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Novel approach to vaginal calculus in a girl with urogenital sinus anomaly

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ABSTRACT

Isolated urogenital sinus can cause distended bladder and/or vagina and may present with an abdominal mass and sepsis during infancy. Older children may present with recurrent urinary tract infections and hematocolpos. We describe a 3-year-old girl with recurrent urinary tract infections thought to be secondary to vesicoureteric reflux. On further investigation, an isolated urogenital sinus anomaly with a calculus inside one of the hemivaginae was noted. She was managed expectantly with a plan to intervene at puberty. At puberty, during removal of the stone, the hemivaginal introitus was found to be stenotic. Gradually increasing sizes of Amplatz type graduated renal dilators were introduced from the introitus of the urogenital sinus into the hemivaginal stone until a size 22F Amplatz sheath could be passed easily. Size 10F cystoscope was passed through this channel, and the stone was fragmented using electrohydraulic lithotripsy. At a later date, she underwent staged anterior sagittal transvulval mobilization of the urogenital sinus.

Primary vaginal stones are reported in association with vesicovaginal and urethrovaginal fistulae and, in conditions causing narrowing of the vaginal introitus [1-4] Urinary stasis and bacterial proliferation appear to contribute to the formation of calculi [5,6]. There are only 4 case reports describing primary vaginal stones in the pediatric literature [6-8].

In urogenital sinus (UGS) anomaly, urine refluxing from the common channel into the vagina tends to stagnate predisposing to infection. Isolated UGS anomaly may present with an abdominal mass and sepsis [6] in infancy and with recurrent urinary tract infections (UTI) or urinary incontinence in childhood. UGS anomaly may be associated with Mullerian duct anomalies including duplication of uterus and vagina [9].

We describe a 3-year-old girl with an isolated UGS anomaly presenting with recurrent UTI. These episodes were initially thought to be secondary to vesicoureteric reflux. On further investigation, duplication of uterus and vagina with a stone in one of the hemivaginae was identified. To our knowledge this is the first report of a vaginal calculus associated with UGS anomaly.

1. Case report

A 3-year-old girl was referred from a local hospital with a calculus identified on an ultrasound scan (USS) thought to be in a bladder diverticulum.

She had previously presented with a UTI at the age of two months. Ultrasound scan had identified a horseshoe kidney and a micturating cystourethrogram had shown bilateral grade II reflux. Conservative expectant management of reflux had been started. More recently USS now suggested a calculus possibly in a bladder diverticulum.

There was no obvious urogenital abnormality on examination and further investigations at our institution included cystovaginoscopy and examination under anesthesia. This showed a urogenital sinus anomaly with 2-separate hemivaginas and a common channel measuring approximately 2.5 cm. An 8-mm-sized calculus was identified in the right hemivagina. The bladder and both ureteric orifices were otherwise normal. It was decided not to intervene at that stage but to investigate her pathology further. Further investigations confirmed female karyotype and excluded any endocrinopathy. Urinary tract infections responded to antibiotic prophylaxis. A decision was made to manage the vaginal calculus expectantly with a plan for vaginoplasty and stone removal at puberty.

She was, however, a poor clinic attendee but remained well until 12 years of age. She then represented with recurrent abdominal pain. Ultrasound scan and magnetic resonance imaging scan now showed

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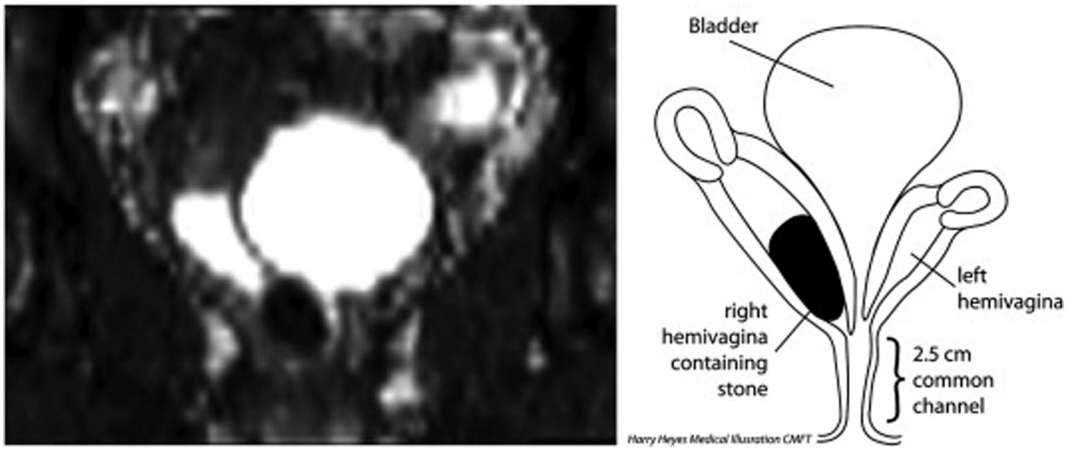

Fig. 1. Representative CT scan and schematic diagram of patient's anatomy.

distended right hemiuterus and hemivagina with a 14×8 mm size calculus in the right hemivagina (Fig. 1).

The common channel was examined using a 10F cystoscope but could not be passed beyond the introitus of the right hemivagina. A guidewire was therefore passed and dilatated with graduated Amplatz type "renal" dilators until it was possible to insert a 22F sheath. A 10F cystoscope could then be passed to allow lithotripsy. The stone was duly fragmented by electrohydraulic lithotripsy as an ultrasonic lithotripter was not available. All stone fragments were removed and confirmed by post-operative endoscopy 2 days after surgery.

Subsequently, after undergoing widening and dilation of each hemivaginal orifice, she underwent staged anterior sagittal transvulval mobilization of the urogenital sinus [10]. She is currently well except for occasional episodes of daytime wetting.

2. Discussion

Vaginal stones usually present with symptoms of vaginal bleeding, discharge or abdominal/perineal pain. Primary vaginal stones in the pediatric population are rare and to our knowledge only 4 cases have been previously reported. Eton et al [7] reported a case of a 7-year-old girl who presented with a vaginal calculus associated with an ectopic ureter opening into the vagina. Bissaad and Hanash [8] reported a large vaginal stone in a 12-year-old girl with urinary incontinence due to neuropathic bladder and urinary incontinence. Plaire et al [6] described 2 cases: a 4-year-old girl with bladder exstrophy who had an asymptomatic stone in vagina and a 13-year-old girl who had undergone previous vaginoplasty and developed narrowing of the introitus.

We are not aware of any reports of vaginal calculi associated with an isolated UGS anomaly. In our case, the long length of the narrow UGS channel leading to the hemivaginal cavity made the surgical procedure complex and, hence, affected the decision regarding timing of the surgery.

Reconstructive surgery and removal of the calculus was postponed until puberty according to our surgical practice of the time favoring late reconstruction of UGS. It is likely, however, that earlier removal of the calculus would have avoided the later complications due to the increased size of the stone.

Endoscopic intracorporeal lithotripsy was completed successfully by using Amplatz type graduated renal dilators allowing introduction of a size 22F Amplatz sheath. Although we were satisfied with electrohydraulic lithotripsy we feel that an ultrasonic lithotripter would probably have been a better mode for the lithotripsy as evacuation of stone fragments would have been easier.

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