Case Report

Spachelated Intussusception a Forgotten Entity: Historical Review and a Case Report

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Intussusception of one or another type is frequently seen in children and is the most common cause of acute bowel obstruction. This pathology is usually promptly recognized and treated accordingly. But if untreated, this entity can have either of the two outcomes. The first one is death from peritonitis and resulting toxemia, which is the most common one. The second one is spontaneous elimination followed by auto-anastomosis and is extremely infrequent. We report one such case of this rare progression who was found to have old healed small bowel intussusception with luminal narrowing on exploration for acute bowel obstruction. We have also reviewed the world literature to understand the mechanism and circumstances under which this progression occurs.

Keywords: Auto-anastomosis, intussusception, spachelated, spontaneous elimination

INTRODUCTION

Intussusception is one of the most common causes of acute bowel obstruction in infants and toddlers. Timely management is of utmost importance to prevent grave outcomes. It is a well-known fact that a transient intussusception can be managed conservatively, but a fixed intussusception which is neither reduced nor resected carries fatal outcome. However, surprisingly, there are exceptions to this. Rarely, circumferential adhesions can form at the neck of intussusception, and gangrenous intussusceptum known as spachelus can slough out and may pass per rectum. Continuity of the bowel is maintained at this site of auto-anastomosis but with a luminal narrowing.[1] There have been sporadic case reports of such progression in the historic times. However, to the best of our knowledge, this is the first report of a secondary complication occurring after a spontaneously healing small bowel intussusception in the pediatric population in the past 25 years. With this intent, we report a case of spachelated intussusception and review the available literature.

CASE REPORT

A 10-month-old male child, born out of normal vaginal delivery, with no history of consanguinity and a smooth perinatal transition, reported to the emergency room (ER) with a 3-day history of severe cramping abdominal pain, bilious vomiting, and nonpassage of stools. There was a marked exacerbation of symptoms just before admission. On examination, the patient was lethargic and dehydrated. The apex beat was located in the 5th intercostal space, mid-axillary line anteriorly. There was no evident cyanosis and murmur. There was no respiratory embarrassment. On local examination, abdomen was markedly distended with visible bowel loops, with no signs of peritonitis. There was no fecal or blood staining on digital rectal examination. The electrolytes were deranged. Coagulation profile and liver function tests were within normal limits. Septic screen was negative.

The patient was rehydrated, and electrolyte imbalance was corrected. The patient underwent laparotomy after...
necessary optimization. Exploration of the abdomen was done through supraumbilical transverse incision, and it revealed dilated small bowel loops with collapsed terminal ileum [Figure 1a]. There were no other positive findings on inspection. On bowel walking, a soft to firm mass was palpated 5cm proximal to ileocecal junction (ICJ). Unsure about the nature of mass, 5cm ileum both proximal and distal to mass and ICJ was resected and ileoascending anastomosis was performed in two layers. On opening up the resected specimen, the mass was appreciated as an intraluminal soft tethered prominence [Figure 1b]. Postoperative course was uneventful. The patient passed stool on the postoperative day (POD) 4. Nasogastric tube was removed on the same day. The patient was allowed orally on POD-5 and was discharged on POD-7.

Pathological examination of the specimen showed dense cicatrix surrounding a 2-mm lumen, with mucosa on both sides with lymphoid proliferation. Underlying stroma showed edema with dense inflammatory infiltration. No evidence of granuloma or malignancy was found [Figure 1c].

On realizing that probably, we have removed an old strictured auto-anastomosis of a previously sloughed intussusceptum, and child’s history was reviewed more thoroughly. His mother revealed that at approximately 5 months of age, the patient had an attack of severe gastroenteritis lasting for 4 weeks. After treatment with oral hydration and antibiotics at home, his diarrhea subsided. No tissue had been passed through the rectum. At present, the child is doing well on a close follow-up of 1 year.

**DISCUSSION**

Intussusception develops with antegrade peristalsis when the proximal bowel (intussusceptum) drags along with its mesentery into the distal bowel (intussuscipiens). Compression of mesentery leads to edema of intussusceptum, causing impairment of lymphatic drainage and venous congestion and stasis and hence outpouring of blood and mucus. If untreated, ischemic changes amount to bowel necrosis in the intussusceptum and can be potentially fatal. Rarely, necrosed intussusceptum may get separated and expelled spontaneously, followed by auto-anastomosis of bowel ends. This entity can be defined as spachelated or stenotic old intussusception.[2] The exact incidence remains unclear. However, in 1958, Aird has estimated the incidence of this infrequent natural progression to be <2%.[3]

Although very rare even in historic times, this atypical pathophysiological progression was relatively more frequently observed about 2 centuries ago than the present times, when the operative mortality used to be high and waiting for spontaneous resolution used to be the best alternative.[4] Progression of intussusception in this manner has markedly reduced and has not been reported in pediatric population in the past 25 years. This can be ascribed to early diagnosis and intervention in the present times.

Treves is credited with the first detailed description of this atypical entity.[5] He described that if the process of vascular obstruction and gangrene is less rapid, adhesions may form between the peritoneal coats of the inner layer and outer layer or intussuscipiens at the neck of the intussusception, hereby preventing perforation at this site. This event is heralded by sloughing of intussusception, which may or may not be noticeable in stools, as in the index case, and restoration of bowel continuity by auto-anastomosis. Hence, it can be inferred that the spontaneous cure of intussusception depends on the presence of appropriate time for allowing the adhesion formation between the inner and outer layer; before the sloughing of necrosed intussusceptum [Figure 2]. This time window is more apparent in ileoileal intussusception, as in the index case, than in ileocolic intussusception where constricting effect of ileocecal valve will lead to rapid onset of vascular occlusion, gangrene, and perforation. Hence, these kinds of intussusceptions are generally

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**Figure 1:** (a) Intraoperative image showing dilated small bowel loops with collapsed terminal ileum. (b) An intraluminal soft tethered prominence. (c) Histopathological examination showing mucosa (black arrow) on both sides with lymphoid proliferation (red arrow) in between
ileocolic. Lichenstern has suggested that this separation occurs 11 to 21 days after intussusception. Separated intussusceptum can sometimes cause a ball valve effect and can cause recurrent obstruction at the narrowed area.

With this surgical curiosity in mind, we have attempted to review world literature as old as 145 years. For our ease of understanding, we have divided this time frame further into – Lichenstern era (1873-1877) and post-Lichenstern era (1878-present). We reviewed 2293 cases of intussusception in total. Out of these, 593 cases date back to Lichenstern era, and 1700 cases belong to post-Lichenstern era.

We have tabulated our observations in Table 1.

Walten reported a series of intussusception in 1911, which comprised 239 cases, but only one case terminated into spontaneous elimination (which was the same as reported by Sherren). In 1908, Fitzwillliams reported a series of 1000 cases of intussusception, with not even a single case undergoing spontaneous elimination. Similarly, Perrin and Lindsay reported a series of 400 cases of intussusception, spanning over a period of 19 years; but spontaneous elimination was not reported in any case.

Going by above data, we found that 310 cases of this entity have been reported in world literature in the past 145 years. The maximum number of such cases, i.e., 80% of cases (n = 249) were seen in Lichenstern era amounting to an incidence of 42%. This was an era when clinicians used to rely on waiting for intussusceptum to slough and auto-anastomosis to occur. Interestingly, operative interventions were also directed toward spontaneous cure in the past. Historic literature review finds the mention of the Barker’s operation or Jesset’s procedure where seromuscular sutures were placed at the neck of intussusception, and intussusceptum was then divided through an enterotomy in the intussuscipiens. Another procedure worth a mention has been described by Bayard in which the base of a sigmoid intussusceptum was banded by gaining access through the anus into the rectum to encourage intraluminal autoamputation.

On pondering over post-Lichenstern era, only 20% cases (n = 61) of such progression have been reported. This infers that the incidence of this entity presently is only 3.5%. Out of these 61 cases, 70.5% of cases (n = 43) have been reported in adults, and only 29.5% of cases (n = 18) were reported in pediatric population; that too none after 1995. This brings down the incidence of spachelated intussusception in pediatric population to just 1%.

This interesting observation can be explained by earlier diagnosis and treatment of intussusception in children, in whom the clinical picture is more striking than in adults. Second, an untreated intussusception in a child will end fatally within a few days due to marked fluid and electrolyte loss which is not well tolerated by this age group; whereas, adults can tolerate fluid and electrolyte disturbances better and hence have a better chance of survival for a sufficiently long period to allow the possibility of spontaneous healing. Third, adults are more likely to have small bowel intussusception as contrary to ileocolic intussusception in pediatric population, which leads to subacute obstruction and hence more chances of spontaneous healing. This has already explained by Treves and our study validates the same.

In the index case, child had a history of acute gastroenteritis lasting for 4 weeks which was most probably an overlooked small bowel intussusception. The most acceptable explanation for survival in this case was subacute intussusception leading to sloughing of intussusceptum and auto-anastomosis of the bowel ends. Five months later, the patient presented with intestinal obstruction with luminal narrowing at the site of auto-anastomosis and this has been correlated histopathologically too. Our findings are consistent with a case reported by Richard et al.

Rarity of this entity coupled with the age of the patient and topography of intussusception prompted us to report the case.
### Table 1: Literature review on spontaneous sloughing and auto-anastomosis in intussusception

| Year | Author | Comments | Precipitating factor/lead point |
|------|--------|----------|-------------------------------|
| **Lichenstern era** | | | |
| 1877 | Lichenstern et al. | First series of intussusception comprising 593 cases. 249 cases resolved spontaneously with sloughing of spachelus with auto-anastomosis | Not available |
| **Post Lichenstern era** | | | |
| 1894 | O'Connor | A case report of intussusception in an adolescent resolving spontaneously with sloughing followed by auto-anastomosis | Not available |
| 1894 | Sutcliffe | A similar case report of spachelated intussusception in a teenager | Not available |
| 1899 | Treves | Credited with the first description of pathophysiology of spachelated intussusception | Not available |
| 1906 | Sherren | A case report of spontaneous elimination of intussusceptum in a woman, following a surgery for reduction of hernia | Adhesions post hernia surgery |
| 1906 | Marnoch | A 3-year-old boy with sloughing of intussusception with restoration of bowel continuity | Not available |
| 1921 | Kingsford | Reported a case of spachelated intussusception in a 46-year-old male | Meckel’s diverticulum |
| 1922 | Martin | Reported a similar case in a 26-year-old female | Intestinal diverticulum with feculoma |
| 1927 | Thompson | Reported a case of spachelated intussusception in a 4-year-old male | Tapeworms (Taenia saginata) |
| 1931 | Silvermann | Reported a case of spontaneous elimination of intestine in intussusception with temporary recovery | Not available |
| 1931 | Grami | Reported a case of spontaneous elimination of intestine followed by auto-anastomosis in a 4-year-old female | Not available |
| 1932 | Mayo | Reported a case of spontaneously resolving intussusception with sloughing of intussusceptum in a 6-year-old male | Not available |
| 1932 | Sapre | Reported a similar case in a 50-year-old male, initially mistaken as gastroenteritis | Not available |
| 1932 | Czyzewski | Reported a case of spachelated intussusception in a 19-year-old male, which was initially mistaken as tubercular peritonitis | Not available |
| 1936 | Bockoven | Reported a case of spontaneously healing intussusception in a 17-year-old male | Meckel’s diverticulum |
| 1938 | Izashvili | Reported a similar case in a 25-year-old male, initially mistaken as acute gastroenteritis | Not available |
| 1939 | Segall | Reported a case of self-healed intussusception in a 40-year-old male with previous history of acute appendicitis | Adhesions postacutaneous appendicitis |
| 1939 | Podetti | Reported a similar case in a 40-year-old female, after acute appendicitis | Scarring on mesenteric border |
| 1942 | Szlavik | Reported a case of spontaneous separation and healing of intussusception in an 8-year-old female, with clinical picture mimicking acute gastroenteritis | Not available |
| 1947 | Finestone | After a literature review of 20 years, reported a single case of intussusception following previous ileosigmoidostomy | Not available |
| 1949 | Louw | Reported a similar case in a 30-year-old male | Roundworms |
| 1950 | Forrester et al. | Reported a case in a 2.5-year-old female, initially misdiagnosed as pyelitis | Not available |
| 1951 | Grant and Bowden | Reported a case of spontaneous expulsion of sequestrated transverse colon per anum in a 47-year-old female, following total gastrectomy and partial colectomy | Bowel denuded of mesocolon |
| 1951 | Plotegether | Reported a case of spachelated small bowel obstruction in a 52-year-old male | Not available |
| 1952 | Coe et al. | Reported a similar case in a 76-year-old female | Not available |
| 1952 | Jacobson et al. | Reported a case in a 49-year-old male with history of Inferior vena cava thrombosis | Not available |
| 1952 | Welsch | Reported a case in a 76-year-old | Polyp |
| 1953 | Benson et al. | Reported 2 cases of spachelated intussusception | Pregnancy? trauma |
| 1954 | Tropea et al. | A case report of 63-year-old male intussusception of ileum with expulsion of intussusceptum per anum, initially mistaken as diverticulitis | Not available |
| 1954 | Scioli | Reported a case of spachelated intussusception in an 28-year-old male | Not available |
| 1955 | Becker | Reported a case in a 10-day-old male | Overfeeding |

*Contd...*
Table 1: Contd...

| Year | Author          | Comments                                                                 | Precipitating factor/lead point               |
|------|-----------------|---------------------------------------------------------------------------|----------------------------------------------|
| 1958 | Hansbrough[4]   | Reported a case in a 45-year-old male                                     | Bowel trauma                                 |
| 1962 | Robb and Souter[6] | A series of 28 patients with spontaneous sloughing and healing of intussusception. Out of these, 7 were pediatric patients, and 21 were adults | Not available                                 |
| 1963 | Peck et al.[17] | One similar case has been reported as “stenotic old intussusception”     | Not available                                 |
| 1995 | Richard et al.[2] | A case report of an 8-year-old boy with luminal narrowing after auto-anastomosis and intraluminal slough in an intussusception | Not available                                 |
| Not known | Garlock[16] | Reported two cases of spontaneous sloughing and healing of intussusception; in previously operated cases of terminal ileitis | Postoperative adhesions                      |

**CONCLUSION**

This case report intent to draw attention toward the entity of sloughed intussusceptum as a rare but possible etiology of partial bowel obstruction in childhood, especially in the cases of conservatively managed pain abdomen and altered bowel habits. It is also imperative to take a proper clinical history and meticulous examination intraoperatively and communication with the pathologist so that such interesting findings do not get missed.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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