Case report

Cecal duplication: A mimicker of intussusception
A case report and review

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ABSTRACT

Introduction: Cecal duplication is a rare congenital anomaly and to the best of our knowledge, only 43 cases have been reported in the literature till date. Most of them present within the first year of life. They can mimic intussusception, and the delay in diagnosis can lead to high morbidity.

Case report: A five-year boy presented with pain abdomen for a week. He was found to have ileocolic intussusception. The intussusception could only be partially reduced by the hydrostatic method. On laparotomy, a submucosal solid mass was found in the cecum with multiple enlarged lymph nodes. Mass was resected with clear margins and lymph nodes sampled. Histopathology was conclusive of cecal duplication. Post-operative course was uneventful, and the child is thriving well, last reviewed at three-year follow-up.

Conclusion: Incomplete reduction of intussusception, intussusception with atypical presentation or intussusception in atypical age group should alert to the possibility of cecal duplication.

1. Introduction

The cecum is one of the rarest sites for duplication cysts [1]. They vary greatly in their presentation [2]. The frequent presentations include pain abdomen, distension of abdomen, palpable mass, and bleeding per rectum. The majority of the duplications are diagnosed within two years of life, with most of them diagnosed antenatally [2]. To the best of our knowledge, there are only 43 cases of cecal duplications have been reported in the literature till date [3]. We present a case of a five-year male child with duplication of cecum who presented to our institution (a tertiary center) with intussusception. The work has been reported in line with the SCARE criteria [4].

2. Case report

A five-year-old male child presented with a history of pain abdomen for a week, with no history of distension of abdomen or vomiting. There was no history of blood in stools, loose stools or constipation. His pulse rate, blood pressure, and respiratory rate were unremarkable. On abdominal examination, no mass was palpable. Rectal examination revealed no mass or bleeding. Ultrasonography of abdomen & pelvis showed an ileocolic intussusception. The intussusception could only be partially reduced by the hydrostatic method (as per protocol used by Bai YZ et al. [5]), hence we proceeded to laparotomy. Intra-operatively, a submucosal solid mass measuring 4 cm x 4 cm was found in the cecum with multiple enlarged lymph nodes. The mass was resected with clear margins and lymph nodes sampled. Histopathology was conclusive of cecal duplication. Post-operative course was uneventful, and the child is thriving well, last reviewed at three-year follow-up. Consent was obtained from parents for possible publication.

3. Discussion

Gastrointestinal duplications are rare congenital malformations which can occur in any part of the alimentary tract from mouth to anus [3]. The word “intestinal duplication” was first used by Fitz and popularized by Ladd in 1930s. Gross classified intestinal duplication in 1950 [3].

As per the Ladd and Gross, characteristics of intestinal duplications [1–3] are-
Four (10%) patients had associated anomalies with two of them had duplication of the whole colon, one duodenal duplication and the other had a vestibular fistula and thoracic meningocoele. Our patient didn't have any associated anomaly.

Enteric duplication cyst can present at any age, but 80% present within two years of life [1]. Our review showed 67% presented within nine months of life. Only two (5%) cases of cecal duplications have been diagnosed antenatally to date.

Females are most commonly affected (67%) than males.

Symptoms differ depending on the size, morphology, and location of the cysts [6]. Vomiting was the most common presentation and was seen in 25 (57%) patients. Twenty (45%) patients presented with a palpable mass, 18 (41%) had abdominal distension, 15 (34%) had pain abdomen, 8 (18%) had blood in stools, and 6 (14%) had constipation. They may present with intestinal obstruction due to the cyst acting as a lead point for intussusception (10 patients), volvulus around the cyst (1 patient) or the cyst itself completely obstructing the lumen (1 patient) [1,3,6,7]. They can have malignant transformation occasionally, and there are two reported cases of adenocarcinoma [8,9] and a case of carcinoid from the cyst wall.

Communication between the gastrointestinal tract and the duplication cyst was found in 5 (11%) cases, and ectopic gastrointestinal mucosa was found in only 1 (2%) patient.

Ultrasoundography is the investigation of choice with a specificity of more than 95% and a positive predictive value of 85–100% [3]. But it's highly operator dependent. There were four cases, apart from our case, reported in the literature with preoperative ultrasonography showing intussusception but same couldn't be found intra-operatively [2,3]. There were a total of ten cases of cecal duplications, which presented with radiological intussusception [2,3]. Among these, four cases underwent hydrostatic reduction, and all these had either a failed reduction or incomplete reduction. And the rest of the patients underwent laparotomy on suspicion of a lead point. Computed tomography is not required in most of the cases as ultrasonography has high specificity.

Technetium pertechnetate scan is helpful to pick up the ectopic gastric mucosa present in the duplication cyst [7].

Eighteen patients (41%) were diagnosed to have cecal duplication either during surgery or after examining the resected specimen. The differential diagnoses include Appendicitis, intussusception, mesenteric cyst, ovarian cyst, choledochal cyst, pancreatic cyst, and lymphoma.

Resection and end to end anastomosis is the treatment of choice. Excision was possible in only one case. The prognosis is very good. Two mortalities have been reported in the literature; both were secondary to sepsis.

4. Conclusion

Duplication cyst of the cecum is a rare entity with only a few cases reports reported previously. It's more common in females. Incomplete reduction of intussusception, intussusception with atypical presentation or intussusception in atypical age group should alert to the possibility of cecal duplication. Resection is the optimal treatment, and it has an excellent long-term outcome and quality of life.

Ethical approval

It’s a case report. No study was conducted. Consent was taken from parents of all the patients for publication.

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“The manuscript has been read and approved by all the authors.”
Author contribution

Dr. Veerabhadra conceptualized and designed the study, conducted the analyses, drafted the initial manuscript, and made final revisions based on critical feedback received from Dr. Ashok Rijhwani and Dr. Bhushan Jadhav.

Dr. Ashok Rijhwani conceptualized the study in collaboration with Dr. Veerabhadra and Dr. Bhushan, reviewed the results, and reviewed and provided critical feedback for the manuscript.

Dr. Bhushan Rao Jadhav conceptualized the study in collaboration with Dr. Veerabhadra and Dr. Ashok Rijhwani, reviewed the results, and reviewed and provided critical feedback for the manuscript.

All the authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Conflicts of interest

The authors have no conflicts of interest relevant to this article to disclose.

Research registration number

Not applicable.

Guarantor

Veerabhadra Radhakrishna.

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