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Double Appendix with Acute Inflammation of Both Appendices in a 60-Year-Old Man: A Rare Anomaly Not Suspected on Preoperative Ultrasonography

[1]Dr. Amol Vanketrao Chavare, General Surgeon, Hospital-Bhakt Diagnostic and Multispecality Hospital, Nanded, (MH), India

[2]Dr.Vinod Naik, Professor and Director, Department of General surgery, KEM Hospital, Pune

[3]Dr. Vikas Rathod, Radiologist, Hospital-Bhakt Diagnostic and Multispecality Hospital, Nanded, (MH), India

Corresponding Author: Dr. Amol Vanketrao Chavare, General Surgeon, Hospital-Bhakt Diagnostic and Multispecality Hospital, Nanded, (MH), India

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Abstract

Background: Duplication of the vermiform appendix is an exceedingly rare congenital anomaly, generally found by chance at operation or on histology. A second appendix that goes unrecognised can leave behind a source of persisting or recurrent sepsis and cause diagnostic confusion.

Case presentation: A 60-year-old man with clinical features of acute appendicitis underwent abdominal and pelvic ultrasonography, which showed a single non-peristaltic, non-compressible tubular structure (internal diameter 8.7 mm) in the right iliac fossa, with surrounding inflammatory change and several intraluminal appendicoliths, the largest about 12 mm. At open appendectomy through a McBurney incision, two inflamed appendices arising from the caecum were found, one containing an appendicolith. The appendicular artery of each was secured separately, both were ligated at their bases and both were excised. Histology of both specimens showed transmural acute inflammation, colonic-type mucosa and lymphoid follicles, consistent with acute appendicitis in a duplicated appendix (modified Cave–Wallbridge type B).

Conclusion: Ultrasonography gave no hint of duplication. Whenever one inflamed appendix is encountered, the caecum should be inspected deliberately for a second, because an inflamed duplicate left in situ can sustain sepsis after surgery.

Keywords: Appendiceal Duplication, Double Appendix, Acute Appendicitis, Appendicolith, Cave–Wallbridge Classification, Case Report

Introduction

Acute appendicitis is the commonest abdominal surgical emergency, carrying an estimated lifetime risk of 7–8%.[1] Appendectomy is consequently among the operations performed most often, and the surgeon enters the abdomen expecting a single inflamed appendix springing from the caecal base, as predicted by the history, examination, laboratory tests and imaging. When the anatomy departs from this pattern, both the conduct of the operation and the postoperative course may be affected.

Developmental variants of the appendix include absence, duplication, triplication, malposition and, more recently described, the horseshoe appendix.[2] Duplication is the variant reported most often, with a quoted incidence of about 0.004%.[2] Large specimen series, such as Collins' study of 50,000 appendices, underpin estimates of how often these anomalies occur.[3] Up to 2015 fewer than 100 cases had been published, and fewer than 15 of them were complicated by acute appendicitis.[4]

The anomaly spans a spectrum from a branching appendiceal trunk to a fully formed second appendix situated elsewhere along the colon.[2] It may occur alone or together with duplication of the caecum,[4] and a single case of triplication has been documented.[5] The pathogenesis is unsettled. Aberrant development of the caecum during midgut rotation, with persistence of more than one appendiceal bud, is the usual explanation,[1] and an embryological aetiology was proposed by Coker and colleagues.[4]

The classification in current use was introduced by Cave,[6] extended by Waugh[7] and Wallbridge,[8] and later modified to accommodate caecal duplication[9] (Table 1). Briefly, type A is a single caecum bearing one partly or completely divided appendix with a common base; type B is a single caecum bearing two separate appendices, placed symmetrically on either side of the ileocaecal valve (B1) or with the second lying along the taenia coli (B2); and type C is a pair of caeca, each carrying its own appendix.[4,10] Among classifiable published cases, type B2 predominates.[2] For histological confirmation, Akhtar et al. require an intact appendiceal structure, tip included, in both specimens.[11]

The clinical weight of the anomaly lies in missing it. A second appendix is seldom suspected beforehand and is often first noticed in theatre or on the pathology slide; overlooking it can lead to treatment failure and to medicolegal consequences.[2,12] Inflammation may involve only one of the pair,[10] and both rupture of a second appendix after removal of the first[13] and sequential appendicitis after an earlier appendectomy[1] have been reported. Nageswaran et al. advise exploring the caecum, including the retrocaecal space, whenever duplication is suspected.[2] Cross-sectional imaging has demonstrated a true duplication in an isolated report,[10] but it is not part of routine diagnostic pathways.

Congenital variants are largely absent from guidelines and meta-analyses on acute appendicitis, high-level evidence for their management is lacking, and wider reporting has therefore been encouraged.[1] We describe a 60-year-old man in whom ultrasonography showed a single inflamed appendix, yet two inflamed appendices arising from the caecum were found at open appendectomy. The report follows the CARE guidelines.[14]

Table 1: Modified Cave–Wallbridge classification of appendiceal duplication and placement of the present case

Type	Anatomical description	This case

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A	Single caecum with one appendix that is partly or completely duplicated; common base	
B1	Single caecum with two separate appendices, symmetrically on either side of the ileocaecal valve	Type B confirmed; subtype not recorded
B2	Single caecum with a normally sited appendix plus a second along the taenia coli (most frequent in classifiable reports)	Type B confirmed; subtype not recorded
C	Duplicated caeca, each with its own appendix	—

Compiled from references 2, 4, 6–10.

Case Presentation

Patient information

- A 60-year-old man was referred by his treating physician for ultrasonography of the abdomen and pelvis because of features suggestive of acute appendicitis, and was then operated on at our institution. Periumbilical pain that migrates to the right iliac fossa, with anorexia, nausea, vomiting and low-grade fever. There was no previous abdominal surgery or comorbidity, and no regular medication, drug allergy or relevant family history.
- Clinical findings: the patient was stable and ill-looking, with tenderness at McBurney's point, guarding, rebound and a positive Rovsing's sign. Digital rectal examination was unremarkable and the chest and cardiovascular examinations were normal.

Timeline

Time point	Event
Presentation	Clinical features of acute appendicitis; referred for imaging
Pre-operative	Ultrasonography of abdomen and pelvis: single inflamed tubular structure with appendicoliths; impression of acute appendicitis
Day of surgery	Open appendectomy under spinal anaesthesia via McBurney incision; two inflamed appendices found and removed
Post-operative	Both specimens sent separately for histopathology; acute appendicitis with two appendices (reported with a query)

Diagnostic assessment

Ultrasonography of the abdomen and pelvis. The key findings are listed below.

- An elongated tubular structure in the right iliac fossa that was non-peristaltic and non-compressible, with marked inflammatory change in the surrounding tissue; internal diameter approximately 8.7 mm.
- Several large intraluminal appendicoliths, the largest about 12 mm.
- Minimal free fluid between bowel loops, with oedematous, dilated loops in the right and left para-umbilical regions.
- Incidental: spleen 11.8 cm with normal echotexture (borderline splenomegaly). Liver, biliary tree, portal vein, gallbladder, pancreas, both kidneys (right 82 × 36 mm, left 88 × 38 mm), bladder and prostate were unremarkable.

The radiologist's impression was acute appendicitis, with advice for clinical–pathological correlation or CT of the abdomen. Only one tubular structure was described, and no second appendiceal structure was reported.The working diagnosis was acute appendicitis with appendicolith, and the patient was taken to theatre.

Therapeutic intervention-

Open appendectomy was performed under spinal anaesthesia with the patient supine. After skin

preparation and draping, a McBurney incision was made; the external oblique, internal oblique and transversus abdominis were split in the line of their fibres and the peritoneum was opened. Two appendices were delivered into the wound (Figure 1). Both arose from the caecum, both were inflamed, and one contained an appendicolith. The appendicular artery to each was identified and controlled separately, each appendix was ligated at its base and excised, and haemostasis was confirmed. The peritoneum and muscle layers were closed with 2-0 polyglactin (Vicryl), followed by subcutaneous and skin closure and a dressing. The operation note recorded the diagnosis as appendiceal duplication with appendicitis and appendicolith. The site of origin of each appendix relative to the ileocaecal valve and taenia coli was not documented. Both specimens were sent for histopathology.

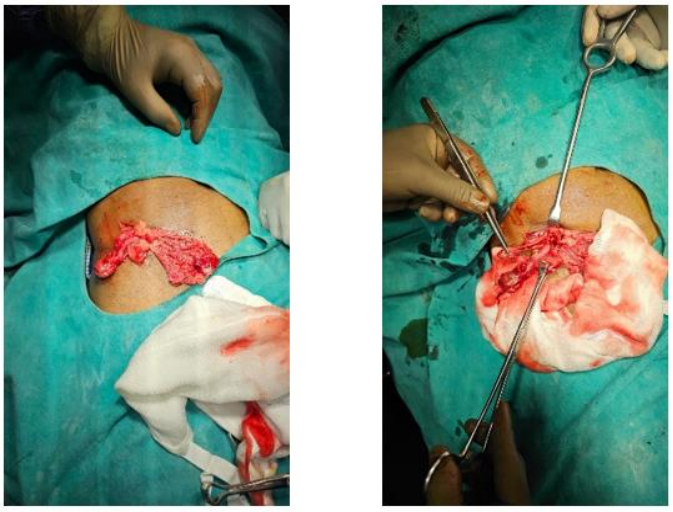


Figure 1: Intra-operative findings at open appendectomy. (A) McBurney incision with inflamed appendiceal tissue delivered into the wound. (B) Operative field with the inflamed appendices held on instruments for dissection, control of the appendicular vessels and ligation of the bases.

Histopathology

Two specimens, labelled A and B, were examined (Table 2, Figure 2). Both were lined by colonic-type mucosa with lymphoid follicles in the lamina propria and showed acute inflammation extending through the full thickness of the wall to the serosa. The pathologist's impression was acute appendicitis with two appendices; this was expressed with a query, and no faecolith was mentioned in the report.

Table 2: Gross and microscopic features of the two excised specimens

Feature	Specimen A	Specimen B
Gross size	3 × 0.5 cm tubular structure	5 × 0.5 cm tubular structure; 2 cm wide at one end, tapering to 0.5 cm at the other
Lumen	Open lumen on sectioning	Faecal material present on opening
Mucosa	Colonic-type epithelium, unremarkable	Colonic-type epithelium, focally ulcerated and covered by neutrophilic exudate
Lymphoid tissue	Numerous lymphoid follicles in lamina propria	Lymphoid follicles in lamina propria
Wall	Oedema with dense neutrophilic infiltrate reaching the serosa; a few congested vessels	Neutrophils and eosinophils in submucosa, muscularis propria and serosa
Serosa	Neutrophilic infiltrate reaching the surface	Covered by exudate; a few congested vessels
Diagnosis	Acute (transmural) appendicitis	Acute (transmural) appendicitis



Figure 2: Gross specimens. (A) Excised appendiceal specimens on the sterile drape beside artery forceps. (B) The two excised appendices displayed together on a gloved hand.

Discussion

Confirming the diagnosis. The diagnosis of duplication in this patient rests on three observations: two separate tubular structures arising from the caecum at operation; gross features compatible with appendices; and histology of both specimens showing colonic mucosa, lymphoid follicles and a muscular wall with transmural acute inflammation. Strictly, Akhtar's criterion calls for an intact appendiceal structure along the whole length of both specimens, tip included.[11] Here the second specimen was dilated and contained faecal material, and the pathologist reported with reserve. We therefore regard the evidence as strongly supportive rather than absolute. The principal alternative, an inflamed caecal diverticulum or other caecal outpouching, was implied by the pathologist's cautious wording but cannot be excluded entirely from the description available, although lymphoid follicles and a muscularis propria favour an appendix.

Classification. On the modified Cave–Wallbridge scheme,[4] two separate appendices with their own bases on a single caecum constitute type B. Allocation to B1 or B2 depends on where each base lies in relation to the ileocaecal valve and taenia coli, which the operative record does not state; the case is therefore type B, subtype undetermined. Type B2 is the form reported most often,[2] and recent reports of sequential appendicitis after a previous appendectomy have also described type B2.[1]

Embryology and associated anomalies. The origin remains uncertain. Duplication is generally ascribed to abnormal development of the caecal bud during midgut development, so that more than one appendiceal outpouching persists.[2,6] Nageswaran et al. suggest considering types B1 and C in patients who have had earlier surgery for congenital abnormalities,[2] and a person with a duplicate appendix may reasonably merit attention to other congenital anomalies.

Limits of preoperative imaging. The case is instructive for two reasons. First, the ultrasound findings, one inflamed tube with appendicoliths, could not tell a single appendix from a double one, and a preoperative diagnosis of duplication is rarely achievable.[2] Imaging can reveal a second structure in selected cases, but ultrasound is operator dependent and a normal-looking second appendix is easily overlooked.

Both appendices were inflamed. Second, and unusually, both appendices in our patient were inflamed. Appendicitis in a duplicated appendix has been described in several case reports,[4,15] including inflammation confined to one appendix of the pair[10] and rupture of the second appendix after removal of the first.[13] Appendicitis in the remaining appendix after an apparently successful operation has caused recurrent

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right iliac fossa pain and delayed diagnosis,[1] and the implications for emergency physicians have also been discussed.[16] Mahmood et al. recommend removing the second appendix even when only one is inflamed, since both carry the same risk;[10] this is a reasonable policy. In open or laparoscopic surgery, Nageswaran et al. advise a deliberate search along the taenia coli and around the ileocaecal junction whenever an appendix looks small or unusually placed, or the findings do not match the clinical picture.[2] Because triplication has also been described,[5] finding two appendices should not end inspection of the caecum.

Strengths and limitations. Strengths of this report are that both appendices were removed, each appendicular artery was controlled individually, both specimens were submitted separately for histology, and the operation was documented photographically. Limitations are the single-case design; the absence of a recorded anatomical relationship of each appendix to the ileocaecal valve and taenia coli; the lack of comment on a faecolith in the histology report, although appendicoliths were seen on ultrasonography and at operation (the specimen may not have been examined for one, or it may have been lost during handling)

Conclusion

Appendiceal duplication should be borne in mind at every appendectomy, even when imaging shows a single inflamed appendix. Systematic inspection of the caecum for a second appendix, removal of both when present, and separate histological examination of each specimen reduce the risk of missed pathology.

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