A persistent urethral diverticulum in pregnancy: Case report and review of the literature

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ABSTRACT

Objective: A urethral diverticulum (UD) is a localized evagination of the urethral mucosa that forms a herniation that is continuous with the lumen of the urethra. We report the case of a patient who presented with a persistent UD during consecutive pregnancies and present a review of the current literature of other cases of UD during pregnancy.

Methods: A literature search was undertaken to identify previous research and case reports on the clinical presentation and management of UD in pregnancy using the search terms "pregnancy" and "urethral diverticulum". Medline, PubMed and Cinahl were used as search engines.

Results: Six publications that described UD in pregnancy were identified with a total of nine cases documented within the literature. The articles reviewed showed that UD during pregnancy can be managed conservatively with expectant management, antibiotics and incision and drainage if required.

Conclusion: Overall, it is important for clinicians treating women in pregnancy to be aware of the rare diagnosis of UD, especially in those women who present with vague urinary symptoms refractory to other treatments.

Brief Summary: A case presentation of UD and literature review of UD in pregnancy.

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1. Introduction

A urethral diverticulum (UD) is a localized evagination of the urethral mucosa that forms a herniation that is continuous with the lumen of the urethra [1]. We report the case of a patient who presented with a persistent UD during three consecutive pregnancies. A literature review was undertaken and highlighted that there is no consensus on the initial investigations or treatment. Nevertheless, the articles reviewed showed that UD during pregnancy can be managed conservatively with expectant management, antibiotics, and incision and drainage if required. Multiple journal articles advise against diverticulectomy during pregnancy [2].

During her next pregnancy, the patient presented to the hospital at 26 weeks of gestation. A 1 x 1 cm mass within the anterior vagina was causing discomfort and urinary incontinence. The mass was tender and reducible, and the patient was managed with analgesia. At 39 weeks of gestation, the patient presented to hospital as the cystic lesion was causing increasing pain and had grown to 3 x 3 cm. A bedside needle aspiration was performed for symptomatic relief, and a total of 15 mL of cream-coloured fluid was aspirated. The patient experienced immediate symptomatic relief; however, the cyst reaccumulated over the course of a day. A pelvic ultrasound showed a thick-walled cystic lesion along the anterior wall of the vagina, located between the urethra and the vagina and measuring 22 x 14 x 20 mm, with some echogenic sludge but with no vascularity seen. The pain continued, with consequent difficulty for the patient in walking and sitting. She underwent a spontaneous vaginal delivery at term with no complications. Following the patient's second pregnancy she was referred to a urologist for further treatment and investigation of her UD. The patient, however, developed severe renal colic and multiple renal stones, which were treated with a lithotripsy and ureteric stents, and her UD was never further treated until her next pregnancy.

During the patient's third pregnancy the mass again increased in size and caused pain. A decision was made to continue with conservative management and undertake surgical management following pregnancy. This pregnancy was complicated by renal colic. A repeat kidney, urinary, bladder ultrasound showed three renal stones.

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The patient had an uncomplicated term vaginal delivery and was reviewed in the hospital's gynaecology department 11 weeks later. On this post-partum review, the patient could feel the diverticulum but with no associated pain or dysuria. She reported difficulty with sexual intercourse due to pain. Examination demonstrated a urethral diverticulum posterior to the urethral meatus. It was approximately 1 x 1 cm and was soft and fluctuant to palpate. The patient subsequently underwent an excision of the UD five months post-partum. The operation was uncomplicated, and the intraoperative findings showed a urethral diverticulum opening into the posterior urethra close to the vesicoureteric junction. The patient was discharged home with an indwelling catheter and subsequently passed her trial of void. The patient was reviewed 2 months following the operation and there were no signs or symptoms suggestive of a fistula and the patient had recovered well.

3. Discussion

UD is an uncommon condition within the general gynaecological population, and even more uncommon within the pregnant population. It is described as a localised evagination of the urethral mucosa that forms a sac that is continuous with the lumen of the urethra [1,3]. The prevalence ranges from 0.6% to 6% in women of reproductive age; the incidence within pregnancy is unknown, but likely even lower [1,4,5]. The pathophysiology of acquired diverticula is unknown. One theory suggests that they may originate from repeated urinary tract infections obstructing the periurethral (Skene’s) glands, resulting in the obstruction rupturing into the urethral lumen, creating local herniation [2]. The main infective agents proposed include Gonococcus, Chlamydia and Escherichia coli [5]. Another theory suggests that once an obstructed periurethral gland becomes infected it causes an abscess that eventually ruptures into the urethral lumen, causing an ostium [6].

In the case reported above, in all three of the patient’s pregnancies she presented with a genital mass that was subsequently diagnosed as a UD. She nevertheless proceeded to have normal vaginal deliveries at term.

UDs are often difficult to diagnose. The literature documents a classic triad of dyspareunia, dysuria and urinary incontinence. Most patients with UD present with vague urinary symptoms or pelvic pain, often refractory to treatment. Other symptoms that may be associated with diverticula are dyspareunia, urethral discharge and urinary dribble. The differential diagnoses include Gartner duct cyst, vaginal wall inclusion cyst, ectopic ureterocoele, Skene gland abscess and urethral carcinoma or adenoma [3]. Complications of UD include stone formation, recurrent urinary tract infections and, rarely, malignancy [5].

Just as there is no consensus on the correct treatment, the initial investigations recommended within the literature also vary. It is recommended that a mid-stream urine specimen and urethral swab is taken in all suspected cases. UD should be considered whenever a patient exhibits non-specific, unresolved urinary problems. High-resolution magnetic resonance imaging and videourodynamic are the best modalities for investigating UD [5,6].

The definitive treatment of UD is a complete transvaginal excision, which can be complex and difficult surgery. Firstly, the tissues within the region are highly vascular and often the full extent of the herniation can be hard to appreciate intraoperatively. Multiple journal articles advise that diverticulectomy during pregnancy is unwise [2]. Complications of this surgery include urethral stenosis, stress incontinence, urethra-vaginal fistula and incomplete excision [2]. Alternative surgical management includes endoscopic deroofing to create a wide-mouth diverticulum and incision and drainage, although this carries a higher chance of re-accumulation [5]. Conservative management includes simple analgesia, antibiotics and fine-needle aspiration [3].

The treating clinician needs to be aware of the potential complication of obstruction during the second stage of labour with a large diverticulum [1]. Carswell et al. hypothesise that a cyst may be aspirated to allow for a vaginal delivery, depending on the diameter and location of the diverticulum [5]. They suggest that when the foetus passes through the true pelvis, a proximal periurethral diverticulum would be compressed anteriorly against the pelvic pubic symphysis, causing urinary tract rupture or dystocia, in which case cyst drainage may be beneficial for labour [1]. However, a distal periurethral diverticulum would be less likely to obstruct the labour passage due to the softer surrounding tissues, suggesting that drainage would not be required.

A literature search was undertaken to identify previous research and case reports on the clinical presentation and management of UD in pregnancy using the search terms “pregnancy” and “urethral diverticulum”. Medline, PubMed and Cinahl were searched, with no limits on the years of publication, although the articles were limited to those in the English language. Six publications that described UD in pregnancy were identified, with a total of nine cases documented, with one patient having recurrence of her UD during a subsequent pregnancy [1–6].

Of these cases, two patients received no treatment during their pregnancy and two patients were commenced on oral antibiotics alone [2,3]. A further two patients started antibiotics and also had their UD aspirated during the pregnancy [1,2]. One patient had multiple cystoscopic drainages and self-catheterisation during her pregnancy [5]. Lastly, two patients underwent a diverticulectomy during their pregnancy [4,6]. Of the seven patients who did not undergo a diverticulectomy during their pregnancy, all underwent diverticulectomy, either at the time of a caesarean section or at varying times post-partum. Four patients had an uncomplicated delivery, three patients had an elective caesarean section (either due to concerns regarding the size of the UD or due to an alternative indication such as breech presentation) and one patient underwent an emergency caesarean section for failure to progress.

4. Conclusion

In the articles reviewed there is a consensus was that UD during pregnancy can be managed conservatively with expectant management, antibiotics, and incision and drainage if required [6]. The definitive treatment of UD is a diverticulectomy; however, during pregnancy this operation is not without its risks [2]. A cystourethroscopy is an essential part of the evaluation at the time or prior to diverticulectomy [2].

Overall, it is important for physicians to be aware of the rare but documented diagnosis of UD, especially in those women who present with vague urinary symptoms refractory to other treatments.

Contributors

All authors contributed equally to the preparation of this case report and saw and approved the final manuscript.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

The patient described in this case study provided written informed consent. The report presented here was reviewed by the chair of the relevant ethics committee and deemed to constitute a case study that does not require further ethical review.

Provenance and Peer Review

This case report was peer reviewed.
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