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CASE REPORT

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Appendiceal duplication an unusual cause of abdominal pain: A case report

Yohannis Derbew Molla[1] | **Menarguachew Atanaw Sisay**[1] | **Samuel Addisu Abera**[2] | **Bewketu Abebe**[2] | **Girma Damtew Adisu**[2]

[1]Department of Surgery, College of Medicine and Health Sciences, Gondar, Ethiopia

[2]Department of Pathology, College of Medicine and Health Sciences, Gondar, Ethiopia

Correspondence
Yohannis Derbew Molla, Department of Surgery, College of Medicine and Health Sciences, Gondar, Ethiopia.
Email: yderbew73@gmail.com

Key Clinical Message

Appendiceal duplication is exceedingly rare, with a prevalence of 0.004% to 0.009% in appendectomy specimens. Appendiceal duplications can occur alone or in conjunction with cecal duplication. The persistence of the temporary embryologic second cecal appendix is hypothesized to cause appendiceal duplications. We present a case of appendiceal duplication in a 26-year-old Ethiopian female patient who had been experiencing abdominal pain in the right lower quadrant for 1 week. She developed anorexia, a loss of appetite, and a low-grade fever as a result of this. She reported direct and rebound mild discomfort in the right lower quadrant on abdominal examination. She was then operated on and she had an appendiceal duplication intraoperatively. As a result, an appendectomy was performed, and the patient was discharged with improved health. To avoid unfavorable patient outcomes and medicolegal difficulties, surgeons and surgical trainees who conduct several appendectomies throughout their training should be aware of the likelihood of appendiceal duplication.

KEYWORDS

pathology and laboratory medicines, surgery

1 | INTRODUCTION

One uncommon congenital anomaly that has not been extensively discussed in the literature is appendiceal duplication. Agenesis, duplication, the horseshoe anomaly of the appendix, triplication, and the abnormal location of a single appendix are all examples of appendiceal anomalies.[1] The incidence of appendiceal duplication has been previously reported to be approximately 0.004%–0.009%.[2] Adults account for the majority of appendix anomalies identified, and the majority of these cases were discovered during surgery, which was primarily done for the other reasons such as GI and gynecologic surgery.[3] Though the exact cause of the appendiceal duplication is unknown, it is assumed that the persistence of a transient, normally developing second cecal appendix is to blame.[4] Appendicitis is the most frequent pathology affecting the appendix; however, pathology in abnormal appendices is significantly less frequent, with only a few reported cases.[5] In patients with right lower quadrant pain, appendiceal duplications present a challenging clinical picture. The clinical characteristics resembled an appendix with one or more duplications. The majority of cases of appendiceal duplication are diagnosed intraoperatively or during

a pathological examination, despite the fact that preoperative diagnosis is possible with the help of radiological studies like CT scans, barium studies, and abdominal ultrasound.[6] Here we present a case of appendiceal duplication in a young patient.

2 | CLINICAL PRESENTATION

A 26-year-old Ethiopian female presented with abdominal pain in the right lower quadrant area for 1 week. Associated with this, she had anorexia, loss of appetite, and a low-grade fever but no history of vomiting, diarrhea, or abdominal distension. She had a similar episode a year ago, but it subsided by itself at that time. Otherwise, she had no history of trauma to the abdomen, pain during urination, frequency, or urgency. She had regular menses, and she was not on any form of contraceptive. Her urine HCG was negative. She had no history of diabetes, hypertension, or asthma. She had no history of drug intake. She had no history of any known allergies.

On examination, her vital signs were within the normal range. She had a clear and resonant chest. On abdominal examination, she had mild direct and rebound tenderness in the right lower quadrant. There was voluntary guarding in the right lower quadrant but no mass. Otherwise, the rest of the examinations were unremarkable. Subsequently, she was investigated with a complete blood count, which was normal with a white blood cell count of 8700/microliter of blood and a neutrophil count of 59.4%, and her hemoglobin was 2.6 g/dL. Abdominal ultrasound showed a thickened appendix with an anteroposterior diameter of 9 mm but no signs of perforation, lymphadenopathy, or fluid collection. Subsequently, the patient was resuscitated and operated on under general anesthesia and in a supine position.

An open appendectomy was done with a right lower quadrant transverse incision since our patient had symptoms and signs localized to the right lower quadrant. However, in patients with generalized abdominal tenderness and other types of intestinal duplications, midline incisions can be used. Intraoperatively, there was an enlarged appendix with a bifurcation at the tip but no perforation or fecolith (Figure 1). There was minimal reactive fluid in the peri-appendiceal area but no pus. Therefore, the mesoappendix was identified and ligated, an appendectomy was done, local mopping was done, and the abdomen was closed in layers. The sample was sent for histopathologic analysis. Postoperatively, she was put on ceftriaxone 1gm IV twice a day and metronidazole 500 mg IV three times a day for 24 h. She was also given diclofenac 50 mg IV twice a day. The patient started feeding after 12 h of the operation, and she tolerated it well.

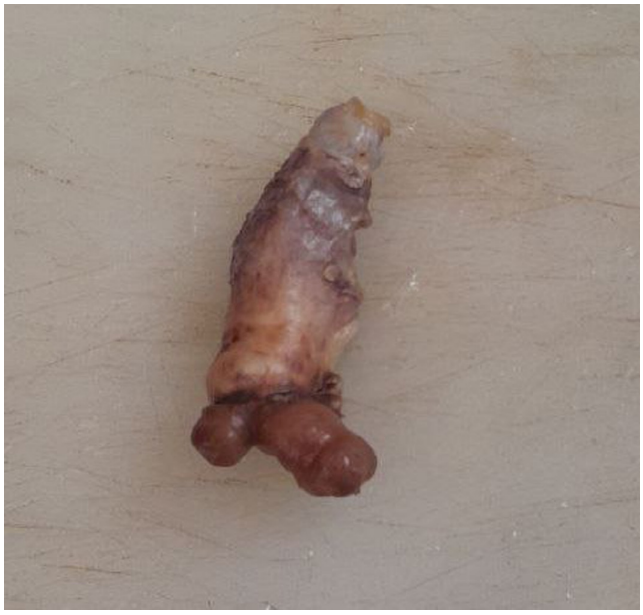

FIGURE 1 Gross appearance of the appendix after surgical removal.

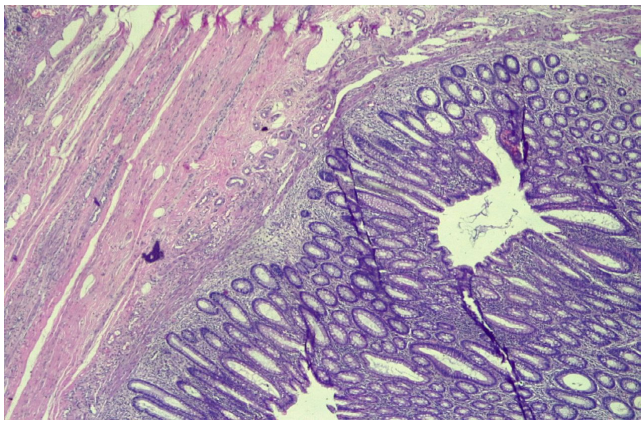

FIGURE 2 Histopathology showing appendiceal duplications which shared some muscularis propria with the appendix and lined by similar epithelium of the appendix.

Therefore, she was discharged from the hospital after 24 h of hospitalization. Histopathology showed normal columnar-lined epithelium with mildly lymphoplasmacytic, eosinophilic, and neutrophilic infiltrated lamina propria and normal muscularis propria, and serosa. There was attached tissue that shared the same muscularis propria with the appendix and was lined by similar epithelium with the appendix, with the index of appendiceal duplication, Wallbridge type A (Figures 2 and 3; Data S1). On subsequent follow-up

at the surgical referral clinic, the patient had no recurrence of symptoms.[7]

3 | DISCUSSION

Duplications of the gastrointestinal tract can occur anywhere, from the mouth to the anus. They are a rare type of congenital anomaly. The mucosa of the digestive tract lines single, variable-sized duplications that are more spherical than tubular in shape (Figure 4). The adjacent bowel and the duplicates typically share a smooth muscle wall and blood supply, which allows for communication.[8] In appendectomy specimens, the incidence of appendiceal duplication ranges from 0.004% to 0.009%.[9] Only two such cases were discovered by Collins in the 50,000 appendices.[4] The possibility of appendiceal duplication should be known to surgeons and surgical residents who perform numerous appendectomies during training in order to avoid adverse patient outcomes and medical-legal concerns. Appendiceal duplication is a very uncommon clinical diagnosis, so misdiagnosis and poor management are frequent occurrences. However, diagnosis may not be difficult in type A duplication. A second appendix may take longer to diagnose, which could increase the risk of gangrene and perforation. In addition to colonic adenocarcinoma, epiploic appendagitis, Meckel's diverticulum, stump appendicitis, gastroenteritis, acute mesenteric adenitis, intussusception, inflammatory bowel disease, and genitourinary pathology, a differential diagnosis for appendiceal duplication should include congenital cecal diverticulum.[10]

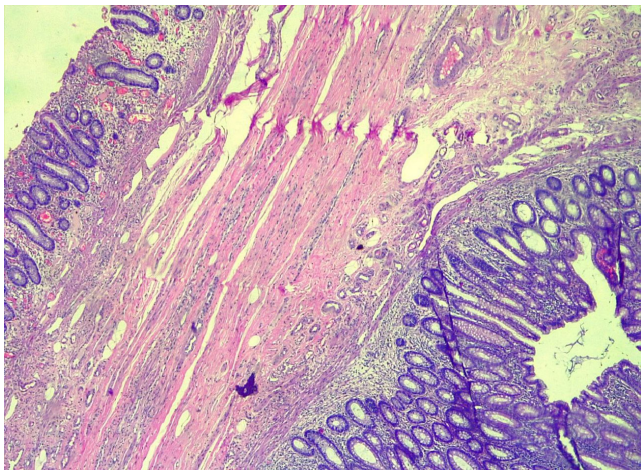

FIGURE 3 Histopathology showing appendiceal duplications which shared some muscularis propria with the appendix and lined by similar epithelium of the appendix.

In 1963, Wallbridge classified appendiceal duplication into the following categories (Figure 5).[11]

Biermann in 1993 classified appendiceal anomalies by the following classification (Table 1).[12]

In addition to the classification above, there have been reports of triple appendix and a horseshoe appendix in which the appendix has two openings into the common cecum.[13] Table 1 illustrates Biermann classification of appendiceal anomalies.

On histopathologic examination, the diagnosis of appendix duplication is only made when both specimens show an intact structure (including the tip) with lumens lined by typical appendiceal mucosa, lymphoid follicles, and two layers of musculature.[14] A single diverticulum of the cecum and appendix duplication must be distinguished from one another; histological examination is the best method for doing so. On the inside of the ileocecal angle, a cecal diverticulum is typically present, and under a microscope, it has no lymphoid tissue in its wall.[15] Diverticulosis of the appendix, which is seen in 0.004%–2.1% of specimens and in 1% of appendectomies, is another pathology that needs to be taken into account in the

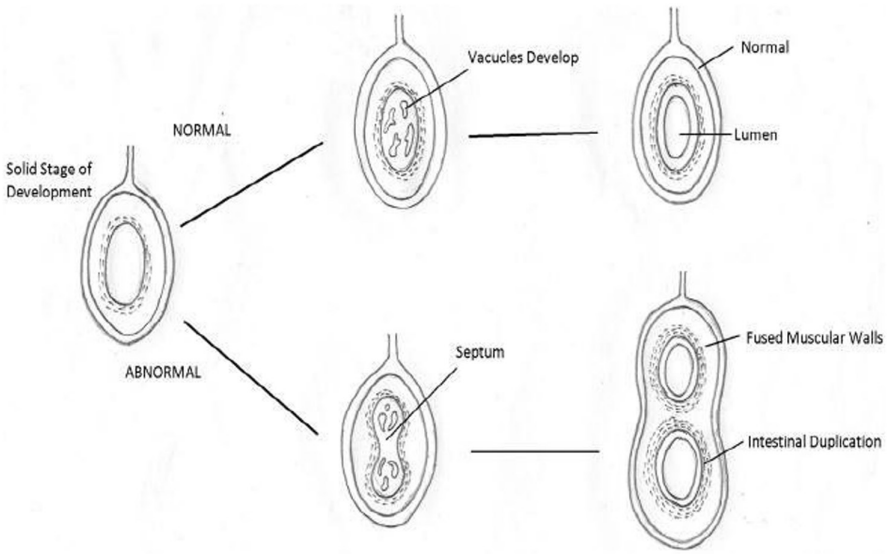

FIGURE 4 Schematic representation showing intestinal duplication (reproduced from Ganesh (11)).

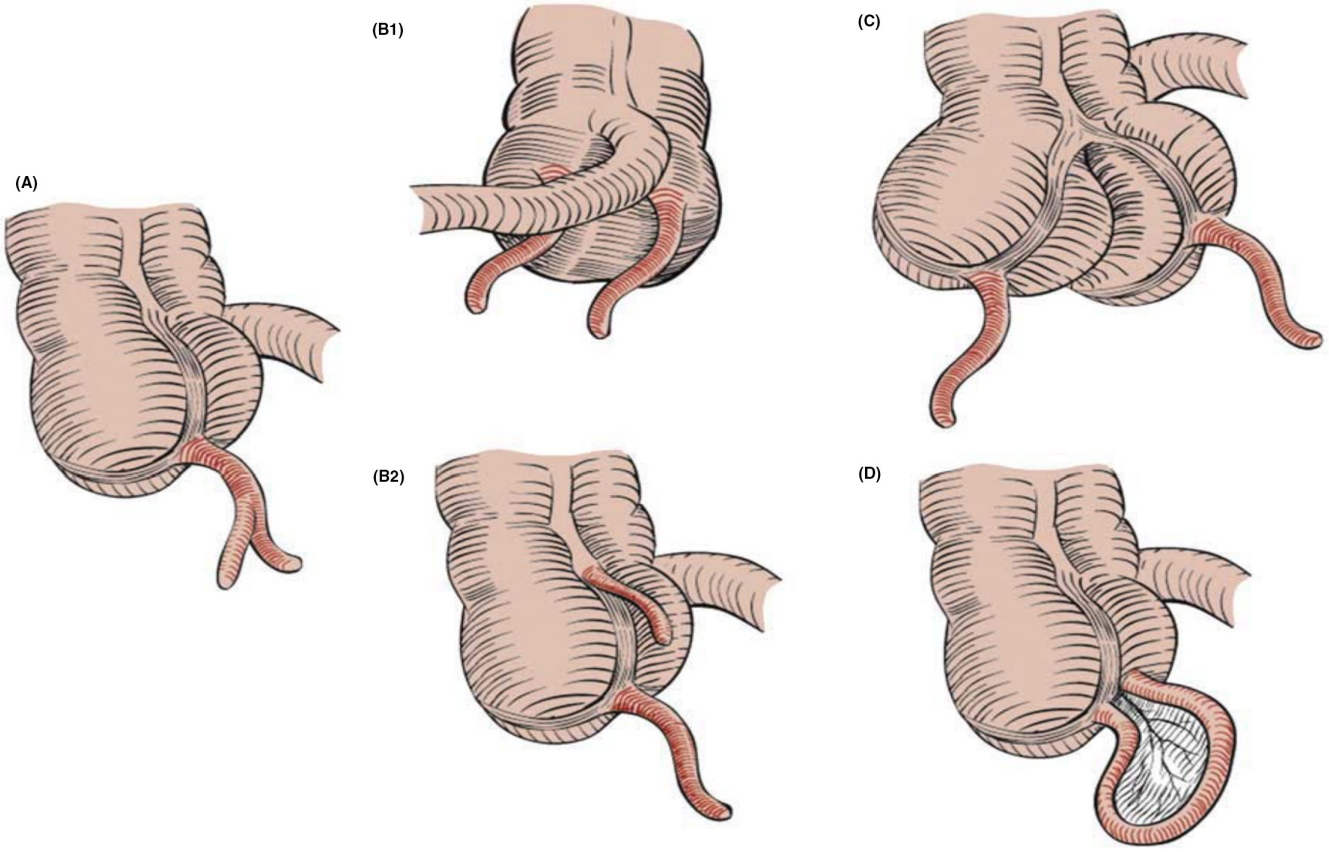

FIGURE 5 Wallbridge classification of appendiceal duplication.

TABLE 1 Biermann classification of appendiceal anomalies.

Type		Morphologic characteristics (appearance)
Type A		Partial duplication of the appendix on a single cecum, the patient this type of malformation
Type B		Two completely separate appendices with a single cecum with two subtypes
	B1	"bird-like appendix": Two appendices symmetrically placed on either side of the ileo-cecal valve and it usually found in birds. If it is found in humans, it is associated with intestinal and/or genitourinary anomalies
	B2	"Taenia-coli type": One appendix arises from the usual site on the cecum with another rudimentary arising from the cecum along the tenia of the cecum
	B3	The second appendix is located along the tenia of the hepatic flexure of the colon
	B4	The second appendix is located along the tenia of the splenic flexure
Type C		Double cecum, each bearing an appendix. This type is usually association with hindgut mal-development (ileum, colon, and anus) and other lower vertebral column and genitourinary anomalies
Type D		Three completely separate appendices with or without other anomalies

Note: N.B. Type B2, B3, and B4 are usually not associated with other congenital anomalies.

differential diagnosis. However, all of the reported cases of appendiceal diverticulosis involved older adults who had persistent symptoms and smaller-sized masses.[16] A duplex appendix should be regarded as an unlikely but possible diagnosis when a patient with a history of appendectomy exhibits clinical appendicitis. Congenital abnormalities, particularly gastrointestinal or genitourinary anomalies, should be suspected in patients with Wallbridge type B1 or type C duplication. The most frequent type of duplication is type B2, and to prevent misdiagnosis, careful examination of the cecal pole and retrocecal space should be performed when an anterior appendix is discovered with

inflammation along the right paracolic gutter or a normal appendix in the presence of convincing appendicitis.[17]

4 | CONCLUSION

A rare but clinically and medico-legally significant clinical condition is appendiceal duplication. All medical professionals (surgeons and residents) performing appendectomies should be aware of the possibility of appendiceal duplication given the prevalence of appendectomies. Even in patients who had appendicectomies, lower abdominal pain should always be investigated for appendiceal duplications.

AUTHOR CONTRIBUTIONS

Yohannis Derbew Molla: Conceptualization; writing – original draft. **Menarguachew Atanaw Sisay1 Atanaw Sisay:** Conceptualization. **Samuel Addisu Abera:** Writing – review and editing. **Bewuketu Abebe:** Writing – review and editing. **Girma Damtew Adisu:** Writing – review and editing.

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Not applicable.

CONFLICT OF INTEREST STATEMENT

No potential conflict of interest relevant to this article was reported.

DATA AVAILABILITY STATEMENT

The authors of this manuscript are willing to provide any additional information regarding the case report.

ETHICS STATEMENT

Not applicable.

CONSENT

Written informed consent was taken from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review for the editor-in-chief of this journal.

ORCID

Yohannis Derbew Molla https://orcid.org/0000-0001-7881-0538

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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