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Intraoperative discovery of complete colonic duplication with duplicated caeca, appendices, vaginal orifices, and anorectum in a dizygotic twin: a case report

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Case Report

Keywords: Complete colonic duplication, Caecal duplication, Appendiceal duplication, Anorectal malformation, Case report

Posted Date: November 4th, 2026

DOI: https://doi.org/10.21203/rs.3.rs-11352799/v1

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Additional Declarations: No competing interests reported.

Abstract

Background

Complete colonic duplication is an exceptionally rare congenital anomaly that, when it occurs, often falls within the caudal duplication spectrum alongside vaginal and anorectal malformations. We report a rare case of complete colonic, caecal, and appendiceal duplication combined with duplicated vaginal orifices and anorectal malformation in a female dizygotic twin, incidentally discovered during surgical repair.

Case Presentation

A female infant, one of dizygotic twins, presented at birth with stool passing through an abnormal perineal orifice, clinically diagnosed as anorectal malformation of the vestibular type. Due to poor general condition in the neonatal period, primary surgical correction was deferred; the infant was managed conservatively with rectal saline irrigation from day nine of life, followed by right transverse colostomy at four months of age. At 7 months, during planned anorectoplasty under general anaesthesia, the surgical team incidentally identified two anal orifices and two vaginal orifices. Intraoperative extension of the procedure through a lateral abdominal incision revealed complete duplication of the colon, caecum, and appendix. No preoperative imaging had been performed; the entire duplication anomaly was thus an unexpected intraoperative discovery. The patient underwent anorectoplasty, vaginoplasty, and revision of the right transverse colostomy. Postoperatively she was managed with intravenous antibiotics, analgesia, and wound care, with satisfactory recovery. Her dizygotic male co-twin was clinically normal with no evidence of any congenital anomaly.

Conclusion

This case represents an exceptionally rare combination of complete colonic, caecal, and appendiceal duplication with concomitant duplicated vaginal orifices and anorectal malformation in a dizygotic twin. It underscores the importance of systematic intraoperative exploration in resource-limited settings, where complex duplication anomalies can masquerade as simple anorectal malformations until surgery reveals otherwise.

1. Background

Alimentary tract duplications are rare congenital lesions normally diagnosed in newborns and children; are generally classified morphologically as cystic (approximately 80%) or tubular (approximately 20%) lesions [1, 5]. This anomaly occurs anywhere from the mouth to the anus with a reported incidence of approximately 0.022 percent (about 1 in 4,500) of births [1]. Colonic duplication is exceedingly rare, which brings diagnostic as well as therapeutic challenges to attending surgeons [2]. They often require surgical operation in the neonatal, infant or older child [1]. Due to the complicated anatomy and common blood supply shared between the duplication and associated its own bowel, appropriate management requires a familiarity with the anatomy and clinical characteristics of this entity [3]. About two-thirds of these duplications are in the abdominal cavity, and more than half are in the jejuno-ileal segments. Affected sites are the thorax or thoraco-abdomen (4%), the foregut (extremely rare), the stomach (7%), duodenum (5%-7%), the pylorus (extremely rare), biliary tract (extremely rare), the small intestine (44%), the appendix (extremely rare) and the colon (15%). Associated anomalies are frequently present in patients with gastrointestinal tract duplications [1]. When appendiceal duplications are detected in childhood, almost all the patients have serious associated intestinal, genito-urinary or vertebral malformations [4].

2. Case Presentation

2.1 Patient Demographics and Presenting Complaint:

A female infant weighing 2300 g was delivered as one of a dizygotic twin pair. She cried immediately after birth. Her male co-twin was born in good condition and had no clinically apparent congenital anomaly during the reported follow-up period. According to the mother, stool was observed emerging from an abnormal perineal orifice on the ninth day of life, prompting hospital admission and evaluation.

2.2 Initial Assessment and Conservative Management:

Clinical findings at presentation demonstrated a female infant with a perineal orifice through which stool was being passed, suggestive of a diagnosis of anorectal malformation of the vestibular type. Because the infant's general condition was poor in the neonatal period, primary surgical correction was considered high-risk and was therefore postponed. From the ninth day of life, to facilitate faecal evacuation, the infant was managed cautiously with rectal saline irrigation. This was continued until the infant achieved sufficient weight and clinical stability for operative intervention. The second anal and vaginal orifices were not identified on initial neonatal perineal examination, likely due to their close approximation to the primary orifice and the limited scope of examination feasible in a critically unwell neonate — underscoring the need for careful perineal inspection under anaesthesia even when an anorectal malformation diagnosis appears straightforward.

2.3 Right Transverse Colostomy (Four Months of Age):

When the infant was 4 months old, with a weight of approximately 4 kg, she underwent right transverse colostomy for faecal diversion under general anaesthesia. The procedure was performed safely and there were no complication. Postoperative recovery was stable and the infant continued to gain weight day by day and develop satisfactorily. At the time of colostomy formation, the abdomen was not explored beyond the sigmoid colon, and the

coexisting caecal, appendiceal, and colonic duplication therefore remained undetected until the definitive repair three months later, a pattern also described by Piplani et al. [16].

2.4 Preoperative Investigations:

Prior to definitive surgical correction, investigation results are shown in Table 1.

Table 1. Preoperative investigation results.

Investigation	Result	Clinical Significance
Haemoglobin	10.5 g/dL	mildly reduced for the laboratory's reference range.
Total WBC	12,730/mm^3	Normal (Infant: 6,000-18,000/mm^3)
Platelet count	509,000/mm^3	Reactive thrombocytosis
S. Creatinine	0.4 mg/dL	Normal renal function
Blood group	A positive	Pre-operative crossmatch
Chest X-ray	Right-sided patchy opacity	Suggestive of right pneumonitis that was managed preoperatively
Pre-operative blood arranged	80 mL fresh whole blood	Arranged for anticipated blood loss

No preoperative imaging of the abdomen or pelvis including ultrasonography, contrast enema, or MRI was performed in advance of the definitive surgical procedure, reflecting the resource constraints of the treating institution.

Figure 1. Preoperative anteroposterior chest radiograph (A) and radiological report (B), showing a right-sided patchy pulmonary opacity interpreted as suggestive of pneumonitis.

2.5 Definitive Surgery and Intraoperative Findings (Seven Months of Age):

At the Seven Months of Age, the infant weighing approximately 6 kg underwent a planned anorectoplasty and vaginoplasty under general anaesthesia. During the perineal phase of the procedure, the surgical team discovered two distinct anal orifices and two distinct vaginal orifices which had not been previously recognised clinically or radiologically. This incidental discovery led to intraoperative extension of the procedure with a lateral abdominal incision to assess the extent of the anomaly.

Intraoperative abdominal assessment confirmed the following findings:

- Complete duplication of the entire colon, extending from the caecum to the rectosigmoid junction, with both colonic limbs being well-vascularized and of normal calibre.
- Complete duplication of the caecum, with both caecal pouches identified intraoperatively.
- Duplicated appendices, with an accessory appendix originating from the duplicated caecum.
- Both colonic limbs were found to share a common mesenteric base.
- Two distinct vaginal orifices corresponding to the two vaginal structures identified during perineal examination.
- Presence of two anal orifices, consistent with anorectal duplication.

The patient subsequently underwent anorectoplasty, vaginoplasty, and colostomy revision. Intraoperative blood loss was managed with transfusion of pre-arranged fresh whole blood. The duplicated colonic limbs were not definitively resected during this procedure and were therefore retained for planned staged reconstruction after further anatomical assessment. A resected sigmoid colon segment was submitted for histopathological examination; however, the histopathological report was not available at the time of manuscript preparation.

Intraoperative photographs were obtained with parental consent, demonstrating the duplicated colonic segments and duplicated appendix (Figures 2A),two anal orifices and anorectal operative field following anorectoplasty (Figure 2B). the double vaginal orifices (Figure 2C).

Figure 2. Intraoperative findings: duplicated colon, caecum, and appendix (2A); duplicated anus and rectum (2B); double vaginal orifice (2C); and the dizygotic twins shown in (2D).

2.6 Postoperative Course:

Postoperative management included intravenous ceftriaxone, metronidazole, analgesia, local wound care, and temporary urinary catheterisation. Diet was commenced with expressed breast milk and then subsequently proceeded to soft complementary feeds as tolerated.

At eight months of age, the infant weighed 6.3 kg and was receiving breast milk with age-appropriate complementary feeding. She remained clinically stable following the reported surgical procedure, with satisfactory wound healing and no postoperative complications documented during the available follow-up period. Further staged surgical intervention, including colostomy closure and definitive reconstruction of the colonic anatomy, was planned after assessment of the pelvic and spinal anatomy with pelvic ultrasonography and spinal MRI. Because these investigations and subsequent reconstructive procedures had

not yet been completed at the time of manuscript preparation, long-term bowel, continence, urinary, reproductive, and neurological outcomes cannot yet be assessed.

2.7 Co-twin Status:

The male dizygotic co-twin was evaluated clinically and found to be completely normal, with no evidence of any anorectal, genitourinary, gastrointestinal, or spinal anomalies. His anthropometric measurements and developmental milestones were appropriate for his age.

2.8 Clinical Timeline:

Table 2 summarises the presentation, investigations, interventions, and follow-up for the reported case.

Table 2. Clinical Timeline

Age / Date	Event / Clinical Finding	Clinical Action / Management
Birth	2300 g female infant delivered as one of a dizygotic twin pair; cried immediately at birth.	Routine newborn care.
Day 9 of life	Mother noted stool passing through an abnormal perineal orifice.	Hospital admission and clinical evaluation; clinically diagnosed as anorectal malformation, vestibular type. Primary surgical correction deferred due to poor general condition.
Day 9 of life – 4 months	Second anal and vaginal orifices not identified on initial neonatal perineal examination.	Conservative management with rectal saline irrigation to facilitate faecal evacuation until sufficient weight/clinical stability is achieved.
4 months of age (4 kg)	No complication during procedure; abdomen not explored beyond sigmoid colon, so coexisting caecal/appendiceal/colonic duplication remained undetected.	Right transverse colostomy performed under general anaesthesia for faecal diversion. Stable postoperative recovery; continued weight gain.
Pre-definitive-surgery workup	Mild anaemia (Hb 10.5 g/dL); WBC and platelet counts within/above normal paediatric range; right-sided patchy opacity on chest X-ray (pneumonitis).	Preoperative investigations completed (Table 1); pneumonitis treated preoperatively; 80 mL fresh whole blood arranged; no abdominal/pelvic imaging performed (resource-limited setting).
7 months of age (6 kg)	During planned anorectoplasty, two distinct anal orifices and two distinct vaginal orifices identified intraoperatively; not previously recognised clinically or radiologically. Lateral abdominal incision revealed complete duplication of colon, caecum, and appendix, sharing a common mesentery.	Definitive surgery: anorectoplasty, vaginoplasty, and revision of colostomy performed under general anaesthesia. Intraoperative blood loss managed with pre-arranged transfusion. Histopathological findings of the resected sigmoid segment were also unavailable at the time of manuscript preparation.
Postoperative period	Satisfactory wound healing; uncomplicated recovery.	IV antibiotics (ceftriaxone, metronidazole), IV analgesia (pethidine, paracetamol), local wound care, temporary urinary catheterisation. Diet advanced from expressed breast milk to soft complementary feeds.
8 months of age	Weight 6.3 kg; exclusively breastfed with age-appropriate complementary feeding; fully immunised per national schedule.	Growth and developmental follow-up. Further staged surgery (colostomy closure, definitive colonic reconstruction) planned, pending pelvic ultrasonography and spinal MRI.
Throughout (co-twin)	Male dizygotic co-twin clinically normal throughout the reporting period; no anorectal, genitourinary, gastrointestinal, or spinal anomaly; anthropometrics and milestones age-appropriate.	Routine clinical evaluation only; no imaging performed (asymptomatic).

3. Discussion

3.1 Rarity of the Anatomical Combination

The coexistence of complete colonic duplication, caecal duplication, appendiceal duplication, duplicated vaginal orifices, and anorectal malformation constitutes an exceptionally rare anomaly complex, with few documented in existing literature [~~2~~2]. Colonic duplication accounts for approximately 13–15% of all gastrointestinal duplications in paediatric series [~~1~~1,~~6~~6], although its true frequency within the colon-specific literature is difficult to establish given the rarity of the anomaly and heterogeneity of reporting. In one adult surgical series, colorectal duplication accounted for only 6.8% of gastrointestinal duplications overall [~~7~~7], underscoring that colonic duplication is rare across both paediatric and adult populations, with figures varying by series and denominator used. Gastrointestinal duplications are generally identified during childhood, although some remain silent and present later on in life. The presentation varies according to the age of the patient as well as the location of the duplication. Some may be totally asymptomatic, being identified on routine physical examination or during incidental investigations [~~1~~1]. Holcomb et al. reviewed 96 patients with 101 alimentary tract duplications identified over a 37-year period (through 1989), of which 21 duplications were confined to the thorax, three were thoracoabdominal, and 77 were abdominal; 74 patients presented as infants under two years of age, and 22 presented later in childhood [~~6~~6].

In the present case, complete colonic duplication was supported by the operative identification of duplicated proximal colonic structures and duplicated distal anorectal openings, together with the operative documentation of "Total colonic duplication".

3.2 Classification: Caudal Duplication Syndrome (CDS) or Distinct Entity?

The term Caudal Duplication Syndrome (CDS) was first defined by Dominguez et al. in 1993 and refers to a spectrum of congenital anomalies characterized by the association of partial or complete duplication of hindgut structures, duplication of lower urogenital tract, spinal cord and vertebral anomalies occurring at heterogeneous degrees of severity, presumably resulting from developmental disturbances at different stages of embryogenesis [8].

Our patient satisfies the hindgut duplication criteria like complete colonic, caecal, and appendiceal duplication, and urogenital involvement like duplicated vaginal orifices and anorectal malformation. Formal imaging to evaluate the spine, vertebrae, kidneys, urinary tract, and pelvic organs remained pending at the time of reporting. The constellation of findings is consistent with the caudal duplication spectrum.

3.3 The Dizygotic Twin: Embryological Significance

The occurrence of this complex congenital anomaly in one member of a dizygotic twin pair, while the co-twin remained clinically normal, is a notable feature of the present case. Dizygotic twins share, on average, approximately half of their genetic material, similar to non-twin siblings; however, the discordant phenotype observed here does not by itself establish a specific genetic, post-zygotic, or vascular mechanism [5]. No karyotyping or genetic testing was performed in the present case, and therefore the underlying mechanism of the discordant presentation cannot be determined. The normal clinical status of the co-twin is reported as an important phenotypic observation rather than evidence for a particular embryological mechanism.

3.4 Intraoperative Discovery in a Resource-Limited Setting

Although preoperative diagnosis is sometimes possible with barium enema, ultrasonography, or CT, most cases, including ours, are diagnosed at surgery. No preoperative imaging was available at our institution, a resource constraint that made systematic intraoperative exploration the sole means of detecting this anomaly. Definitive treatment is surgical, aiming to resect the duplication and reconstruct the anatomy while preventing the complications noted above, though operating on such cases remains technically demanding. Outcomes of operation are generally excellent when the condition is identified and managed properly [2]. Similar cases reported in the literature highlight varied surgical strategies, which often involve staged reconstruction, and emphasize the importance of individualized planning [14].

3.5 Staged Surgical Management

Because gastrointestinal duplications are rare and can present with a wide range of clinical manifestations, or may even be encountered intraoperatively as in the present case, surgical management must be individualised to the anatomy encountered. The treatment is surgical, with total resection performed when possible as the aim of the intervention. Incomplete treatment may be associated with recurrence, depending on the lesion and residual mucosa. Surgical complications are related to the size and location of the duplication, communication with the gastrointestinal tract or vertebral canal, presence of heterotopic gastric mucosa and involvement of mesenteric vessels. Because of the mesenteric location of most duplications, they share a common blood supply with the normal intestine. Therefore, surgical dissection must be precise [1]. During surgical correction, the duplicated colon is excised, and a posterior sagittal anorectoplasty is most often performed. This mirrors the experience of other reported cases: Subedi et al. described an intraoperative finding of a blind-ending tubular colonic duplication filled with impacted stool, which explained their patient's abdominal distension and constipation and was managed by surgical resection [2]. Such reports reinforce the recurring theme that complete colonic duplication is frequently an unanticipated intraoperative finding rather than a preoperatively diagnosed condition.

3.6 Comparison with previously reported cases

Complete colonic duplication associated with anorectal and/or genitourinary anomalies has been described in a limited number of paediatric reports, with considerable variation in the extent and anatomical configuration of duplication. Previous cases have included complete or tubular colonic duplication associated with anorectal malformations, duplicated anal openings, rectovestibular or rectovaginal communications, and genital anomalies. Some duplications were recognised only during operative exploration, highlighting the potential for delayed diagnosis.

The cases reported by Kaur et al. 2004 [11], Jellali et al.2012 [13], Acer et al.2013 [14], AbouZeid et al.2019 [15], and Piplani et al. 2022 [16] are particularly relevant to the present case because of their combinations of colonic duplication with anorectal and/or genital anomalies. Jellali et al. and Acer et al. also described caecal and appendiceal duplication, while Piplani et al. demonstrated that colonic duplication may remain undetected during initial colostomy and be recognised during subsequent surgery.
In the present case, the duplication was unexpectedly identified during definitive anorectal surgery after previous colostomy. Intraoperative findings included two caecal pouches, two appendices, duplicated colonic limbs sharing a common mesenteric base, two anal orifices, and two vaginal orifices. The occurrence in a dizygotic twin with a clinically normal co-twin further distinguishes the clinical context. Table 3 summarises the most relevant previously reported cases and their similarities to the present case.

Table 3. Most relevant previously reported cases and their similarities to the present case

No	Reference	Age / Sex	GI / colonic findings	Anorectal anomaly	Genital anomaly	Associated anomalies	Relevance to the Present Case
1	Yucesan S, et al. 1986. J Pediatr Surg [9]	1y, F	Complete tubular colonic duplication	—	2nd orifice; duplicated colon–vagina fistula	None reported	Early report of complete colonic duplication with genital communication
2	Prasil P, et al. 2000. J Pediatr Surg [10]	7mo, F	Cystic/tubular recto-sigmoid duplication; pelvic mass	Recto-vaginal fistula (delayed)	Not reported	Solitary kidney; sacral anomalies; tethered cord with lipoma	Same age as ind case; highlights for spinal screer
3	Kaur N, et al. 2004. Pediatr Surg Int [11]	3mo, F	Tubular duplication of entire large bowel, caecum, appendix, terminal ileum	ARM with rectovestibular fistula	Not reported	None reported	Close anatomica comparison
4	Banu T, et al. 2007. J Pediatr Surg [12]	42d, F	Total colonic duplication	2 non-communicating anal openings	2 vaginas, 1 vulva, 1 clitoris, 1 urethra	Polydactyly; pelvicalyceal dilatation; duplicated kidneys/ureters	Very close comparison
5	Jellali MA, et al. 2012 (3 cases). J Pediatr Surg [13]	2d F; NB M; 7h F	Complete tubular colonic duplication (all 3); double caecum/appendix in Case 2	Anal atresia; urethral fistulae	Split vaginal orifice (Case 1); vaginal atresia (Case 3)	Sacral hemivertebra/scoliosis; duplicated bladder/urethra; ureteral dilatation	Closest structura parallel (double caecum/append complete colonic duplication + AR
6	Acer T, et al. 2013. J Pediatr Surg [14]	NB, F	Omphalocele; duplex appendix/colon/sigmoid/caecum; malrotation	Duplex anal dimple, imperforate anus, bilateral rectovestibular fistulae	Duplex vagina, duplicated external genitalia, duplex uterus/tubes	Vertebral dysplasia; bifid L5/sacrum	Closest full cauc duplication anal but with omphalocele/ve defects absent i index case
7	AbouZeid AA, et al. 2019. Eur J Pediatr Surg Rep [15]	7mo, F	Complete colonic/rectal duplication with narrow proximal communication	Suspected recto-vaginal fistula; late-diagnosed ARM	Fistula between duplicated rectum and lower vagina	Solitary kidney; sacral anomalies	Same age and delayed-diagnos pattern — closes presentation ma
8	Piplani R, et al. 2022 (3 cases). Ann Pediatr Surg [16]	NB F; 18mo F; 1y F	Duplicated colon (Case 1); entire colon proximal/distal to colostomy duplicated (Cases 2–3)	Rectovestibular/H-type fistulae	Not separately reported	None reported	Shows colonic duplication can l missed at initial colostomy, recog later — as in inde case
Index Case	—	**7mo, F (dizygotic twin)**	**Operative "total colonic duplication"; 2 caecal pouches, 2 appendices, common mesentery**	**2 anal orifices; anorectal duplication; anorectoplasty performed**	**2 vaginal orifices; vaginoplasty performed**	**Right pulmonary opacity (pneumonitis)**	**Rare combinatio duplicated caeca/appendic common mesen base, 2 vaginal + anal orifices in a dizygotic twin**

Abbreviation: ARM, anorectal malformation; F, female; M, male; NB, newborn; d, day(s); mo, month(s); y, year(s).

3.7 Limitations

This report has several limitations. No preoperative cross-sectional or contrast imaging was available, reflecting resource constraints, and the anomaly was diagnosed intraoperatively rather than through planned imaging.The proximal and distal operative photographs were obtained from separate surgical fields, and no single image demonstrates the entire duplicated colon. The classification of complete colonic duplication was based on the combined operative assessment and the OT note documenting "total colonic duplication." Formal imaging to evaluate the spine, vertebrae, kidneys, urinary tract, and pelvic organs remained pending at the time of reporting. Karyotyping was not performed. Genetic testing was not performed, and long-term bowel, continence, urinary, reproductive, neurological, and developmental outcomes remain unknown. Histopathological findings of the resected sigmoid segment were also unavailable at the time of manuscript preparation.

4. Conclusion

This case demonstrates an exceptionally rare complex hindgut duplication identified unexpectedly during definitive surgery for anorectal malformation in a female dizygotic twin. The coexistence of complete colonic duplication with duplicated caeca and appendices, two anal orifices, and two vaginal orifices highlights the importance of careful perineal examination and systematic intraoperative assessment when unexpected duplicated structures are

encountered. In settings where preoperative abdominal and pelvic imaging is limited, intraoperative recognition may be critical for appropriate surgical planning. Further imaging and staged reconstruction are required before the patient's definitive anatomical and functional outcomes can be determined.

Declarations

A. Ethics and Consent to Participate:

This case report was conducted in accordance with the ethical standards of the institutional research committee and with the 1964 Declaration of Helsinki and its later amendments. Formal approval from the Institutional Review Board was not required for publication of a single retrospective case report.

B. Consent to publish:

Written informed consent was obtained from the patient's parent or legal guardian for publication of this case report and accompanying images. The consent form was reviewed and updated to ensure consistency with the final manuscript title, and all identifying and descriptive elements correspond to the present submission. The consent form is available for review by the journal editor upon request.

C. Availability of Data and Materials:

The data and materials that support the findings of this study, including primary clinical records, the operative notes and admission documentation, are available from the corresponding author upon reasonable request, subject to institutional data protection policies.

D. Competing interests:

The authors declare that they have no competing interests as defined by BMC or other interests that might be perceived to influence the results and/or discussion reported in this paper.

E. Funding Statement:

The authors have nothing to report. No external funding was received for this case report.

F. Author contributions:

M.N.H. and T.S.S. (Topu Shree Saha) conceptualized the report, managed data collection, and supervised the project. M.N.H. also prepared the clinical visuals. All authors (M.N.H., T.S.S. [Topu Shree Saha], M.T.K.S., and T.S.S. [Tonu Shree Saha]) participated in the clinical investigation and literature review. M.N.H., T.S.S. (Topu Shree Saha), and M.T.K.S. drafted the primary manuscript. Critical revision and editing were performed by M.N.H., T.S.S. (Topu Shree Saha), and M.T.K.S., while T.S.S. (Tonu Shree Saha) assisted with the review. All authors read and approved the final manuscript.

G. Acknowledgments:

The authors thank the patient's parent or legal guardian for providing consent for publication. The authors also acknowledge the Department of Pediatric Surgery, Jashore Medical College for providing the laboratory documentation and clinical records necessary for this report. The authors further express gratitude to Dr. M.A. Golam Kibria, Professor Dr. Ansar Ali, Dr. Musfiqur Rahman, and Dr. Sharifuzzaman, of the Department of Pediatric Surgery, Jashore Medical College and Hospital, Jashore, Bangladesh for their expert review and supervision of this manuscript.

H. Patient perspective:

A formal patient perspective was not obtained because the patient was an infant at the time of reporting. The patient's parent or legal guardian provided written informed consent for publication of the clinical details and accompanying images.

I. CARE checklist:

This case report has been prepared in accordance with the CARE (Case Report) guidelines. The completed CARE checklist is submitted as Additional File 1.

J. Clinical trial number:

Not applicable

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Figures

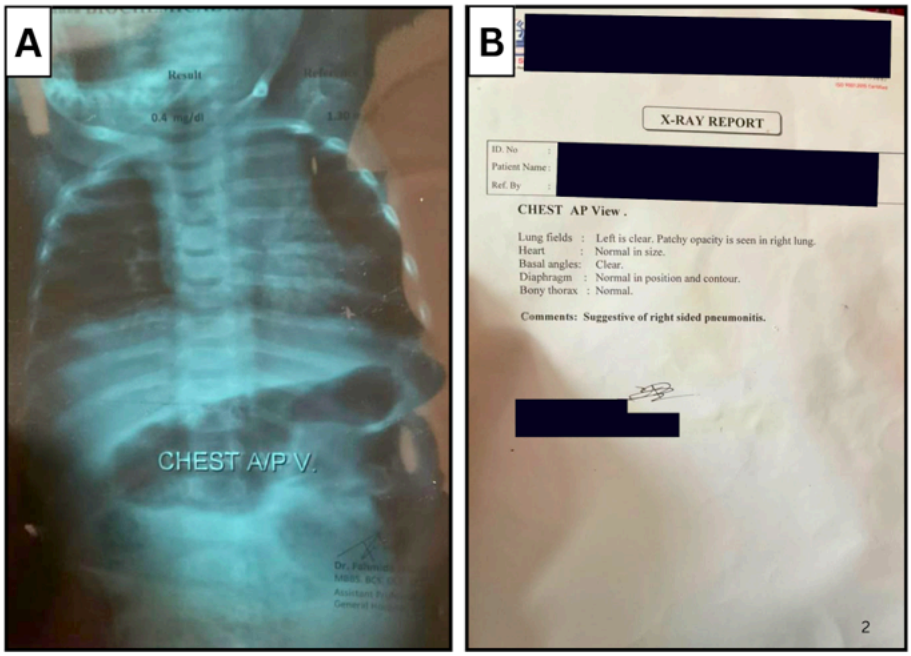


Figure 1

Preoperative anteroposterior chest radiograph (A) and radiological report (B), showing a right-sided patchy pulmonary opacity interpreted as suggestive of pneumonitis.

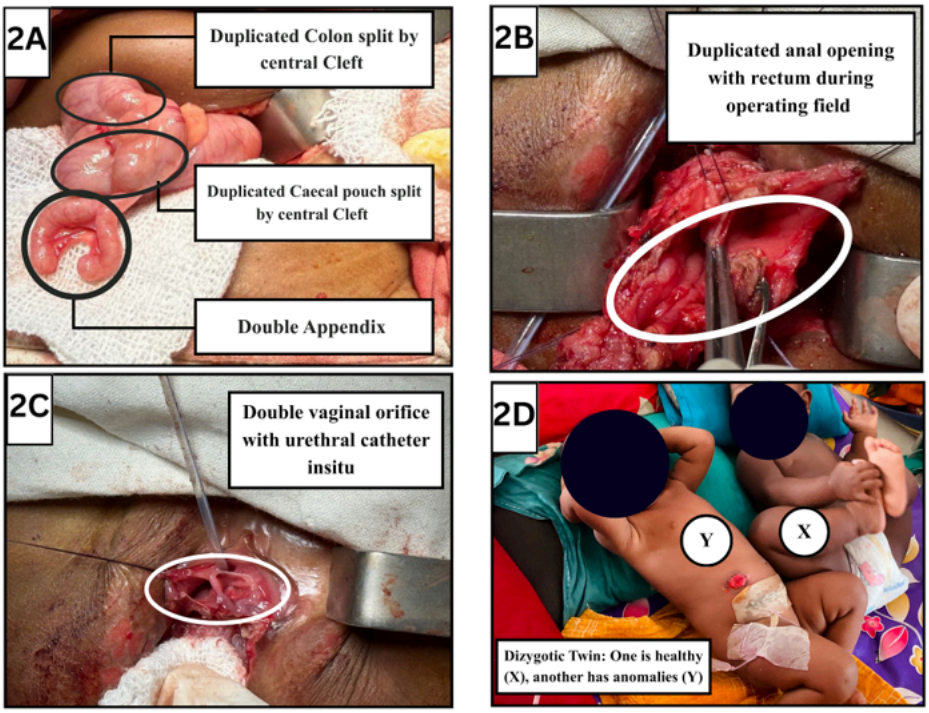


Figure 2

Intraoperative findings: duplicated colon, caecum, and appendix (2A); duplicated anus and rectum (2B); double vaginal orifice (2C); and the dizygotic twins shown in (2D).

Supplementary Files

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