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Case report

Sigmoid colon duplication associated with urogenital malformations: A rare case report

Luiz G. Freitas Filho *, Alexandre Augusto Medeiros Tonel, Bárbara dos Santos Rego, Maria Fernanda Brum Ribeiro, André Henrique Teruaki Kato, Maria Cristina Gomes Abreu

Pediatric Urology and Pediatric Surgery Department, Hospital Santa Marcelina, Chile

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ABSTRACT

Introduction and importance: Sigmoid colon duplication is a rare congenital anomaly, particularly when associated with complex urogenital malformations such as bladder and urethral duplication. These anomalies present significant diagnostic and surgical challenges. We previously reported this patient's case focusing on her genitourinary anomalies. During follow-up, the patient developed symptoms that led to the diagnosis of an undetected sigmoid colon duplication with a fistulous connection to the vagina, necessitating further surgical intervention.

Presentation of case: A female neonate was diagnosed with bladder exstrophy, urethral and bladder duplication and ambiguous genitalia at birth. At 7 months of age, she developed intermittent fecal discharge through the vagina, initially suspected to be a rectovaginal fistula. Imaging studies, including contrast-enhanced CT, revealed a duplicated sigmoid colon with a fistulous connection to the vagina. At 3 years old, she underwent laparotomy, which confirmed two sigmoid segments sharing a common mesentery. The anterior duplicated segment forming the fistula, was surgically resected, and the fistula was closed, THE patient had an uneventful postoperative recovery, with resolution of symptoms.

Clinical discussion: Sigmoid colon duplication is a rare entity that can remain undiagnosed for years. It may present with diverse symptoms, including obstruction, infection, or fistulization to adjacent organs. The association with bladder and urethral duplication suggest a possible common embryological origin. Accurate diagnosis requires advanced imaging techniques, particularly contrast studies from multiple entry points. Surgical intervention is the treatment of choice, aiming to remove the non-functional duplicated segment while preserving bowel integrity.

Conclusion: This case highlights the importance of long-term follow-up in patients with complex congenital anomalies. Sigmoid colon duplication should be considered in cases of unexplained vaginal fecal discharge, particularly in individuals with associated urogenital malformations. Early recognition for preventing and surgical treatment are crucial for preventing complications and improving patient outcomes.

1. Introduction

Gastrointestinal and urogenital duplications are rare congenital anomalies that result from abnormal embryological development. While intestinal duplications can occur anywhere along the digestive tract, sigmoid colon duplication is extremely uncommon [1]. Its association with bladder exstrophy, urethral and bladder duplication, and uterine anomalies makes this case unique.

We previously reported this patient's case [2] describing her complex congenital malformation, including bladder exstrophy, urethral and bladder duplication, and ambiguous genitalia. At that time, no intestinal anomalies had been identified. However, during long-term follow-up, the patient developed symptoms suggestive of an underlying gastrointestinal malformation, which led to the diagnosis of sigmoid colon duplication with a fistulous connection to the vagina. This follow-up report highlights the diagnostic challenges and surgical management of this additional finding, further contributing to the understanding of these rare anomalies.

* Corresponding author at: Rua Batista Cepelos 87 – ap 61, 04109-120 São Paulo, Brazil.
E-mail address: luiz.freitas@unifesp.br (L.G. Freitas Filho).

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2. Case report

The work has been reported in line with the SCARE criteria [3,4].

A 7-day-old female neonate was referred to our Pediatric Surgery Department with a diagnostic of omphalocele and ambiguous genitalia. She was born via cesarean section at 37 weeks of gestation to a mother with diabetes and hypertension. Prenatal investigations included an abdominal ultrasound (unremarkable), a transfontanellar ultrasound (normal), an echocardiogram (showing atrial and ventricular septal defects without hemodynamic compromise), and an abdominal X-ray (demonstrating pubic symphysis diastasis).

On physical examination, the omphalocele was found to be associated with bladder exstrophy, and no apparent urethral orifice was observed. The external genitalia had a bifid clitoris and were phenotypically female. On the 8th day of life, the patient underwent posterior iliac osteotomy with correction of the omphalocele and bladder exstrophy. Intraoperatively, two separate bladders, each with its own urethra, were identified and surgically unified while maintaining both urethras [2]. The patient had an uneventful postoperative recovery and was discharged without complications.

At 7 months of age, during a follow-up visit, the mother reported intermittent fecal discharge from the vaginal region, particularly when the child was constipated or experienced tenesmus. Under general anesthesia an examination with methylene blue instillation suggested a rectovaginal fistula. The patient underwent posterior sagittal anorectoplasty for better visualization, but no fistula was identified intraoperatively.

Despite continued outpatient follow-up, the mother persisted in reporting episodes of fecal discharge through the vaginal region. At 3 years of age, a second examination under anesthesia with contrast studies through rectum and vaginal raised suspicion of sigmoid colon duplication with a fistulous connection to the vagina (Figs. 1 and 2). A contrast-enhanced CT scan confirmed an apparent duplication of the proximal sigmoid colon, forming two loops: a posterior loop with a normal anatomical course contiguous with the rectum, and an anterior loop communicating with the anteriorsuperior vaginal wall, containing fecal-like material.

The patient underwent elective laparotomy, revealing urethral and bladder duplication, a didelphic uterus, and a sigmoid colon duplication. The sigmoid colon had a single mesocolon, but catheterization through both the rectum and vagina with a Hegar dilator demonstrated two separate lumens divided by a central membrane (Fig. 3). The anterior segment ended in a blind pouch with a fistulous connection to the anteroposterior vaginal wall, while the posterior segment followed the normal rectal course. The anterior segment was completely resected, and the fistula was closed.

Postoperatively, the patient recovered well, tolerated oral feeding, had regular bowel movements, and experienced no further episodes of fecal discharge from the vaginal region.

3. Comment

Sigmoid colon duplication is an exceptionally rare entity, especially in association with bladder exstrophy and genitourinary anomalies [5]. The embryological mechanism underlying these malformations remains unclear, though abnormal splitting of the embryonic cloaca or disturbances in the notochord development may contribute to such complex presentations [6].

Most intestinal duplications present in childhood with symptoms such as obstruction, infection, or, in rare cases, fistulous connections to adjacent structures, as seen in our patient. The diagnostic process can be challenging, particularly when the symptoms are intermittent, as in this case, where initial examinations failed to reveal the underlying duplication. Contrast imaging, especially CT or MRI with contrast administration through multiple orifices, can be instrumental in identifying such anomalies.

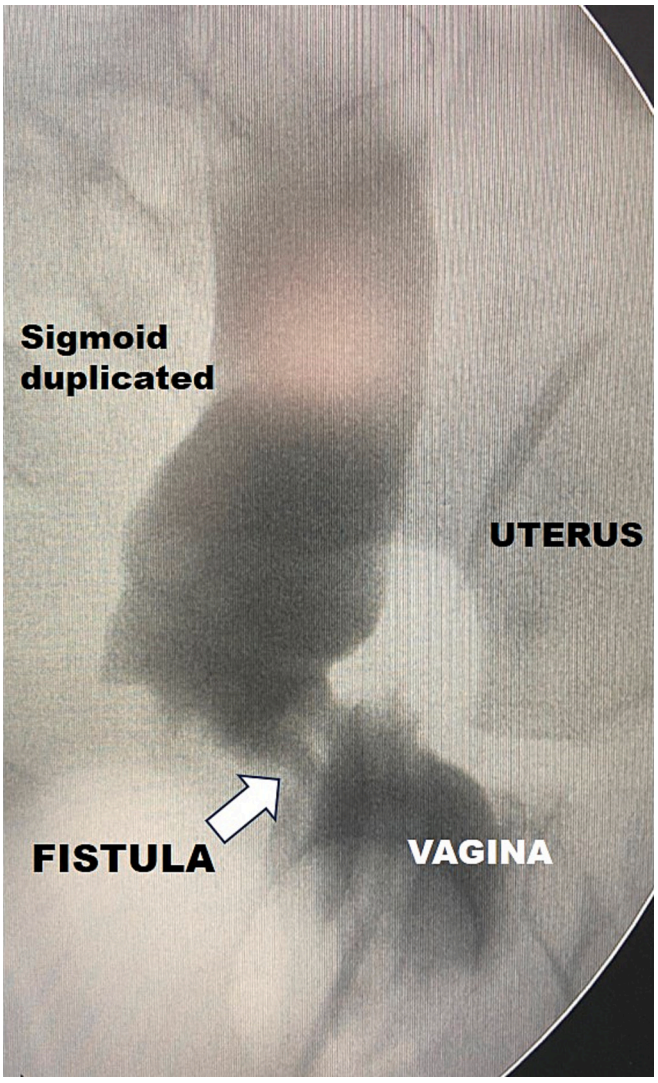

Fig. 1. Iodinated contrast introduced through the vagina, highlighting the fistulous tract between the vagina and duplicated sigmoid colon.

Surgical management aims to remove the non-functional duplicated segment while preserving normal bowel function. In this case, complete resection the anterior sigmoid duplication with fistula closure was curative. Long-term follow-up is essential to monitor for potential complications, including bowel dysfunction, recurrent fistulas, or urinary issues related to the associated urogenital anomalies.

We would also like to refer to a previously published article by our team, where the management of this patient's other urogenital anomalies, including bladder and urethral duplication, was detailed. This article provides a more comprehensive view of the surgical management of these complex malformations [2].

It is important to note that a contrast fistulogram might have been a useful diagnostic tool to confirm the fistulous connection prior to performing the laparotomy. This procedure could have provided further clarity in the assessment of the rectovaginal fistula and sigmoid colon duplication.

4. Conclusion

This case highlights the complexity of multiple congenital malformations involving the gastrointestinal and urogenital systems. Sigmoid colon duplication with a fistulous connection to the vagina is extremely rare and present diagnostic and surgical challenges. A thorough

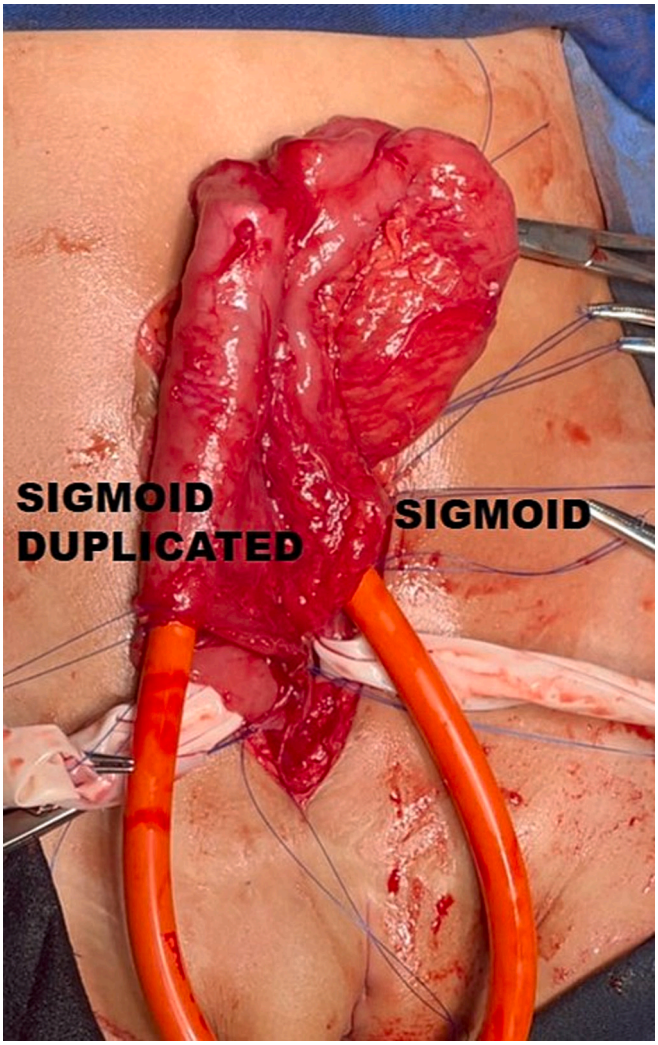

Fig. 2. Sigmoid colon duplication with a fistulous connection between the sigmoid and the vagina.

evaluation with advanced imaging and careful surgical planning is crucial for optimal patient outcomes. Long-term follow-up is necessary to assess functional recovery and detect any late complications.

CRediT authorship contribution statement

All authors of the manuscript **"Sigmoid colon duplication associated with urogenital malformations: a rare case report"** contributed to the design, data collection, problem interpretation, and manuscript writing.

Freitas Filho, LG – He contributed to the design, data collection, problem interpretation, and manuscript writing.

Rego, AAM - He contributed to the design, data collection, problem interpretation, and manuscript writing.

Rego, BS - She contributed to the design, data collection, problem interpretation, and manuscript writing.

Ribeiro, MFB - She contributed to the design, data collection, problem interpretation, and manuscript writing.

Kato, AHT - He contributed to the design, data collection, problem interpretation, and manuscript writing.

Abreu, MCG - She contributed to the design, data collection, problem interpretation, and manuscript writing.

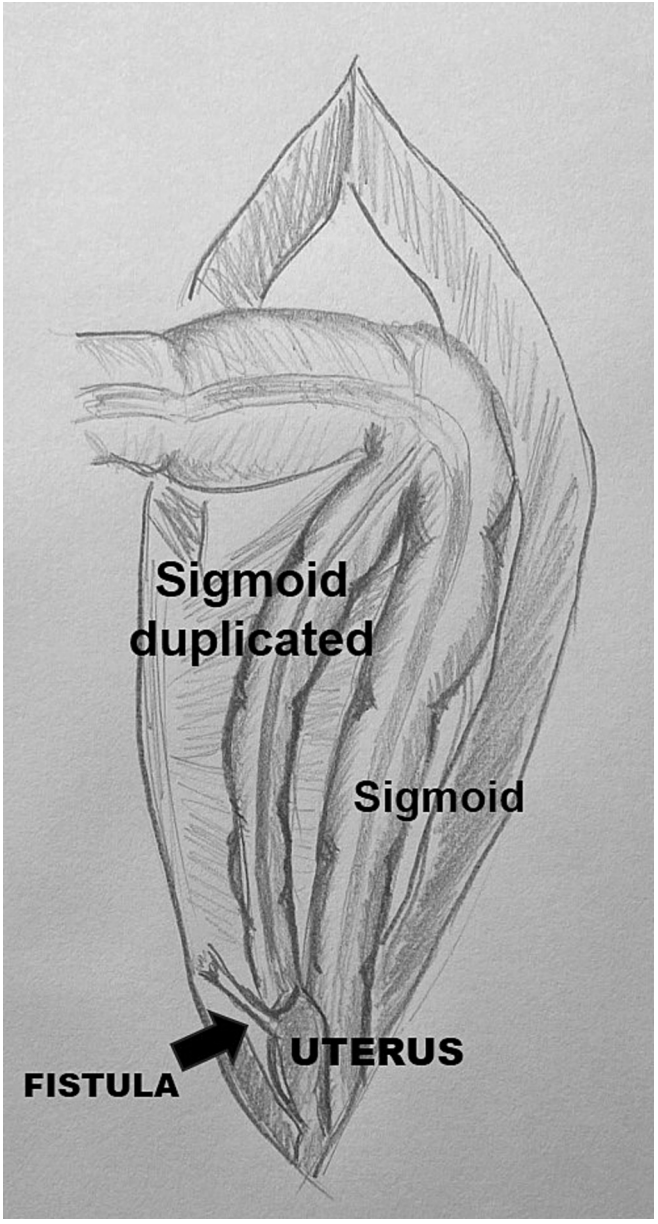

Fig. 3. Intraoperative view showing the two sigmoid colons with a common mesentery.

Informed consent

Written informed consent was obtained from the parents of the patient for publication of this case report accompanying images.

Ethical approval

We would like to confirm that our manuscript **"Sigmoid colon duplication associated with urogenital malformations: a rare case report"** has been reviewed and approved by the Ethics Committee of "HOSPITAL SANTA MARCELINA".

Comitê de Ética do Hospital Santa Marcelina: Protocolo 2025- AP 120-AE.

Author contribution

Guarantor

Luiz G. Freitas Filho

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Declaration of competing interest

Luiz G Freitas Filho – I declare that I have no financial or personal relationships with any organizations or individuals that could inappropriately influence or appear to influence the content of the manuscript

Alexandre Augusto Medeiros Tonel - – I declare that I have no financial or personal relationships with any organizations or individuals that could inappropriately influence or appear to influence the content of the manuscript

Bárbara dos Santos Rego - – I declare that I have no financial or personal relationships with any organizations or individuals that could inappropriately influence or appear to influence the content of the manuscript

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André Henrique Teruaki Kato - – I declare that I have no financial or personal relationships with any organizations or individuals that could inappropriately influence or appear to influence the content of the manuscript

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