Reducible appendiceal intussusception: A case report and review the literatures

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ABSTRACT

Appendiceal intussusception is a very rare cause of abdominal colic in childhood. This paper reports a reducible appendiceal intussusception in the barium enema in a girl and reviews articles in PubMed.

Introduction

Appendiceal intussusception is a rare condition which has been diagnosed in case reports. 1,2, 3 The incidence rate of appendiceal intussusception is 0.01% in 11000 human appendix specimens. 1, 2 This condition primarily occurs in children. 2, 3 In most cases, intussusception is limited to appendix but in some conditions cecocolic or ileocolic intussusception may occur. 4 A variety of underlying anatomical and physiopathological causes as lead point are described. 2, 9 So far, several forms of clinical presentations and classifications have been reported. 2, 3 Diagnosis of appendiceal intussusception is by ultrasound, barium enema, CT scan and colonoscopy 2, 10-13 and the main routes of treatment are hydrostatic reduction and surgery. A complete review of appendiceal intussusceptions of childrens in pubmed also was performed. The age, sex, presentation, treatment and pathology were reviewed on childrens. 14-25

Case presentation

Case report

A four-year old girl referred to our hospital with a history of one week abdominal pain, intermittent nausea, agitation, vomiting and

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Appendiceal intussusception was reduced. The patient was followed and by resolving the symptoms (after four days) she was discharged. She was not improvement of symptoms after one year follow up.

**Results**

The age, sex, clinical findings, diagnostic imaging, treatment and pathology were evaluated at case reports. The patient age varies from 2month to 15years with average age of 6.9 years (table-1). Male patients (71.4 %) more affected than female patients (29.6 %)(Table2). The most clinical findings were abdominal pain (90%) (Table3). The most of case reports were diagnosed at preoperative imaging procedures (83 %).The most common pathologic finding was inflammation (42%)(Table-4). Appendectomy was performed in most of the case reports. Surgical Procedures are open (mostly) or laparoscopic .Partial colectomy also wereachieved at patient in appendiceal intussusception without reduction or relapse.

**Discussion**

Appendiceal intussusception is very uncommon\(^1,2\) which is seen more in the first two decades of life.\(^26\) The first case of appendiceal intussusception was reported in 1858. Half of pediatric cases occur in children less than 10 years of age and the boy/ girl ratio occurrence is 4-5/1. Intussusception of the appendix probably occurs by the same mechanism and pathogenesis as intussusception elsewhere. Appendiceal intussusception mostly simple (primary) form intussusceptions of appendicular mucosa (ie, only spontaneously invagination of appendix into cecum) in some instances might be associated with secondary cecocolic or ileo colic intussusception.\(^1, 26, 27\) Primary type occure by either partial and complete invagination of the appendix into itself.
<table>
<thead>
<tr>
<th>Author</th>
<th>G</th>
<th>Age</th>
<th>Time of onset</th>
<th>Clinical findings</th>
<th>Diagnostic procedure</th>
<th>Treatment</th>
<th>pathology</th>
<th>year</th>
</tr>
</thead>
<tbody>
<tr>
<td>Dunavant</td>
<td>M</td>
<td>8mo</td>
<td>6d.</td>
<td>Hematochezia, Inversion of appendix with anal protrusion</td>
<td>*AI</td>
<td>Surgical reduction, Appendectomy</td>
<td>-</td>
<td>1950</td>
</tr>
<tr>
<td>Atkinson</td>
<td>F</td>
<td>2.5y</td>
<td>1mo.</td>
<td>Cramp/constipation, RLQ tenderness</td>
<td>*BE: AI without reduction</td>
<td>Appendectomy</td>
<td>Lymphoid Hyperplasia</td>
<td>1976</td>
</tr>
<tr>
<td>&quot;</td>
<td>F</td>
<td>4.5yr</td>
<td>3wk</td>
<td>Cramping /vomiting</td>
<td>*US: Soft tissue/mass in RLQ BE: AI without Appendix filling</td>
<td>Right hemicolecotomy inverted Appendix</td>
<td>&quot;</td>
<td>1976</td>
</tr>
<tr>
<td>Kleinman</td>
<td>M</td>
<td>15yr</td>
<td>4wk</td>
<td>Cramping/ RLQ tenderness</td>
<td>*SBT: Filling defect in cecum BE: Reduction with Appendix filling</td>
<td>Ba.reduction</td>
<td>-</td>
<td>1979</td>
</tr>
<tr>
<td>Casteel</td>
<td>F</td>
<td>14yr</td>
<td>-</td>
<td>Recurrent /abdominal pain/ vomiting</td>
<td>BE: Spiral shell sign (AI)</td>
<td>Appendectomy</td>
<td>-</td>
<td>1986</td>
</tr>
<tr>
<td>Bailey</td>
<td>M</td>
<td>7yr</td>
<td>-</td>
<td>Recurrent abdominal pain</td>
<td>Colonoscopy Inverted appendix</td>
<td>Polypectomy, Appendectomy</td>
<td>Juvenile cecal polyp</td>
<td>1987</td>
</tr>
<tr>
<td>Galatito</td>
<td>F</td>
<td>27m</td>
<td>-</td>
<td>RLQ pain</td>
<td>US: Target sign in RLQ</td>
<td>Laparoscopic Reduction</td>
<td>-</td>
<td>1999</td>
</tr>
<tr>
<td>Pumberger</td>
<td>M</td>
<td>4yr</td>
<td>3wk</td>
<td>Colicky /RLQ mass, RLQ Tenderness</td>
<td>US: RLQ, multicentric Ring sign WSCM enema reduction (AI)</td>
<td>Appendectomy after recurrence, Chronic inflammation</td>
<td></td>
<td>2000</td>
</tr>
<tr>
<td>Pumberger</td>
<td>5yr</td>
<td>2dy</td>
<td>Pain/ Hematochezia</td>
<td>US: Target sign, Wsmcavenua No reduction(AI)</td>
<td>Appendectomy</td>
<td>Chronic inflammation</td>
<td></td>
<td>2000</td>
</tr>
<tr>
<td>Gupta</td>
<td>M</td>
<td>9yr</td>
<td>1mo</td>
<td>Hematochezia</td>
<td>Colonoscopy /AI</td>
<td>Appendectomy, Partial cecectomy</td>
<td>Resolving infectious process</td>
<td>2000</td>
</tr>
<tr>
<td>Koumanidou</td>
<td>M</td>
<td>6yr</td>
<td>1wk</td>
<td>RLQ pain /Nausea/ vomiting</td>
<td>US: Donut – Like mass in RLQ BE: Coilspring /nonfilling appendix</td>
<td>Appendectomy</td>
<td>Lymphoid Hyperplasia</td>
<td>2000</td>
</tr>
<tr>
<td>Koumanidou</td>
<td>M</td>
<td>5yr</td>
<td>5dy</td>
<td>RLQ pain /Diarrhea / RLQ Tenderness</td>
<td>US: Donat Like mass in RLQ BE: Coilspring</td>
<td>Appendectomy</td>
<td>Lymphoid Hyperplasia</td>
<td>2000</td>
</tr>
<tr>
<td>Koumanidou</td>
<td>M</td>
<td>5yr</td>
<td>7wk</td>
<td>Periumblical pain/ RLQ tenderness</td>
<td>AXX: Normal US: Donat Like mass in RLQ BE: AI</td>
<td>Appendectomy</td>
<td>Lymphoid Hyperplasia</td>
<td>2000</td>
</tr>
</tbody>
</table>
### Table 1: Case Reports of Appendiceal Intussusceptions in Children

<table>
<thead>
<tr>
<th>Author</th>
<th>G</th>
<th>Age</th>
<th>Onset</th>
<th>Clinical Findings</th>
<th>Diagnostic Procedure</th>
<th>Treatment</th>
<th>Pathology</th>
<th>Year</th>
</tr>
</thead>
<tbody>
<tr>
<td>Koumanidou</td>
<td>F</td>
<td>7yr</td>
<td>3dy</td>
<td>Abdominal pain/ Nausea/ Vomiting/ RLQ tenderness</td>
<td>AXR-Bowel obstruction US: Donut Like mass in RLQ Ba – Reduction</td>
<td>Appendectomy</td>
<td>Maltoma</td>
<td>2005</td>
</tr>
<tr>
<td>Karabulut</td>
<td>M</td>
<td>8yr</td>
<td>24hr</td>
<td>Abdominal pain/ Nausea/ Vomiting/ RLQ tenderness /RLQ tenderness RLQ mass</td>
<td>AXR-Normal US: AI Ba – AI irreducible</td>
<td>Appendectomy</td>
<td>Maltoma</td>
<td>2005</td>
</tr>
<tr>
<td>Ram</td>
<td>M</td>
<td>6yr</td>
<td>8wk</td>
<td>Periumblical pain/ Constipation/ vomiting/ Hematochezia /RLQ mass</td>
<td>US: No mass colonoscopy: mass Diathermy (with/relapse) Right hemicolecetomy CF(inverted appendix)</td>
<td>Appendectomy Distal cecectomy</td>
<td>-</td>
<td>2005</td>
</tr>
<tr>
<td>Luzier</td>
<td>M</td>
<td>8yr</td>
<td>1wk</td>
<td>Periumblical pain/ Constipation/ vomiting/ RLQ mass</td>
<td>US: Donut like mass in RLQ</td>
<td>Laparoscopic Appendectomy</td>
<td>-</td>
<td>2006</td>
</tr>
<tr>
<td>Baeza-Herrea</td>
<td>F</td>
<td>2mo</td>
<td>-</td>
<td>Sever RLQ pain</td>
<td>US: RLQ mass</td>
<td>Appendectomy</td>
<td>Appendix torsion</td>
<td>2006</td>
</tr>
<tr>
<td>Lipskar</td>
<td>M</td>
<td>6yr</td>
<td>1dy</td>
<td>Cramp/ RLQ pain/ Tenderness/ Nausea/ Vomiting</td>
<td>US: AI CT: RLQ mass</td>
<td>Appendectomy partial cecectomy (After recurrence)</td>
<td>Lymphoid Hyperplasia</td>
<td>2008</td>
</tr>
<tr>
<td>Wang</td>
<td>M</td>
<td>10yr</td>
<td>10dy</td>
<td>Periumblical pain/ Periumblical tenderness</td>
<td>AXR: RLQ mass US: Swellenappedexi/AI</td>
<td>Appendectomy</td>
<td>Burkitt's Lymphoma</td>
<td>2010</td>
</tr>
<tr>
<td>Chang</td>
<td>M</td>
<td>30mo</td>
<td>1dy</td>
<td>Fever/ vomiting/ RLQ mass/ RLQ tenderness</td>
<td>Air contrast enema Reduce AXR: Appendicolsits US: Target sign/ Appendicitis</td>
<td>Appendectomy (After recurrence of AI)</td>
<td>Appendicitis perforation</td>
<td>2011</td>
</tr>
<tr>
<td>Echert</td>
<td>M</td>
<td>12yr</td>
<td>-</td>
<td>RLQ Pain/ Hematochezia</td>
<td>Us: Target sign in RLQ</td>
<td>&quot;</td>
<td>Ulcerative colitis</td>
<td>2012</td>
</tr>
<tr>
<td>Epifanio</td>
<td>M</td>
<td>6yr</td>
<td>15hr</td>
<td>Abdominal pain/ vomiting/ RLQ tenderness</td>
<td>US: Target sign in RLQ BE: AI reduction Colonoscopy: polypoid lesion</td>
<td>&quot;</td>
<td>Lymphoid Hyperplasia</td>
<td>2012</td>
</tr>
<tr>
<td>Aalae</td>
<td>F</td>
<td>4yr</td>
<td>1wk</td>
<td>Abdominal pain/ Nausea agitation/ vomiting/ Diarrhea</td>
<td>AXR: Bowel distention US: Target lesion BE: coil spring with reduction</td>
<td>Reduction</td>
<td>-</td>
<td>2013</td>
</tr>
</tbody>
</table>

a. AI: Appendiceal intussusception; b. RLQ: Right Lower quadrant; c. BE: Barium enema; d. SBT: Small Bowel Transit; e. US: Ultrasound; f. WSCM: water soluble Contrast Media; g. CDUS: Color Doppler Ultrasound
The etiology of appendiceal intussusception is mostly idiopathic, but a series of underlying pathophysiologic and anatomic causes are encountered. The 10% cases of appendiceal intussusceptions is a intraluminal mass have lead points such as fecaliths. Anatomical factors are non-fixed mobile appendix, fetal type of cecum with the appendix originating from its tip, narrow, thin mesoappendix, mobile cecum, wide appendicular lumen and funnel – shaped appendix. Idiopathic appendiceal intussusception also occur with no underlying abnormality of the appendix. Pathophysiologically factors are follicular lymphoid hyperplasia, helminthiasis, fecaliths, foreign bodies, enteric infection such as adenovirus, polyps, mucocoele, postappendiceal stump, endometriosis, ulcerative colitis, crohn's D, cystic fibrosis, peutz – jeghers syndrome, melanosis coli, and Tumors (adenomas, cystadenomas, carcinoids, papilloma, adenocarcinomas, maltaoma). The most common causes of appendiceal intussusceptions in children include fecalith, foreignbody, parasite, lymphoid hypertrophy and polye. Appendiceal intussusception classified in four types is based on the level of appendiceal invagination (Mcswain classification). In type 1, appendix tip is the intussusceptum, and the more proximal portion is the intussuscipiens. In type 2, the base of appendix is the intussusceptum, and the cecum is intussuscipiens. In type 3, the proximal part of the appendix forms the intussusceptum and the distal portion is the intussuscipiens and in type 4, there is a complete inversion of the appendix into the cecum. Most reports are partial invagination of the appendix in to the cecum. In case with ileocolic intussusceptions that do not reduce in enema must be rule out appendiceal intussusception. Clinical presentation of appendiceal intussusception include four types asymptomatic, intussusception, recurrent intermittent right lower quadrant pain with vomiting and melena, and acute appendicitis. Diagnosis of appendiceal intussusception is often difficult. Most appendiceal intussusception are founded at the time of operation of the patients suspected to have appendicitis. Ultrasound is diagnostic imaging modality of choice. A few cases in asymptomatic patients have been incidentally diagnosed by barium enema, colonoscopy, CTcolography and endoscopic sonography. In

### Table 2: Demographics of Intussusception of appendix

<table>
<thead>
<tr>
<th>Sex</th>
<th>N (percentage)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male</td>
<td>20 (71.4%)</td>
</tr>
<tr>
<td>Female</td>
<td>8 (28.2%)</td>
</tr>
<tr>
<td>Total</td>
<td>28 (100%)</td>
</tr>
</tbody>
</table>

### Table 3: Clinical findings in Intussusception of appendix

<table>
<thead>
<tr>
<th>Clinical findings</th>
<th>N (percentage)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Abdominal pain</td>
<td>20 (90%)</td>
</tr>
<tr>
<td>Vomiting</td>
<td>13 (46%)</td>
</tr>
<tr>
<td>Diarrhea</td>
<td>12 (42%)</td>
</tr>
<tr>
<td>Constipation</td>
<td>6 (21%)</td>
</tr>
<tr>
<td>RLQ tenderness</td>
<td>5 (17%)</td>
</tr>
<tr>
<td>Hematochzcia</td>
<td>5 (17%)</td>
</tr>
<tr>
<td>RLQ mass</td>
<td>2 (17%)</td>
</tr>
</tbody>
</table>

### Table 4: Pathology of the appendiceal intussusception

<table>
<thead>
<tr>
<th>Pathology</th>
<th>N(percentage)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Inflammation</td>
<td>12 (42%)</td>
</tr>
<tr>
<td>Cystic fibrosis</td>
<td>3 (1%)</td>
</tr>
<tr>
<td>Lymphoma</td>
<td>2 (0.7%)</td>
</tr>
<tr>
<td>Fecal material</td>
<td>2 (0.7%)</td>
</tr>
<tr>
<td>Polyp</td>
<td>1 (0.3%)</td>
</tr>
<tr>
<td>Ulcerative colitis</td>
<td>1 (0.3%)</td>
</tr>
<tr>
<td>Torsion</td>
<td>1 (0.3%)</td>
</tr>
<tr>
<td>Total</td>
<td>22 (71%)</td>
</tr>
</tbody>
</table>
multiple cases appendiceal intussusceptions was mistaken for cecal polyp in colonoscopy. Sonographic examination demonstrated a lead point within a characteristic multiconcentric ring sign, and longitudinal sonograms showed the inverted appendix protruding into the cecal lumen.\textsuperscript{14,18} CT scan is also a modality for diagnosis of appendiceal intussusceptions.\textsuperscript{13,21} The majority of cases reported in literature were demonstrated in colonoscopic and surgical approaches.\textsuperscript{31} Few cases were managed through laparascopical surgery.\textsuperscript{32,34} Appendectomy is the treatment of choice unless there is a concern for a neoplasm. Appendiceal intussusception may be reducible in cases without lead point. An intussuscepted appendix may be normal and does not need bowel resection but torsion and perforation of the vermiform appendix can be associated with appendiceal intussusception.\textsuperscript{16,24,35,36} Appendectomy is choice treatment of appendiceal intussusception but it may relapse in spite of previous appendectomy. In these cases partial cecectomy also may be curative. In this case appendiceal intussusception was recognized by barium enema that shows coil-Spring Sign in cecum with filling defects of appendiceal lumen (Figure). Although surgical removal of appendix is treatment of choice, but in this case hydrostatic reduction of appendiceal intussusception occur.\textsuperscript{10} Probably filling defects of appendiceal lumen is due to fecoliths. We recommend diagnostic and therapeutic barium enema in childhood longstanding, persistent abdominal colic, to rule out clinical suspicion of appendiceal intussusception.

Conflict of Interest
None declared.

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None declared.

References
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