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Surgical Decision-Making in Adult Appendiceal Intussusception: A Case of an Occult Neuroendocrine Tumor

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Female, 25-year-old
Final Diagnosis: Complete appendiceal intussusception associated with a well-differentiated grade 1 appendiceal neuroendocrine tumour with regional lymph node metastases (pT3 pN1)
Symptoms: Intermittent colicky abdominal pain • right iliac fossa pain • rectal bleeding • nausea • vomiting
Clinical Procedure: Diagnostic laparoscopy converted to open right hemicolectomy with side-to-side ileocolic anastomosis
Specialty: Surgery
Objective: Rare disease
Background: Appendiceal intussusception is rare, identified in approximately 0.01% of appendicectomy specimens, and may be associated with an underlying pathological lead point. This single case describes an appendiceal neuroendocrine tumor (NET) that was radiologically occult despite computed tomography (CT) correctly identifying the appendiceal intussusception. It illustrates how concerning intraoperative findings can influence the extent of resection when preoperative imaging does not identify the underlying cause.
Case Report: A woman in her mid-20s presented with intermittent lower abdominal pain and hematochezia and re-presented 48 hours after discharge with worsening right iliac fossa pain, darker bleeding, nausea, and vomiting. CT demonstrated appendiceal intussusception into the cecum with invaginated mesoappendiceal fat but no definite lead-point mass or lymphadenopathy. At laparoscopy, difficult retraction of the intussuscepted appendix and mesenteric lymph nodes raised concern for occult pathology and contributed to conversion to open right hemicolectomy. Histopathology demonstrated a 19-mm grade 1 NET with focal subserosal extension and metastasis in 2 of 16 regional lymph nodes (pT3 pN1, AJCC version 9 stage III). Ga-68 DOTA-peptide positron emission tomography showed no somatostatin-receptor-expressing residual or metastatic disease.
Conclusions: A radiologically occult appendiceal NET can present as adult appendiceal intussusception. In this single case, difficult appendiceal retraction and mesenteric lymph nodes influenced the extent of resection when CT showed no lead-point mass or lymphadenopathy. This observation is illustrative and does not establish criteria for right hemicolectomy.
Keywords: Intussusception • Appendiceal Neoplasms • Neuroendocrine Tumors • Colectomy • Computed Tomography • X-Ray • Decision Making
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Introduction

Appendiceal intussusception is rare, with an estimated incidence of approximately 0.01% in appendectomy specimens; the true incidence of symptomatic presentation is unknown [1]. Appendiceal intussusception can present acutely, intermittently, or chronically, including with abdominal pain or/and gastrointestinal bleeding [1,2]. This condition should be distinguished from adult intestinal intussusception more broadly, which is frequently associated with an underlying pathological process, including neoplasia [3,4]. Reported proportions vary across series and should not be extrapolated to appendiceal intussusception.

Contrast-enhanced computed tomography (CT) is important in adult intestinal intussusception and may demonstrate a target-like or bowel-within-bowel configuration [5,6]. Evidence for detecting the underlying lead point is heterogeneous, particularly for appendiceal intussusception. Kangaspunta et al reported limited preoperative CT detection of appendiceal tumors in 5224 appendectomies for suspected acute appendicitis [7]; these findings should not be extrapolated quantitatively to intussusception. Coursey et al similarly found that none of 10 appendiceal carcinoid tumors imaged preoperatively was prospectively identified on CT [8]. These data support the possibility of radiologically occult appendiceal neoplasia but do not establish CT sensitivity for neoplastic lead points in appendiceal intussusception.

Appendiceal neuroendocrine tumors (NETs) are uncommon and are frequently diagnosed incidentally after appendectomy [9,10]. Neoplasia is recognized among the pathological conditions associated with appendiceal intussusception [2]. We report a single case in which CT identified the intussusception but not the appendiceal NET or nodal disease. Its educational value lies in the specific combination of a radiologically occult tumor that was a plausible lead point, difficult appendiceal retraction and mesenteric lymph nodes at surgery, and a 19-mm node-positive NET on histopathology. The case is presented as an illustrative boundary condition rather than evidence of a new association or a general management rule.

Case Report

Presentation

A woman in her mid-20s presented with intermittent lower abdominal cramping, and small-volume bright-red rectal bleeding with defecation. She had recently completed 4 days of prednisolone and amoxicillin-clavulanic acid for a lower respiratory tract infection; the prednisolone dose was unavailable in the records reviewed. She had previously received corticosteroids

for asthma, without gastrointestinal bleeding, and denied regular non-steroidal anti-inflammatory drug use, prior abdominal surgery, inflammatory bowel disease, or a family history of colorectal cancer or inflammatory bowel disease.

She was hemodynamically stable, with a heart rate in the 70s beats/min, blood pressure 110/72 mm Hg, respiratory rate 16 breaths/min, and oxygen saturation greater than 94% on room air. The abdomen was soft with mild lower abdominal tenderness and no guarding or palpable mass. Digital rectal examination showed a small amount of blood without hemorrhoids, skin tags, or another obvious anorectal source. Initial clinical and laboratory assessment was reassuring. The working impression was that the rectal bleeding might be related to the recent prednisolone course; prednisolone was ceased, and pantoprazole commenced. A broader differential diagnosis at the initial attendance was not documented in the available medical record. Cross-sectional imaging was not performed because of the initially small-volume bleeding, reassuring assessment, and her young age. The reason ultrasound was not obtained was not specifically documented. She was discharged with advice to return if bleeding, pain, or other symptoms worsened.

She re-presented 48 hours later with worsening lower abdominal pain, predominantly in the right iliac fossa, darker and increased rectal bleeding mixed with stool, poor oral intake, nausea, and vomiting. She appeared pale and uncomfortable but remained clinically stable; her heart rate was approximately 100 beats/min, systolic blood pressure was approximately 130 mm Hg, and she was afebrile with oxygenation maintained on room air. Examination demonstrated marked right iliac fossa tenderness with additional right-sided and suprapubic tenderness, without guarding or peritonism. The white blood cell count was $11.5 \times 10^9/L$, C-reactive protein level was less than 1 mg/L, venous lactate level was 1.3 mmol/L, renal and liver functions were within the reference range, and serum β -hCG was less than 2 IU/L. Differentials included colitis and other gastrointestinal causes of bleeding, while an acute appendiceal cause was initially considered less likely. The clinical course is summarized in **Figure 1**.

Imaging

Given her age and otherwise reassuring physiological and laboratory findings despite several days of symptoms, ultrasound was selected initially to limit ionizing radiation. Pelvic ultrasound showed no uterine, ovarian, or adnexal abnormality. Focused right iliac fossa ultrasound demonstrated a thick-walled, dilated, non-compressible appendix with an internal echogenic component and trace adjacent free fluid (**Figure 2**).

Contrast-enhanced CT performed the same day showed a focal bowel-within-bowel, targetoid lesion at the cecal pole and

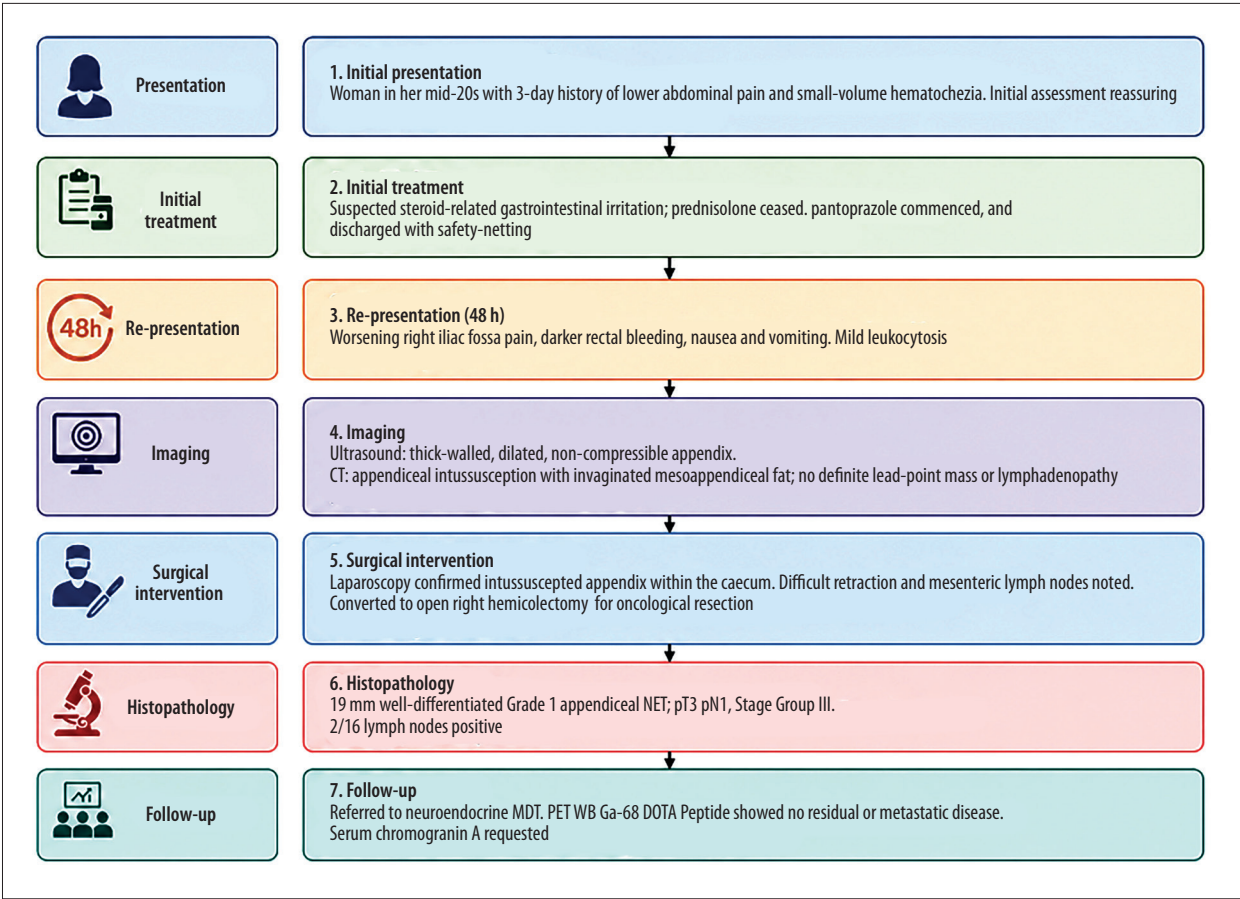


Figure 1. Clinical management pathway. This flowchart illustrates the key stages in the patient's clinical course, including presentation, investigations, operative management, histopathological findings, and follow-up.

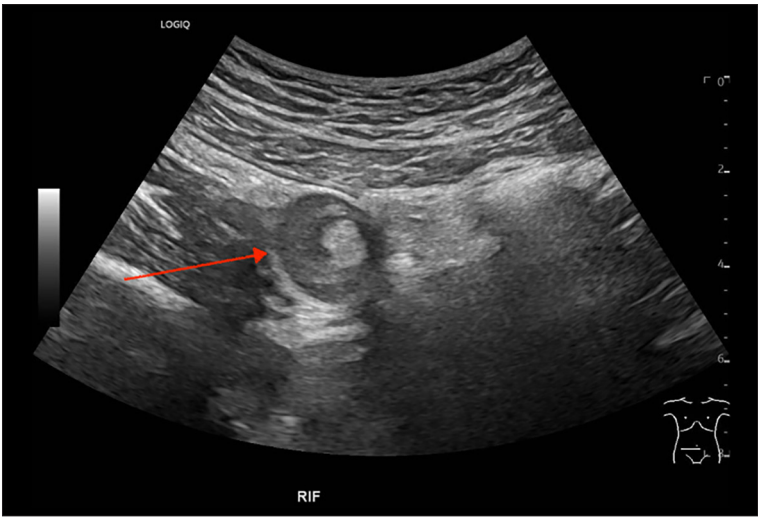


Figure 2. Transabdominal ultrasound demonstrating a thick-walled, dilated appendix with trace adjacent free fluid; the appendix was non-compressible on sonographic examination.

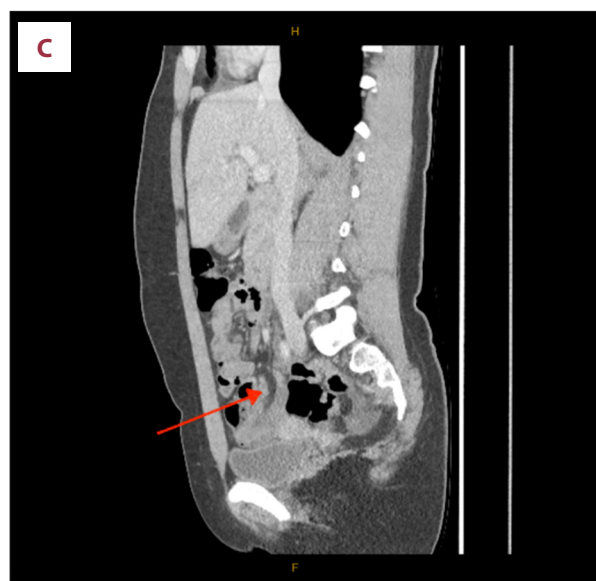
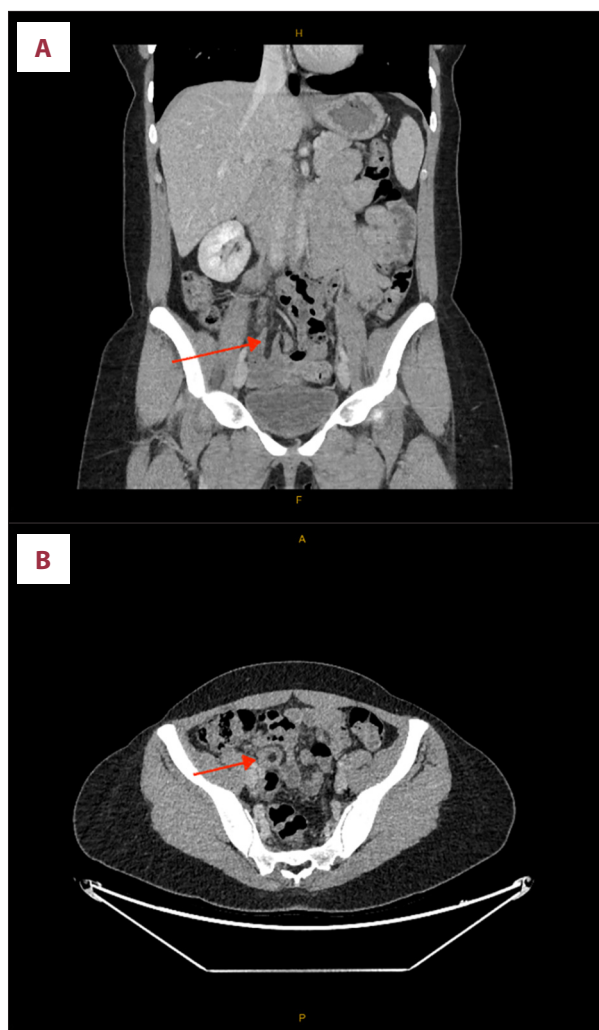


Figure 3. Contrast-enhanced CT of the abdomen and pelvis demonstrating appendiceal intussusception at the caecal pole with a bowel-within-bowel configuration and invaginated mesoappendiceal fat: (A) coronal, (B) axial, and (C) sagittal views.

expected appendiceal orifice. The appendix was not separately visualized, and internal fat density was consistent with invaginated mesoappendiceal fat (Figure 3A-3C). No discrete enhancing lead-point mass, appendicolith, or lymphadenopathy was seen. There was trace pelvic free fluid but no obstruction, perforation, abscess, or free air. The impression was appendiceal intussusception into the cecum with no definite lead point, although an underlying lesion could not be excluded.

Treatment

The patient was admitted and underwent surgery the following day. Laparoscopy demonstrated appendiceal intussusception within the cecum. Retraction of the intussuscepted appendix was difficult, and mesenteric lymph nodes were noted intraoperatively, increasing concern for an underlying pathological process. The number, size, and macroscopic appearance of these nodes were not further documented. No discrete cecal-base mass, induration, or friability was documented, and the



Figure 4. Macroscopic surgical specimen showing complete invagination of the appendix into the cecum, consistent with type V appendiceal intussusception.

operative note did not describe a formal reduction manoeuvre beyond the difficulty encountered with retraction.

These intraoperative findings contributed to the decision to proceed with right hemicolectomy for oncological resection. The right colon was mobilized laparoscopically before conversion to an open approach; mesenteric division incorporated the ileocolic pedicle and right branch of the middle colic vessels, with resection of high mesenteric nodes. Bowel continuity was restored with a stapled side-to-side ileocolic anastomosis. No formal assessment for distant metastatic disease

was documented, and frozen section was not documented as performed or considered. An image of the specimen sent for histopathology is shown in **Figure 4**.

Outcome and Follow-up

Recovery was uncomplicated. Histopathology demonstrated a 19-mm unifocal, well-differentiated grade 1 NET arising at the appendiceal tip with focal subserosal extension. The mitotic rate was 1.2 mitoses per 2 mm², and Ki-67 was approximately 1%. Perineural invasion was present; lymphovascular invasion was not identified unequivocally. Resection margins were clear. Tumor cells stained for synaptophysin, chromogranin-A, AE1/AE3, and CAM5.2. Two of 16 regional lymph nodes contained metastatic NET, with deposits up to 0.7 mm. The final stage was pT3 pN1, stage group III according to the American Joint Committee on Cancer (AJCC) version 9 (2024) [11]. Histology also confirmed complete appendiceal inversion with ulceration and active inflammation, without well-defined perforation.

The patient was informed and referred for specialist neuroendocrine multidisciplinary review. Serum chromogranin A was requested, but the result was unavailable at manuscript preparation. Whole-body positron emission tomography with Gallium-68 DOTA peptide imaging using Gallium-68 DOTA-octreotate showed no somatostatin-receptor-expressing residual disease at the bowel anastomosis and no somatostatin-receptor-expressing regional or distant metastatic disease. The duration of subsequent follow-up, current disease status beyond this postoperative staging assessment, and a documented long-term surveillance strategy were not available in the records reviewed.

Discussion

This report provides illustrative, case-bounded evidence from one patient. CT identified appendiceal intussusception but no lead-point mass or lymphadenopathy; difficult appendiceal retraction and mesenteric lymph nodes raised intraoperative concern; and histopathology demonstrated a 19-mm grade 1 NET with regional nodal metastases. The value of the report lies in this specific combination. It shows a boundary condition in which preoperative imaging did not identify the cause of the intussusception and concerning intraoperative findings influenced the extent of resection. It does not establish the diagnostic performance of CT, a general hierarchy between imaging and operative assessment, or criteria for right hemicolectomy.

Pathological Mechanisms and Classification of Intussusception

Intussusception occurs when one bowel segment invaginates into another, drawing its mesentery with it; progressive venous

and lymphatic congestion may impair arterial perfusion and lead to ischemia, necrosis, or perforation [3,12]. In adults, an underlying structural lesion may act as a lead point [3,12]. Appendiceal intussusception is classified by the site and extent of invagination. Type V denotes complete invagination of the appendix into the cecum [2]. Operative and pathological findings in this case were consistent with type V intussusception (**Figure 4**). The distal appendiceal NET provides a plausible pathological association, although its precise mechanical role as the lead point cannot be established from a single case.

Preoperative Imaging and Diagnostic Uncertainty

CT is well established for diagnosing adult intestinal intussusception and may suggest a pathological lead point [5,6], although appendiceal-specific evidence remains limited [2]. Available appendiceal tumor studies indicate that small neoplasms may remain radiologically occult [7,8]. In this patient, CT correctly identified appendiceal intussusception and invaginated mesoappendiceal fat but did not demonstrate the underlying NET or regional nodal disease. This single case illustrates a boundary condition: accurate CT recognition of appendiceal intussusception did not exclude a radiologically occult appendiceal neoplasm. It does not establish a defined limitation or miss rate for CT in appendiceal intussusception.

Risk Profiling of Appendiceal Neuroendocrine Tumours

Appendiceal NETs are usually well differentiated, low grade, and incidentally diagnosed [9,13]. Tumor size, grade, proliferative activity, resection status, and invasive features inform postoperative risk assessment [13]. European Neuroendocrine Tumor Society 2023 clinical practice guidelines recommend completion right hemicolectomy for tumors greater than 2 cm and incomplete (R1/R2) resection; for completely resected tumors of 2 cm or less, it is not generally recommended; and tumors measuring 1 to 2 cm should be considered individually according to grade and patient factors [13]. AJCC is used for staging rather than treatment recommendations; AJCC version 9 (2024) was used here [11].

Despite measuring 19 mm and being grade 1 with low proliferative activity and clear margins, this tumor had regional nodal metastases and perineural invasion. These postoperative findings provide retrospective context but did not determine the intraoperative extent of resection because the NET was unknown at surgery. They should not be interpreted as evidence that all appendiceal NETs between 1 and 2 cm require right hemicolectomy.

Comparison with Previously Reported Cases

NET-associated appendiceal intussusception has been reported previously [14-16]. Kantarovsky et al described intussusception

identified during surgery for presumed appendicitis, with carcinoid tumor diagnosed histologically [14]. Thomas et al reported a 70-year-old man with weight loss and an apparently malignant cecal lesion on colonoscopy; CT did not show the tumor, surgery identified a cecal mass and enlarged mesenteric node, and extended right hemicolectomy demonstrated complete appendiceal intussusception with a well-differentiated neuroendocrine neoplasm and 1 of 11 positive nodes [15]. Jenkin et al reported an asymptomatic 69-year-old woman with a positive fecal occult blood test, a protruding appendiceal-orifice lesion on colonoscopy, no discrete mass on CT, and a normal-appearing cecal base at surgery; limited stapled cecectomy revealed a 6-mm grade 1 distal appendiceal NET [16].

The present case adds incremental evidence to these reports. Its value lies in the specific combination of CT-demonstrated appendiceal intussusception without a visible tumor or nodal disease, intraoperative difficulty with appendiceal retraction and mesenteric lymph nodes, and a 19-mm node-positive NET. This case does not show that these findings predict malignancy or that similar findings require right hemicolectomy.

Extent of Resection in the Present Case

Published evidence on the extent of resection for appendiceal intussusception consists mainly of case reports and retrospective reviews [2]. Appendicectomy or limited cecal resection may be appropriate when a satisfactory resection can be achieved without compromising the ileocecal junction and there are no gross features strongly suggesting malignancy [16]. More extensive oncological resection may be considered when operative findings raise concern for neoplasia or when limited resection is not technically appropriate [2,16].

Broader adult-intussusception literature supports operative exploration in selected patients with gastrointestinal bleeding, obstruction, a palpable mass, or a CT lead point [17,18]. These data are not validated appendiceal-specific criteria and do not determine the extent of resection in an individual case.

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In this patient, difficult appendiceal retraction and mesenteric lymph nodes, rather than a mass or lymphadenopathy on CT, raised concern about an occult pathological process and contributed to right hemicolectomy. The node-positive NET found on histopathology provides retrospective context only. This single case supports considering intraoperative findings alongside imaging, technical feasibility, and patient factors; it does not validate any specific operative finding as an indication for oncological resection.

Conclusions

A radiologically occult appendiceal NET can present as adult appendiceal intussusception. In this single case, CT identified the intussusception but not the tumor or nodal disease, whereas difficult appendiceal retraction and mesenteric lymph nodes influenced the decision to perform right hemicolectomy. The report supports integrating concerning intraoperative findings with imaging when individualizing the extent of resection, but it does not establish operative criteria or a general preference for operative assessment over imaging.

Department and Institution Where Work Was Done

Department of General Surgery, Royal Perth Hospital, Perth, Western Australia, Australia.

Patient Consent for Publication

Written informed consent was obtained from the patient for publication of the clinical details and accompanying images. The signed patient-consent form is supplied separately with the revised submission.

Declaration of Figures' Authenticity

All figures submitted have been created by the author who confirms that the images are original with no duplication and have not been previously published in whole or in part.

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