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Novel use of a tracheobronchial stent in a patient with uterine didelphys and obstructed hemivagina

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Abstract

Objective: To describe a novel use for a tracheobronchial stent to maintain patency after vaginal septum excision in a patient with an obstructed hemivagina and uterine didelphys.

Design: Description of a novel technique

Setting: University-affiliated Children's Hospital

Patient: One patient with an obstructed hemivagina and uterine didelphys who presented with hematometria and hematocolpos.

Intervention: In order to maintain patency and decrease stenosis risk after vaginal septum excision, a coated tracheobronchial stent was deployed and left in place for 6 weeks.

Main Outcome Measure: To evaluate ease of stent placement and removal, re-epithelialization and patency of the neo-vagina, and post-operative assessment of pain and recurrent obstruction.

Results: The tracheobroncheal stent was easily positioned and deployed with vaginoscopic guidance. Six weeks later it was removed without any tissue in-growth or granulation tissue noted. The vaginal walls were nicely epithelialized. Twelve months post-operatively the patient remained pain free with regular cycles and no evidence of obstruction or abnormality on ultrasound.

Conclusion: Use of a coated tracheobronchial stent to maintain patency after septum excision in a patient with an obstructed hemivagina presents a safe, easy and effective option to diminish stenosis risk and avoid infectious complications or hysterectomy.

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Capsule:

Use of a tracheobronchial stent to maintain patency after septum excision in a patient with an obstructed hemivagina and uterine didelphys is a safe, easy and effective procedure.

Keywords

Obstructed hemivagina; Uterine didelphys; Stenosis after vaginal septum excision; Hematocolpos; Müllerian anomaly; Herlyn-Werner-Wunderlich syndrome (HWWS); Obstructed hemivagina and ipsilateral renal anomaly (OHVIRA); Stent

Introduction

The processes of lateral fusion, vertical fusion, and resorption are all intricately involved in the creation of the uterus and vagina, beginning in the early embryo. Furthermore, the close relationship between the urinary and reproductive systems (müllerian and metanephric ducts) leads to the coexistence of anomalies in the renal collecting system and the reproductive tract. Failure of fusion of the müllerian ducts can result in a uterine didelphys in some cases and further failure of canalization or resorption in the uterovaginal canal or urogenital sinus can lead to a vaginal septum (1, 2). Specifically, the combination of uterine didelphys, obstructed hemivagina and ipsilateral renal agenesis is a variant of the broad spectrum of müllerian anomalies that is being diagnosed and reported more frequently with increasing awareness and advances in radiologic imaging. This variant has been referred to as the Herlyn-Werner-Wunderlich syndrome (HWWS) as well as the obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome in the literature (3, 4, 5, 6). Young adolescents with this disorder usually present with abdominal pain shortly after menarche.

Excision of the vaginal septum is the treatment of choice, usually through a single-stage surgery, in order to drain the hematometrocolpos. Many surgeons wait to perform the vaginal septum resection until a large hematocolpos builds up to distend and thin the septal tissue and allow for an easier excision. In these patients, the bulging hemivagina is easily seen and palpated. If the vaginal septum is fully excised, the outflow track is likely to remain patent and normal function of the ipsilateral uterus will often resume. If the septum is inadequately excised, re-obstruction may occur, as well as recurrence of hematometra and/or hematocolpos. Yet, if the hematometra distends the uterus significantly while awaiting surgery or it continues to recur, uterine function may ultimately be compromised (7). If the patient's symptoms warrant intervention before a bulging mass is presenting low in the vagina or the obstructed hemivagina is located high in the pelvis, excision of the vaginal septum can be challenging. The high location of a thick septum, the small caliber of the adolescent vagina, and the intact hymenal tissue contribute to the surgical difficulty. The hematocolpos may be located very close to the cervix of the unobstructed uterine corpus. Given the difficulty of an adequate septum resection and the collapsed nature of the vaginal walls, post-operative vaginal stenosis of the septal orifice is a significant possibility. Even high transverse septae across the entire vagina without associated uterine anomalies have been reported to have risks of contracture and stenosis within weeks to months postoperatively. Some surgeons encourage specific operative techniques, such as a "Z"-plasty, as well as pre- and/or postoperative vaginal dilation to reduce contracture or stenosis risks (8, 9). More recently, some have even suggested use of a vaginal mold secured to an elastic belt, used while sleeping, or a Penrose drain (10, 11). Dilation or specific operative techniques become more difficult in the situation of small or high hemivaginal septae. We

present a specific case using a novel technique and instrumentation to maintain patency after septum excision and decrease the risk of stenosis and reoperation post-operatively.

Materials and Methods

CP is a 13-year-old G0, otherwise healthy young adolescent, who presented to her primary care physician with dysmenorrhea, abdominal and rectal pain. She had recently undergone menarche and had three uneventful periods. It was during her fourth menstrual cycle when she presented with pain. An ultrasound was performed and reported a “hematocolpos, possible imperforate hymen.” A local gynecologist, upon physical exam, recognized a more complicated müllerian anomaly. On pelvic MRI, two uterine bodies were identified. The right side had a normal endometrial cavity, a cervix and a single vagina. The left uterine horn was enlarged and hematometria and hematocolpos were noted. The ovaries were normal except for a simple left ovarian 3 cm cyst, and left renal agenesis was noted. The MRI imaging studies were forwarded to multiple consultants. The patient was given a single injection of a GnRH agonist to prevent another menses awaiting the recommendation of the consultants. Unfortunately, 2-3 weeks later the patient started another period and presented to the emergency room with intractable pain. She was treated with narcotics and eventually air-evacuated to St. Louis Children’s Hospital for consultation with the Pediatric Gynecology service. The next morning the patient was taken to the operating room where a distended obstructed hemivagina was not found by an exam under anesthesia. A single normal appearing cervix was seen. Upon bimanual examination induration was noted in the left vaginal fornix and a large left abdominopelvic mass was palpated. An 18 gauge spinal needle mounted on a saline filled syringe was directed into the left fornix and dark brown fluid returned. An initial incision was made and over 500 ml of old blood was drained. Ultimately, a 2 cm³ section of vaginal septal tissue was removed, and dilation of the neo-vaginal opening was attempted. Despite resection of as much tissue possible, the opening remained significantly narrow and could not be extended due to apposition of the right cervix and left pelvic sidewall. Following the vaginal procedure, a diagnostic laparoscopy was performed to evaluate persistence of the pelvic mass. The findings included widely divergent uterine horns, an enlarged 10 cm left ovarian cyst, which spontaneously drained clear fluid on manipulation, and no evidence of pelvic endometriosis. At the conclusion of the surgery, vaginal packing with estrogen cream was left in the neo-vaginal opening at the site of the septum resection and the patient was awakened.

Although the patient was pain free, the attending surgeon was concerned that the newly drained hemivagina and hematocolpos were at risk of obstruction, reaccumulation of menses and potential infection if the opening did not remain patent. The family was informed of the concern, and a plan to re-examine the patient was made to place a stent to maintain patency. The patient returned to the operating room three days later, and an 18 × 40 mm AERO™ tracheobronchial stent (Alveolus, Inc.) was deployed with vaginoscopic guidance (see Figures 1, 2, 3) after consultation with the cardiothoracic service. This metal and silicone stent was chosen due to its biologically inert covering that reduces granulation and tissue in-growth. Furthermore, its design allows easy deployment and repositioning once released. It radially expands to resist compression, and is removable and radiopaque. The procedure itself was very brief and the patient was discharged the following day in stable condition

without complaints. The patient was counseled to contact the pediatric gynecology service for fever or pain. The stent was to remain in place for six weeks to allow for reepithelialization of the vaginal walls and healing.

Results

Six weeks later the patient returned to the operating room for stent removal. At this time vaginoscopy and diagnostic hysteroscopy were performed, documenting the normal (right) cervix and a patent well epithelialized tract leading to the left uterine horn. The coated stent was easily removed.

Three months and two menstrual cycles after stent removal a pelvic ultrasound was performed. The two uterine horns were normal size with normal endometrial thickness, and no obstruction was noted. One year later the patient is doing well and has no complaints.

Discussion

Surgical management of obstructing vaginal septae varies in the degree of difficulty. A distended low lying vaginal septum is relatively easy to resect and should remain patent if an adequate septal resection was accomplished. Mid-to-high vaginal transverse septae or high obstructing hemivaginal septae present challenging operative cases. The thick septae, narrowed area of resection, collapsing sidewalls, and high location increase the post-operative risk of contracture, stenosis, infection and need for reoperation (10). Vaginal packing is only a brief post-operative solution and stenosis can occur weeks to months after the initial operation. Vaginal dilators and molds are effective but are more difficult to place correctly on an intermittent basis and often very uncomfortable for the patient.

Use of this stent allowed a young adolescent to return to normal activity while healing and re-epithelialization occurred without stenosis of the narrowed area. The silicone coating of the stent allows easy placement and removal without tissue in-growth or granulation. Menstrual flow continues through the hollow lumen of the stent and the radial expansion of the device secures it in place. Based on this experience, we would recommend placement of such a stent at the conclusion of a procedure where patency of a drained viscus is at risk for stenosis. Stents are used in any lumen to maintain patency and prevent stricture in cardiovascular, pulmonary, urinary or gastrointestinal procedures. This case represents the first use of a vascular stent to maintain patency of an obstructed uterus and vagina.

Further study and use is necessary to validate this surgical technique and off-label use of this device. Yet, it presents a safe, effective, and well-tolerated option for these difficult surgeries. Improving surgical outcomes in these young adolescents and reducing post-operative complications is valuable. As with any treatment or use of a new device, long-term follow-up is required.

References

1. Laufer, MR.; Goldstein, DP.; Hendren, WH.; Emans, SJ.; Laufer, MR.; Goldstein, DP. Pediatric & Adolescent Gynecology. 2nd. Lippincott Williams & Wilkins; Philadelphia: 2005. Structural abnormalities of the female reproductive tract; p. 334-416.

2. Sadler, TW.; Langman, J.; Leland, J., editors. Langman's Medical Embryology. 8th. Lippincott Williams & Wilkins; Philadelphia: 2006. p. 229-256.
3. Smith NA, Laufer MR. Obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome: management and follow-up. *Fertil Steril*. 2007; 87:918–22. [PubMed: 17320871]
4. Orazi C, Lucchetti C, Schingo P, Marchetti P, Ferro F. Herlyn-Werner-Wunderlich syndrome: uterus didelphys, blind hemivagina and ipsilateral renal agenesis. Sonographic and MR findings in 11 cases. *Pediatr Radiol*. 2007; 37:657–65.
5. Gholoum S, Puligandla P, Hui T, Su W, Quiros E, Laberge J. Management and outcome of patients with combined vaginal septum, bifid uterus, and ipsilateral renal agenesis (Herlyn-Werner-Wunderlich syndrome). *J Pediatr Surg*. 2006; 41:987–92. [PubMed: 16677898]
6. Nguyen T, Lawrence L, Ratts V, Odem R, Siegel C, Merritt D. Diagnosis and management of didelphic uterus, obstructed hemivagina and ipsilateral renal agenesis (Herlyn-Werner-Wunderlich syndrome): The Washington University Experience. *Fertil Steril*. 2007; 88(1):S54.
7. Donnez O, Jadoul P, Squifflet J, Donnez J. Didelphic uterus and obstructed hemivaginal: recurrent hematometra in spite of appropriate classic surgical treatment. *Gynecol Obstet Invest*. 2007; 63:98–101. [PubMed: 17016051]
8. Wierrani F, Bodner K, Spangler B, Grunberger W. “Z”-plasty of the transverse vaginal septum using Garcia's procedure and the Grunberger modification. *Fertil Steril*. 2003; 79(3):608–12. [PubMed: 12620448]
9. Jurst BS, Rock JA. Preoperative dilation to facilitate repair of the high transverse vaginal septum. *Fertil Steril*. 1992; 57(6):1351–1. [PubMed: 1601164]
10. Lacy J, Correll G, Walmer D, Price T. Simple vaginal mold for use in the postoperative care of patients with a transverse vaginal septum. *Fertil Steril*. 2007; 87(5):1225–6. [PubMed: 17418828]
11. Patterson D, Mueller C, Strubel N, Rivera R, Ginsburg H, Nadler E. Laparoscopic neoos creation in an adolescent with uterus didelphys and obstructed hemivagina. *J Pediatr Surg*. 2006; 41(10):E19–22. [PubMed: 17011253]

Figure 1.

Deployment of the AERO™ (Alveolus, Inc.) tracheobronchial stent 3 days after initial left hemivaginal septum excision. The stent is inserted and deployed with a one-handed technique under vaginoscopic guidance.

Figure 2.

Vaginoscopic view of the tracheobronchial stent after deployment. Radial expansion keeps the stent securely in place and prevents stenosis of the area. The length of the stent extends from just caudal to the area of septal excision up to the level of the left cervix.

Figure 3.

View of the AERO™ (Alveolus, Inc.) stent after deployment outside of the patient. It has a biologically inert coating to prevent tissue in-growth and granulation.