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Case Report

Familial imperforate hymen in an adolescent presenting with acute urinary retention and haematocolpometrosalpinx

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ABSTRACT

Imperforate hymen rarely presents as a familial disorder while most of the patients with the anomaly are asymptomatic until the menstrum above the imperforate hymen results in mechanical effect. An uncommon presentation of imperforate hymen is acute urinary retention in young girls. We report a case of acute urinary retention due to massive hematocolpometrosalpinx in a 12-year-old girl whose elder sister had surgery for imperforate hymen in the past. She presented with a progressively increasing perineal bulge and acute urinary retention of one month and seven hours respectively. Ultrasound revealed hematocolpo-metrosalpinx and the retention was rapidly relieved with size 10 Foley catheter. She subsequently had hymenectomy and drainage of the accumulated menstrum. She did well and was discharged three days later. Familial occurrence of imperforate hymen is uncommon more so presenting as acute urinary retention. Enlightenment of parents on imperforate hymen may prevent complication in later life.

Keywords: Urine retention, inperforate hymen, haematocolpos, haematometra, hematocol-pometrosalpinx, adolescent.

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Introduction

Untreated imperforate hymen is a rare but recognized cause of acute urinary retention. Imperforate hymen has an approximate incidence of 0.05-0.1%[1] and urinary retention is an uncommon presenting symptom.[2] Imperforate hymen is thought to occur due to absence of degeneration process that causes hymen anomalies. Most of the patients will be asymptomatic until the accumulation of menstrum above the imperforate hymen results in mechanical effect on the bladder, urethra and sometimes the

rectum leading to obstructive urinary symptoms and constipation respectively.[2] We present a case of an adolescent girl with imperforate hymen that presented with acute urinary retention.

Case History

A 12-year-old girl presented with inability to pass urine and constipation, lower abdominal pain and vulval swelling of 7 hours, 5 days, 3 days and 1 month respectively. She was yet to see her menses, however had developed secondary sexual characteristics.

The bulge on her vulva started a month prior to presentation, gradually increased in size but became noticeable two days prior to her presentation at our facility. The vulva bulge was painless, but associated with discomfort, as she could not close her legs while in sitting position. She had been apparently well despite her initial symptoms until she started having constipation five days prior to presentation. This was associated with lower abdominal pain three days prior to her presentation. The pain was colicky and intermittent, severe enough to affect her routine chores. There was initial difficulty in urination, but stream improved with straining, until 7 hours prior to presentation when she could not pass urine even with straining.

There was no history of fever, headache or vomiting and she was yet to attain coitache. Her elder sister had similar problem and had surgery for cryptomenorrhea at the age 16 years.

She was found to be in painful distress. Her BMI was 22.8 kg/m2 with a Tanners' stage III breast and pubic hair developments. The patient's vital signs were normal. There was a smooth, slightly tender, suprapubic mass corresponding to 18 weeks sized uterus. Pelvic examination revealed a bulging bluish membrane covering (Figure 1) while digital rectal examination revealed normal anal sphincter, empty rectum with smooth mucosa. The anterior rectal wall was bulging.

The laboratory results revealed haemoglobin estimation 11.5 g/dl, urinalysis was negative for glucose and proteins and urine microscopy and culture revealed no pathogens.

Pelvic ultrasound scan showed vagina and uterus filled with mixed echogenic fluid extending into the fallopian tubes (Figure 2).

She was diagnosed to have acute urinary retention due to massive hematometrocolposalpinx secondary to imperforate hymen. The urinary retention was relieved immediately with a size 14 Foley urethral catheter and drained 500ml of amber coloured urine. The patient and her parents were counselled on surgical management (hymenectomy) which they consented for. A Hymenectomy was performed under subarachnoid block. A total of 400mls of dark menstrum was evacuated from the vagina. The remnant hymen was sutured by evaginating it using Vicryl 2.0 (Figure 3). Antibiotics were given prophylactically.

Repeat pelvic ultrasound scan done 48 hours after the procedure revealed a slightly bulky uterus with a defined endometrial plate and vagina. The patient was discharged on the third postoperative day and resumed normal menses after one month.

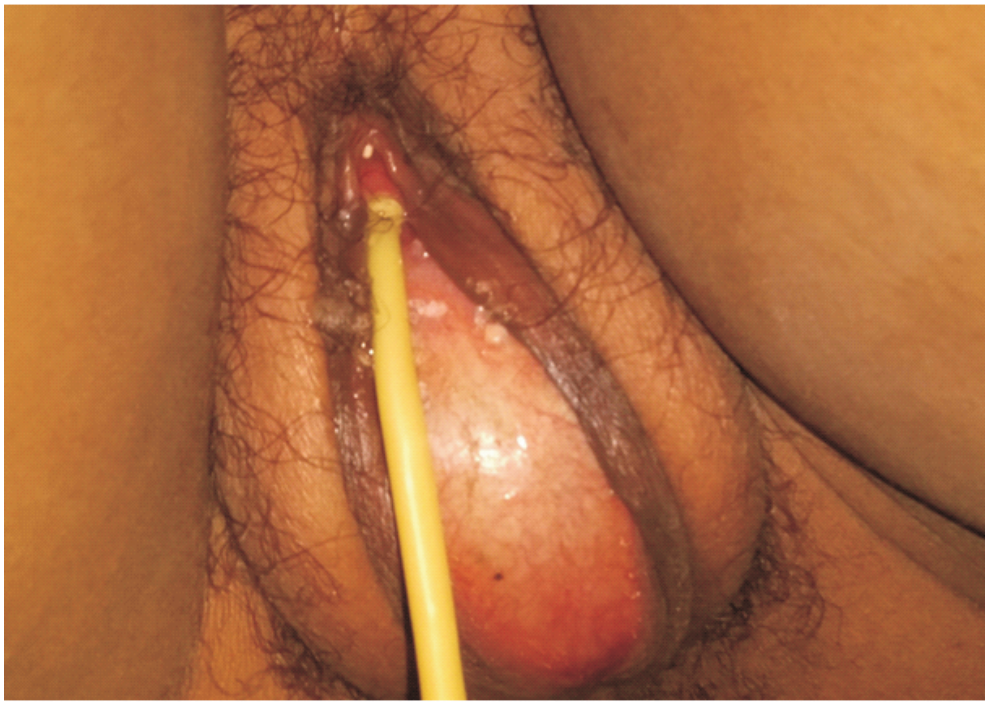

Figure 1: A bulging hymen with a Foley catheter in situ

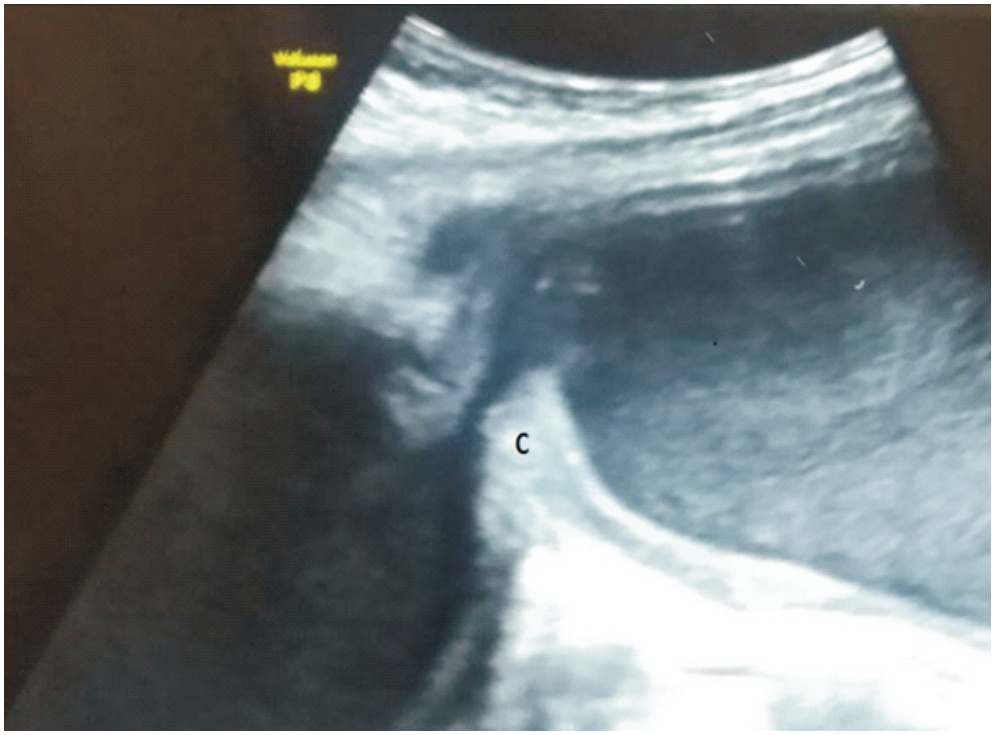

Figure 2: Ultrasound scan showing haematocolpos on the lower left, haematometra on the upper right and the cervix (c)

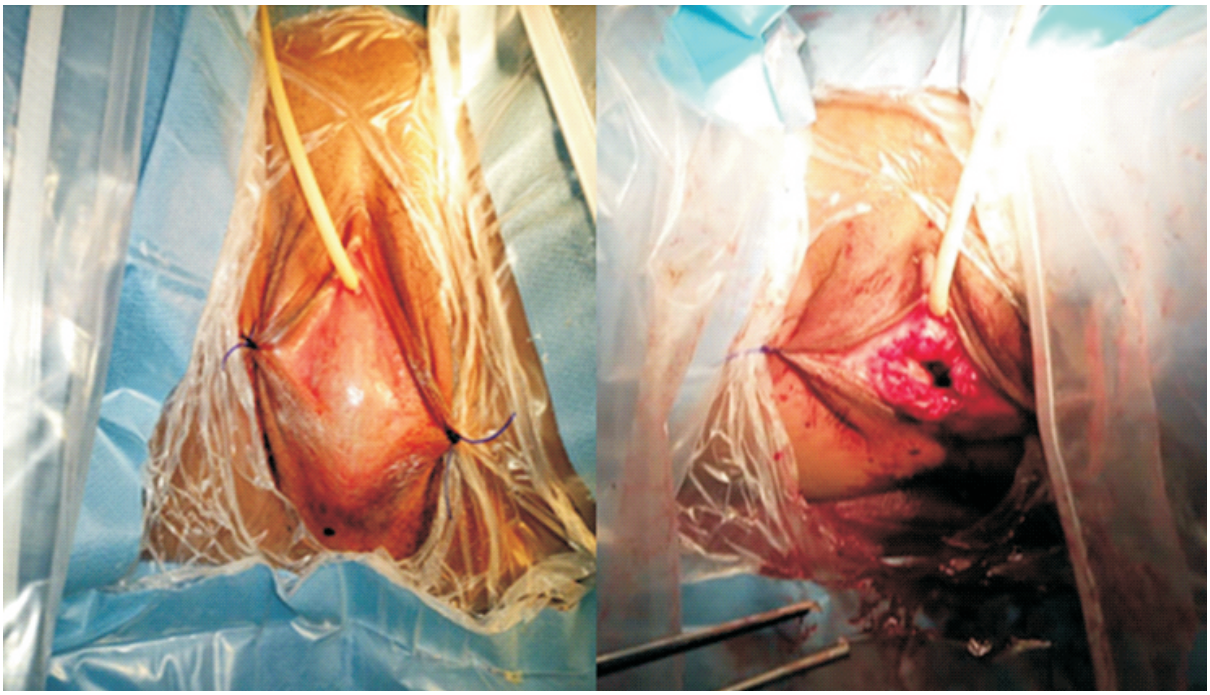

Figure 3: Imperforate hymen before (left) and after surgery (right)

Discussion

The hymen is an embryological derivate of the inferior part of the sinovaginal bulb which canalizes to give rise to varying anatomical configurations of the hymen. Imperforate hymen is a rare form of congenital vaginal obstruction.[1] Untreated imperforate hymen is a rare but recognised cause of acute urinary retention. Bladder outlet obstruction is one of the least seen presentation in patients with imperforate hymen with an incidence.[1] Presentation varies from during neonatal life with an incidence of 0.4% to other manifestations in later life where cyclical abdominal pain due to crypto-menorrhea or primary amenorrhea, constipation and urinary obstruction could occur.[1-4]

In the case presented there was family history of similar problem on account of which surgery was done. Imperforate hymen usually occurs sporadically but can be rarely familial.[5] There are very few cases reported of familial imperforate hymen.[5-8]

The mean age at presentation though was 10.7 years.[1] The young girl presented was just turning 12 years of age, given the age of presentation around the time of establishment of secondary sexual characteristics. We posited that menstruation had begun quite long before her presentation. Haematocolpometria could result in acute urinary retention due pressure on the bladder from the distended uterus causing upward angulation of the bladder neck and kinking of the urethra, direct tamponade effect on the urethra by the distention of vagina, bulging hymen causing distention of the vagina and cephalad angulation and stretching of the urethra worsening the tamponade.[9,10]

The management of acute urinary retention with imperforate hymen include urethral catheterization temporarily to relieve obstruction and improve symptoms. Hymenectomy as done for the patient presented is one of the common surgical approach for imperforate hymen. Cruciate incision on the hymen along its diagonal diameters is done. This avoids injury to the urethra and can be

enlarged by removal of excess hymenal tissue. To prevent scarring and stenosis, the hymenal tissues is excised too close to the vaginal mucosa.[11] The hymenal ring is then evaginated by suturing it to the vaginal epithelium using absorbable sutures to secure haemostasis as was done to this patient.

Conclusion

Acute urinary retention due to haematocolpometrosalpinx in young females is rare. Familial occurrence of imperforate hymen is also rare. With good history and examination especially at birth, early diagnosis and treatment can be instituted. This will prevent complications in the young girls and anxiety in their parents.

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