Cureus
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Open Access Case Report

Appendicitis in a Spontaneously Closed Malone Appendicostomy: A Case Report and Review of the Literature

Review began 10/27/2026
Review ended 11/06/2026
Published 11/10/2026

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DOI: 10.7759/cureus.119406

Krishna Vikneson [1, 2], Phillip J. Whiley [1, 2], Juliana Mai [1], Krishanth Naidu [1, 2]

1. General Surgery, Canberra Hospital, Canberra, AUS 2. School of Medicine and Psychology, Australian National University, Canberra, AUS

Corresponding author: Krishna Vikneson, krishna.vikneson@act.gov.au

Abstract

The Malone appendicostomy is an established treatment for refractory constipation in children with neurogenic bowel dysfunction or anorectal malformation. Once the conduit is no longer required, there is no published guidance on managing the redundant tract, and practice remains varied. Appendicitis arising within a spontaneously closed appendicostomy has been reported only three times. An 18-year-old patient with spina bifida, neurogenic bowel dysfunction, and a ventriculoperitoneal shunt presented with right iliac fossa pain, fever, and purulent discharge from a Malone appendicostomy created at eight years of age and unused for five years. Inflammatory markers were only modestly elevated, without leukocytosis. Computed tomography (CT) demonstrated an inflamed appendicostomy with surrounding fat stranding and subcutaneous collections, without a radiopaque fecolith. Open appendicectomy was performed through the appendicostomy site, and histopathology confirmed suppurative appendicitis with a 3 mm intraluminal fecolith. Three published cases of appendicitis following spontaneous closure and one of luminal obstruction within an actively used conduit were reviewed alongside the present case. Spontaneous closure of a Malone appendicostomy may leave a blind-ending appendix susceptible to luminal obstruction, and a normal white cell count and the absence of a radiopaque fecolith, both of which fall within the limits of routine investigation, should not exclude the diagnosis. As the first generation of appendicostomy recipients reaches adult life, this presentation may be encountered more often in adult surgical practice.

Categories: General Surgery, Pediatric Surgery, Gastroenterology
Keywords: acute appendicitis, appendicostomy, case report, faecolith, malone antegrade continence enema (mace), neurogenic bowel dysfunction, spina bifida, transition of care, ventriculoperitoneal (vp) shunt

Introduction

The Malone antegrade continence enema was first described in 1990 and uses the appendix as a continent, catheterizable conduit between the skin and cecum through which antegrade colonic enemas are administered [1]. It is an established treatment for refractory constipation in neurogenic bowel dysfunction and anorectal malformation. Complication rates of 30 to 80% have been reported in pediatric cohorts undergoing the procedure for anorectal malformation [2]. Most complications are minor, but some patients require revision surgery, and stomal stenosis is the most common reason, with an average reported rate of 16.5% [3]. A conduit may fall out of use once antegrade enemas are no longer required, and the cutaneous tract may then close without formal excision. No published guidance exists on the management of conduits that are no longer used, and practice remains variable.

Appendicitis within such a conduit differs from conventional appendicitis because the appendix has been mobilized and tunneled to the skin, so pain and inflammation tend to localize to the former stoma site rather than follow the classical migratory pattern. Acute appendicitis arising within a Malone appendicostomy is exceedingly rare, with only three cases reported, alongside a fourth report of luminal obstruction without appendiceal inflammation in an actively used conduit [4-7]. A further case arising within a spontaneously closed conduit in a patient with a ventriculoperitoneal (VP) shunt is reported here. The published cases are also reviewed alongside a discussion of why this uncommon presentation may become relevant to adult surgical units as patients who underwent appendicostomy formation in childhood reach adulthood.

Case Presentation

An 18-year-old patient presented to the emergency department with a 24-hour history of nonmigratory right iliac fossa pain, nausea, and new blood-stained mucoid discharge from the site of a previous Malone appendicostomy. The appendicostomy had been created at eight years of age as part of constipation management secondary to neurogenic bowel dysfunction and spina bifida. Five years later, the patient's bowel symptoms were manageable without antegrade enemas; the conduit was no longer accessed; and the cutaneous tract subsequently closed. The medical history also included a lipomyelomeningocele causing hydrocephalus, for which a VP shunt was inserted at five weeks of age.

On presentation, the patient was febrile at 37.8°C and tachycardic at 136 beats/minute but normotensive. Examination demonstrated peritonism in the right iliac fossa, and the previously closed cutaneous tract had broken down with purulent discharge from the former stoma site. The white cell count was 8.4 × 10^9/l (reference range: 4.0-11.0 × 10^9/l), and the C-reactive protein was 51.9 mg/l (reference range: <5 mg/l). Contrast-enhanced computed tomography (CT) demonstrated an inflamed appendicostomy with mucosal hyperenhancement, surrounding fat stranding, and two subcutaneous fluid collections measuring up to 30 mm in maximal axial diameter, without a radiopaque fecolith (Figure *1*). Stoma-site cellulitis and intra-abdominal abscess were also considered, but CT established the diagnosis. The clinical course is summarized in Table *1*.

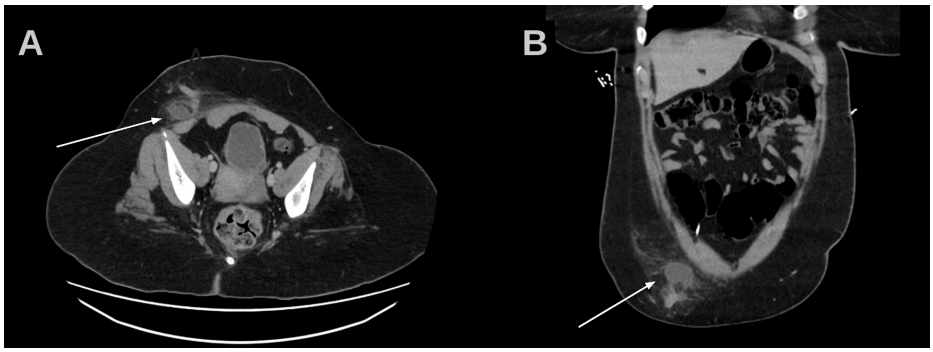


FIGURE 1: Contrast-enhanced computed tomography at presentation demonstrating features consistent with appendicitis and a fluid collection at the old appendicostomy site

(A) Axial view. (B) Coronal view. Arrows indicate the site of appendicitis and the associated fluid collection

Time point	Clinical events and findings	Investigations and interventions
Age 5 weeks	Lipomyelomeningocele with hydrocephalus	Ventriculoperitoneal shunt inserted
Age 8 years	Refractory constipation secondary to neurogenic bowel dysfunction and spina bifida	Malone appendicostomy created
Age 13 years	Bowel symptoms manageable without antegrade enemas	Appendicostomy no longer accessed, cutaneous tract subsequently closed
Age 18 years, day 0 (24 h of symptoms)	Nonmigratory right iliac fossa pain, nausea, blood-stained mucoid discharge from the appendicostomy site, febrile 37.8°C, tachycardic 136 bpm, normotensive, right iliac fossa peritonism	White cell count of 8.4 × 10^9/l, C-reactive protein of 51.9 mg/l, contrast-enhanced CT demonstrating an inflamed appendicostomy and two subcutaneous collections up to 30 mm in maximal axial diameter, intravenous amoxicillin-clavulanate commenced
Day 1	Imaging-confirmed appendicitis within the redundant conduit	Open appendicectomy through the appendicostomy site, intraperitoneal lavage avoided, Penrose drain placed
Postoperative day 1	Afebrile, tolerating diet, pain controlled	Discharge home with drain in situ and 5 days of oral amoxicillin-clavulanate
Postoperative day 5	Wound healing satisfactorily	Penrose drain removed
Histopathology	Suppurative appendicitis with a 3 mm intraluminal fecolith	No further intervention required
4 weeks postoperatively	Return to normal preoperative function, no evidence of recurrence or shunt infection	No further follow-up required

TABLE 1: Timeline of significant clinical events from appendicostomy formation to postoperative follow-up

bpm: beats per minute; CT: computed tomography

Intravenous amoxicillin-clavulanate was commenced preoperatively. Given the superficial location of the inflamed conduit, definitive management was undertaken by open appendicectomy through the previous appendicostomy site. An elliptical skin incision encompassing the appendicostomy exit site was made, and dissection continued down to the anterior fascia. The existing 3 cm × 3 cm fascial defect was extended to facilitate identification of the appendiceal base. The appendix was inflamed without intraperitoneal perforation, and the previously identified subcutaneous collections were uncomplicated seromas without intraperitoneal extension. No microbiological samples were sent, as definitive surgical source control was achieved at the same operation. Intraperitoneal lavage was deliberately avoided to minimize the risk of VP shunt contamination. The appendix base was doubly suture ligated prior to division; the peritoneum and fascia were closed in layers, and a Penrose drain was placed in the subcutaneous space prior to skin closure.

The patient was discharged on postoperative day one with a further five days of oral amoxicillin-clavulanate, and the drain was removed on postoperative day five. The gross specimen (Figure *2*) comprised the resected appendix with attached mesoappendix and surrounding fibrofatty tissue demonstrating dense fibrosis and marked thickening. Histopathological examination confirmed suppurative appendicitis with a 3 mm intraluminal fecolith that had not been visible on preoperative CT (Figure *3*). Sections through the appendicostomy site demonstrated keratinizing squamous epithelium at the skin surface with appendiceal mucosa tracking down toward it (Figure *4*). At four weeks, the patient remained well, with a healed wound, no recurrence of pain or discharge, and no evidence of VP shunt dysfunction or infection. No intraoperative or postoperative adverse events occurred.



FIGURE 2: Macroscopic pathological specimen of the resected appendiceal conduit

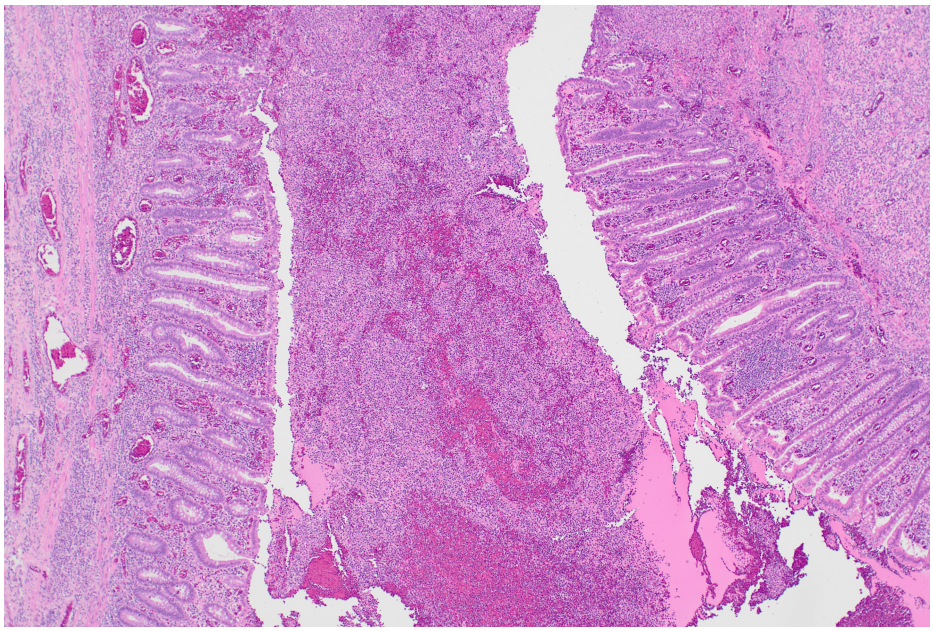


FIGURE 3: Histopathological slide of the resected appendiceal conduit demonstrating acute suppurative appendicitis

Hematoxylin and eosin, original magnification ×40. The lumen (center) is filled with purulent exudate. There is mucosal ulceration on either side and a transmural acute inflammatory infiltrate extending through the appendiceal wall

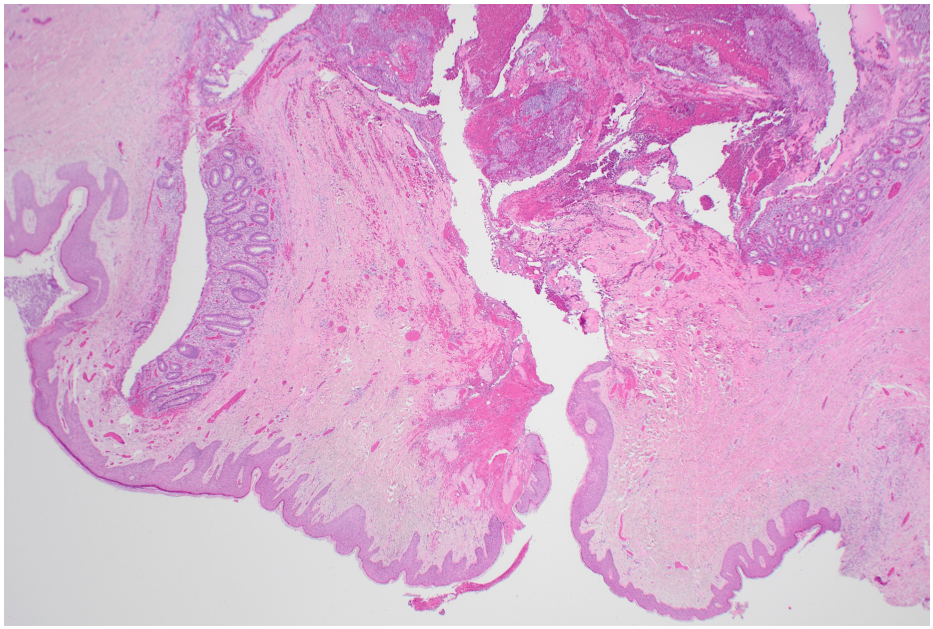


FIGURE 4: Low power histopathological slide of the appendicostomy site

Hematoxylin and eosin, original magnification ×20. Keratinizing squamous epithelium of the skin surface is seen at the lower border, with appendiceal mucosa tracking down towards it and inflamed granulation tissue within the intervening tract

Discussion

Suppurative appendicitis arose within a Malone appendicostomy that had closed after five years of disuse and was managed by open appendicectomy through the former stoma site. We propose, as a hypothesis rather than an established mechanism, that closure of the cutaneous tract contributes to this presentation. While the conduit remains patent, communication between the skin and cecum provides a route by which luminal contents can drain. Once the tract closes, that route is lost, and the appendix again behaves as a blind-ending structure in which obstruction cannot decompress. The cecal junction was not formally assessed operatively or histologically, so this mechanism is inferred rather than demonstrated. Histological sections through the appendicostomy site showed appendiceal mucosa tracking towards keratinizing squamous epithelium at the skin surface (Figure *4*), which confirms the anatomy of the conduit although it cannot establish when the tract lost continuity. The purulent discharge at presentation did not indicate a persistently patent conduit. The tract had been closed for five years and broke down during the acute episode, allowing the obstructed conduit to decompress through the old tract to the skin. The report of a partially obstructing fecolith within an actively used, patent conduit [7] shows that obstruction can arise irrespective of patency, so what closure plausibly alters is not whether obstruction occurs but whether it can decompress. The presenting features, investigations and management of the published cases are summarized alongside the present one in Table *2*. The report by Abbes Orabi et al. is included for completeness but is pathophysiologically distinct, describing noninflammatory luminal obstruction within an actively used conduit rather than appendicitis after spontaneous closure. Of the three cases of appendicitis following closure, only one describes the operative approach in detail [4], and each was managed differently.

First author, year	Appendicostomy status	Presentation	Imaging	Laboratory findings	Management strategy
Bosley et al., 2023 [4]	Spontaneously closed	Abdominal pain and erythema, no hemodynamic compromise or fever	CT: appendicitis with fecolith	No leukocytosis	Appendicectomy via minimally invasive circumferential dissection
Lacrosse and Dragean, 2022 [5]	Spontaneously closed	Pain, fever, periumbilical inflammation, nausea and diarrhea	CT: appendicitis with large fecolith	Discrete rise in CRP	Initial conservative management, interval appendicectomy after recurrent symptoms
McAndrew et al., 2002 [6]	Not used, had spontaneously closed	Abdominal pain, vomiting, diarrhea and tender abdominal lump	US: superficial lump suggestive of abscess	Not reported	Appendicectomy (method not described) plus antibiotics
Abbes Orabi et al., 2011 [7]	Active (patent conduit, noninflammatory obstruction)	Reflux of enema liquid	CT: fecolith in appendicostomy	Not reported	Cecotomy with fecolith removal via low midline laparotomy
Present case	Spontaneously closed	Right iliac fossa pain, nausea, purulent discharge from the appendicostomy site, fever	CT: appendicitis without radiopaque fecolith, two 30 mm subcutaneous collections	CRP: 51.9 mg/l; no leukocytosis	Open appendicectomy through the appendicostomy site; lavage avoided (VP shunt in situ)

TABLE 2: Reported cases of appendicostomy-associated appendicitis or luminal obstruction

CRP: C-reactive protein; CT: computed tomography; US: ultrasound; VP: ventriculoperitoneal

The Malone appendicostomy was described only in 1990 and has been performed predominantly in children, so its earliest recipients are now well-established in adulthood [1]. Survival into adult life in conditions such as spina bifida has also improved substantially [8]. Adult surgical services may therefore encounter conduits created in childhood and later abandoned, often in units unfamiliar with the original operation and without access to paediatric operative records. The present patient had a conduit created at eight years of age and abandoned at thirteen, which illustrates this scenario. Neurogenic bowel dysfunction secondary to spinal dysraphism would not be expected to resolve, and the reasons a conduit falls out of use are often poorly documented. In this patient, the conduit was abandoned once bowel symptoms were manageable without antegrade enemas rather than because of any demonstrated recovery of intrinsic bowel function. Previous appendicostomy formation should be recognized as relevant surgical history in any patient presenting with pain or inflammation at a childhood stoma site.

Presentation appears to differ from conventional appendicitis because of the altered anatomy. Among the three previously reported cases arising after spontaneous closure, pain and inflammatory change were predominantly localized to the former stoma site [4-6]. Systemic inflammatory responses were modest where reported [4,5]. Although the previously reported cases imaged with CT demonstrated a fecolith, none was identified preoperatively in this patient. A 3 mm noncalcified fecolith lies at or below the threshold at which CT reliably resolves such a lesion, so its nonvisualization reflects a recognized limitation of imaging rather than an atypical feature of this presentation. A normal total white cell count is sometimes seen in acute appendicitis. Neither finding should be taken to exclude the diagnosis when the clinical picture is compatible.

Because the underlying pathology appears structural and involves a conduit predisposed to recurrent obstruction, management should aim at definitive removal of the source rather than a temporizing measure. One previous case was managed conservatively with antibiotics and required interval appendicectomy years later after recurrent episodes of appendicitis [5]. The other three underwent upfront surgery by subumbilical laparotomy and cecotomy for fecolith removal, by minimally invasive circumferential dissection, and by an undescribed approach, all with satisfactory outcomes [4,6,7]. Although the evidence is limited to isolated case reports, definitive appendicectomy appears reasonable in surgically fit patients because conservative treatment leaves the anatomical substrate for recurrent obstruction in situ.

The VP shunt added a further consideration to the operative approach. Intra-abdominal infection may increase the risk of shunt infection, particularly in the setting of perforated appendicitis [9]. Whether shunt externalization is required remains uncertain, and the available evidence is derived largely from small

pediatric series [10,11]. Uncomplicated appendicitis in this patient permitted early definitive source control without externalization. Intraperitoneal lavage was avoided to minimize potential contamination, although evidence supporting this strategy is lacking.

The present case also raises an unanswered question regarding the long-term management of redundant Malone appendicostomies. Current practice varies between formal surgical excision and spontaneous tract closure, and no guidelines exist to inform decision-making. Routine prophylactic appendicectomy cannot be recommended on the limited available evidence, but retained conduits remain susceptible to subsequent pathology, and whether formal excision should follow permanent abandonment warrants further study. These observations are drawn from a single patient and four heterogeneous published cases, and the absence of incidence data means that any change in the frequency of such presentations cannot be quantified. The comparison presented here is narrative rather than systematic, as no predefined search protocol, databases, or screening criteria were applied, and further reports may exist that were not identified. Its value lies in bringing together the previously reported cases, which have not been reviewed collectively, and in proposing a mechanism that can be tested against future reports.

Conclusions

Spontaneous closure of a Malone appendicostomy may not be a benign anatomical endpoint, as it can leave a blind-ending appendix in situ and, with it, a persistent baseline risk of appendicitis. In this patient, the closed cutaneous tract broke down during the acute episode, which accounted for the discharge at the former stoma site. Appendicitis should be considered in any patient presenting with pain or inflammation at a previous appendicostomy site, even many years after closure, and a normal white cell count or the absence of a radiopaque fecolith on CT, both of which fall within routine clinical and radiological limits, do not exclude it. As patients who underwent appendicostomy formation in childhood may increasingly present to adult surgical care, the optimal management of redundant appendiceal conduits warrants further study.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Krishna Vikneson, Phillip J. Whiley, Juliana Mai, Krishanth Naidu

Acquisition, analysis, or interpretation of data: Krishna Vikneson, Phillip J. Whiley, Juliana Mai, Krishanth Naidu

Drafting of the manuscript: Krishna Vikneson

Critical review of the manuscript for important intellectual content: Krishna Vikneson, Phillip J. Whiley, Juliana Mai, Krishanth Naidu

Supervision: Juliana Mai, Krishanth Naidu

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. ACT Health Human Research Ethics Committee issued approval N/A. The institutional policy of Canberra Health Services exempts single-patient case reports from Human Research Ethics Committee review. The patient provided written informed consent for the publication of their clinical data and images, and the signed consent form has been retained by the department and can be provided on request. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

Acknowledgements

The authors thank Dr. Lee Bornman from the Anatomical Pathology Department, The Canberra Hospital, Australia, for his histopathological analysis and for providing the gross specimen image. The authors would like to thank Dr. Ailene Fitzgerald for her clinical expertise and care of the patient described in this case report. Claude (Anthropic, San Francisco, USA) was used to assist with manuscript formatting, language editing, and restructuring of author-written content to meet journal-specific requirements. No artificial intelligence tool was used to generate clinical content, data, or their interpretation. All authors reviewed and approved the final manuscript and take full responsibility for its content.

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