Case Report

Barium stone formation in the vagina of a child with persistent cloaca following a distal colostogram

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Abstract

Persistent cloaca is a severe form of malformation of the anorectum. A distal colostogram is important in the planning of its treatment. We report herein a case of a girl who had barium stone formation within her vagina following a distal colostogram. Water soluble contrasts are recommended for this procedure.

Key words: Barium stones, distal colostogram, persistent cloaca, water soluble contrast

INTRODUCTION

Persistent cloaca is a complex congenital anomaly in which the rectum, vagina, and urethra are united into a common channel which presents externally as a single perineal opening which discharges feces and urine. This rare defect occurs in 1:250,000 neonates (Holschneider and Scharbatke, 2006). The anomaly is believed to occur very early in embryogenesis, that is, between the 3rd and 5th weeks of intrauterine life (Warne *et al*., 2011). Treatment of cloacas represents a significant technical and anatomical challenge with the goals that the patient should have urinary control, bowel control, sexual function, and obstetrical potential at maturity (Levitt and Peña, 2010; Begum *et al*., 2011).

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We report herein the challenges we faced in the management of a girl with persistent cloaca.

CASE REPORT

The patient, a 2-year-old girl, first presented to our unit at age 8 days on account of ambiguous external genitalia, and was diagnosed with persistent cloaca for which she had a diverting sigmoid colostomy 2 days later. She had a distal colostogram done at age 1 year with barium contrast. Due to financial constraints, her surgery was delayed for another 1 year. At subsequent follow-up visits after the colostogram, her parents complained that her urine was cloudy and that she often cried during micturition. There was no history of fever. A urine microscopy analysis yielded *Escherichia coli*, and the patient was placed on oral antibiotics (cefuroxime) to treat her urinary tract infection. At the definitive surgery, posterior sagittal ano-recto vagino-urethroplasty, she was noted to have a markedly distended vagina which was inadvertently ruptured during the dissection. The common channel of the anomaly measured about 4 cm. There was a high rectal implantation into the vagina. Four multifaceted whitish stones each measuring about 2 cm in its longest axis was found within the lumen of the vagina [Figure 1]. The stones were extracted, and the procedure completed. Her colostomy was closed 8 weeks later. Recovery was uneventful. The patient currently has good bowel control but often dribbles urine.

DISCUSSION

Persistent cloacae represent a wide spectrum of anomalies of fusion of the vagina, urethra, and rectum, creating a common channel that opens into a single perineal orifice at the position where the urethral opening is normally located (Levitt and Peña, 2010) [Figure 2].

These anomalies pose a challenge to the pediatric surgeon and makeup 13.6% of the cases of anorectal malformations (Shakya *et al*., 2008).

The length of the common channel measured endoscopically, has been used to categorize persistent cloacae into those with a short common channel (<3 cm), and those with a long common channel (>3 cm) who more commonly pose a difficult reconstructive challenge (Warne *et al*., 2011).

Pena *et al*. reported that 62% of their patients with persistent cloaca had a common channel < 3 cm as well as a significantly lower incidence of associated anomalies, while 38% had a common channel longer than 3 cm, with a more higher incidence of associated anomalies (Pena *et al*., 2004).

Our patient did not have an endoscopic evaluation of her common channel, as this facility is currently not available in our center. The length of her common channel as visualized during the corrective surgery was about 4 cm.

Associated anomalies in these patients may occur as part of the vertebral, anal, cardiac, tracheoesophageal, renal, and limb defects association. The most critical to recognize however are the urological abnormalities which include absent kidney, vesicoureteral reflux, horseshoe kidney, ectopic ureters, double ureters, hydronephrosis, and megaureters which may result from a vesicoureteral reflux or ureterovesical obstruction (Levitt and Peña, 2010). Different variations of anomalies of the rectum, vagina, and urethra have been described (Holschneider and Scharbatke, 2006). Our patient had a high rectal implantation into the posterior wall of the vagina [Figure 3].

Inability to empty is the main type of bladder malfunction seen in this abnormality, and this causes the patient to start dribbling urine as an overflow phenomenon after the urinary bladder is completely full (Levitt and Peña, 2010). Our patient was noted to have frequent episodes of dribbling of urine as reported by her parents on postcorrection follow-up visits.

The diagnosis of persistent cloaca is a clinical one made by identifying a single perineal orifice (Levitt and Peña, 2010). It is important that a careful perineal examination be done, as an incorrect diagnosis may lead to the repair of only the rectal component (Levitt and Peña, 2010; Shakya *et al*., 2008).

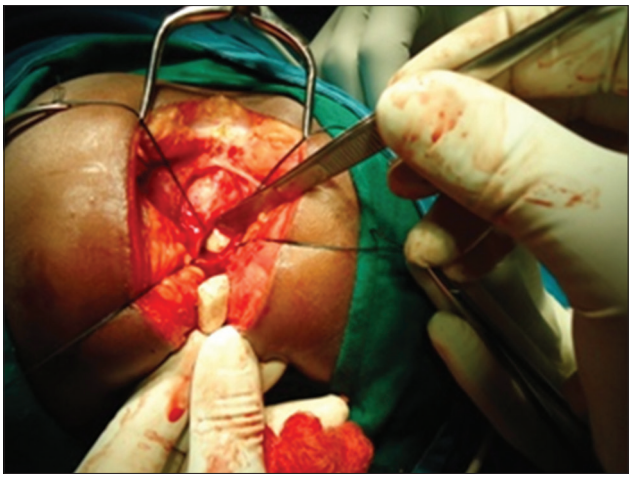

Figure 1: Barium stones found at surgery

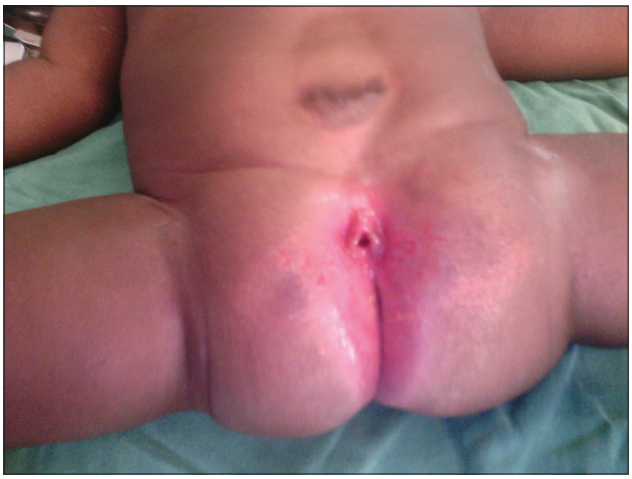

Figure 2: Child with persistent cloaca

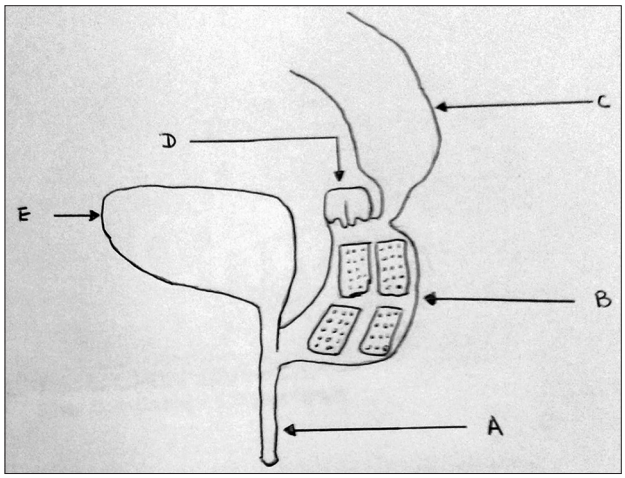

Figure 3: Schematic diagram of anomaly found at surgery. (A) Common channel. (B) Dilated vagina with barium stones. (C) Rectum implanted high on the vagina. (D) Uterus. (E) Urinary bladder

The surgical management of persistent cloaca is a planned procedure. After the initial diverting colostomy in the newborn period, an endoscopic evaluation of the common channel is commonly done in a separate setting outside of the newborn period (Levitt and Peña, 2010). A contrast study, that is, a distal colostogram is usually done some weeks before the definitive repair to delineate the precise type of anatomic defect in order to help plan the treatment (Warne *et al*., 2011; Peña and Levitt, 2003). This is a very important investigation in the management of this anomaly and can now be done with computer assistance to generate three-dimensional images, that is, three-dimensional cloacagram (Levitt and Peña, 2010; Peña and Levitt, 2003; Horsirimanont *et al*., 2004).

Keiller is believed to have been the first to describe the distal loopogram, having used barium sulfate to delineate the distal colon in anorectal malformations (Rahalkar *et al*., 2010). Perfection of the technique by later workers led to the development of the augmented pressure distal colostogram in which a water-soluble contrast is injected into the distal stoma with enough hydrostatic pressure to ensure the opacification of any distal fistula (Peña and Levitt, 2003; Horsirimanont *et al*., 2004; Rahalkar *et al*., 2010). The use of water-soluble contrasts is preferred to barium sulfate which is known to inspissate in the cavities occupied (Levy 1963). This was the case in our patient in whom barium sulfate was used as a contrast. Given that persistent cloaca is a rare anomaly, complications such as this may not be frequently reported.

In resource-constrained settings like ours, financial considerations may prompt the use of barium sulfate as a contrast agent in distal colostography. This report, however, buttresses the need to ensure the use of only water soluble contrast media in this procedure.

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