SURGICAL MANAGEMENT OF PATHOLOGICAL AEROPHAGIA – A CASE REPORT

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Abstract

Aerophagia, excessive air swallowing, is a rare clinical condition usually associated with neurologically abnormal children. A case of a 5-year-old Caucasian male, who presented with an acute surgical abdomen and worsening symptoms of abdominal pain and increasing abdominal distension, is discussed. Symptoms failed to resolve with conservative management therefore an exploratory laparotomy was performed but revealed no pathological cause. At follow-up the child was noted to be air swallowing using a plastic straw and he was subsequently diagnosed with pathological aerophagia. Five previous cases of aerophagia requiring surgical intervention have been published in the literature. Interestingly, the majority of these children were neurologically normal. Aerophagia is an important differential diagnosis to consider in children presenting with worsening abdominal distension and the key to diagnosis is observation in order to identify the air swallowing behaviour.

Keywords: aerophagia, air swallowing, abdominal distension

Background

Aerophagia is a rare functional gastrointestinal disorder that is characterised by excessive air swallowing. The causes and optimal treatment of this disorder are unknown. Ongoing aerophagia can lead to gross bowel distension, which in turn can result in significant complications such as intestinal volvulus, ischaemia and perforation. Many cases of pathological aerophagia in the paediatric population are initially misdiagnosed and the diagnosis is only made in retrospect [1].

Case report

A 5-year-old Caucasian male presented with a 3-week history of intermittent abdominal pain, distension and vomiting. During the history, his parents describe excessive flatulence but denied repetitive belching or any change in his bowel habit, such as constipation or diarrhoea. His past medical history was unremarkable and his neurological development was normal. Physical examination revealed a severely distended abdomen with visible bowel loops and generalised tenderness without peritonism. Hyper-active bowel sounds were heard on auscultation. His temperature was 36.5 °C, blood pressure 120 / 70 mmHg and pulse rate 96 beats per minute. Standard blood tests were unremarkable including coeliac screen. Stool microscopy and culture were negative. An abdominal radiograph demonstrated a large air-distended stomach and grossly dilated loops of small and large bowel (Fig. 1 and 2). He was treated conservatively with a nasogastric and rectal tube but these measures failed to decompress his abdomen. A gastroscopy and colonscopy were performed but showed no macro- or microscopic abnormalities. Over the following days his abdominal pain worsened despite conservative treatment and so exploratory surgery was undertaken. At laparotomy the entire bowel was grossly dilated although no transition point or fistulae was

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found. A gastrostomy and defunctioning ileostomy were fashioned to treat his distension. He made a successful post-operative recovery. Of note it was observed that his stoma produced around 8-10 litres of gas per day, with a further 2-3 litres of gas being aspirated from the gastric tube. Histology from the specimen sent at the time of surgery demonstrated normal bowel. At follow-up his parents reported that gas production was greatest at the end of the day and flatulence worse whilst he slept. On more detailed questioning, it came to light that he usually drank through a plastic straw and was likely to be air swallowing; therefore the diagnosis of aerophagia was suspected. His parents were advised to avoid drinking using straws and over the next 3 months his gas production improved. He was referred for psychotherapy but no emotional triggers were identified. A lower gastrointestinal contrast study, done prior to the reversal of his ileostomy, identified a colonic stricture. He subsequently had his ileostomy closed and during this procedure the colonic stricture was resected and an ileocolic anastomosis performed. Additionally, a gastroplexy was also performed in order to prevent a future gastric volvulus due to his very mobile stomach. Two years later he is healthy and has suffered no further complications.

Discussion
Aerophagia is a rare disorder in children, which involves excessive air swallowing. The Rome III criteria (RIIC) suggest a diagnosis can be made if 2 or more of the following features are present for 2 months:

1. Air swallowing
2. Abdominal distension because of intraluminal air
3. Repetitive belching or increased flatulence [2]. A prospective study has recently shown that these criteria can diagnose aerophagia in 15% of children who are referred to general paediatric clinics with recurrent abdominal pain [3]. A number of retrospective studies have also reported other associated symptoms such as abdominal pain, vomiting, diarrhoea or constipation [1, 4, 5]. The diagnosis appears particularly challenging in children with concurrent constipation [5]. In our patient clinical features of aerophagia developed within 3 weeks, and so, were incompatible with the RIIC. Nonetheless, it is highly probable that our patient suffered from aerophagia because once he avoided drinking straws (i.e. air swallowing) his clinical symptoms resolved. Moreover, it has been argued that the time-to-diagnosis is superfluous in such patients because early diagnosis and treatment can prevent unnecessary investigations [4, 5] and interventions. Aerophagia is a functional gastrointestinal disorder [2]. Current treatment involves educating the child and parents and avoiding precipitating factors such as drinking straws, carbonated beverages and chewing gum. In those children with associated psychological stress psychotherapy may prove helpful. In the acute setting abdominal decompression with nasogastric and rectal tube is often necessary. Surgical intervention is restricted to those cases in which life-threatening complications may develop, such as a bowel perforation. We conducted a literature search using
Ovid MEDLINE to identify all cases of childhood aerophagia that had required surgery. The search term used was “aerophagia” and a limit of “humans” was applied to identify 5 previous reported cases (summarised in Table 1). Aerophagia is more common in children with neurodevelopment delay and it is currently believed that these children are more likely to experience life-threatening complications secondary to aerophagia [6]. However, three of the five cases reported in the literature, described children that were neurologically normal. This case series also illustrates the challenges in diagnosing aerophagia, as it can often initially be confused for late-onset Hirschsprung’s disease: although an unlikely diagnosis in the absence of constipation [6-8]. In our patient, failed conservative management necessitated exploratory laparotomy but revealed no underlying pathological cause. Although the reversal of his defunctioning ileostomy was successful, a colonic stricture had developed and required colonic resection and ileocolic anastomosis. This complication presumably resulted from severe dilatation of the transverse colon (Fig. 1 and 2) with vascular compromise in the immediate post-operative period. Indeed, aerophagia was only fortuitously diagnosed following visual air swallowing witnessed in the outpatient clinic. Additionally in our patient, gastric distension secondary to air swallowing led to the development of a hyper-mobile stomach, which put him at high risk of gastric volvulus and therefore required a gastropexy for prevention.

In conclusion, aerophagia is an important differential diagnosis in children who present with a severely distended abdomen and symptoms of a functional bowel obstruction. In those patients in whom the diagnosis is being considered it is important to attempt to observe air swallowing as an inpatient, as this may prevent further unnecessary investigations or intervention.

<table>
<thead>
<tr>
<th>Author</th>
<th>Gender</th>
<th>Age (years)</th>
<th>Neurological status</th>
<th>Initial diagnosis</th>
<th>Operative findings</th>
<th>Surgical intervention</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gauderer et al [7]</td>
<td>Male</td>
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<td>Normal</td>
<td>Gastric outlet obstruction</td>
<td>Unknown</td>
<td>Gastrostomy and pyloroplasty</td>
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<tr>
<td>Gauderer et al [7]</td>
<td>Female</td>
<td>4</td>
<td>Normal</td>
<td>Hirschsprung’s disease</td>
<td>Unknown</td>
<td>Colostomy then gastrostomy</td>
</tr>
<tr>
<td>Trillis et al [8]</td>
<td>Female</td>
<td>7</td>
<td>Mild neurological impairment</td>
<td>Hirschsprung’s disease</td>
<td>Colonic volvulus</td>
<td>Colonic resection</td>
</tr>
<tr>
<td>Komuro et al [9]</td>
<td>Male</td>
<td>8</td>
<td>Normal</td>
<td>Aerophagia</td>
<td>Gastric volvulus</td>
<td>Laparoscopic gastropexy</td>
</tr>
</tbody>
</table>

Table I: Reported cases of pathological aerophagia in children requiring surgery

REFERENCES