



Contents lists available at ScienceDirect

## International Journal of Surgery Case Reports

journal homepage: [www.casereports.com](http://www.casereports.com)

## Double intestinal duplication and incidental neuroendocrine tumor of appendix, a rare case of acute abdomen



G. Bellanova<sup>a,\*</sup>, P. Valduga<sup>a</sup>, A. Costa<sup>b</sup>, M. Barbareschi<sup>c</sup>, N. De Carli<sup>c</sup>, G. Giannelli<sup>d</sup>,  
A. Di Sipio<sup>a</sup>, C. Prezzi<sup>a</sup>, F.A. Ciarleglio<sup>a</sup>, P. Beltempo<sup>a</sup>, S. Marcucci<sup>a</sup>, D. Giacomini<sup>e</sup>,  
G. Depretis<sup>e</sup>, A. Brolese<sup>a,\*</sup>

<sup>a</sup> General Surgery and HBP unit Pathology Surgery, S. Chiara Hospital, Trento, Italy

<sup>b</sup> Emergency Department, S. Chiara Hospital, Trento, Italy

<sup>c</sup> Pathology Unit, S. Chiara Hospital, Trento, Italy

<sup>d</sup> Radiology Department, S. Chiara Hospital, Trento, Italy

<sup>e</sup> Gastroenterology Unit, S. Chiara Hospital, Trento, Italy

### ARTICLE INFO

#### Article history:

Received 30 March 2015

Received in revised form 13 June 2015

Accepted 19 June 2015

Available online 6 July 2015

#### Keywords:

Intestinal duplication

Multiple enteric duplication

Appendiceal neuroendocrine tumor

Acute abdomen

Double intestinal duplication

Laparoscopy

### ABSTRACT

**INTRODUCTION:** Intestinal duplication is rarely reported in adulthood and often remains undiagnosed until onset of complications. We describe the case of a 39 year old woman who came to our observation for acute abdomen due to a combination of double intestinal duplication (colon and ileum) and an incidental neuroendocrine tumor of the appendix.

**MATERIALS AND METHODS:** A 39 year old woman who was admitted at with upper abdominal pain.

Multisliced spiral CT scan showed a cystic lesion suggestive of an inflamed Meckel's diverticulum. The patient was underwent an urgent explorative laparoscopy.

The intraoperative findings revealed a cystic lesion of the anti-mesenteric side of transverse colon, apparently dissectable from the bowel and a second lesion with a strongly adherent and unresectable from the anti-mesenteric aspect of the small bowel. A combined appendectomy was also performed.

The histological diagnosis was consistent with a typical intestinal duplication for both intestinal lesion and an incidental 2 mm carcinoid tumor was also found in the appendix.

The postoperative course was uneventful and the patient was discharged on p.o. day 5. At the present-time she is well and following a regular oncologic follow-up.

**DISCUSSION:** The rarity of this case is due to the concomitant presence of an incidental, synchronous, appendiceal NET.

The elective treatment is surgical resection.

**CONCLUSION:** Intestinal duplication in the adulthood is extremely rare and may either have an acute presentation as acute abdomen or represents an incidental finding of mass.

We suggest that, once the diagnosis is suspected patient must undergo surgery.

© 2015 The Authors. Published by Elsevier Ltd. on behalf of Surgical Associates Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

### 1. Introduction

Intestinal duplication is rarely reported in adulthood and often remains undiagnosed until onset of complications. Multiple duplication is an extremely rare event and a double colonic duplication was firstly reported in 1895 [1].

In 1937 Ladd defined “alimentary tract duplications” as congenital malformations that involve the mesenteric side of alimentary tract and share a common blood supply with the native bowel [2].

Herein we describe the case of a 39 year old woman who came to our observation for acute abdomen due to a combination of double intestinal duplication (colon and ileum) and an incidental neuroendocrine tumor (NET) of the appendix.

### 2. Materials and methods

A 39 year old woman who had been having abdominal pain for the 3 previous weeks was admitted at our emergency department (ED) in September 2013. Her upper abdominal pain worsened in the last 72 h. No other comorbidities were reported.

Physical examination revealed achiness in the epigastrium with no sign of peritoneal defense.

Plain abdomen X-ray showed no significant findings whereas abdominal ultrasound (US) showed a 55 mm mesogastric cystic

\* Corresponding authors at: Department of Surgery “Chirurgia 2”, S. Chiara Hospital, Largo Medaglie d'Oro, 9, 38122 Trento, Italy. Fax +39461903735.

E-mail address: [giovanni.bellanova@apss.tn.it](mailto:giovanni.bellanova@apss.tn.it) (G. Bellanova).

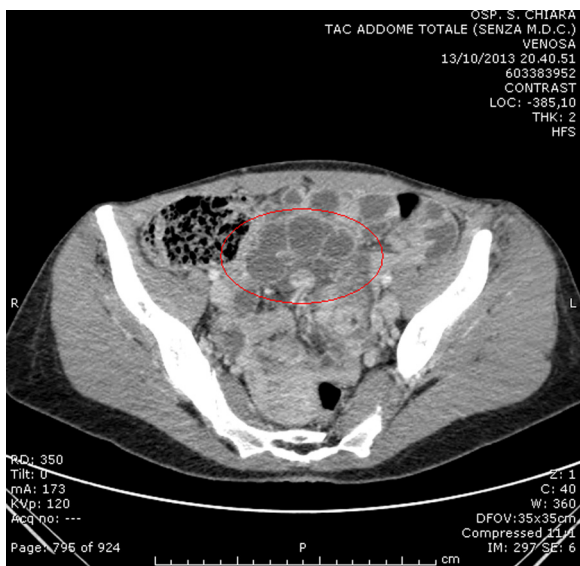


Fig. 1. Small bowel duplication—CT scan image.

lesion and a dilated small bowel loops in the lower abdominal quadrant.

Multisliced spiral CT scan showed a 55 × 45 mm, smoothly rounded, cystic lesion located between the duodenum and the gallbladder, close to the transverse colon, filled by mucinous-like fluid consistent with a mesenteric cyst, along with a narrowing of the terminal ileum which appeared dilated with thickening and convolution of the intestinal folds. Moreover, above the stenotic segment an overdistended, blind loop was evident. Its fluid-air content, wall thickening and communication with the true intestinal lumen, were suggestive of an inflamed Meckel's diverticulum (MD).

The patient was then admitted to our surgical unit and because of fever with an elevated WBC (11,000/dl) and worsening of her abdominal pain she underwent an urgent explorative laparoscopy.

The intraoperative findings revealed a cystic lesion of the anti-mesenteric side of transverse colon, apparently dissectable from the bowel and a second lesion with a strongly adherent and unresectable from the anti-mesenteric aspect of the small bowel. Then, upon conversion to a small midline laparotomy, we performed the complete isolation of the cystic lesion of the large bowel containing white “jelly-like” material and the resection of 10 cm of the distal ileum (40 cm from the ileocecal valve) with total excision of the mass (Fig. 1).

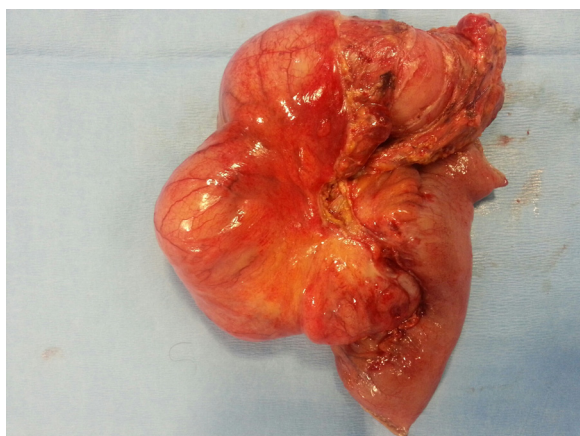


Fig. 2. Small bowel duplication—intraoperative.



Fig. 3. Small bowel duplication—gross pathology.

Given the peculiarities of the intraoperative findings and the particular anatomical condition of the appendix (subserosal and posterior sided), a combined appendectomy was also performed.

The gross appearance of the ileum showed a cystic lesion placed into the meso, without continuity with the lumen of the bowel and the perivisceral fat tissue also revealed necrotic areas (Figs 2 and 3), while a mucoid content was found inside the 7 cm cystic lesion arised in the meso of the transverse colon.

The histological diagnosis was consistent with a typical intestinal duplication for both intestinal lesions and an incidental 2 mm carcinoid tumor was also found in the appendix.

Microscopically the wall of the two specimens was formed by a layer of smooth muscle lined by a layer of submucosae and mucosa of colonic type, without epithelial dysplasia, even after an extensive sampling.

On other hand the histological examination of the appendix had detected an incidental well differentiated neuroendocrine tumor (NETG1 – appendiceal carcinoid) limited to the wall of the appendix (Fig. 4). This tumor was classified according to the criteria proposed by WHO (2010).

The postoperative course was uneventful and the patient was discharged on p.o. day 5. At the present time she is well and following a regular oncologic follow-up (30 months from surgery).

3. Discussion

There are several theories on the genesis of intestinal duplications, but little is known with certainty and its embriogenesis still remains controversial. Duplication of alimentary tract involves

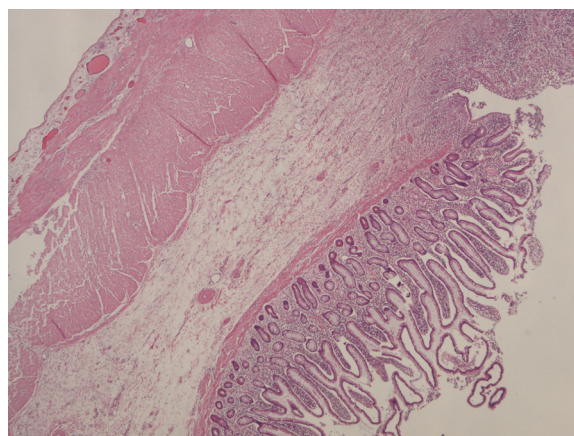


Fig. 4. Appendiceal net—Histology.

most frequently the distal tract of ileum up to the ileo-colic region [2]. Histologically they consist of at least one external muscle layer and an inner gastrointestinal mucosa which is often of gastric type [3]. Colo-rectal duplications occur in 6,8% of cases. However, malignant changes are uncommon and whenever they occur, they are most often found in the colon [4].

The rarity of this case is due to the concomitant presence of an incidental, synchronous, appendiceal NET (G1), which usually represent less than 10% of cancer of the appendix [5] but, according to the literature, is not associated with intestinal duplication.

In the absence of complications or neoplasm, the diagnosis of intestinal duplication is quite difficult because its radiological imaging do not differ from mesenteric and omental cystic disease with which it is often misdiagnosed because of their location along the mesenteric line [3].

The elective treatment is surgical resection of the enteric duplication lesion with the adjacent bowel segment if this is not-completely dissectable from vascular vessels [2] as in the case herein reported.

#### 4. Conclusion

Intestinal duplication in the adulthood is extremely rare and it is difficult to find an unambiguous description of its clinical and morphological characteristics in the literature due to the small number of reported cases.

In adults this disease may either have an acute presentation as acute abdomen or alternatively represent an incidental finding of tumor mass. The only sensible investigation is abdomen CT scan with contrast medium that nevertheless often fails to differentiate intestinal duplication from other mesenteric diseases.

We would like to suggest is that, once the diagnosis is suspected or established patient must undergo surgery. Complete excision is strongly recommended because it is the only definitive treatment that may prevent the occurrence of life-threatening complications as well as the high incidence of malignant transformation.

At our knowledge, no other reports or studies have previously described an association between enteric duplication and NET of appendix.

#### Conflicts of interest

All authors disclose any financial and personal relationships with people or organization about the case report.

#### Sources of funding

No sponsor was researched to pay this publication. Every authors use own money.

#### Consent

Consent acquired.

#### Author's contribution

G.Bellanova, P.Valduga, M.Barbareschi—study concept and design.

C.Prezzi, FA Ciarleglio, P.Beltempo, S.Marcucci, D.Giacomin, G.Depretis—data collection.

A. Costa, A.Di Sipio—data interpretation.

G.Bellanova, N. DeCarli, G.Giannelli, A.Brolese—writing the paper.

#### Guarantor

G.Bellanova.

A.Brolese (director).

#### References

- [1] S.J. Diaz-Cano, F. Rivera-Hueto, A. Mesa-Navarro, Duplication in a non rotational colon study of a case associated with mucinous adenoma, *Pathol. Res. Pract.* 191 (June 5) (1995) 415–419.
- [2] S.T. Ildstad, D.J. Tollerud, R.G. Weiss, D.P. Ryan, M.A. McGowan, L.W. Martin, Duplications of the alimentary tract. Clinical characteristic preferred treatment and associated malformations, *Ann. Surg.* 208 (2) (1998) 184–189.
- [3] S. Abhishek, S. Jimin Du Yutian, C. Dianbo, Dynamic change of intestinal duplication in an adult patient: a case report and literature review, *Case Rep. Med.* 2012 (2012) 297585.
- [4] N. Mourra, N. Chafai, B. Bessoud, V. Reveri, A. Werbrouck, E. Tiret, Colorectal duplication in adults: report of seven cases and review of literature, *J. Clin. Pathol.* 63 (December 12) (2010) 1080–1083.
- [5] M. Yilmaz, M. Kanlioz, R. Emre, N. Sahiu, Routine Histopathologic examination of appendectomy specimens: retrospective analysis of 1255 patients, *Int. Surg.* 98 (October–December 4) (2013) 354–362.

#### Open Access

This article is published Open Access at [sciedirect.com](http://sciedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.