

Bleeding Barrett's ulcer as a complication of GERD in physically and intellectually disabled children – report of two cases

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Abstract

Gastroesophageal reflux disease (GERD) is a problem frequently occurring among physically and intellectually disabled individuals. In this group of patients GERD is often overlooked, since the symptoms are usually non-specific. We present two cases of disabled children, who developed complications of GERD in the form of Barrett's esophagus, Barrett's ulceration and bleeding, the life-threatening events which were not preceded by typical GERD complaints.

Key words: Barrett's esophagus, disabled individuals.

Introduction

Gastroesophageal reflux disease (GERD) is a problem frequently occurring among physically and intellectually disabled individuals but is often overlooked, since the symptoms are usually non-specific [1]. The development of Barrett's esophagus (BE), a premalignant condition occurring with transformation of normal esophageal squamous epithelium into specialized columnar epithelium containing goblet cells, is a severe complication of GERD [2]. We present two cases of disabled patients, who developed complications of GERD in the form of Barrett's esophagus, Barrett's ulceration and bleeding, the life-threatening events which were not preceded by typical GERD complaints.

Case 1. The boy was well until the age of 4, when he had

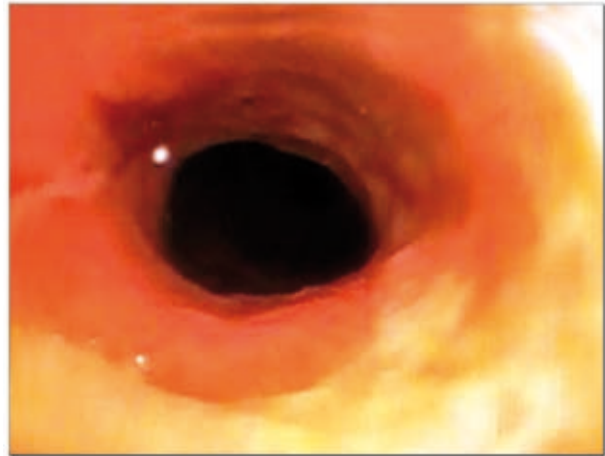
a car crash and developed spastic paralysis of the lower extremities, followed by severe scoliosis. When the boy was 14, he was treated for anemia with iron agents. At that time, gastroesophageal reflux was diagnosed by barium swallow study, but, as the patient was asymptomatic, he did not receive any treatment. At the age of 16, he was admitted to our department due to bleeding from the alimentary tract. On admission, his general state was fairly good, he weighed 35 kg. On the physical examination we found the boy in a wheelchair, with severe spasticity in the lower extremities, deformation of the thorax and skin pallor. His pulse was 90 beats/min and his blood pressure was 110/75 mm Hg. Laboratory evaluation disclosed a hemoglobin concentration of 10 g/dl, iron 36 µg/dl. Stool tests were positive for occult blood. On the third day of hospitalization, the boy had an episode of hematemesis. Endoscopy revealed fresh blood in the esophagus and stomach, deep and slightly bleeding ulceration of the distal esophagus, esophageal diverticulum filled with blood and esophageal hernia. CT scans showed the presence of an esophagomediastinal fistula in the distal esophagus, with thickening of periesophageal tissues. The patient required transfusion of two units of packed red blood cells. Under fasting, intravenous feeding, antibiotic therapy and PPI application, closure of the fistula was achieved. He was discharged home in good general condition. He did not continue treatment with medications.

The patient returned to hospital 6 months later due to sialosis, dysphagia and odynophagia. Results of laboratory tests were within normal range. Magnification endoscopy was done, revealing a long segment of salmon-pink mucosa extending almost to the upper esophageal sphincter (UES) and ulceration in the proximal esophagus. An irregular Z line was seen 3 cm below the UES. The fistula detected in the previous endoscopy was completely healed. The long-segment columnar epithelium was visualized by chromoendoscopy with methylene blue. Histopathological investigation of esophageal biopsy confirmed the BE. Therapy with PPI was instituted. Currently, the boy is chronically treated with PPI and is in good condition.

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Figure 1. Patient 1. Long-segment Barrett's esophagus**Figure 2. Patient 2. Bleeding ulceration of Barrett's esophagus**

Case 2. The second patient, a 17-year-old boy with cerebral palsy diagnosed in the first year of life, treated with anticonvulsant drugs, was admitted to our department due to upper GI tract bleeding. Two weeks prior to admission, feeding problems appeared. The boy refused to eat, was restless and anxious, and suddenly developed hematemesis. On admission, we found a spastic quadriplegic boy with severe scoliosis. He was malnourished, with body mass of 28 kg. Except for typical neurological findings, physical examination did not reveal any other abnormalities. There were no signs of dental erosions. Laboratory tests showed anemia (hemoglobin – 9.1 g/dl, iron – 45 µg/dl). Upper gastrointestinal endoscopy was done under general anesthesia, revealing salmon-pink velvety mucosa over half of the esophagus, indicating long-segment Barrett's esophagus. Bleeding ulceration, Schatzki ring and hiatal were detected in the esophagus. Biopsy showed goblet cell metaplasia confirming BE. Under short-term intravenous feeding and PPI administration, the patient improved. The treatment with PPI was continued, the boy was qualified to anti-reflux surgery.

Discussion

Gastroesophageal reflux disease (GERD) is a common complication in patients with severe motor and intellectual disabilities. Previous studies have estimated that up to 10-25% of institutionalized patients have symptoms of vomiting, regurgitation or rumination. Gastroesophageal reflux GER occurs in up to 70% of children with cerebral palsy [3,4].

In the general population, heartburn is the most common symptom of GERD. In intellectually disabled individuals (IDI) it is possible to define the risk of GERD based on non-specific symptoms. Children with GERD may present with feeding difficulties, failure to thrive, recurrent vomiting, choking attacks, anemia, wheezing, hematemesis, rumination, dental erosions, recurrent pneumonia, aggression, fear, episodes of screaming [5].

Several factors are considered to be responsible for the high prevalence of GER in IDI, including anticonvulsant drugs, cerebral palsy, constipation, scoliosis, non-ambulatory [1,6]. In physically and intellectually disabled the risk factors for GERD are already present in childhood. Tovar [7] demonstrated a decreased lower esophageal sphincter pressure and high percentage of non-propulsive waves in children with severe brain damage as compared to healthy subjects. Also delay of gastric emptying was shown in this group [8]. Scoliosis by displacing the stomach and stretching the lower esophageal sphincter can be responsible for malposition of the cardia and fundus, hernia, and may therefore promote GERD [9].

Esophageal complications of GERD include erosive esophagitis, esophageal stricture, Barrett's esophagus and adenocarcinoma. BE is a metaplastic condition in which columnar epithelium containing goblet cells replaces the normal squamous esophageal mucosa. This specialized intestinal metaplasia is noted in about 15% of patients suffering from GERD and is associated with more than 50-fold increase in the risk for the development of adenocarcinoma of the esophagus [10,11]. Since the prevalence of GERD in disabled individuals is very high, this group is particularly at risk for developing BE. The prevalence of BE in IDI is estimated as 12-26% [1,12]. For a long time, symptoms may be minimal or absent due to impaired sensitivity of the columnar lining to acid [13]. The diagnosis of GERD and BE is considered when serious complications, such as hematemesis, occur. Barrett's ulcer, which develops within BE, is frequently responsible for bleeding. Perforation is a rare but often fatal complication of Barrett's ulcer. The risk of esophageal carcinoma in IDI is about three times higher, as compared to that in the general population [14].

Reliable diagnosis of BE depends on the endoscopic recognition, followed by histologic sampling to screen for intestinal-type metaplasia. Chromoendoscopy is a new method that can enhance endoscopic diagnosis, including the detection of intestinal metaplasia through more accurate targeting of biopsy specimens. The diagnosis of BE is established when intestinal

metaplasia is found in biopsy specimens obtained from the esophageal mucosa.

PPIs are the most effective pharmacological agents in the management of patients with BE in respect to symptom control, ulcer healing and stricture prevention. The normalization of intraesophageal acid exposure can decrease cellular proliferation and possibly reduce the risk of cancer [15]. Another option is anti-reflux surgery and indications for such a treatment should be established in each case.

Conclusion

Considering the high prevalence of GERD and Barrett's esophagus in disabled children, early diagnosis and proper treatment are essential to prevent complications.

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