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Horseshoe Appendix: Report of Extremely Rare Congenital Anomaly with Review of Literature

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Abstract: ***The horseshoe anomaly of the vermiform appendix is extremely rare. Preoperative confirmation of this anomaly is difficult; therefore, routine procedures, such as appendectomy, may become unexpectedly challenging when such anomalies are encountered during the surgical process. Here, we report a 25 year old who underwent emergency appendectomy. Intraoperatively both the ends of the appendix were found to be communicating with the cecum with two separate base or stump - the so called "horseshoe appendix".***

Keywords: horseshoe appendix, vermiform appendix anomaly, emergency appendectomy, surgical challenges, cecum communication

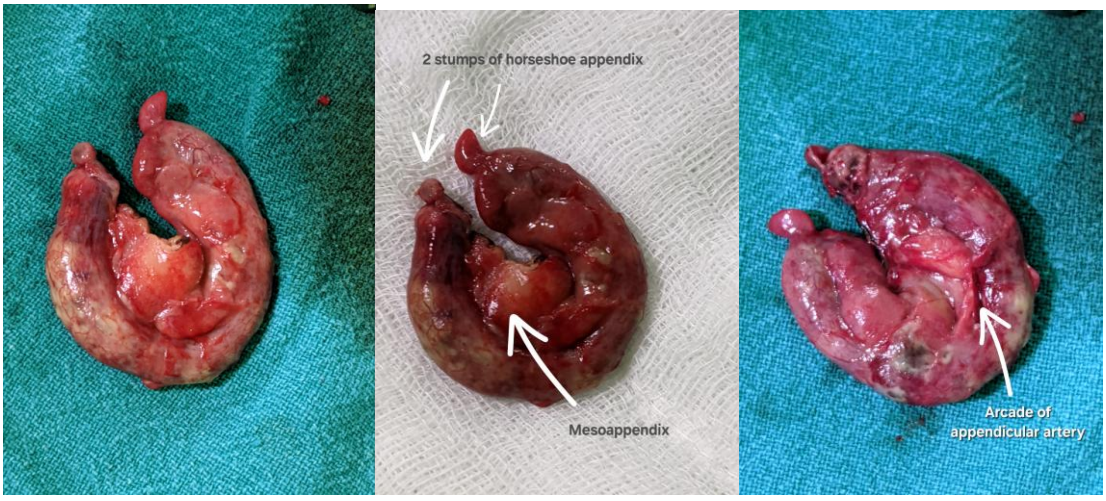
1. Introduction

Acute appendicitis is a very common disease [1] Appendiceal anomalies are extremely rare malformations that are usually found incidentally. In this report, we present a case of a horseshoe appendix in 25 year old male that was incidentally found during emergency appendectomy.

2. Case Report

A 25 year old male came to emergency department with 2 days history of pain in the abdomen associated with vomiting and fever since 1 day. On examination abdomen was soft, non-distended with rebound tenderness in right iliac fossa. There was pain in lower abdomen on coughing. On investigations Total leucocyte count was 16000, Ultrasound suggestive of blind, non - compressible tubular structure measuring 10 mm in diameter with surrounding inflammatory changes. Diagnosis of acute appendicitis was established and patient was planned for emergency surgery

The taenia coli were traced down to its confluence and the base was identified. Attempt was made to release the tip but found out that it was communicating with the cecum by another stump. The appendix was supplied by a single blood vessel in the mesoappendix as shown in the figure. An appendectomy with control of both appendicular bases was performed. There were no complications intraoperatively and in the post op period. Patient was discharged on the 5th postoperative days on oral antibiotics



3. Discussion

Anomalies of the appendix are extremely rare, and a horseshoe appendix is even rarer. Duplication of the vermiform appendix is an extremely rare malformation with an estimated incidence of 0.004% [2]. It was first described by Picoli in 1892 [3]. The aetiology of double appendix or horseshoe appendix is still unclear because of its rarity and the extremely limited cases that are reported so far. Some theories that has been put forward is that the horseshoe appendix may develop from the fusion of the tip of the normal appendix with that of the variant appendix which is at the abnormal site thereby giving rise to the so called horseshoe anomaly or, the other possible explanation could be that the horseshoe appendix develops from the fusion of the tip of the normal appendix with another part of the caecum which later on become the second base. However, these theories don't seem to explain why the appendix was supplied by a single blood vessel in the mesoappendix with its tributaries spreading out rather than forming an arcade on the inner aspect of the horseshoe as seen in the cases reported. Thus, the most likely explanation could be that it is due to some abnormality arising during the embryologic period, which could be perhaps during the embryologic life, the base of the appendix somehow split in two, and during the course of development and cecal growth gets separated further leading to a double - based, yet single structure [4], [5]

In 1963 Wallbridge updated and modified Cave's classification [6]. However, horseshoe appendix doesn't appear in this classification system. Wallbridge classified appendiceal duplication into;

- Type A: A single appendix base with various degrees of duplication of the tip.
- Type B: Two separate appendixes from one caecum.
- B_1 - Two appendixes on either side of the ileocecal valve, similar to the arrangement found in birds "Bird like". This type of appendiceal abnormality is usually associated with other gastrointestinal (Including hindgut) mal development (ileum, colon, anus) and urinary malformation.
- B_2 - There are two appendixes, one at the normal site, and the other usually on the taenia in the caecum at a varying distance from the first, also known as "Taenia - coli type". This type of anomaly is not usually associated with other congenital anomalies.
- Type C: Two caecums, each with its own appendix.

Biermann in 1993 classified appendiceal anomalies into [7]:

- A: Partial duplication of the appendix on a single cecum;
- B: Two completely separate appendices on a single cecum with two subtypes:
- B1: "bird - like appendix" or "avian type": Two appendices symmetrically placed on either side of the ileocecal valve. This type is found normally in birds. In humans it is found associated with intestinal and/or genitourinary anomalies.
- B2: "Taenia - coli type": One appendix arises from the usual site on the cecum with another rudimentary arising from the cecum along the tenia of the cecum.
- B3: The second appendix is located along the tenia of the hepatic flexure of the colon.
- B4: The second appendix is located along the tenia of the splenic flexure

The later three (B2, B3 and B4) are usually not associated with other congenital anomalies.

- C: Two caecum, each bearing an appendix. This type occurs in association with hindgut mal development (ileum, colon, anus) and other anomalies of genitourinary tract and lower vertebral column.
- D: Three completely separate appendices with or without other anomalies.

Calota F et al., classified appendiceal anomaly into "Number anomalies" and "Shape Anomalies" [8].

Number Anomalies

1) Congenital Agenesis
2) Multifarious appendices
   a) A: Appendiceal duplication (Partially) - "Y shaped"
   b) B: Duplex Appendix on a single cecum:
      - B_1 - "Avain type" with intestinal and/or genitourinary anomalies
      - B_2 - "Tenia - coli cecum type"
      - B_3 - "Tenia coli hepatic flexure type".
      - B_4 - "Tenia coli splenic flexure type".
   c) C: Duplex Appendix on two cecum (with hindgut, genitourinary tract, lower vertebral column maldevelopment.
   d) D: Triplex appendix:
      - "Newborn type" with/without other congenital anomalies.
      - "Adult type" without other congenital anomalies.

Shape Anomalies

Horseshoe shaped appendix:

- With Frontal disposal
- With Sagittal disposal

The evaluation of the cecum must be rigorously performed to avoid any forensic problems if a new inflammation affects the remaining annex [9, 10].

In our case, the radiology team suspected acute appendicitis, and on surgical exploration found horseshoe appendicitis with a single vessel on the mesoappendix.

4. Conclusion

Surgeons should be aware that the appendix can have rare and unusual abnormalities. These anomalies can significantly affect the outcome of surgical procedures

This case describes a very rare and exceptional case of Horseshoe appendix. As it is extremely rare, most surgeon will never encounter this unusual condition, which promotes better understanding and optimal patient care

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