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CASE REPORT

Adult idiopathic hypertrophic pyloric stenosis



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Received 7 March 2012; received in revised form 8 May 2012; accepted 4 July 2012

KEYWORDS

adult;
circular muscle;
gastric outlet
obstruction;
idiopathic
hypertrophic
pyloric stenosis

Idiopathic hypertrophic pyloric stenosis (IHPS) is a predominantly infantile disease. The adult type of IHPS is extremely rare but it has been well recognized since the 19th century. We report a case of a 47-year-old male patient who presented with postprandial nausea and vomiting. He underwent upper gastrointestinal endoscopy, and gastric outlet obstruction was discovered. The upper gastrointestinal barium study showed a distended stomach with delayed gastric emptying due to pyloric stenosis. The abdominal computed tomography revealed thickening of the distal stomach. Since gastric malignancy could not be excluded, he underwent antrectomy with Billroth I anastomosis. The pathology revealed no malignancy but showed hypertrophy and hyperplasia of the inner circular muscle of the pylorus, which was compatible with IHPS. We reported the case to remind young physicians of this rare disease.

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Introduction

Idiopathic hypertrophic pyloric stenosis (IHPS) is a predominantly infantile disease, whose incidence is between 0.1% and 0.8%.¹ Infantile IHPS is always diagnosed and treated during the first 2 months of life. The adult type of IHPS is so rare that most physicians nowadays are not aware of it. In the French

atlas of pathologic anatomy *Anatomie pathologique du corps humain*, published in 1842, Cruveilhier first described the pathologic anatomy of adult IHPS.² Maier confirmed it as a clinical disease entity based on a series of pathologic findings at necropsy in 1885.³ However, there are less than 300 cases reported in the literature.^{4–6} We report a case of adult IHPS to remind young physicians of this rare disease.

Case report

The patient is a 47-year-old male Taiwanese. He complained of epigastric fullness and easy satiety for 1 month. The symptoms persisted and worsened, leading to

Conflicts of interest: The authors have no conflicts of interest relevant to this article.

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postprandial nausea and vomiting lately. He had no specific medical or surgical history. He had no history of peptic ulcer disease or diabetes mellitus. He visited our outpatient department and underwent upper gastrointestinal (UGI) endoscopy. The endoscopy showed a distended stomach with a constricted pylorus (Fig. 1). The scope passed the pylorus with slight resistance, and mild superficial erosions at the area of narrowing were noted. He was admitted under the diagnosis of gastric outlet obstruction. Physical examination revealed a soft and flat abdomen. No tenderness or palpable mass was detected. He had no leukocytosis or anemia. He underwent UGI barium study, which showed delayed emptying of the stomach and an elongated narrowing pyloric channel (Fig. 2). Abdominal computed tomography (CT) was performed. Thickening of the wall in distal stomach was noted (Fig. 3). Because gastric malignancy could not be excluded and the symptoms persisted, he underwent surgery. The pylorus was thickened and no adhesion was noted in laparotomy. He underwent antrectomy with Billroth I reconstruction. The surgery was uneventful. The pathology revealed no malignancy but showed thickening of the pyloric muscle (Fig. 4). Prominent hypertrophy and hyperplasia of the inner circular muscle layer of pylorus were noted. Hypertrophy of muscularis mucosae with muscle strands within the lamina propria was also noted. The pathologic picture was consistent with IHPS in adults. His symptoms were much improved after surgery.

Discussion

Hypertrophic pyloric stenosis (HPS) could be classified as primary and secondary. The secondary type are more common and may be induced by many causes, including healing of previous gastric or duodenal ulcers, hypertrophic gastritis, carcinomas, gastrointestinal stromal tumors, bezoars, vagal hyperactivity, and postoperative extrinsic



Figure 1 The stomach is filled with much undigested food residues. The pylorus is constricted and had no relaxation during examination.

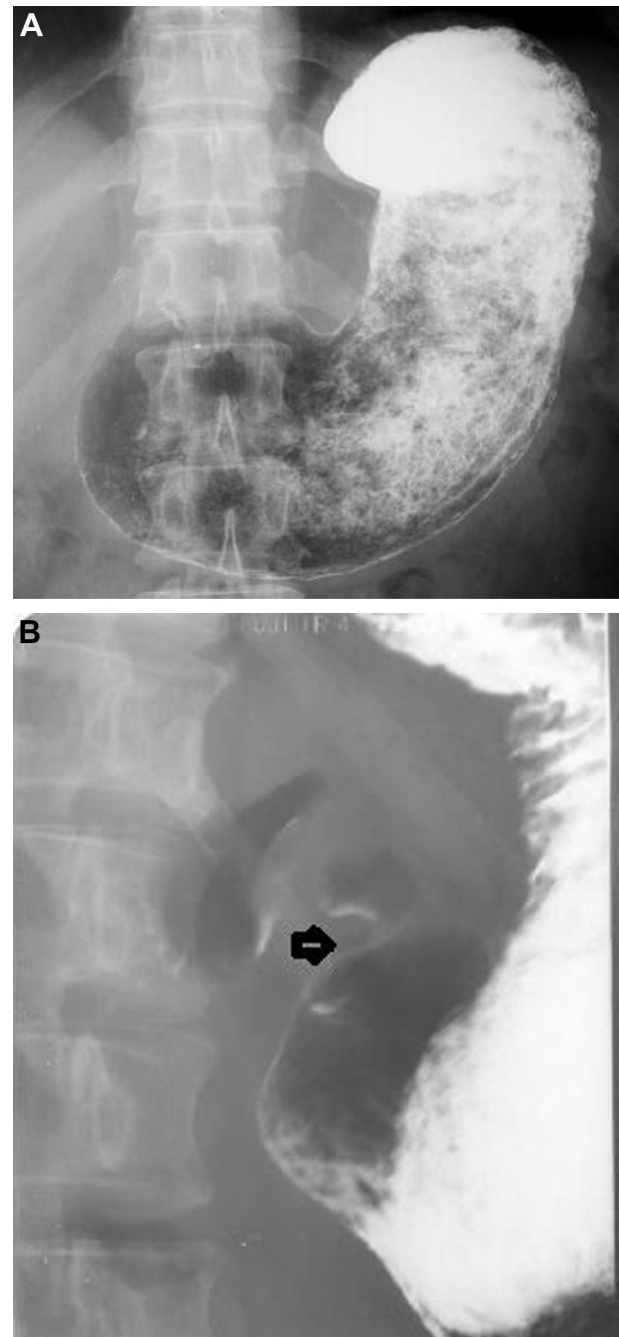


Figure 2 (A) The UGI series shows a marked distended stomach filled with much food debris. (B) The pylorus is elongated and narrowed (arrow).

adhesions.⁶⁻⁸ This type of HPS has no or only mild hypertrophy of the pyloric muscle. The muscle fibers are usually replaced by fibrous tissues. Development of HPS without predisposing factor is defined as primary or idiopathic type, and is characterized by hypertrophy and hyperplasia of pyloric muscle. IHPS is always diagnosed in infants, and the adult variant of IHPS is extremely rare. The exact occurrence of adult IHPS cannot be estimated accurately, since some of the cases may remain asymptomatic. With no known reason, adult IHPS occurs more commonly among middle-age males,^{9,10} as is the case with the patient in this report.

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