Case Report

The Dangers of Hymenotomy for Imperforate Hymen: A Case of Iatrogenic Pelvic Inflammatory Disease with Pyosalpinx

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A B S T R A C T

Background: Complications associated with imperforate hymen include cyclical abdominal pain, acute urinary retention, endometriosis, and even iatrogenic infections.

Case: A 14-year-old young woman was diagnosed with an imperforate hymen, hematocolpos, and right hematosalpinx. A hymenotomy was performed, followed by a hymenectomy 3 days later. On postoperative day 7, she was admitted for pelvic inflammatory disease with a right pyosalpinx. The infection was refractory to intravenous gentamicin, ampicillin, and clindamycin so the patient underwent computed tomography-guided drainage of the pyosalpinx. Two days later, she was discharged home in good condition.

Summary and Conclusion: Small incisions and punctures into imperforate hymens without immediate definitive management should be avoided because inoculation of the newly introduced bacteria can ascend the gynecologic tract and lead to serious infections.

Key Words: Adolescent, Amenorrhea, Hematocolpos, Hymen/imperforate, Pelvic inflammatory disease, Salpingitis, Vagina/abnormalities

Introduction

Imperforate hymen is likely the most common obstructive anomaly of the female reproductive tract. Embryologically, the hymenal membrane separates the vaginal lumen and the urogenital sinus, then ruptures before birth. An imperforate hymen results from a failure of the urogenital sinus to canalize. Ideally, the diagnosis should be made during the newborn period, but many adolescents initially present with primary amenorrhea, cyclical abdominal pain, and a blue bulging hymen.[1]

Complications associated with imperforate hymen include acute urinary retention secondary to a hematocolpos mass effect[1–3] and endometriosis from retrograde menstruation.[4,5] In 1 case, a female adolescent presented with an acute abdomen secondary to a ruptured hematosalpinx from her imperforate hymen.[6] Infants can present with hydrocolpos and mucocolpos secondary to increased vaginal secretions from maternal estrogen stimulation. The mucus typical resorbs, but unusual cases of pyocolpos in infants have been reported.[7,8] We present, to our knowledge, the first report of iatrogenic pelvic inflammatory disease (PID) in a female adolescent with an imperforate hymen.

Case

A healthy 14-year-old virginal girl presented to her pediatrician for a 6-month history of constipation associated with abdominal pain. The patient eventually disclosed a bulge in her vagina and was referred to a gynecologist. The gynecologist elicited a history of primary amenorrhea and noted a large blue bulging membrane at the level of the hymen. An office hymenotomy was performed for symptomatic relief of her hematocolpos. The hymen was cleansed with betadine, and a sterile scalpel was used to create a 5-mm incision and drain a large amount of chocolate-like fluid. The patient was then referred to an adolescent gynecologist for further management.

The following day, the patient was seen by an adolescent gynecologist who elicited a history of thelarche at age 11 and adrenarche at age 12. On exam, an intact bulging membrane was noted again. The previous incision site was unable to be visualized because of spontaneous resealing. A transabdominal pelvic ultrasound examination revealed a thin obstructing structure at the introitus with 350 cc of hematocolpos and a $7 \times 3 \times 3$ cm right hydrosalpinx (Figs. 1 and 2). Ultrasound examination findings were consistent with an imperforate hymen, unlikely a low transverse vaginal septum or Müllerian agenesis of the lower vagina. Definitive surgical management was declared urgent because of the patient's stability and absence of active infection, and insurance preauthorization was immediately submitted.

Two days later, the patient was brought to the operating room for a hymenectomy. Doxycycline 100 mg was administered prophylactically. A cruciate hymenal incision was made with a scalpel, draining 150 cc of hematocolpos. The freed apexes of the incision were sutured in the cephalad direction to the vagina with 3-0 polyglactin in a simple interrupted fashion (Fig. 3).[1]

On postoperative day 7, the patient presented to the emergency department for fever, chills, and right lower quadrant abdominal pain. Her temperature was 104.2°F, blood pressure 124/64 mm Hg, and heart rate 143 beats per minute. The right lower quadrant of her abdomen was

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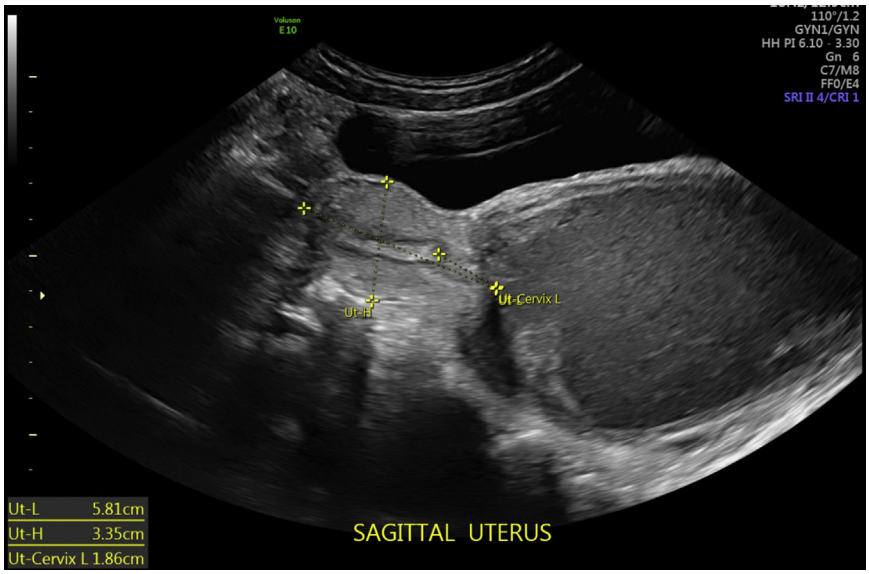

Fig. 1. Pelvic ultrasound status post hymenotomy, postprocedural day 1: 350 cc of hematocolpos. Ut-L, uterine length; Ut-H, uterine height; Cervix L, cervical length.

tender to palpation without guarding or rebound tenderness. Visual inspection of the hymenotomy confirmed patency. The patient was admitted for PID. White blood cell count was 15.8 with a left shift. Imaging was ordered to visualize the gynecologic tract, in addition to evaluation of the renal system. Magnetic resonance imaging was chosen to minimize radiation exposure and revealed an $8 \times 6 \times 7$ cm right pyosalpinx and normal renal system (Fig. 4). The patient's abdominal pain and leukocytosis improved with intravenous gentamicin, ampicillin, and clindamycin but she continued to have intermittent fever despite conservative management (Fig. 5). On hospital day 3, she underwent computed tomography-guided drainage of her pyosalpinx, and 10 cc of purulent material was evacuated. Fluid culture was negative for bacterial growth. The patient remained afebrile for more than 36 hours and was discharged home on hospital day 5 in good condition with a 14-day course of oral doxycycline 100 mg twice daily.

Three days after discharge, the patient was seen in the office for a follow-up visit. She had no complaints and denied any fevers, chills, or abdominal pain. The patient was instructed to complete her doxycycline and follow-up in 8 weeks with a repeat pelvic ultrasound to confirm resolution of the pyosalpinx.

Summary and Conclusion

To date, this case is the first report of iatrogenic PID secondary to hymenotomy. This was confirmed by a PubMed and Google Scholar literature search in English

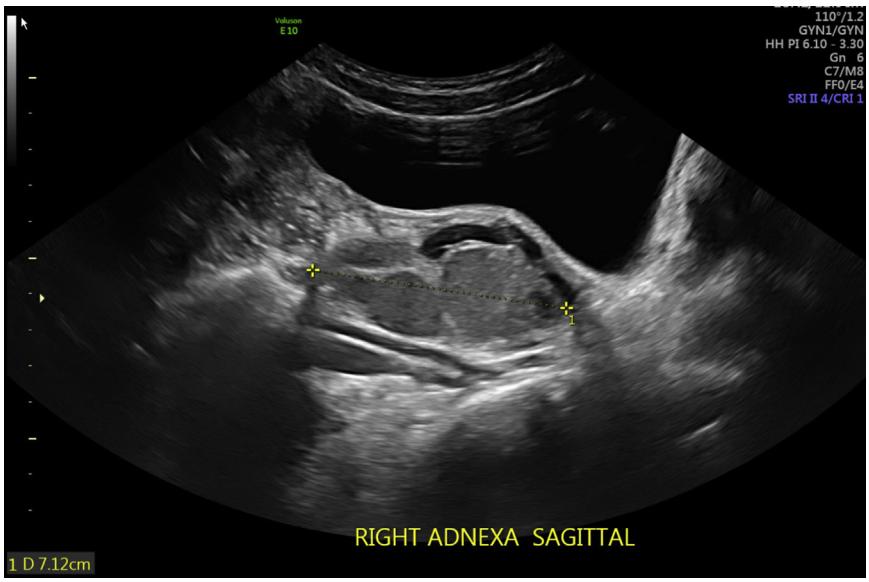

Fig. 2. Pelvic ultrasound status post hymenotomy, postprocedural day 1: $7 \times 3 \times 3$ cm right hydrosalpinx. 1 D. Ut-L, uterine length; Ut-H, uterine height; Cervix L, cervical length.

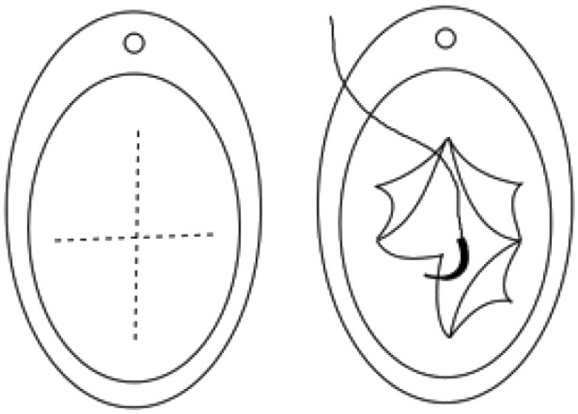

Fig. 3. Hymenectomy with a cruciate incision.

Fig. 4. Magnetic resonance imaging status post hymenectomy, postoperative day 7: 8 × 6 × 7 cm right pyosalpinx.

from the years 1988 to 2019. In 2001, Hung Lok et al reported a case of a female adolescent with iatrogenic pyocolpos secondary to needle puncture of an imperforate hymen.[9] These cases show the dangers associated with hymenotomy for imperforate hymen, even when performed under sterile conditions. Small incisions and punctures into the hymen without immediate definitive surgical management should be avoided. A small perforation does not allow the hematocolpos to fully drain because of the high viscosity of old blood, in addition to introducing bacteria into a previously sterile environment.[1] Hematocolpos, hematometra, and hematosalpinx provide nutrient-rich media for the inoculation of ascending bacteria.

In this case, definitive surgical management was delayed for insurance preauthorization by 3 days, which was enough time to develop a pyosalpinx. Definitive surgical management was deemed urgent because of the patient's stability and absence of active infection upon initial presentation. However, because of the patient's failed hymenotomy and presence of a hematosalpinx, there should have been strong consideration for definitive surgical management on the day of presentation. By declaring a procedure emergent, insurance preauthorization can be avoided. Clinicians should not hesitate to perform an emergent hymenectomy for a failed hymenotomy, even in a stable-appearing patient, because of the severity of the silently inoculating sequelae associated with delayed definitive management.

The most common method of definitively managing an imperforate hymen is hymenectomy. A cruciate or elliptical incision is created in the hymen with complete evacuation of the vaginal mucous or blood. Care should be taken to prevent painful scarring of the tissue and vaginal stenosis. Estrogen is a key component in the prevention of vaginal stenosis, so surgery should be deferred until puberty.[1] If surgery is required before puberty, the patient could receive supplementation of topical estrogen. Vaginal dilators can also be considered to promote healing and avoid stenosis. Other novel management methods with reported success include hymenal incision with placement of a Foley catheter for 2 weeks[10] and hymenal excision using a carbon dioxide laser.[11]

Prophylactic antibiotics are not routinely recommended for hymenectomy.[1] However, antibiotics could be considered for patients at high risk for infection, such as patients status post failed hymenotomy and with hematosalpinx. Adequate drainage of the hematosalpinx would be difficult to achieve with hymenectomy because of the indirect connection between the vagina and fallopian tube. No controlled trials have been performed on antibiotic use for imperforate hymen. With limited data available, doxycycline 100 mg twice daily could be considered because of its success in treating PID.[12]

The treatment of pyosalpinx ranges from conservative intravenous antibiotics and image-guided drainage to laparoscopic aspiration, salpingostomy, and salpingectomy.[13] Patients who fail to improve after 48-72 hours of antibiotic treatment require additional management. For hemodynamically stable patients, the authors prefer minimally invasive image-guided drainage because of its efficacy, low complication rate, and avoidance of surgery.[14] Pelvic infections are infamously challenging surgeries because the extensive inflammation causes anatomic distortion and friable tissues. Prompt surgical intervention by a skilled gynecologic surgeon is recommended if the patient is hemodynamically unstable, image-guided drainage is not feasible, fever and/or severe pain persist despite image-guided drainage, or malignancy is suspected.

An appropriate evaluation of the renal system should be considered because of the increased risk of renal injury and anomalies associated with imperforate hymens. A comprehensive systematic review of imperforate hymens showed that urinary retention and renal failure were present in 20% and 2% of imperforate hymens, respectively.[15] These complications are likely secondary to obstruction from an enlarged blood-filled uterus. Although most imperforate hymens result from isolated sporadic

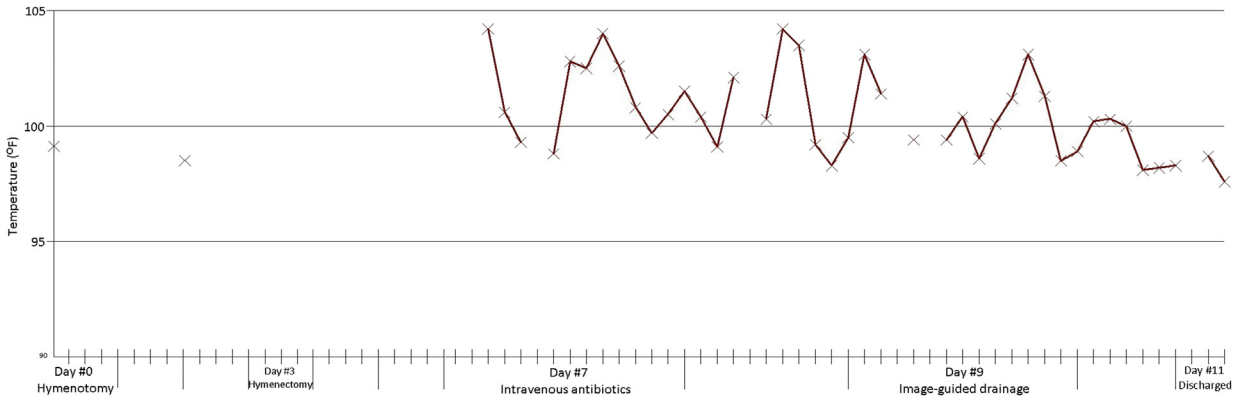

Fig. 5. Temperature curve.

mutations, persons with imperforate hymens are at increased risk of congenital renal anomalies including ipsilateral renal agenesis, bilateral duplex ureter, and ectopic ureter.[15] The rate of renal anomalies associated with imperforate hymens is less than that of other congenital gynecologic tract anomalies, such as Müllerian anomalies, but the rate is still higher than that of the general population. Therefore, evaluation of the renal system should be considered upon diagnosis of an imperforate hymen.

In conclusion, small incisions and punctures into an imperforate hymen without immediate definitive management should be avoided because inoculation of the newly introduced bacteria can ascend the gynecologic tract and lead to serious pelvic and intraabdominal infections. Although imperforate hymen is likely the most common obstructive anomaly of the female reproductive tract, a paucity of data exist on the management and complications of this condition, and the literature is limited to primarily case reports. Additional studies are needed on the management of imperforate hymen.

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