Current Trends in Internal Medicine

Ng OH and Skaife P. Curr Trends Intern Med 6: 168. www.doi.org/10.29011/2638-003X.100068 www.gavinpublishers.com





Case Report

A Case Study and Literature Review on Idiopathic Adult Intussusception

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Citation: Ng OH, Skaife P (2022) A Case Study and Literature Review on Idiopathic Adult Intussusception Curr Trends Intern Med 6: 168. DOI: 10.29011/2638-003X.100068

Received Date: 26 September 2022; Accepted Date: 30 September 2022; Published Date: 04 October 2022

Abstract

Aim: To review existing literature on adult intussusception and idiopathic adult intussusception, and to summarise current recommendations for the management of adult intussusception and idiopathic adult intussusception.

Methods: An electronic search of PubMed was performed with search term "intussusception", "adult intussusception", and "idiopathic intussusception". Studies concerning the paediatric population was excluded. Resultant searches were reviewed and studies which were felt to be relevant were identified with full text retrieved. The references of all retrieved texts were searched to identify any further relevant studies.

Background: Intussusception is a rare diagnosis in adult patients, with only 5% of intussusceptions occurring in adults, and only 1-5% of bowel obstructions in adults occurring as a result of intussusception. While historically associated with malignancy, idiopathic intussusceptions accounts for 8-20% of cases of adult intussusceptions.

Case Presentation: This report presents the case of a 54-year-old male who presented with PR bleeding who was found to have intussusception on radiological imaging with suspicion of intraluminal lesion and underwent laparotomy with no lead point found intraoperatively. Decision was made against performing bowel resection. He went on to have normal post-operative imaging.

This case study is followed by a review of existing literature on adult intussusception and a discussion regarding the latest recommendations for management of this rare condition.

Conclusions: Current recommendations advise surgical oncological resection without reduction. From this case study, we would add that even in cases without a palpable lead point intraoperatively, if there is a suspicion of malignancy clinically or radiologically, it may be prudent to undertake resection rather than risk enterotomy and potential seeding of tumour.

Keywords: Intussusception; Idiopathic Adult Intussusception

Case presentation

A 54-year-old Caucasian male presented to our tertiary hospital with 6 episodes of small volume fresh red blood per rectum associated with several hours history of mild central abdominal pain and loose stool. He had no abdominal distension, was not vomiting and was passing flatus. On examination, he had a soft, non-tender abdomen with normal bowel sounds. There was no palpable mass on digital rectal examination, however some dark red blood was present on the glove. Of note, he had significant skin changes (known history of eczema for which he is under the care

of the dermatologist), but had not noticed any new or concerning skin lesions. On systemic enquiry he had no history of weight loss or change in bowel habits.

Laboratory investigations showed blood haemoglobin 101 g/L (normal: 140-180 g/L), WCC 18.8 x 10*9/L (normal: 4-11 x10*9/L) with neutrophil 17.01x10*9/L (normal: 1.9-8 x10*9/L) and lymphocyte 0.7 x10*9/L (normal: 0.9-4.5 x10*9/L), C-reactive protein of 18 mg/L (normal: 0-10 mg/L).

CT abdomen pelvis with contrast revealed a polypoidal, hypoattenuated, non-enhancing, intraluminal lesion measuring 4cm x 3cm in the proximal jejunum causing jejuno-jejunal

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Curr Trends Intern Med, an open access journal

intussusception with high density contents in the lumen suggestive of blood. A further short segment of small bowel intussusception is present more distally with no discernible lead point. The involved bowel loops appeared thick walled and oedematous with minimal surrounding fat stranding with no features of bowel ischaemia. Prominent lymph nodes were noted in both groins measuring 14mm. Hypodense lesions noted in L1 and L2 vertebral body as well as focal 1cm hypoattenuation in liver adjacent to falciform ligament raised possibility of metastases Figure 1.





Figure 1: Computed tomography abdomen and pelvis with contrast demonstrating jejuno-jejunal intussusception.

Given the radiological findings, the decision was made for emergency laparotomy. On exploration, there was evidence of reduced jejunal intussusception with some bruising and thickening of an 18cm segment of jejunum about 50cm from the ileocaecal valve with an adjacent reactive lymph node. No visible or palpable mass lesion on close external examination, hence an enterotomy was performed along the point of intussusception. There was some evidence of mucosal ischaemic changes with sloughing of mucosa, however no mass lesion was seen or felt intraluminally. As no lesion was found and there was no concerns regarding the viability of the bowel, the intraoperative decision was made against performing any bowel resection. The reactive lymph node in the adjacent mesentery was excised and sent for biopsy.

The patient had an uncomplicated post-operative period and was discharged several days later. Histopathology of the mesenteric lymph node showed reactive changes without evidence of neoplasia. An MRI small bowel was performed 3 months following discharge which showed a non-enhancing 20cm segment of proximal jejunum with diffuse bowel wall thickening and residual fat stranding. No underlying aetiology was identified. At follow-up at 4 months following discharge, the patient was symptom-free and remained well in himself.

Discussion

ISSN: 2638-003X

Intussusception in adults remains a rare diagnosis. Only 5% of intussusceptions are estimated to occur in adults while only 1-5% of bowel obstructions in adults are caused by intussusception and intussusception accounts for only 0.003%-002% of all adult hospital admissions [1-3]. 70-90% of adult intussusceptions have

a demonstrable lead point. According to Marsicovetere, et al. [4], two thirds of lead points are secondary to neoplasms, 50% of which are malignant. One third are secondary to pathologies such as infection, adhesion, Crohn's granuloma, ulcer, or congenital abnormalities such as Meckel's.

A meta-analysis of 1229 patients over 40 case series by Hong KD et al found that 49.5% of intussusceptions were enteric, 29.1% ileocaecal, and 19.9% colonic [5]. Overall, malignant lesions accounted for 32.9% of intussusceptions, benign 37.4% and idiopathic 15.1%. However, the risk of malignancy was two times higher in colonic intussusception then enteric (enteric: 22.5% malignant; colonic: 46.5% malignant). This was in keeping with Marsicovetere et al's findings, whereby 50-75% of enteric lead points were benign, whereas colonic intussusception was an independent predictor of malignancy [4]. Ileocolic intussusception was identified as a unique variant, as almost 100% has a malignant lead, most commonly caecal adenocarcinoma involving the ileocaecal valve.

In 1981, Nagorney, et al. [6] recommended primary oncological resection without reduction for all intussusceptions regardless of site and suspected aetiology. In 2006, Zubaidi, et al. [2] recommended that while colonic intussusception should be resection without reduction because the underlying pathology was more likely to be malignant, enteric intussusceptions should be reduced prior to resection should the underlying etiology be suspected to be benign, or if the resection that would be required without reduction is determined to be massive. Likewise, both Yakan, et al. [3] and Marinis A, et al. [7] recommended that if the enteric segments were viable and malignancy was not suspected, reduction prior to resection should be attempted.

Of note, there is noted to be one notable exception to the recommendations for resection of the intussuscepted segment, and that is when the intussuscepted segment is <3.5cm in length. Lvoff, et al. [8] suggested that intussusceptions less than 3.5cm in length were likely to be self-limiting and could be managed conservatively. Similarly, Marsicovetere, et al. [4] recommended that entero-enteric intussusception without lead point and short affected segments, defined as <3.5cm-3.8cm could be managed with serial clinical and imaging evaluations to ensure resolution.

Idiopathic Intussusception

Idiopathic intussusception accounts for 90% of paediatric intussusception cases [9] and 8-20% of adult intussusception cases [7]. While paediatric idiopathic intussusception has been theorised to be secondary to anatomical features in the developing gastrointestinal tract [4] or to viral infections [10], there has been no research performed into the possible underlying aetiologies of adult idiopathic intussusception, despite it potentially accounting for up to 1 in 5 adult intussusceptions. Moreover, there were no recommendations made specific to the management of suspected idiopathic intussusception.

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Reflection

In our patient management, the failure to appreciate a tumour within the thickened area of jejunum could have potentially resulted in seeding of tumour cells throughout the abdominal cavity by performing an enterotomy.

While there is diverse opinion on the management of intussusception within the literature reviewed, and all opinions have their merit, the potential for seeding of a tumour may be a reason to recommend primary resection of identified bowel abnormality whether the intussusception remains reduced or not at laparotomy.

Conclusion

Adult intussusception remains a rare diagnosis without established published guidelines for investigation and management. The current recommendations from literature can be summarised as follows: surgical oncological resection without reduction is recommended in colonic lesions, ileocaecal lesions, in enteric lesions suspected of being malignant, and in cases of non-viable bowel. If enteric intussusceptions are suspected to be benign, the bowel is viable, or if the resection without reduction is deemed to be massive, reduction could be attempted prior to resection. If intussusceptions are <3.5cm, there is potentially a role for conservative management with serial examination and imaging to ensure resolution.

From our reflection, we would add that even in cases without a palpable lead point intraoperatively, if there is a suspicion of malignancy clinically or radiologically, it may be prudent to undertake resection rather than risk enterotomy and potential seeding of tumour. In the absence of an oncological suspicion, reduction and further luminal investigation may be an option. Furthermore, research into the aetiology of idiopathic intussusception should be performed and would be useful in

guiding development of viable management strategies for cases of intussusception in which no lead point is suspected or found.

Funding: The authors received no financial support for the study, authorship, and publication of this article

Conflict of interest: The authors do not have any conflict of interest to declare.

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ISSN: 2638-003X