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Tubular Colonic Duplication with Recto-Vestibular Fistula

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I. INTRODUCTION:

Duplication of the colon is a very rare congenital anomaly that can have different presentations depending on its location and size. It may occur in approximately 15% of all gastrointestinal duplications. The double termination of the tubular duplication of the colon in the perineum is even rarer. Over 80% of cases present before the age of 2 years as an acute abdomen or bowel obstruction. The most common site for duplication is the ileum, while colonic duplication is rare, accounting for 6-13% of all gastrointestinal duplications, commonly located in the cecum.

These lesions, if encountered incidentally, should be treated surgically to avoid future complications. Although congenital perineal or H-type fistula is a relatively common anomaly in Asian countries, its association with tubular Y-shaped colonic duplication is rare.

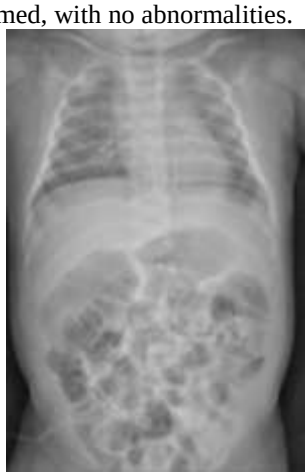
We report a case of colorectal duplication in a neonate with an occlusive syndrome, whose stool initially came from a very small amount of normal anal opening and a fistulous opening in the vestibule.

II. CASE REPORT:

A 2-day-old neonate was admitted to the neonatal intensive care unit for an occlusive syndrome with a bloated abdomen. Clinical examination revealed a vestibule with three openings: a normal urethral and vaginal opening and a recto vestibular fistula (Fig 1), with a normal anus.

Fig (1)

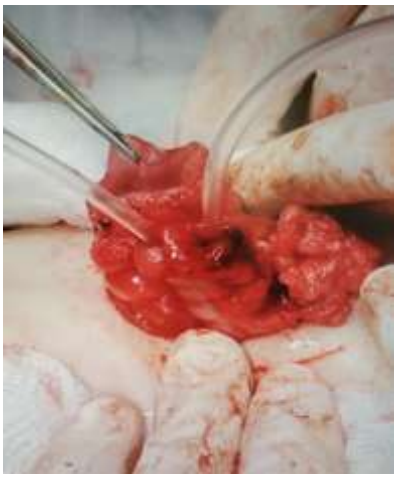
A thoraco-abdominal X-ray was performed, showing distension of the coves without clear visualization of hydroaerobic levels (Fig2). An abdominal, renal and cardiac ultrasound was then performed, with no abnormalities.



Fig(2)

After 24 hours, the patient's condition worsened, prompting the decision to perform an emergency colostomy.

Surgical exploration revealed the presence of a proximal and two distal lumina in the colon (Fig3).



Fig(3)

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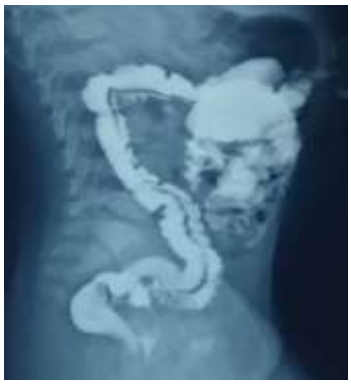
A distal end opacification was requested postoperatively, revealing a colonic duplication with a recto vestibular fistula.



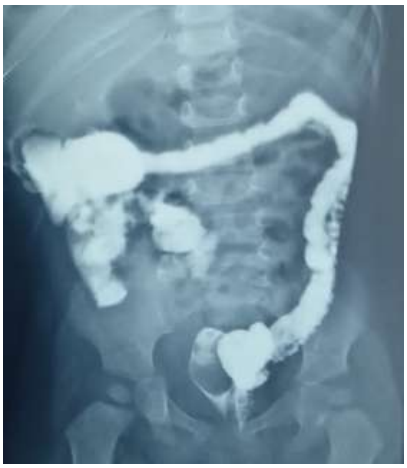


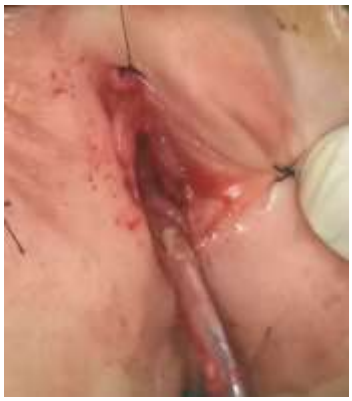
Fig (4)

After diagnosis, the baby was scheduled for surgery after 03 months.

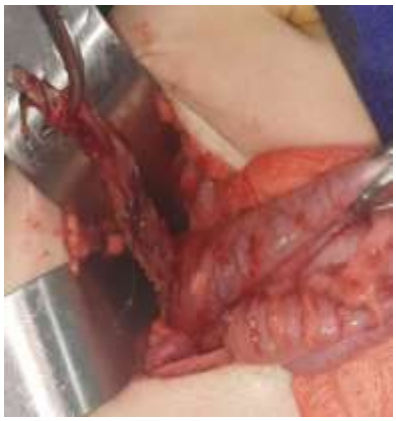
At 4 months of age, the patient was taken up for definitive surgical management. An abdomino-perineal approach was used, in which initially the colostomy was mobilized. For the distal duplicated colonic segment, the mucosal cuff of the colon leading to the vestibular fistula was dissected like a Soave's endorectal pullthrough from the distal stoma to the vestibular fistula and excised. The remaining muscular cuff was plicated and closed.

The proximal duplicated colonic segment shared the vascular supply with the native colon, hence resection was not possible. The duplicated segment ex- tended proximally till the ascending colon and common wall comprised of two layers of mucosa. Therefore, the intervening mucosal septum between the duplicated colon, upto the non-duplicated ascending colon was divided. A colo-colic anastomosis between the proximal unified colon with the distal native colon leading to the normal anus was done. The postoperative course was uneventful and the colostomy was closed after 3 months. At follow up of 6 months post-surgery, the opening in the vestibule had closed, with no bowel complaints.

Fig (5) Probing the recto-vestibular fistula



Fig(6) Sub-mucosal dissection of the fistula via the perineal route



Fig(7) Abdominal dissection and recovery of the distal tip

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Fig(8) Opening of the distal end of the anterior colon and laterally of the posterior colon showing the mucosal partition between the two + End-to-side anastomosis

III. DISCUSSION:

Duplications of the colon are rare malformations, either tubular or cystic in nature. An early aberration in the formation of the primitive hindgut is probably at the origin of a scission or twinning process, which results in a duplication of the terminal intestine with or without duplication of the genitourinary organs [6]. Duplications are double or Y-shaped. They have a double muscular layer and an epithelium similar to the rest of the colon [1,7]. The two lumens may be unobstructed and function normally as two perineal anuses, or terminate distally blind as an anus imperforate with one or both lumens, or the ventral colon terminates in a recto-urinary, recto-vaginal or vestibular fistula[7]

Most authors recommend that once the diagnosis has been made, elective surgery should be performed in the patient's optimal condition to avoid complications: it is the excision of the duplicated colon. Colorectal duplications are essentially benign lesions and radical surgical excision is not necessary. The need for surgery in asymptomatic cases is debatable.

Resection of the split colon is generally not possible because of the common blood supply to both colons via a single mesentery [8]. In our case, separation of the two proximal colonic segments was impossible because, in addition to the common blood supply, the intermediate wall consisted solely of mucosa. In addition, excision of the common wall resulted in a disparity in calibre between the proximal and distal loops.

Possible therapeutic attitudes in unresectable proximal duplications of the colon include division of the septum to convert it into a common colonic duct, long lateral anastomosis of the adjacent duplicated bowel; Excision of the distal common septum with creation of a common duct should prevent accumulation of faecal material in the false necks or performance of a mucosectomy on a limb of the proximal duplication [10-13]

IV. CONCLUSION:

Endorectal resection of the duplicated segment allows the condition to be treated effectively without compromising the continence mechanism.

Resection of the duplicated colon may not be possible due to the common blood supply between the duplicated segments.

Tubular duplication of the colon should be considered as one of the differential diagnoses with perineal canal in cases of vestibular fistula with a normal anus.

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