Adult Intussusception due to a Rare Tumor (Inflammatory Myofibroblastic Tumor of Mesentery): A Case Report

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Adult intussusception occurs infrequently and differs from childhood intussusception in its presentation, etiology, and treatment. Diagnosis can be delayed because of its long-standing, intermittent, and non-specific symptoms and most cases are diagnosed at emergency laparotomy.

We report a case of 23-year-old female with complaints of abdominal pain, constipation, and loss of appetite since 2 months. Computed tomography revealed right colonic obstruction caused by intussusception of the ileocecum. On exploration, a mesenteric mass was seen extending with the intussusceptum following, which ileocecal intussusception was found. The mass was excised. Histopathology examination of the specimen revealed an inflammatory myofibroblastic tumor. These tumors can occur throughout the body, most commonly in the lung, mesentery, and omentum. Complete surgical resection is the treatment of choice. A Review of this rare condition is interesting and should be borne on the mind.

Keywords: Anaplastic lymphoma kinase, Contrast enhanced computed tomography, Inflammatory myofibroblastic tumor, Intestinal obstruction, Intussusception

INTRODUCTION

Intussusception has been defined as the invagination of one division of the bowel into an immediately another adjacent division; the proximal division of the intestinal tract, or the intussusceptum, is carried within the lumen of an adjacent division known as the intussuscipiens.¹ Adult intussusception represents 5% of all cases of intussusception and accounts for only 1-5% of intestinal obstructions in adults.¹ It has been commonly seen in the small bowel of children, where the etiology is benign and non-operative treatment is usually successful.² In contrast, intussusception in adults is uncommon.² Colonic involvement represents up to 50% of cases, and in 70% the underlying cause is a malignant neoplasm.³ A large range of other causes in the colon have been described, including adenomatous polyps, inflammatory bowel disease as found in our patient, mycobacterial infection, and surgical anastomoses.⁴

Due to a significant risk of associated malignancy, which approximates 65%,⁵ radiologic decompression is not addressed preoperatively in adults. Therefore, 70-90% of adult cases of intussusception require definite treatment, of which surgical resection is, most often, the treatment of choice.⁶

Inflammatory myofibroblastic tumor (IMT) is a rare neoplasm usually seen in children and adolescents, mostly occurring between 2 and 16 years.⁷ Females are more commonly affected than males. The diagnosis of this condition can be difficult as symptoms are often non-specific and episodic.

CASE REPORT

A 23-year-old female presented with complaints of chronic abdominal pain, constipation, and decreased appetite since 2 months. Abdominal examination revealed distension in the lower abdomen, the presence of sluggish bowel sounds with minimal tenderness in right lumbar, and iliac fossa. There were no clinical features of peritonitis or abdominal lump. Ultrasonography of the abdomen and pelvis was suggestive of ileocecal junction intussusception that has been confirmed on contrast-enhanced computed tomography (CT) (Figure 1).
All the lab reports were normal except for anemia (hemoglobin 6.5 g/dl).

Exploratory laparotomy was done. Exploration revealed an ileocecal intussusception with a mass on mesentery side and entering with the intussusceptum (Figures 2-5). Loco-regional resection with ileo-ascending colon anastomosis was done in 2 layers (Figure 6). Thorough Peritoneal lavage was given with normal saline and pelvic drain was kept. Post-operative period was uneventful. The patient was discharged on postoperative day 10.

Histopathology of resected specimen showed features of IMT of mesentery (Figure 7).

Follow up after 1, 3, and 6 months showed no recurrence.

**DISCUSSION**

Since its first description in 1674 by Barbette, intussusception has been studied and found to be a disease of infancy and early childhood. Adult intussusception is different from pediatric intussusception. It is rare, the condition being found in <1 in 1300 abdominal operations and 1 in 100 patients operated for intestinal obstruction. Compared to intussusceptions in children where nearly 80% are idiopathic, a demonstrable etiology is found in 90% of cases in the adults. Intussusception occurs when a part of the bowel, the intussusceptum, invaginates into the lumen of the distal bowel, the intussuscipiens. Intussusception in adult patients may be caused by intraluminal, mural, or extra luminal lesions. It can be ileocolic (most common type, 75%), colo-colic, and ceco ileocolic. The basic mechanism by which intussusception is thought to occur is when an intraluminal mass is pulled forward by peristalsis and draws the attached bowel wall segment with it. Pedunculated tumors, such as adenomatous polyps or lipomas, are the examples of this group. As in our patient, it was IMT of the mesentery.

Most adult cases of intussusception occur in the distal small bowel (52-55%) or in the large intestine (38-45%). Clinical presentation in adult intussusception is variable, but mostly associated with the chronic obstruction. The most

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**Figure 1:** Computed tomography abdomen, and pelvis showing target sign

**Figure 2:** Ileocecal intussusception with growth intraoperatively

**Figure 3:** Resected specimen of ileo cecal intussusception with polypoidal growth

**Figure 4:** Excised portion of intussusceptions
commonly used diagnostic tools are ultrasonography and CT, and it is important to diagnose acute intussusception as early as possible. Ultrasonography is used to evaluate suspected intussusception as it is cheap, easily available, and non-invasive. The characteristic features include the “target and doughnut sign” on transverse view and the “pseudo-kidney sign” in the longitudinal view. The disadvantages of ultrasonography are operator dependency and difficulty in image interpretation in the presence of air, which is often present in cases of obstruction. The preoperative diagnostic accuracy of ultrasonography in cases of palpable abdominal mass is around 86.6%.

CT scan has been reported to be the most useful radiimaging technique, with an accuracy of 58-100% and a specificity of 57-71%. As compared to pediatric patients, where intussusception is primary and benign, preoperative reduction with barium or air is not suggested as a definite treatment for adults’ intussusception. The hypothetical risks of preliminary manipulative reduction of an intussuscepted bowel can lead to intraluminal seeding and venous tumor dissemination, perforation and seeding of microorganisms and tumor cells to the peritoneal cavity, and increased risk of anastomotic complications of the manipulated friable and edematous bowel tissue.

In our patient, a mass showed IMT of the mesentery. In the earlier period, it was referred under different synonymous such as xanthomatous pseudotumor, myofibroblastoma, pseudosarcomatous myofibroblastic proliferation, myofibroblastoma, inflammatory myofibrohistiocytic proliferation, plasma cell granuloma, and most commonly inflammatory pseudotumor. IMT has been originally described in the lungs in 1937, and since then has been seen throughout the body, including the mesentery, liver, mediastinum, stomach, abdomen, retroperitoneum, omentum, and bladder of children and young adults.

Histopathologically made up of cellular, fascicular fibroblastic/myofibroblastic proliferations accompanied by prominent infiltrate of chronic inflammatory cells mainly plasma cells. The spindle cell component has plump focally atypical nuclei and variable mitotic rate.

The etiology of these tumors is not clear. Some have been described them to be associated with Epstein - Barr virus infection and also found in human herpes virus-8 viral sequences in the tumor cells. These tumors have also been described in patients with human immunodeficiency virus infection and chronic granulomatous disease. Others have insisted that a chromosomal rearrangement associated with the anaplastic lymphoma kinase (ALK) gene lead to the activation of a tyrosine kinase receptor and could lead to abnormal expression.

Patients most commonly present with a painless abdominal lump. They may remain completely silent until the lump...
reaches a significant size to cause complications. Patients may also present with abdominal pain without a palpable lump. Sometimes present with diarrhea or intestinal obstruction. Atypical presentations have been reported such as portal venous thrombosis, intussusceptions as it was with our patient, pyrexia of unknown origin, anemia, and leukemoid reaction as the initial presentation of IMT.

Preferred treatment for the IMT surgical resection, which can also be used to confirm this disease. The recurrence rate has been seen from 18% to 40%, and only proper surgical removal of the tumor will avoid recurrence of tumor. Local recurrences were seen with the abdominopelvic site, larger size, and older age, which are seen in about 10-25% cases of abdominal tumors within a year of surgery. Rarely, these lesions undergo malignant transformation and metastasize. The risk of distant metastases is seen in surgically operated cases <5% and the 5-year survival rate is around 87%. There is no specific marker like histological or molecular which can predict the malignant transformation of IMT, cases who are positive for ALK rearrangements have been seen that there has been decreased risk of metastases when compared with ALK-negative IMT. In case of ALK-positive, the role of crizotinib, an ALK inhibitor, has been studied, and trials had shown good results in the majority of cases, although more studies are required to establish selective targeting of ALK as a definitive therapeutic option. In a study by Mossé et al., other treatment modalities have been used in the IMT where complete surgical resection was not feasible. Corticosteroid monotherapy, non-steroidal anti-inflammatory agents, and radiation alone has been tried with good results in the ALK-negative IMT.

CONCLUSION

Intussusception in the adult is relatively uncommon and this, along with the indistinguishable clinical features, makes diagnosis complicated. Ultrasonography and CT have been shown to be successful diagnostic tools. In adults, intussusception with IMT requires surgical resection, and it is a mainstay treatment.

REFERENCES


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