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RESEARCH ARTICLE

A RARE CASE OF APPENDICEAL INTUSSUSCEPTION

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Abstract

Introduction: Appendiceal intussusception is a very rare condition (0.01%) found in patients undergoing appendectomy. It happens when the appendix is pulled into itself or into the caecum. Endometriosis of the appendix is particularly a rare cause of appendiceal intussusception; in the past 50 years, less than 30 cases have been reported in the literature. Clinical symptoms vary and some cases are asymptomatic.

Case Presentation: A 25 years old, female presented with complaints of periumbilical pain for 4-5 days, 6-7 episodes of bilious vomiting and constipation for 3 days. On examination, the abdomen was soft, mild tenderness was present in the periumbilical region. Patient was stable clinically. Ultrasound abdomen showed possibility of Ileo-colic Intussusception and intramural fibroid and CECT abdomen was suggestive of ileo-ileal intussusception and multiple uterine fibroids. Patient underwent Exploratory Laparotomy. Appendiceal intussusception was found in the caecum. Appendix and a part of caecum were found thickened and gangrenous. Ileocaecal junction and rest of the bowels were normal. Patient underwent ileocaecal resection with primary anastomosis.

Results: Histopathological report of specimen was consistent with findings of endometriosis of appendix with peri appendicular abscess and adenomatous changes in caecum.

Conclusion: Appendiceal intussusception to the caecum caused by endometriosis of the appendix is a very rare(0.01%) condition and difficult to diagnose preoperatively. It is usually diagnosed intra-operatively. Appendiceal intussusception due to endometriosis can

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only be confirmed by histopathology.

Introduction:-

Intussusception is telescoping or invagination of one segment of bowel into the adjacent segment. Intussusception can cut off the blood supply to the affected part of the intestine, which can cause infection, infarction or necrosis of bowel tissue or perforation in the bowel. In children younger than 3 years old, the most common cause of intestinal obstruction is intussusception, where the most common cause remains idiopathic. It is rare in adults, but in adults, most cases are due to an underlying medical condition, such as a tumor.

Appendiceal intussusception is a very rare disease, there could be partial telescoping of appendix or entire colon can be involved, where appendicular protrusion from anus can occur. It is found in only 0.01%[**1]** patients who underwent an appendectomy. It is a difficult clinical or radiological preoperative diagnosis, seen more often in the first decade and in males.[**2]** Appendiceal intussusception occurs due to the appendix being pulled into itself or into the caecum. Due to anatomical or pathological factors, there can be abnormal appendiceal peristalsis which leads to its invagination into the caecum. It is very difficult to diagnose in pre-operatively and colonoscopy and CT scan usually provides aid to diagnosis. It should not be tried to reduce by colonoscopy. It can mitigate various abdominal conditions.

Appendiceal intussusception caused due to endometriosis of the appendix is a particularly rare cause, with less than 30 cases in the literature in the last 50 years,[**3]** it can also occur due to mucocele (19%).[4**]** It is due to extragonadal endometriosis, commonly childbearing age group females are affected with a broad spectrum of clinical features. It could be mistaken for a cecal mass, hence it should be kept as differential in such cases. Intussusception. Clinical symptoms are varied, some patients may be asymptomatic, can have vague symptoms or may present with features of intestinal obstruction. We present a case of appendiceal intussusception presenting with vague symptoms of periumbilical pain, nausea, vomiting and constipation.

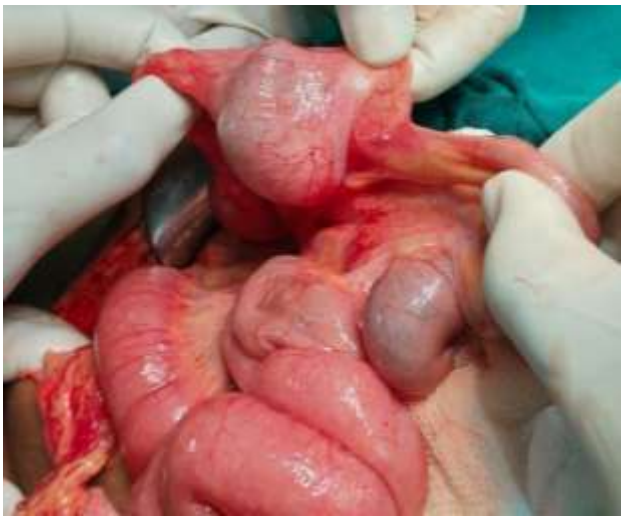
Case Presentation:

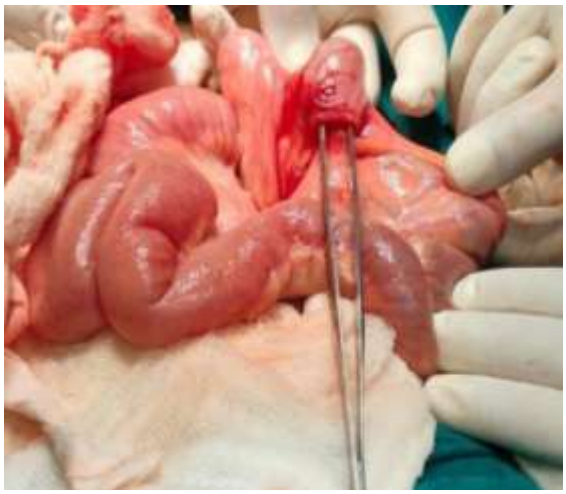
A 25 years old, female patient presented with chief complaints of periumbilical pain for 4-5 days, with nausea, 6-7 episodes of bilious vomiting,and constipation for 3 days. There was no history of abdominal fullness, tuberculosis or other comorbidities. On examination, the abdomen was soft, mild tenderness was present in the periumbilical region. Patient was vitally and clinically stable.

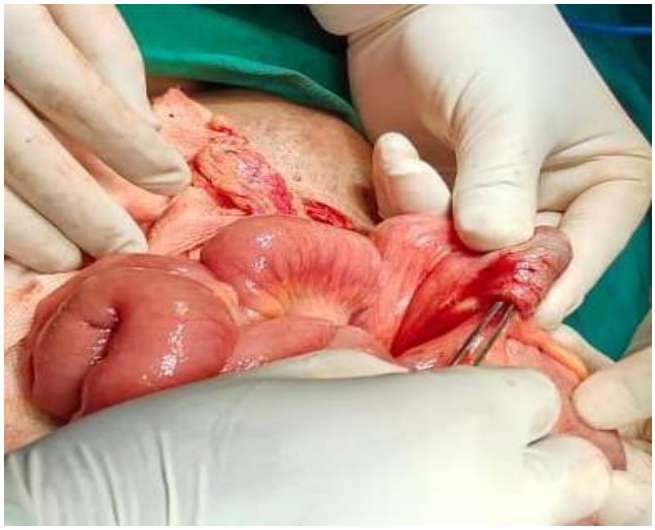
On investigations, complete blood counts, liver function test, renal function test and coagulation profile were within normal limits. Ultrasound abdomen showed approx 3.5cm sized short segmental, small bowel loops (ileal loops) giving bowel in bowel appearance in the right iliac fossa region, suggestive of Ileo-colic Intussusception. There were multiple hypoechoic lesions without internal vascularity in the anterior and posterior wall of the uterus, seen on USG which was suggestive of intramural fibroid. CECT abdomen showed presence of invagination of pelvic ileal loop on right side into the adjacent ileal loops giving rise to target sign, approx length of invaginated segment measure 37mm, no evidence of any bowel obstruction, suggestive of pelvic ileo-ileal intussusception and presence of uterine fibroids was confirmed on CECT.

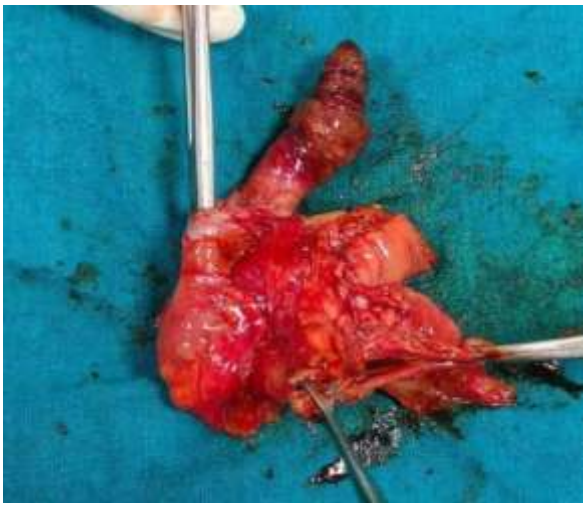
Patient underwent Exploratory Laparotomy, Appendiceal intussusception was found in the caecum. Manual reduction of intussusception was tried, but could not be reduced. Appendix and a part of caecum were found thickened and gangrenous. Ileocaecal junction was normal. All other bowel loops collapsed and no abnormalities were found. Patient underwent ileocaecal resection and primary anastomosis done between ileum and ascending colon.

Post op course was uneventful. Histopathological report of specimen showed appendix endometriosis with peri appendicular abscess and adenomatous changes in caecum.

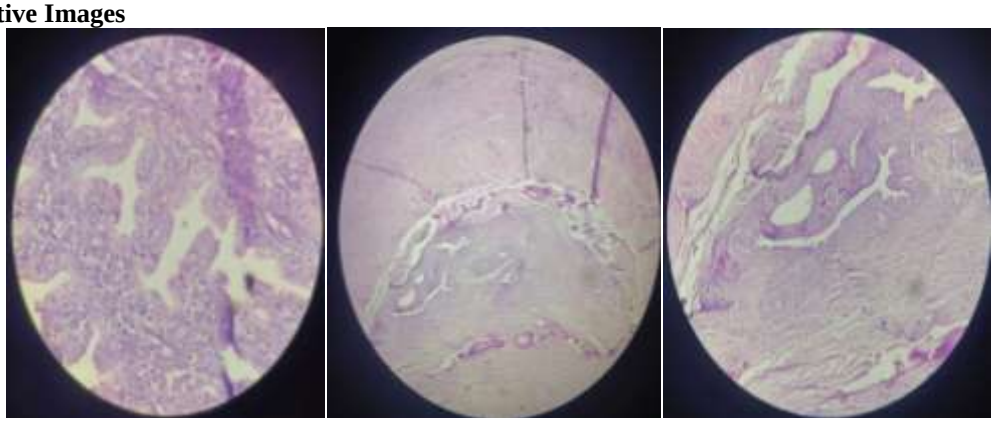




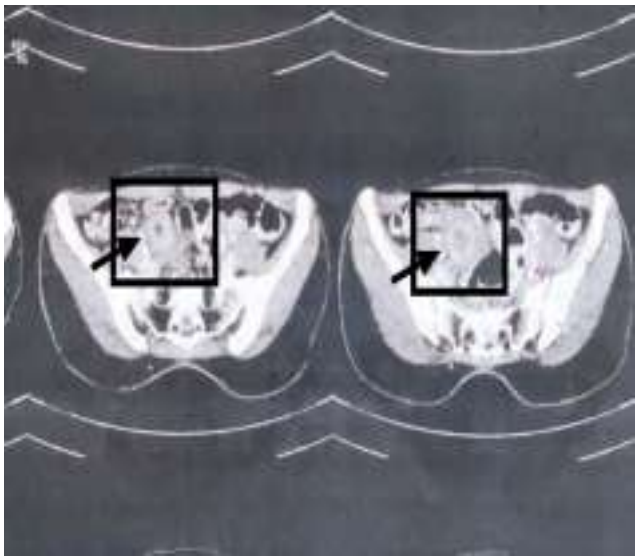




Intraoperative Images



Histopathological Images



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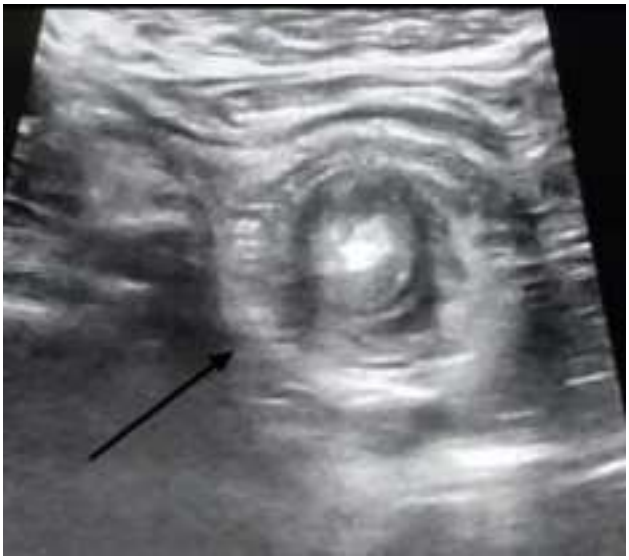

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Discussion:-

Appendiceal intussusception is a rare disease with an incidence of 0.01% as reported by Collins.[**1]** Appendiceal intussusception occurs due to the appendix being pulled into itself or into the caecum. Due to anatomical or pathological factors, there can be abnormal appendiceal peristalsis which leads to its invagination into the caecum. This condition is characterized by abnormal appendiceal peristalsis due to anatomical or pathological factors that lead to invagination of the appendix into the caecum. Mesoappendix can be mobile, thin appendix or there can be proximal wider lumen of appendix, which constitute anatomical factors.[**5,6,7]** Adenocarcinoma, carcinoid tumor, mucocele, polyps, parasites, endometriosis and lymphoid hyperplasia may be the pathological factors for intussusception acting as lead point.[1,7-**9]**

Clinical features varies from some cases being asymptomatic to some having acute or non-specific chronic (63%) symptoms. Most patients present with abdominal pain (78%), vomiting (26%) and melaena or hematochezia (23%).[**8]** Incidence of endometriosis involving gastrointestinal tract is 15-37%, rectum and the rectosigmoid junction (72%) being the most commonly affected, however appendix is involved in only 3% of cases.[**10,11]** Serosa and muscular layer of the intestinal tract is often affected by endometrial tissue, which shows presence of

stroma, fibrosis and hemosiderin-laden macrophages. Fibrosis and adhesions can occur due to serosal involvement and implants can lead to hyperplasia of the muscular layer, which leads to constriction of the appendiceal lumen and can mimic a mass. Hyperperistalsis can occur due to it, and it becomes a lead point, resulting to appendiceal intussusception into the caecum.[**10,12]** Appendiceal intussusception is a diagnostic challenge for surgeons, and is often diagnosed intraoperatively or postoperatively. Pre-operative imaging is important in deciding surgical management. Pathognomonic signs include a target sign or a concentric central mass, or bull's eye sign on CT, while ultrasonography will show an onion-skin like lesion, or pseudokidney sign or multiple concentric hyperechoic and hypoechoic rings, Barium enema shows typical claw sign or coiled spring sign (pincer end). On colonoscopy, lesion appear as a polypoid mass covered in normal mucosa and a central dimple at the appendiceal orifice.[**10,12]** In some cases, diagnosis is made only during surgery, in such cases good surgical judgment becomes important.

The chances of developing secondary ileocaecal intussusception increases due to appendiceal intussusception, it should be managed surgically. [**6]** The most common procedure performed is Appendectomy (42%), 2nd most commonly done is ileocecectomy (27%) and often right hemicolectomy (21%) is needed.[**8]** When appendix seems to be benign and malignancy is not suspected, usually the treatment of choice is appendicectomy. [**13,14]** However, when appendiceal mass or caecal malignancy is suspected then exploratory laparotomy should be done. When the histopathological report is not available and intraoperatively suspicion for malignancy is there, right hemicolectomy should be done. [**13]**

Endometriosis can lead to an ileocolic intussusception also, which shows the same clinical presentation as of appendiceal intussusception and approach to management remains same, as documented in a few published articles, and thus being a differential diagnosis. Patients presenting with lower abdominal or hypogastric pain, a palpable mass and stool for occult blood positive, should provide high suspicion for ileocolic intussusception, as surgical entity is implied by these signs and symptoms. Of all cases of ileocaecal intussusception due to endometriosis, two patients required ileocaecal resection, as there was large polypoid tumor below the ileocecal valve, in the medial part of the caecum, which was revealed as a cecal polyp on colonoscopy, and was taken as an inverted appendix with a tumor, as computed tomography also showed an invaginated appendix into the lumen of ceacum, the other two underwent right hemicolectomy, where appendiceal intussusception due to appendiceal mucocele into the caecum and secondary distal ileum invagination was found, the tumor was found to be involving the whole appendix till its base. Both appendiceal and ileocolic intussusception due to endometriosis are rare conditions, both requiring the same approach for diagnosis and surgical management.

Conclusion:-

Appendiceal intussusception into the caecum is a rare cause of abdominal pain and difficult to diagnose preoperatively. Most of the cases are diagnosed intraoperatively. Patients may present with vague symptoms or may present with features of intestinal obstruction. Endometriosis as a cause of appendiceal intussusception is a very rare condition that can be diagnosed by histopathology.

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