Intestinal Duplication Management in Adult Patients: A Systematic Review

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Intestinal Duplication Management in Adult Patients: A Systematic Review

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Abstract

Introduction. Gastrointestinal duplication is a rare congenital abnormality found in adults. This intestinal duplication may be associated with complications i.e. perforation, bleeding, obstruction and malignancy. Degeneration to malignancy is a basic consideration to treat a radical surgery, which is preventive. This systematic review aims to provide evidences to evaluate the preventive or conservative surgery in the management.

Method. A systematic review conducted according to preferred reporting items for systematic review and meta–analysis protocols (PRISMA). Literature search proceeded on PubMed and ScienceDirect using keywords "malignancy arising from adult duplication gastrointestinal "AND" treatment ". All articles are selected based on inclusion and exclusion criteria. The data evaluated included postoperative complications, recurrence, follow-up and recovery.

Results. There were eleven articles – which were case reports – analyzed. These articles reported preventive surgery provide a better outcome than conservative one.

Conclusion. Case reports in the study showed that 63% of intestinal duplication were developed to malignancy, requiring preventive surgery (level of evidence IV). However, this should be judge with a careful clinical assessment.

Keywords: Malignancy developed from adult duplication gastrointestinal; gastrointestinal duplication in adult; treatment

Introduction

Gastrointestinal duplication is a congenital abnormality found in 65–85% two–years–old children.1 Most of these cases occur in ileocecal, with the most common location in the ileum (30%). Duplication can be in the form of a cyst (>85%) or tubular (10–15%) and is located on the side of the mesentery. Clinical manifestations of intestinal duplication in children can vary, in the form of symptoms of obstruction, nausea, vomiting, abdominal pain, abdominal mass, intussusception, which can be accompanied by congenital abnormalities.2

The occurrence of intestinal duplication as the cause of intussusception in adult is an unusual pathologic condition which is rarely found.2 In adults, the clinical manifestations of intussusception are not typical as in pediatric and may associated with complications such as intestinal perforation, GI bleeding, intestinal obstruction and degeneration to malignancy.

The diagnosis is seldom be established preoperatively, as the patient frequently admitted due to strangulated intestinal obstruction requiring immediate surgical intervention. The decision of appropriate surgical intervention should be made, particularly when such anomaly is found intraoperatively. There are two approaches in the management, namely preventive surgery or conservative one. Preventive means radical surgery as there a great possibility to develop malignancy, while as conservative means the management with a goal to overcome the intussusception and conserved the intestine physiologically. As there were lack of data, then a systematic review was conducted to find out the highest evidence. A case report is provided to draw the clinical problem.

Case report

Male of 24–years–old came to emergency department of Tangerang General Hospital with the symptoms of intestinal obstruction and colicky pain at the right lower abdomen quadrant and history of currant jelly stool for two days. No history of previous intestinal obstruction.

On physical examination, patient was found alert, tachycardia with normal breath per minutes and normothermia. No anemic nor icteric. Distended abdomen found with tenderness at the lower right quadrant. Signs of peritonitis were absent. Leukocytosis found on peripheral blood specimen, and abdominal x–rays indicating intestinal obstruction at upper GI level. On the laparotomy, dilated ileum was noted, and intussusception in the jejunum 300 cm orally to ligament of Treitz. Resection was decided with end to end ileo–ileal anastomosis. When intussusception was cut, a tubular mass measuring 14 cm long and 2 cm in diameter with hematoma, necrotic tissue and mucus filled the lumen was macroscopically shown. Microscopically, mucosa of the ileum and the area of duplication have a complete muscular layer, with bleeding and necrotic area involves all intestinal layers. There was no sign of malignancy. The patient discharged on the sixth postoperative day and came for follow up seven days after being discharged; no complain and he proceeded normal activities.

Method

A systematic review conducted to find out the management of intestinal duplication that have a great potency to malignant
degeneration, as found in adult. The study proceeded in accordance with preferred reporting items for systematic review and meta-analysis protocols (PRISMA). The literature search proceeded on PubMed and ScienceDirect using keywords "malignancy arising from adult duplication gastrointestinal" AND "treatment". All articles discussing the management of intestinal duplication in adult cases in the last ten years using English or Indonesian were included in this study. Those discussing degeneration to malignancy were included. While as those in pediatric, correspondence, or did not mention the outcomes of the treatment were excluded from the study. These articles were critically appraised using critical appraisal tool for systematic review. Relevant data to research question were analyzed in those articles of the highest level of evidence.

**Results**

There were total of 133 articles available, comprised of 88 articles in PubMed and 24 articles in ScienceDirect. On filtering, eleven articles were in lined to the criteria.

In a critical review of patient’s information, there were no information about demographic characteristics in all articles, but the symptoms of obstruction including medical history and history of illness related to the symptoms. In the diagnosis, the method used to diagnose were described, including the use of imaging (ultrasound, CT scan and MRI). Each of article describing the interventions that were carried out, changes in interventions due to different intraoperative findings or difficulties enfaced. Reoperation for recurrence completed with chemotherapy were described. These eleven articles also discussed the evaluation including clinical assessment before and after the intervention. The compliance of follow–up were the focus on each article. The outcome of overall treatments was addressed in these articles. In the discussion, no article explaining the strength and weakness of the intervention carried out, but comparisons to other studies and the rationale as well as the benefits of interventions were addressed. Not a single article describing the prognosis and patient’s perspectives. Five articles reported preventive surgeries, and four articles (80%) reporting recovery as the outcome, an article (20%) reporting recurrent and mortality. Other six articles proceeded conservative surgery and found four articles (50%) associated with mortality and two articles (33%) recovery, and three articles (67%) associated with recurrent (table 1 and 2).

In these articles, patients complained of abdominal mass (45.5%) and abdominal pain (45.5%). Preoperative diagnosis established using CT scan (91%) and abdominal ultrasound (9%). On histopathological findings, seven articles (63.6%) of duplication of intestine transformed into adenocarcinoma, and two articles were non–malignant (18.2%), and two other articles transformed into transitional cell carcinoma and GIST.

Out of five articles, the preventive surgery was proceeded in four articles with the outcome was recovered in 80% subjects and died preceded by recurrent in 20% subject. The conservative proceeded in six other articles; four articles reported mortality of 50%, recovery of 33%, and recurrent of 67%.

**Discussion**

The articles subjected to analysis in this study were case reports and those with level of evidence IV. Thus, to decide whether preventive or conservative method when dealing with intestinal duplication based on these studies requiring a very careful judgement. It is reasonable as intestinal duplication is an infrequent case. The malignant changes that developed from intestinal duplication may be found in vary, namely adenocarcinoma (the most common type), squamous cell carcinoma and transitional cell carcinoma.23

These changes were reported on seven articles, which is 63% developed to adenocarcinoma. Even though the incidence is infrequent, development of malignancy as much as 63% is quite high and should be of one consideration. There are two surgical interventions in the management of intestinal duplication currently applied, namely conservative– and preventive surgery. Conservative surgery defined as resection of the duplicated and normal intestine to a certain extent, in accordance with the site of duplicated part. While as preventive surgery referred to a surgical resection in accordance with oncologic principles, which is radical, including lymph nodes dissection. The outcome is found in vary, namely recovered with no complication and back to normal activity or postoperative mortality.

Both of treatment requiring critical consideration with individual approach. First, it should be realized that in most articles, the intestinal duplication was suspected prooperatively. In this case, any diagnostic tools to sharpen the diagnosis is justified. However, in emergency setting, the priority in the management is to solve the problem encountered. Intussusception, which is an emergency case of strangulated bowel need an emergency surgery that addressed to prevent intestinal necrosis, where sharpening the diagnosis with abdominal CT is not justified in this setting. In this setting, ultrasound may be of some preference as it is practical and may shows the features of intussusceptions, that surgeons can do.

When dealing with intussusceptions in adults, this unusual entity should be carefully addressed. Careful assessment of possible cause is important. However, it is premature to decide preventive is better than conservative based on these studies since there is many factors to be considered. Should the abdominal assessment represent the stigmata suggesting malignancy such as nodules, lymph node enlargement etc. in addition to history of malignancy symptoms, then surgical resection with oncological approach is justified. In contrast, conservative treatment should be adequately followed up for possible unexpected development of malignant degeneration.
Table 1. Data extraction preoperatively patients with intestinal duplication

<table>
<thead>
<tr>
<th>Article</th>
<th>Study design</th>
<th>Age (years) / gender</th>
<th>Initial symptom</th>
<th>Imaging</th>
<th>Surgical management</th>
<th>Type of duplication</th>
<th>Histology</th>
<th>Level of evidence</th>
</tr>
</thead>
<tbody>
<tr>
<td>Barussaud (2008)</td>
<td>Case report</td>
<td>67 years / M</td>
<td>Abdominal mass</td>
<td>Abdominal CT scan</td>
<td>Preventive</td>
<td>Cyst</td>
<td>Adenocarcinoma</td>
<td>4</td>
</tr>
<tr>
<td>Fletcher (2012)</td>
<td>Case report</td>
<td>28 years / M</td>
<td>Abdominal pain</td>
<td>USG, abdominal CT scan</td>
<td>Conservative</td>
<td>Cyst</td>
<td>Adenocarcinoma</td>
<td>4</td>
</tr>
<tr>
<td>Dalia (2017)</td>
<td>Case report</td>
<td>61 years / M</td>
<td>Abdominal mass</td>
<td>Abdominal CT scan</td>
<td>Conservative</td>
<td>Cyst</td>
<td>Transitional carcinoma</td>
<td>4</td>
</tr>
<tr>
<td>Cabrera (2016)</td>
<td>Case report</td>
<td>71 years / F</td>
<td>Abdominal mass, endoscopy</td>
<td>Abdominal CT scan</td>
<td>Conservative</td>
<td>Cyst</td>
<td>Adenocarcinoma</td>
<td>4</td>
</tr>
<tr>
<td>Zhu (2015)</td>
<td>Case report</td>
<td>62 years / M</td>
<td>Abdominal pain</td>
<td>Abdominal CT scan</td>
<td>Preventive</td>
<td>Cyst</td>
<td>Adenocarcinoma</td>
<td>4</td>
</tr>
<tr>
<td>Yamasaki (2016)</td>
<td>Case report</td>
<td>42 years / F</td>
<td>Abdominal mass</td>
<td>Abdominal CT scan, colonoscopy</td>
<td>Preventive</td>
<td>Cyst</td>
<td>Adenocarcinoma</td>
<td>4</td>
</tr>
<tr>
<td>Habib (2009)</td>
<td>Case report</td>
<td>38 years / M</td>
<td>Abdominal pain</td>
<td>Abdominal CT scan</td>
<td>Preventive</td>
<td>Cyst</td>
<td>Adenocarcinoma</td>
<td>4</td>
</tr>
<tr>
<td>Damiano (2011)</td>
<td>Case report</td>
<td>88 years / F</td>
<td>Hematochezia</td>
<td>Abdominal CT scan</td>
<td>Conservative</td>
<td>Cyst, tubular</td>
<td>Non-malignant</td>
<td>4</td>
</tr>
<tr>
<td>Hee (2012)</td>
<td>Case report</td>
<td>41 years / M</td>
<td>Abdominal mass</td>
<td>USG, abdominal CT scan</td>
<td>Conservative</td>
<td>Cyst</td>
<td>Adenocarcinoma</td>
<td>4</td>
</tr>
<tr>
<td>Kamal (2016)</td>
<td>Case report</td>
<td>20 years / M</td>
<td>Abdominal pain</td>
<td>USG, abdominal CT scan</td>
<td>Preventive</td>
<td>Cyst</td>
<td>Non-malignant</td>
<td>4</td>
</tr>
</tbody>
</table>

Table 2. Outcome of management of intestinal duplication

<table>
<thead>
<tr>
<th>Article</th>
<th>Study design</th>
<th>Age (years) / gender</th>
<th>Initial symptom</th>
<th>Follow up</th>
<th>Postoperative intervention</th>
<th>Result</th>
<th>Level of evidence</th>
</tr>
</thead>
<tbody>
<tr>
<td>Barussaud (2008)</td>
<td>Case report</td>
<td>67 years / M</td>
<td>Liver metastasis</td>
<td>Abdominal CT scan</td>
<td>Chemotherapy</td>
<td>Died</td>
<td>4</td>
</tr>
<tr>
<td>Fletcher (2012)</td>
<td>Case report</td>
<td>28 years / M</td>
<td>Recurrent obstruction</td>
<td>Abdominal CT scan</td>
<td>Relaparotomy mass excision</td>
<td>Died</td>
<td>4</td>
</tr>
<tr>
<td>Dalia (2017)</td>
<td>Case report</td>
<td>61 years / M</td>
<td>Perforation</td>
<td>Abdominal CT scan</td>
<td>Chemotherapy</td>
<td>Died</td>
<td>4</td>
</tr>
<tr>
<td>Cabrera (2016)</td>
<td>Case report</td>
<td>71 years / M</td>
<td>No</td>
<td>Abdominal CT scan, endoscopy</td>
<td>None</td>
<td>Recovered</td>
<td>4</td>
</tr>
<tr>
<td>Zhu (2015)</td>
<td>Case report</td>
<td>62 years / M</td>
<td>No</td>
<td>Abdominal CT scan</td>
<td>None</td>
<td>Recovered</td>
<td>4</td>
</tr>
<tr>
<td>Yamasaki (2016)</td>
<td>Case report</td>
<td>42 years / F</td>
<td>Ascites, metastasis</td>
<td>Abdominal CT scan</td>
<td>Chemotherapy</td>
<td>Died</td>
<td>4</td>
</tr>
<tr>
<td>Habib (2009)</td>
<td>Case report</td>
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</tr>
<tr>
<td>Damiano (2011)</td>
<td>Case report</td>
<td>88 years / F</td>
<td>Metastasis, obstruction</td>
<td>Abdominal CT scan</td>
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<td>Died</td>
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</tr>
<tr>
<td>Hee (2012)</td>
<td>Case report</td>
<td>41 years / M</td>
<td>No</td>
<td>Abdominal CT scan</td>
<td>None</td>
<td>Recovered</td>
<td>4</td>
</tr>
<tr>
<td>Kenoki (2015)</td>
<td>Case report</td>
<td>20 years / M</td>
<td>No</td>
<td>Abdominal CT scan</td>
<td>None</td>
<td>Recovered</td>
<td>4</td>
</tr>
<tr>
<td>Kamal (2016)</td>
<td>Case report</td>
<td>20 years / M</td>
<td>No</td>
<td>Abdominal CT scan</td>
<td>None</td>
<td>Recovered</td>
<td>4</td>
</tr>
</tbody>
</table>
Conclusion
Case reports in the study showed that 63% of intestinal duplication were developed to malignancy, requiring preventive surgery (level of evidence IV). However, this should be judge with a careful clinical assessment.

Conflict of interest
Author disclose there was no conflict of interest.

References