

# Complete cloacal duplication imaged before and during pregnancy


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## ABSTRACT

The authors describe a 31 year-old female who presented emergently with abdominal pain and was found at CT to have complete genitourinary duplication including separate urinary bladders, uteri, cervixes, and vaginas, and also duplication of the rectum. No etiology for abdominal pain was identified. The patient was referred to urology for further evaluation, and an intravenous urographic study was obtained, which confirmed complete lower urinary tract duplication. The patient presented emergently 9 months later during a subsequent pregnancy for further evaluation of abdominal pain. A second CT scan was ordered to rule out appendicitis. Findings consistent with cloacal duplication were again noted. There was also dilatation of the urinary collecting systems, more prominently on the right side. A Cesarean section was performed and confirmed total genitourinary and rectal duplication.

## CASE REPORT

### CASE REPORT

A 31 year-old G0 patient presented with vaginal and left lower quadrant pain to the emergency department at our institution. She reported a remote history of surgery on her anus, performed in Mexico during infancy. A computed tomographic (CT) scan was obtained with intravenous contrast to further evaluate her pain. The CT scan (Figure 1) demonstrated complete genitourinary duplication. Duplicated structures included the uterus, cervix, vagina, and urinary bladder. Complete genitourinary duplication was also present at ultrasound (Figure 2). Physical exam confirmed two urethras, a vaginal septum, and 2 cervixes. Rectal duplication was also noted at CT (Figure 1). The patient appeared to have a single anal orifice at physical examination, with post-surgical

scarring adjacent to the anus. This was correlated on CT with a probable post surgical scar in the right ischiorectal fossa (not shown). The patient was unaware of the details of her prior surgery, as the procedure was performed while she was a young child. Right renal scarring was also noted, which was attributed to probable prior infection. No other etiology for abdominal pain was identified.

The patient received outpatient gynecological follow up for left lower quadrant pain and infertility for 1 year. The urology service was also consulted and ordered an intravenous urogram, which confirmed duplication of the lower collecting system bilaterally (Figure 3). Further referral was also made to the Reproductive Endocrinology and Infertility clinic, although the patient became spontaneously pregnant with a single live

intrauterine pregnancy within the right hemi-uterus, before further treatment for her infertility symptoms was pursued.

Nine months after her initial presentation, the patient presented to labor and delivery with right lower quadrant and back pain. An intrauterine pregnancy in the right hemi-uterus with a clinical age of 32 and 5/7 weeks was confirmed. A presumptive diagnosis of pyelonephritis was made, and the patient was admitted for intravenous antibiotic treatment. When urine cultures failed to confirm a urinary tract infection, a repeat CT scan was performed in order to evaluate for appendicitis or other possible causes of pain. While the long-term effects of radiation to the fetus were acknowledged, the clinicians opted for CT over MRI secondary to the acute nature of the patient's condition. At her second CT scan (Figure 4), the patient was noted to have an intrauterine pregnancy within the right hemi-uterus. There was also dilatation of the ureters bilaterally, worse on the right side compared to the left (Figure 5). The nephrograms were asymmetric, with a slightly delayed right nephrogram. Right renal scarring was again noted. The clinical impression was that her right ureteral dilatation was the cause for her pain.

Over the course of 2 weeks, the patient was observed on the labor and delivery service while antibiotics were administered. On hospital day 13 when the patient was 34 and 5/7 weeks gestation, fetal heart rate decelerations, oligohydramnios and lack of interval fetal growth were noted. The patient was then taken to the surgery for cesarean section. Complete genitourinary and rectal duplication were confirmed at the time of surgery. There were no other complications during the delivery. The mother was discharged on post-operative day 3 and the infant was discharged on day of life 7.

## DISCUSSION

Complete cloacal duplication in females is an exceedingly rare entity characterized by duplication of genitourinary and hindgut structures. 13 patients have been described in the literature with complete genitourinary duplication. Complete cloacal duplication is an even more rare entity, with 3 cases described previously in the literature (1). To our knowledge, there are no reported cases of complete cloacal duplication resulting in a term pregnancy.

This patient's presentation in adulthood is also very unusual. To our knowledge, there has been only one patient published previously who presented in adulthood. The prior case report described a patient who presented with complete genitourinary but without hindgut duplication (2). Successful term pregnancy was not encountered in our review of the literature in any patients with this entity. Greenberg et al demonstrated that patients can undergo successful vaginal delivery after cloacal malformation repair as infants (3). However, the patient presented herein had a different spectrum of anomalies than those reported by Greenberg et al.

Dominguez et al have described a series of 6 patients who presented with duplication anomalies of various cloacal and

caudal neural structures including anomalies of varying severity in the genitourinary system, hindgut, and lower spine and cord. This paper also introduced the term Caudal Duplication Syndrome (4). The current presentation may be among the spectrum of this disease constellation, although the patient's presentation is dissimilar to any of the patients discussed by Dominguez et al.

Although an etiology of caudal duplication is not known, some have espoused a theory of caudal twinning (5). Embryologically, the primordial hindgut forms as a caudal structure, with the lumen formed by vacuolization, giving rise to the distal ileum, colon, rectum, bladder, and urethra. Abnormal vacuolization of the pluripotent hindgut cell mass during caudal growth might create two parallel hindgut channels, thereby duplicating all hindgut structures distal to the point of separation (6).

Patients within the spectrum of urogenital or anorectal duplication have been reported with a variety of presentations and symptoms. Uncommonly, this entity is encountered incidentally (1, 7-11). Children may present with abnormalities of the external genitalia, urinary tract obstruction or infection, urinary incontinence, voiding disorders, and anorectal malformations (2, 12). Uterine abnormalities range from bicornuate uterus to complete uterine and vaginal duplication. Similarly, duplication of the urinary tract can vary in severity from partial to complete duplication of the bladder to and lower urinary tract. Anomalies are often associated and usually involve the hindgut. Spinal cord, or spinal deformities have also been reported (13, 14). Possible associated hindgut anomalies include duplication of the distal gastrointestinal tract, anal atresia, anal stenosis, and imperforate anus (2, 12-14). Based on available clinical information in our patient, it was felt that two anal orifices were likely present at birth, one of which was closed during the patient's initial surgery. This could not be verified however, since outside records could not be obtained.

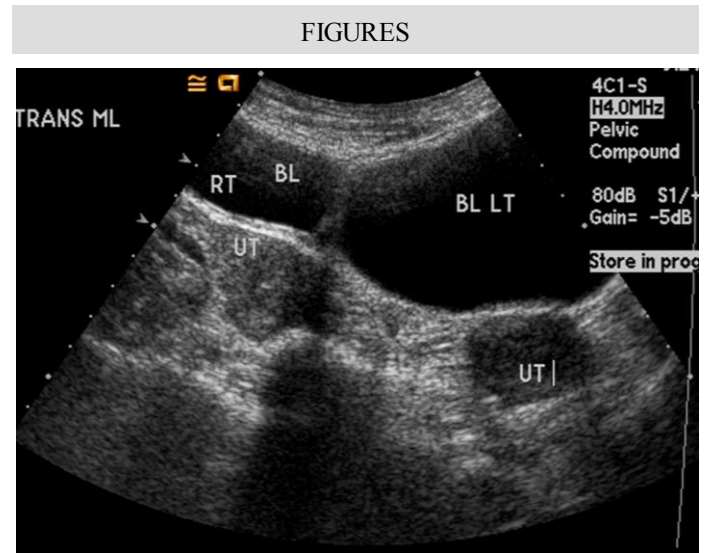
## TEACHING POINT

Cloacal duplication is an extremely rare occurrence, and would likely be diagnosed in infancy. However, patients may progress to adulthood and it is important to identify these patients in order to assess complications and for pregnancy planning.

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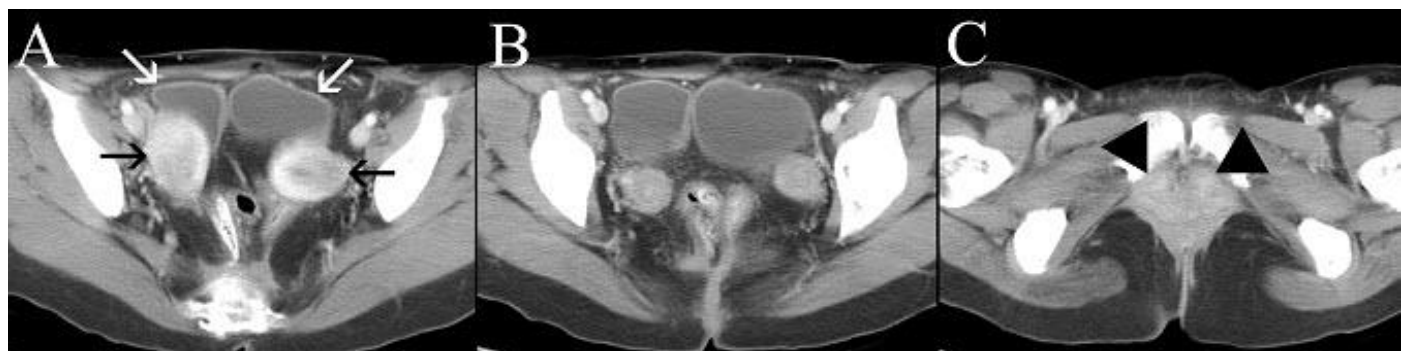
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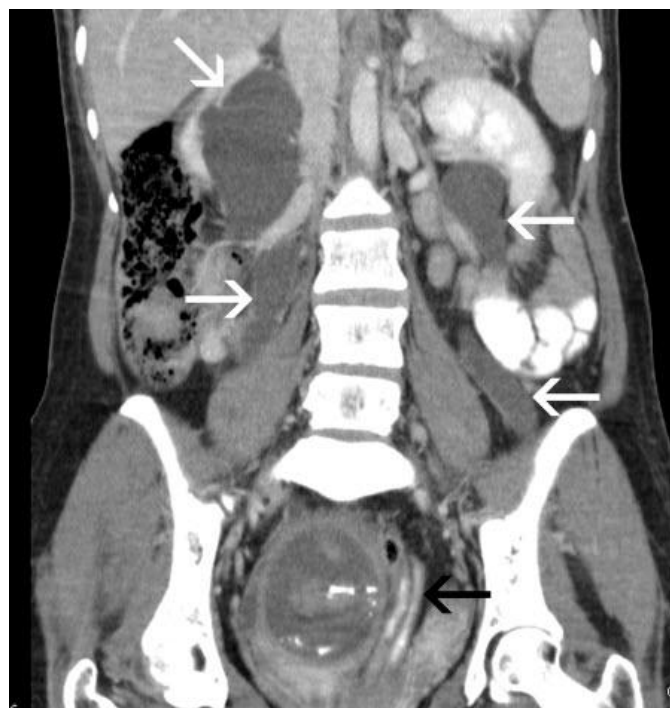
**Figure 2.** 31 year-old female with complete cloacal duplication. A transverse ultrasound image of the pelvis obtained with a 4 MHz curved-array transducer thru the abdominal wall using the urinary bladders as acoustic windows demonstrates the urinary bladders bilaterally (BL) and a hemi-uterus on each side of the pelvis (UT).

**Figure 1 (bottom).** 31 year-old female with complete cloacal duplication. CT images are ordered from cranial (A) to caudal (C). The study is performed after intravenous administration of 100 cc's of Isovue 370 and Gastroview oral contrast. 8 mm slice thickness images are obtained (kVp=130, mA=30). White arrows demonstrate the duplicated urinary bladders. Black arrows demonstrate duplication of the uterus. There were separate urethral and vaginal orifices at the perineum (black arrowheads).

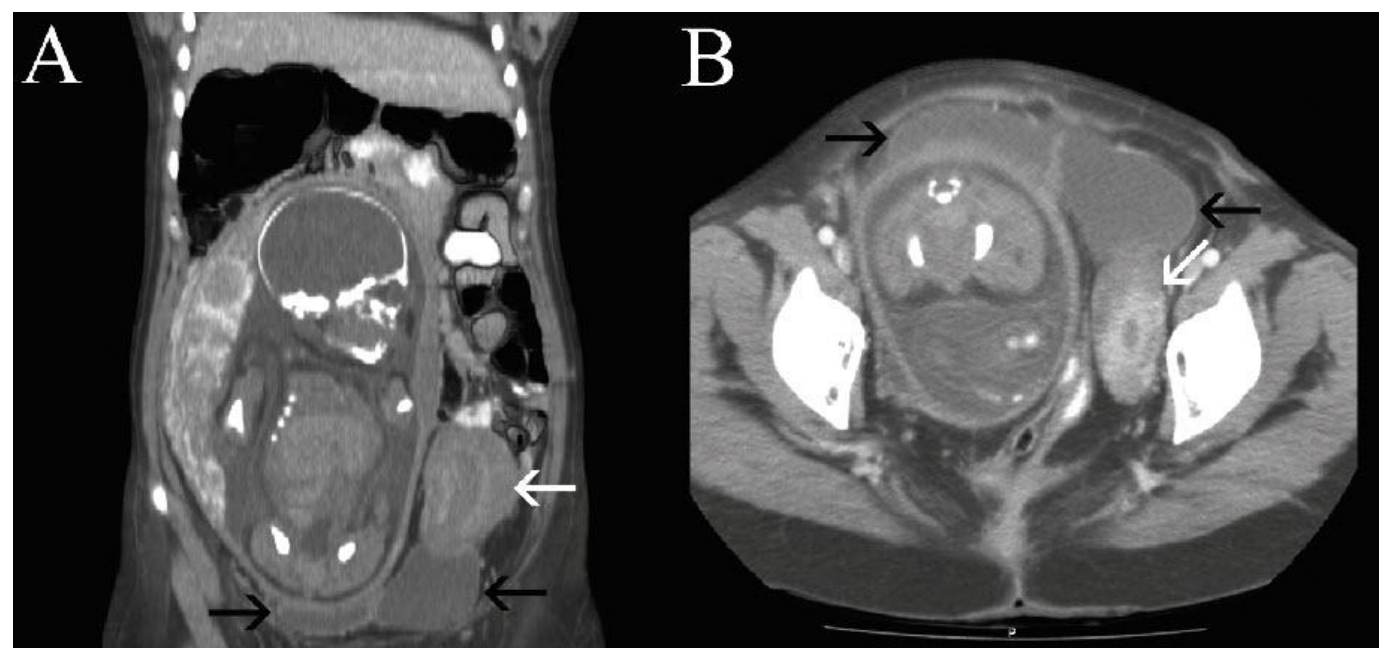




**Figure 3.** 31 year-old female with complete cloacal duplication. A single view of the abdomen obtained as part of an intravenous urogram study 15 minutes after contrast administration with 100 cc's of Isovue 370. Complete duplication of the urinary bladder is demonstrated (white arrows).



**Figure 5.** 31 year-old female with complete cloacal duplication. A coronal reformatted image from the CT scan described above demonstrates dilated pelvis and ureters, bilaterally (white arrows). Note that the duplication of the rectum is visible (black arrow).



**Figure 4.** 31 year-old female with complete cloacal duplication. CT images are obtained at the time of the patient's second presentation to the emergency department. The study is performed after intravenous administration of 100 cc's of Isovue 370 and Gastroview oral contrast. 3 mm slice thickness images are obtained (kVp=120, mA=100). Coronal (A) and transverse (B) images show the gravid uterus on the right, with a smaller, non-gravid uterus on the left (white arrows). Duplication of the urinary bladder is also evident (black arrows).

#### ABBREVIATIONS

CT: Computed Tomography  
kVp: peak kilovoltage  
mA: milliampere  
MHz: megahertz.

#### KEYWORDS

Complete genitourinary duplication, uterine anomalies, complete cloacal duplication, genitourinary duplication, cloacal duplication, caudal duplication syndrome, caudal duplication

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