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Authors
Cheng, Lily S
Courtier, Jesse
MacKenzie, Tippi C

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Anal duplication in a one-year-old girl

Lily S. Cheng, **Jesse Courtier**, and **Tippi C. MacKenzie***

University of California San Francisco, 500 Parnassus Ave., San Francisco, CA 94143, USA

Abstract

We report a case of a patient with anal duplication discovered incidentally at 1 year of age. Pre-operative evaluation excluded any complications or associated anomalies. She underwent surgical excision with an excellent outcome.

Keywords

Anal duplication; Alimentary tract duplication; Anorectal malformation

Anal duplication is a rare congenital malformation with only about 50 cases described in the literature [1–5]. Most cases are diagnosed incidentally in the first year of life, and nearly half of all cases are associated with other midline anomalies [1,2]. The majority of patients are female [1,2]. Surgical resection is recommended to prevent infectious complications and outcomes are generally excellent [1,2,4,6].

1. Case report

A 13-month old Caucasian girl was brought to her pediatrician after her parents noticed what appeared to be a second anal opening while changing her diaper. The opening was 0.5-cm in length and located just posterior to her normal anus in the natal cleft (Fig. 1). She had no history of infections in the area, no passage of stool from the duplicate anus, no constipation or diarrhea. She was developing normally without any other medical problems.

Magnetic resonance imaging (MRI) demonstrated a tubular structure posterior to the normal anus 2-cm in craniocaudal length and possibly communicating with the rectum (Fig. 2). No other anomalies were identified.

The patient was taken to the operating room for examination under anesthesia and resection of the anal duplication. Exploration revealed a blind-ending pouch posterior to the normal anus but within the sphincter complex. It did not communicate with the rectum or any other

* Corresponding author. University of California San Francisco, Department of General Surgery, Division of Pediatric Surgery, 513 Parnassus Ave., Box 0570, San Francisco, CA 94143. Tel.: +1 415 476 2538; fax: +1 415 476 2929. tippi.mackenzie@ucsfmedctr.org (T.C. MacKenzie)..

Consent

Consent is not applicable as no identifiable patient information is evident in the texts or photographs.

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structures. The duplicate anus was excised and an anoplasty was performed by suturing the full thickness native anus to the posterior aspect of the sphincter complex. At the end of the case, a Pena nerve stimulator confirmed normal sphincter function, and a 12-Hegar dilator was easily admitted into the repaired anus. Histological findings were consistent with the diagnosis of anal duplication.

Her postoperative course was uneventful and she was discharged home the following day. At one year follow-up, she presented with constipation, which initially required polyethylene glycol, but has since resolved with diet modification. She is now 5 years old, has excellent anal tone and control and is continent after toilet training.

2. Discussion

Anal canal duplication is a rare congenital anomaly with only about 50 reported cases in the literature since first described in 1956 [1,3,7,8]. More than 90 percent of these cases occur in females, and most diagnoses are made incidentally [1,2].

Anal canal duplication arises from abnormal development of the dorsal cloacal membrane [9]. One theory holds that this occurs early in embryological development due to duplication of the dorsal cloacal membrane [10]. Another theory is that this occurs late in embryological development due to recanalization of excess dorsal cloacal membrane [11]. Histologically, anal canal duplication is typically characterized by a combination of squamous epithelium, transitional epithelium, and smooth muscle cells [7]. However, depending on the length of the anal duplication, all of these elements may not be present.

Patients with anal duplication usually present without symptoms, but some may present with nonspecific abdominal pain, nausea, vomiting and constipation, and some may present with symptoms of local or systemic infection. Many patients present in the first year of life, but age at initial presentation ranges from 0 to 24 years with a mean age of 28 months [1]. Older patients are more likely to present with symptoms. The first case of anal duplication described in the literature presented at the age of 65 with colloid carcinoma of the duplicate anus [6]. No other cases of malignancy associated with anal duplication have been reported.

More than 90 percent of patients present with an anal opening in the posterior midline. By some definitions, anal duplications must occur on the posterior side of normal anal canal [12]. In about 10 percent of cases, the duplicate anus communicates with the normal anus or rectum. About half of patients have other anomalies, usually also in the midline. The most commonly described anomalies associated with anal duplication are: cleft left/palate, congenital heart defects, spina bifida, sacral teratomas, and genitourinary malformations [1,8].

Diagnosis can be made by physical exam, although imaging is commonly performed to confirm the diagnosis and rule out any complications or associated anomalies [13]. Fistulograms, MRI, computed tomography (CT) and ultrasound are commonly used [9]. In our patient, the MRI was helpful in operative planning in that it ruled out a communication between the duplication and the native rectum.

Surgical approach is most often perineal or posterior sagittal excision of the entire duplication. Mucosal stripping of the canal has also been described [13]. Rare complications of surgery include sphincter dysfunction, amenable to surgical repair. Constipation is commonly reported after repairs of other anorectal malformations, and can also be seen in patients with anal duplication. Surgical outcomes are generally excellent [1,8,9].

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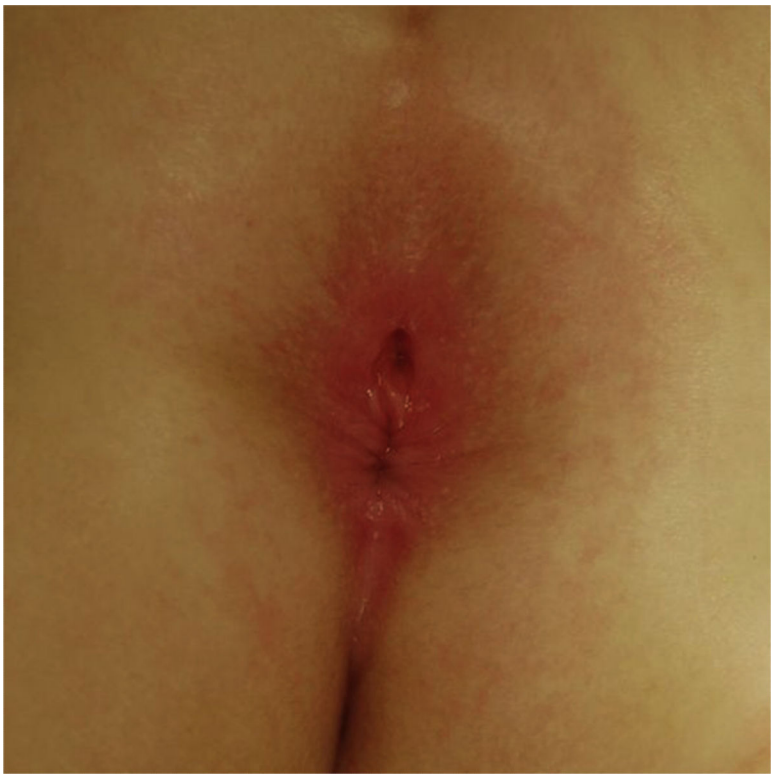

Fig. 1.
Preoperative appearance of the duplicate anus e note the skin defect posterior to the native anus (superior in this photograph).

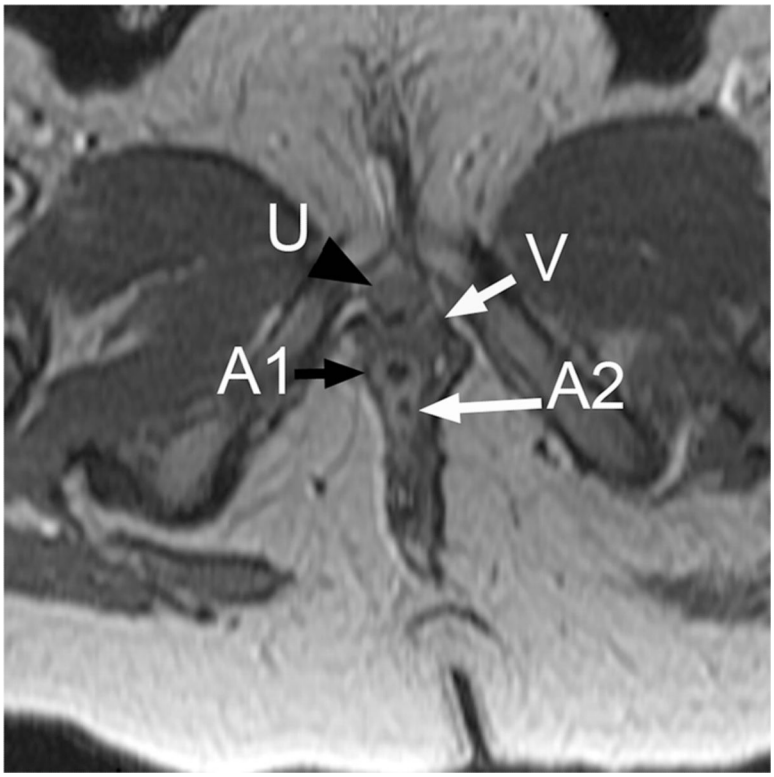

Fig. 2.

Axial magnetic resonance imaging (MRI) at the level of the puborectalis demonstrates the urethra (U), vagina (V), the anterior anal orifice (A1) and posterior anal orifice (A2).