematocolpos. Initially, she was managed surgically for imperforate hymen. However, recurrence of symptoms, led to the diagnosis of multiple vaginal septa as well as stenosis of surgical site postoperatively. Definitive management was done by ultrasound guided drainage ensuring complete patency and tampons were advised postoperatively to prevent adhesion formation and thus recurrent reproductive outflow tract obstruction. Cyclic menstrual bleeding was observed for six months after the third surgery.

Transverse vaginal septa are frequently accompanied by urinary tract, musculoskeletal, gastrointestinal and cardiac abnormalities. A complete septum can be located at various levels, but is more common in the middle (40%) and upper third (46%) of the vagina.[4] Hypoechoic crescent shaped tissue with distended blood-filled uterus on ultrasound may reveal the diagnosis. However, an MRI is recommended, especially for surgical correction as it can help locate and measure the thickness of a septum.[5]

In a similar case reported in Japan, a 16-year-old girl had been surgically managed several times for haematocolpos since the age of 13. Her imperforate hymen was incised, followed by ultrasound-guided puncture and excision of transverse vaginal septum. Re-operation was performed after six months due to recurrence of adhesions. A silicon dilator was then inserted. However, as she was unable to use it at home, she used a tampon instead.[6]

A case reported in Nigeria, suggested the use of high pressure dilatation balloon for the surgical management of transverse vaginal septa, to prevent postoperative

narrowing.[7] Dilation techniques may be used before surgery to improve the outcome, or after, to prevent strictures, scarring, or stenosis postoperatively.[8]

Reoperation in our case could have been prevented if intraoperative ultrasound guidance was used initially. This would have also helped in identifying the cervix which is crucial in ensuring that the vaginal canal is patent throughout its length.

Conclusion

It is important to keep in mind the simultaneous occurrence of imperforate hymen and transverse vaginal septum in young patients diagnosed with haematocolpos. After initial surgical management, recurrent formation of haematocolpos should be anticipated and in such cases ultrasound guidance should be used intraoperatively. Cervix must be palpated for assessment of adequate patency and, if unable to do so, a guide wire should be passed until the cervix is reached. At the time of discharge, the patients should be advised to use tampons to prevent postoperative adhesion formation in the lower genital tract and thus the recurrence of haematocolpos.

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Conflict of interest: One of the co-authors of this manuscript has also signed the ethical review statement.

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