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Imperforate Hymen with Bicornuate Uterus in an Adolescent Patient

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Abstract

A thirteen-year-old patient presenting with chronic pain and amenorrhea was evaluated for ongoing discomfort and worsening symptoms. Pelvic examination showed a vaginal protrusion consistent with imperforate hymen and imaging was consistent with significant hematocolpos. Hymenectomy was performed and a bicornuate uterus was revealed in postoperative imaging. This case report adds to the medical literature exploring the diagnosis of imperforate hymens, which occur in 0.1% of female neonates and may co-occur with a bicornuate uterus (0.4% prevalence in females). It provides a perspective on the rarity of the incidence of imperforate hymen patients with another genitourinary anomaly. It also challenges current ideology regarding imperforate hymens not requiring additional workup to assess other congenital uterine anomalies.

Keywords

Imperforate Hymen, Hematocolpos, Bicornuate Uterus, Abdominal Pain, Back Pain, Amenorrhea, Hymenectomy, Vaginal Reconstruction, Case Report

1. Introduction

The case explores the presentation of an adolescent female with imperforate hymen and her subsequent hymenectomy for treatment. This patient is a unique case in that postoperative pelvic ultrasound revealed a secondary congenital anomaly of

a bicornuate uterus. Two uncommon anomalies occurring together create a unique case report and should raise suspicion for multiple Müllerian anomalies when one is identified.

2. Case Report

2.1. Initial presentation

A morbidly obese thirteen-year-old female presented to the emergency department for chronic abdominal and low back pain. She is accompanied by a family member who assisted in providing the patient's history. She reports that her symptoms have been constant for approximately a year, with intermittent episodes of worsening pain monthly. She was seen at another facility recently and diagnosed with a urinary tract infection. It was treated with antibiotics, but the patient had no improvement in her symptoms. The patient denied fever, chills, chest pain, palpitations, cough, dyspnea, dysuria, or headache. She is alert and in no apparent distress. She had a soft abdomen without distention. She reported suprapubic tenderness. She denied starting her menstrual cycle. Alternative causes of primary amenorrhea were initially considered, including pregnancy, hypothalamic dysfunction, and congenital reproductive tract anomalies. The presence of secondary sexual characteristics and cyclic pain increased suspicion for an outflow obstruction. Her urinary pregnancy test is negative. Her workup in the emergency department started with pelvic ultrasound, lumbar spine X-ray, and urinalysis. After an ultrasound showed an enlarged uterus, her examination expanded to include CMP, CBC, and CT of the abdomen and pelvis. A pelvic exam was performed with consent and revealed a bluish bulge at the opening of the vagina consistent with imperforate hymen. She did not tolerate vaginal exams well. The decision was made to treat with surgical excision and vaginal reconstruction, with risks and benefits discussed with patient and family members.

2.2. Clinical Findings

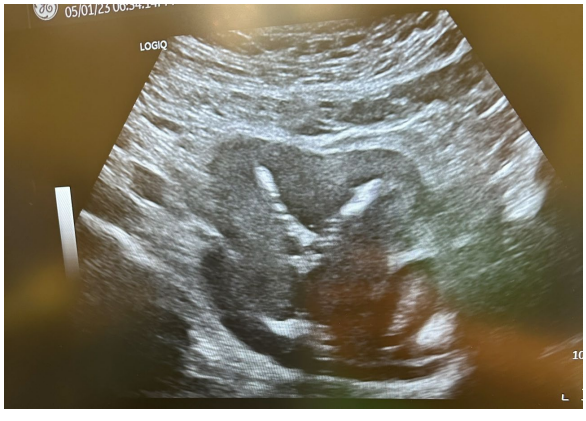

Figure 1. Ultrasonographic view of the pelvis.

The physical exam of the pelvis revealed a notable blue perineal mass in the open-

ing of the vagina consistent with an imperforate hymen. Lumbar spine X-ray showed no acute pathology. Urinalysis was negative for nitrates, leukocyte esterase, glucose, or blood. Pelvic ultrasound (**Figure 1**) revealed a 16 × 11 cm fluid-filled cystic structure in the pelvis and an anechoic cyst of the right adnexa. Due to limitations of body habitus and obscured visualization of the uterus and left ovary, the patient was sent for CT of the abdomen and pelvis with contrast. The CT scan revealed a 20 × 10 cm fluid-filled structure of a dilated uterine cavity and endocervical canal. Cystic changes in the right adnexa were confirmed, and mild bilateral hydronephrosis was reported.

2.3. Therapeutic Intervention

The patient was transferred to the operating room from the emergency department. General anesthesia was managed, and she was prepared according to usual sterile fashion. Her legs were placed in Allen-type stirrups. Betadine was used to cleanse the perineum and vulvar area. Allis clamps were used for manipulation and visualization of the operative field. The imperforate hymen was easily visualized. The bluish color indicated hematocolpos as seen in previous imaging. A Z-plasty was created using a #11 blade scalpel. Approximately 1050 cc of dark blood was immediately expressed and suctioned from the vaginal vault. It was then revealed that the patient had a significantly enlarged cervix, likely due to pressure from the hematocolpos. A weighted speculum was placed along with a right-angle clamp for better cervix visualization. A single-tooth tenaculum was used to manipulate the anterior cervix, and a sound dilated the cervical canal to 6 cm. Ultrasound of the abdomen and pelvis was performed to confirm that the blood was evacuated and to repeat measurements of the uterine size. The patient was revealed to have a bicornuate uterus with endometrial stripe in both horns. The uterus was now approximately 8 cm in size. Mild bilateral hydronephrosis was noted preoperatively due to ureteral compression by the dilated uterine cavity and endocervical canal. Following evacuation, the hydronephrosis resolved. This was confirmed by the normalization of follow-up blood urea nitrogen and creatinine levels, as well as the resolution of flank pain. Vaginal hymen reconstruction was performed with careful consideration to prevent regrowth of the hymen.

2.4. Follow-Up and Outcomes

The patient was reassessed with the family member present on postoperative day one. She was alert and smiling during follow-up. She denied any pain or difficulties with voiding. She was actively eating and drinking and reported minimal vaginal bleeding. The patient and family members were instructed on vaginal estrogen cream usage. She was discharged from the hospital on postoperative day two with instructions on continued estrogen cream usage and avoidance of tampons. She was followed up at the outpatient clinic approximately two weeks post-excision of imperforate hymen secondary to hematocolpos. She reported a minimal bleed for approximately one week after her surgery with no clots and no pain. She was

concerned about possible infection due to dysuria, itching, and vaginal discharge. A urine culture was ordered and showed no growth. On a modified pelvic exam, no sutures were noted. She did have a significant amount of yeast on the external genitalia and odorless vaginal discharge. She was prescribed Diflucan to address these concerns. Patient education was also extensive, with a review of proper hygiene practices, ovulation, and menstruation.

3. Discussion

Literature Review

An imperforate hymen is a congenital malformation of the female genital tract. It presents as a layer of squamous epithelium obstructing the vaginal introitus. Failure of degeneration leads to recognition in three main stages of life: in utero, newborn to childhood, and puberty [1]. When it is not recognized until puberty, as in our case, patients often present with cyclic abdominal pain and primary amenorrhea. In more severe cases, urinary tract obstruction, hydronephrosis, and subfertility may also occur [2]. 38% - 40% of patients experience low back pain due to referred pain from the sacral plexus and vaginal and uterine nerve roots [1]. Mou, Tang, Chan, Tam, and Lee examined three cases of imperforate hymens in adolescent females. All three presented with similar symptoms of lower abdominal pain and multiple physician visits. Cases 1 and 2 both had the distinct "bluish, bulging" imperforate hymens on perineal examination. Case 2 had additional symptoms of tenesmus and dysuria. Case 3 lacked the typical physical exam findings due to "a large pelvic mass" revealed to be a labial adhesion. Examination under anesthesia was necessary to reveal the imperforate hymen above the adhesion. Cases 1 and 2 underwent hymenectomies and maintained patency and normal menses on follow-up, while case 3 had a mini-hymenectomy with 6-month follow-up showing recurrence and another hymenectomy was performed. The mini hymenectomy, or simple cruciate incision of the hymen without excision of membrane, was completed as an attempt to preserve cultural significance of a first postcoital bleed [3].

Kahn *et al.* (1975) reported an incidence of about 0.1% of imperforate hymen cases in female neonates [4]. Chan *et al.* (2011) reviewed congenital uterine anomalies and reported a 0.4% prevalence of a bicornuate uterus in the general population [5]. Generally, imperforate hymen is not associated with concurrent urogenital abnormalities. However, our case and other cases in the literature may encourage reevaluating this ideology. A 2012 case presented an eight-month-old female with restlessness, fever, and abdominal mass [6]. MRI showed significant hydrometrocolpos secondary to imperforate hymen. After cruciate incision and mucoid secretion drainage, repeated ultrasounds were performed over six months to confirm resolution of bilateral hydroureteronephrosis and the incidental finding of bicornuate uterus was discovered [6]. Similarly, Jain, Gupta, Kumar, and Minj described a case of a twelve-year-old girl who presented with one year of pelvic pain

and cramping without menstruation. Examination revealed a blind vaginal pouch and imaging confirmed additional abnormalities of a transverse septate and bicornuate uterus [7]. Levsky and Mondshine were very direct in their case report about investigating uterine malformations. Their case of a fourteen-year-old female is most similar to our case. Their patient presented with a history of urinary tract infection diagnosis and a recent right-sided adnexal mass. A large, circumscribed mass was found on physical examination and imaging confirmed hematocolpos. After hymenotomy, repeat ultrasound revealed a bicornuate uterus [8]. These cases all expressed concerns about imperforate hymens accompanying uterine anomalies. They also recommended additional consideration for concomitant congenital uterine conditions and imaging to assess for anomalies, especially in consideration of long-term fertility [6]-[9].

4. Conclusions

Fertility was addressed with the patient in light of her bicornuate uterus. Now that the hymen has been surgically corrected, she is able to monitor her menstrual cycles. Given the potential implications, research evaluating the relationship between bicornuate uterus and fertility should be explored.

Although hematocolpos is rare in practice, it can be associated with other abnormalities, such as bilateral hydroureteronephrosis, blind vaginal pouch, transverse septate, bicornuate uterus, uterus didelphys [10], and renal agenesis. Awareness of these potential abnormalities is vital to providing a comprehensive evaluation and the appropriate treatment.

In similar cases, we recommend obtaining an intraoperative ultrasound to ensure complete evacuation of retained blood. This should be followed by a CT urogram and MRI of the abdomen and pelvis to assess for normal renal and ureteral anatomy. Currently, there are no established guidelines recommending routine postoperative imaging following hymenectomy. Most professional guidance, including statements from ACOG and ASRM, emphasizes the importance of thorough preoperative imaging when Müllerian anomalies are suspected, as these are frequently associated with renal tract malformations. Pelvic ultrasound and MRI are the primary modalities used in the preoperative setting. However, postoperative imaging may be considered on a case-by-case basis when the anatomy remains unclear or if complications are encountered [11].

This report is limited by its single-case design, which restricts the generalizability of the findings and clinical recommendations. Further large-scale studies are needed to strengthen the evidence base guiding management of imperforate hymen and associated anomalies.

Declaration

Dr. Cheau Williams serves as a surgical consultant for Coloplast in the field of urology. No sources of support were received for this study.

Ethics Statement

This case report was deemed exempt from the Institutional Review Board (IRB) review according to Colquitt Regional's policy. Oral informed consent for publication was obtained from the patient's legal guardian, and assent was obtained from the patient.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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