A case of double appendix and cecum in an infant – case report

Murad Habib*, Sadia Asmat Burki and Muhammad Amjad Chaudhary

Department of Paediatric Surgery, The Children’s Hospital, Pakistan Institute of Medical Sciences, Islamabad, Pakistan
*Correspondence address: Department of Paediatric Surgery, The Children’s hospital, Pakistan Institute of Medical Sciences, Islamabad, Pakistan.
E-mail: muradhabib007@gmail.com

Abstract
The duplication of the cecum and appendix is a rare congenital anomaly found incidentally on exploration for another indication. We present here a case of a female child at 18 months of life, who was a diagnosed case of anorectal malformation with a persistent cloaca and at the time of the surgery, duplication of the appendix and cecum was found. Both the appendices were attached to the cecum with a separate base. Thus, appendectomies and a sigmoid divided colostomy were performed.

INTRODUCTION
The development of a double appendix is an extremely rare occurrence with an incidence of merely 0.004–0.009% [1]. It was in the year 1892 that the first case of the double appendix was ever clinically reported [2]. It is an anomaly that mostly presents in association with other congenital anomalies of the intestinal and genitourinary system [3]. The presence of the double appendix in a neonate has the potential to cause intestinal obstruction and it can even imitate the symptoms of intussusception and other abdominal pathologies [4]. With the congenital duplication of the appendix, there are reported cases of associated duplication of other structures like the colon [5] and other intestinal and genitourinary systems. The authors report a case of a newborn with persistent cloaca and double appendices at laparotomy, and discuss the pathogenesis of appendix duplication [6].

CASE REPORT
A full-term child of 18 months was brought to Children’s hospital with complaints of inability to pass stool through the anal opening, abdominal distension and non-bilious vomiting. There was no family history of malformations, consanguinity and no history of medication to the mother during pregnancy. Physical examination revealed evidence of persistent cloaca as an abdominal distention and a single perineal orifice with partially fused labia and absence of an anal orifice. Routine complete blood count and biochemical tests revealed no abnormalities except a slight increase in white blood cells. All other milestones achieved. Radiographic examination revealed prominent fecal loaded gut loops as well as obscurity of mid-abdomen due to bowel gas shadows.

A sigmoid divided colostomy was made through an oblique lower abdomen incision; a single opening in the vestibule, a free-floating cecum with two bases, a double appendix and a markedly distended distal pouch were found as shown in Fig. 1. The presence of a double appendix was the rare finding of this surgery that was meant to create temporary colostomies.

DISCUSSION
Congenital appendiceal anomalies are an infrequent occurrence. ‘Appendiceal Duplication’ and ‘Complete
Table 1. Cave-Wallbridge classification of duplication of appendix [8]

| Types                          | A                        | B1 (Bird-like)                  | B2 (Tenia coli variant)            | C                        | D (Horseshoe appendix) |
|--------------------------------|--------------------------|---------------------------------|------------------------------------|--------------------------|------------------------|
| No of cecum                    | Solitary                 | Solitary                         | Solitary                           | Duplicated              | Solitary               |
| Appendiceal duplication        | Incomplete               | Duplicated                       | Duplicated                         | Incomplete duplication  | Duplicated            |
| Development                    | Partial duplication of   | Due to failure of proper         | The duplicated appendix is present  | With a common duct,     |                        |
|                                | the appendix-like        | cloacal differentiation, two     | along some colon tenia line, both  | two appendices originate|                        |
|                                | structure with one       | separate appendices originate    | the appendices originate from a    | from two separate       |                        |
|                                | normal appendix is       | from the singular cecum close to | single cecum.                      | appendices originating   |                        |
|                                | present at the base      | ileocecal valve.                 |                                    | from two separate       |                        |
| Related congenital anomalies   | None                     | - Atresia of colon and anus      |                                    | cecum.                   |                        |
|                                |                          | - External genitalia abnormalities|                                    | - Duplication of various  |                        |
|                                |                          | - Ectopic bladder.               |                                    | parts of hindgut, e.g.   |                        |
|                                |                          | Small intestine and              |                                    | ileum, colon, anus,      |                        |
|                                |                          | bladder communication.           |                                    | external genitalia and   |                        |
|                                |                          |                                  |                                    | bladder, etc.            |                        |

Type A: partially duplicated appendix of different extents with only a single cecum.

Type B: two discreet appendices with one cecum; this further has two more types. B1 ('Bird-like' Type) – two individual appendices present on both sides of the ileocecal valve. B2 (Tenia Coli Type) – an anatomically typical appendix along with the one originating along the line of tenia.

Type C: a duplicated cecum with each cecum having its separate appendix [9].

Type B1 and C are the ones that come with an increased incidence of other intestinal and genitourinary malformations and duplications, so if these types are encountered during the surgery special attention to other anatomical structures should be paid [10]. The Cave-Wallbridge classification is also modified in Table 1.

During the sixth week of embryological development, the cecal bud is the structure that lays down the foundation of the development of appendix. This cecal bud itself originates from the inferior end of the primary intestinal pouch. This cecal bud descends from its point of origin to give rise to many structures and it is during this process that appendix develops from its distal end as a slender diverticulum. It is anatomically placed posterior to the cecum and colon as it develops during the descent of the colon [11].

A case of triplication of the appendix has also been reported in a male child of 1 year with other corresponding congenital anomalies of the abdomen [12].

Duplication of the appendix is undoubtedly an infrequent anomaly; however, surgeons should always keep it in mind particularly in the instances when an appendix has been removed and the patient presents with the signs of symptoms of appendicitis or when the apparent appendix seems normal during the surgery, as it can have lethal repercussions for the patient and medico legal problems for the surgeon [13].

CONCLUSION

Duplication of cecum and appendix is extremely rare but a case of anorectal malformation may present with it. Awareness of this condition and thorough intraoperative inspection is critical as not to miss underlying diagnosis and associated anomalies.
CONFLICT OF INTEREST STATEMENT
There are no conflicts of interest.

DECLARATION
This study was reviewed and approved by ethical review board committee of Pakistan Institute of Medical Sciences.

PATIENT’S CONSENT
A written and informed consent was acquired from the guardian (father) regarding names and evidence used in this publication. And he had no objections what so ever.

FINANCIAL SUPPORT
The author received no financial support for this article.

REFERENCES
1. Travis JR, Weppner JL, Paugh JC 2nd. Duplex vermiform appendix: case report of a ruptured second appendix. J Pediatr Surg 2008;43:1726–8.
2. Marshall AP, Issar NM, Blakely ML. Appendiceal duplication in children presenting as an appendiceal tumor and as recurrent intussusception. J Pediatr Surg 2013;48: e9–12.
3. Dubhashi SP, Dubhashi UP, Kumar H, Patil C. Double appendix. Indian J Surg 2015;77:1389–90.
4. Olarinoye-Alebejo MI, Steve K, Ologun GO. Incidental finding of double appendix during laparotomy for intussusception: a case report. Int J Surg Case Rep 2020;75:219–21.
5. Kabay S, Yucel M, Yaylak F, Hacioglu A, Algin MC, Olgun EG, et al. Combined duplication of the colon and vermiform appendix in an adult patient. World J Gastroenterol 2008;14:641–3.
6. Peddu P, Sidhu PS. Appearance of a type B duplex appendix on barium enema. Br J Radiol 2004;77:248–9.
7. Kothari AA, Yagnik KR, Hathila VP. Duplication of vermiform appendix. J Postgrad Med 2004;50:285.
8. Griffiths EA, Jagadeesan J, Fasih T, Mercer-Jones M. Bifid vermiform appendix: a case report. Curr Surg 2006;63:176–8.
9. Wallbridge PH. Double appendix. Br J Surg 1962;50:346–7.
10. Christodoulidis G, Symeonidis D, Spyridakis M, Koukoulis G, Manolakis A, Triantafylidis G, et al. Acute appendicitis in a duplicated appendix. Int J Surg Case Rep 2012;3:559–62.
11. Glover J. The human vermiform appendix—a general surgeon’s reflections. CEN Tech J 1988;3:31œ38.
12. Tinckler LF. Triple appendix vermiformis – a unique case. Br J Surg 1968;55:79–81.
13. Lim KH. Duplication of the vermiform appendix in an adult patient. Ann R Coll Surg Engl 2014;96:e16–7.