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A Case Report of Chronic Dysphagia, Intractable Hiccups, Vomiting, and Sever Gastroesophageal Reflux as Symptoms of a Posterior Cranial Fossa Hemangioblastoma

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Brain Tumor; Case report; Duodenal Switch; Duodenogastric Reflux; Reflux Esophagitis

1. Abstract

1.1. Background

For many years, bile reflux esophagitis has been reported and discussed in the literature. What makes our case unusual is the occurrence of postoperative sudden onset epigastric pain without obvious pathology and development of posterior cranial fossa hemangioblastoma 4 years after the surgery.

1.2. Case report

We report a 35 years old female patient who presented with heartburn, intractable hiccups, and food regurgitation in the last three years with no response to medications. Upper gastrointestinal (GI) endoscopy showed that there are erythema and multiple erosions throughout the esophagus and the gastric area with diagnosis of severe gastroesophagitis due to reflux of duodenal juice into the gastric and esophageal area. Surgical treatment with duodenal switch and partial fundoplication was effective for her symptoms. Postoperative GI endoscopy revealed marked improvement of the reflux, but the patient experienced a little postprandial sudden epigastric pain with no obvious pathology. After 4 years of follow-up, the patient developed brain tumor; she was operated and died after it.

1.3. Conclusion

Brain tumor may manifestants as intractable hiccups leading to severe gastroesophagitis. When encountering a patient presented with this symptom and no improvement after medical and surgical therapy, appropriate CNS imaging images should be included in the evaluation protocol.

2. Introduction

Gastroesophageal reflux disease (GERD) is prevalent among 20-50% of the population in Yemen and also in Western countries [1]. The rising incidence of this disease, as well as its consequences, imposes a substantial burden on the society in terms of hospital stays and related healthcare costs [2]. More notably, GERD-related cancers place an additional burden on patients and family members [2].

Brain tumors are one of the ten most common types of cancer globally [3]. All concerns about a possible link between GERD and esophageal cancer have been dispelled. However, significant concerns remain regarding the relationship between GERD and the presentation of a brain tumor [1,4]. Hence, we report a case of bile reflux esophagitis operated with duodenal switch and partial fundoplication with early presentation of postoperative sudden on-

set epigastric pain and late presentation of brain tumor.

3. Case presentation

Patient Information: The patient was a 35-year-old female who visited our outpatient surgery department due to a history of food regurgitation, intractable hiccups, heartburn, chest pain, and abdominal pain in the last three years. The symptoms were aggravated during pregnancy. She additionally experienced bilious vomiting and hiccups some times, but there was no history of melena or upper gastrointestinal (GI) bleeding. The protease inhibitors and proton pump inhibitors (PPI) were not effective for her symptoms. No history of cancer, smoking, or specific disease was mentioned. No family history of cancer was detected.

3.1. Clinical Findings

Physical examination including neurologic exam, was normal with no positive abdominal signs such as tenderness.

3.2. Diagnostic assessment

The laboratory data showed no abnormalities. Upper GI barium investigations showed that the esophageal folds were continuous, the wall was soft, and peristalsis and evacuation were normal. There was no evidence of stenosis or filling defects. The stomach had limited tension and was hook-shaped, with normal mucosal appearance. Abdominal computed tomography (CT) scan was normal without any pathology findings. We found erythema and multiple erosions in the entire esophagus during an upper GI endoscopy, and she was diagnosed with Grade C reflux esophagitis according to the Los Angeles classification (Figure 1).

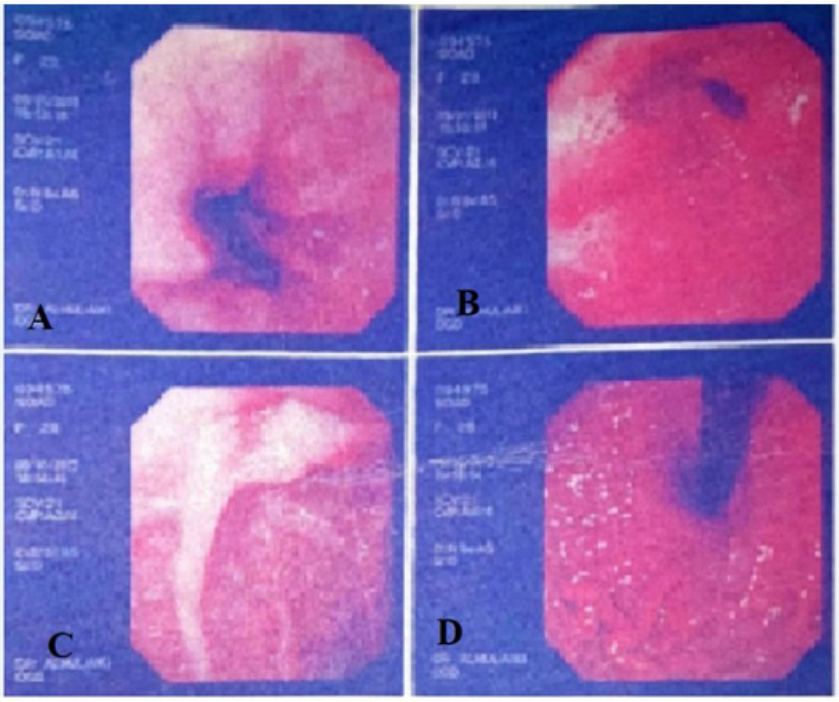

Figure 1: Upper gastrointestinal (GI) endoscopy showed
(A) Sever hyperemic mucosa associated with moderate to severe lower esophageal sphincter incompetence.
(B) Severe erythematous patch involving the most parts of the stomach
(C) Sever GERD.
(D) Normal pylorus part.

3.3. Therapeutic interventions

Because of intractable symptoms, the surgical intervention was discussed with the patient and her relatives. We decided to perform the duodenal switch technique with partial fundoplication to divert the duodenal juice from the stomach and esophageal area. The operative time was 120 minutes with minimal intraoperative blood loss (400cc).

3.4. Follow-up and outcome of interventions

The patient was discharged from the hospital with good condition. The heartburn and other symptoms disappeared, and upper GI endoscopic examination performed one month after the surgery revealed noticeable improvement of the GERD and just mild dyspeptic erythematous lesions which responded to PPI therapy (Figure 2). She was followed for at least four years postoperatively; the patient was free from symptoms including heartburn, and there was a little postprandial sudden epigastric pain with no obvious pathology. After 4 years of follow-up, the patient referred with sudden headache and brain tumor was diagnosed for her based-on brain CT scan and brain MRI radiologic investigations. Brain CT scan showed a well-defined semi-round hyperdense lesion with

homogenous enhancement measuring about (3.5 ×3× 3.5 cm) mass occupying the fourth ventricle and casing mild hydrocephalous and diffuse brain edema associated with periventricular ischemia change, which was highly suspected to be a posterior cranial fossa tumor. The MRI report was a well-defined posterior cranial mass lesion at the inferior cerebellar vermes and the adjacent part of the ventricle and cerebellar hemisphere measuring about (38 ×33 ×36 mm) with high signal at T1 and low in T2 (Figure 3). The patient was operated and unfortunately, she died after it. The histopathology report was hemangioblastoma grade one (WHO grade).

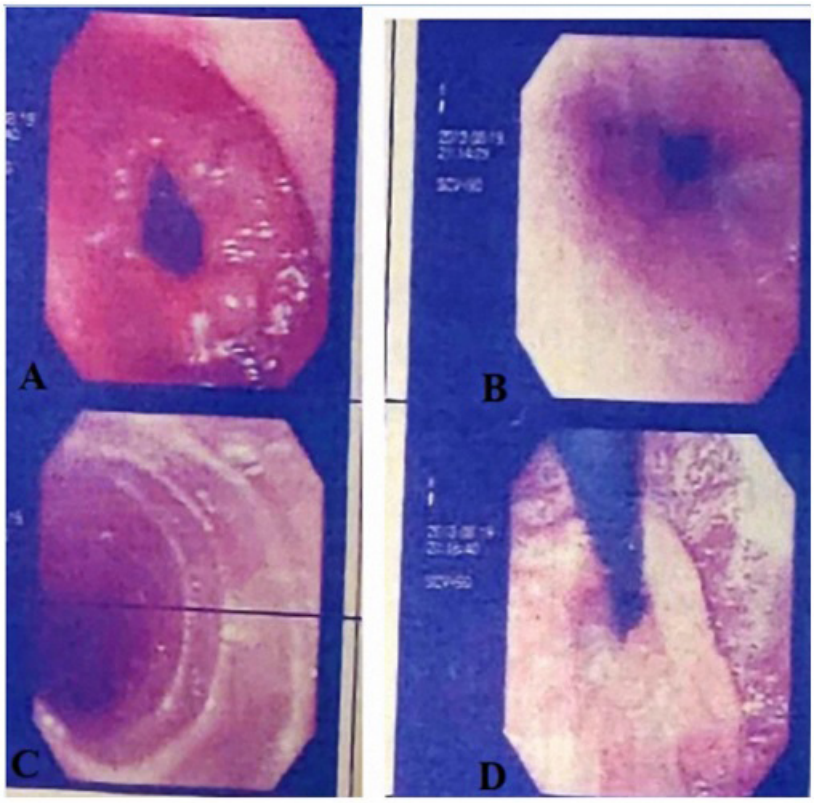

Figure 2: Upper gastrointestinal (GI) endoscopy showed improvement of GERD in all parts, just only mild gastritis.
(A) Normal esophageal mucosa appearance with normal esophageal sphincter competence.
(B) Mild hyperemic mucosa of the stomach.
(C) Normal esophageal mucosa.
(D) Normal pylorus part.

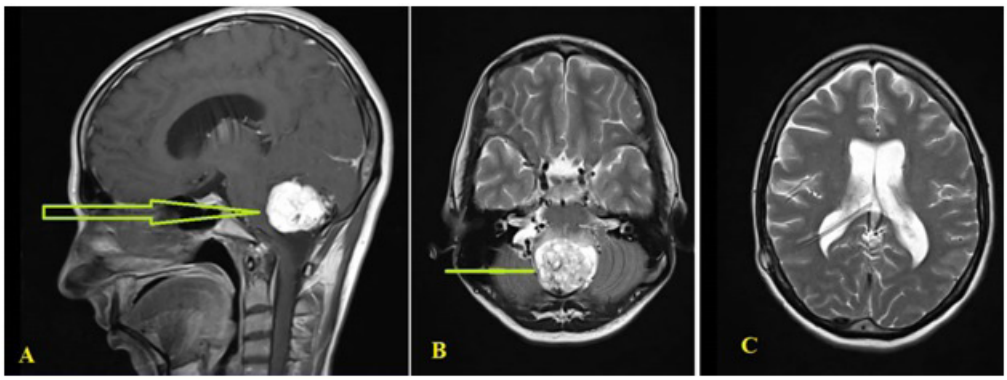

Figure 3: Brain MRI showed well defined semi-round hyperdense lesion with homogenous enhancement measuring about (3.5 ×3× 3.5 cm) mass occupying the fourth ventricle and casing mild hydrocephalous and diffuse brain edema associated with periventricular ischemia change, which was highly suspected to be a posterior cranial fossa tumor.

4. Discussion

Duodenogastric reflux refers to the excessive reflux of duodenal contents into the stomach. It is usually performed as a follow-up to prior gastric surgery in which the pylorus is removed or rendered dysfunctional, whereas duodenogastric reflux performed without prior gastric surgery is referred to as primary duodenogastric reflux. It is thought to be the cause of persistent symptoms after cholecystectomy, making it a significant cause of postoperative symptoms [5]. In our patient, the reflux was thought to be related to alkaline reflux, and not related to the reflux of gastric acid. Indeed, PPI and protease inhibitors were not effective [5].

Anti-reflux surgery attempts to create a barrier to GERD by packing the esophagus with stomach fundus (fundoplication). This is usually done in patients who have little comorbidity and still have reflux symptoms despite being on a PPI. However, the long-term results are disappointing, with a failure rate of 20% after five years [6].

In our patient, the duodenal switch was recommended. However; it was reported that in patients with preserved gastric acid, duodenal switch could not improve GERD, and that reflux esophagitis owing to gastric acid may persist even after this procedure. Furthermore, there is a risk of stomal ulcer after duodenal switch in patients with preserved gastric acid secretion. In patients with severe GERD, pylorus resection during duodenal switch may lead to gastric acid drainage into the distal part of the stomach, and doctors must recommend the use of PPI after this procedure. In our patient, the majority of her symptoms were alleviated. Similarly, previous articles have reported positive clinical outcomes following this procedure [5,7].

Our patient still had a sudden onset of postprandial epigastric pain without obvious pathology and after 4 years she developed brain tumor. We have two hypotheses for this effect; the first one is the sudden contraction of the gallbladder and subsequently bile flow against closed duodenal loop highlights the benefit of simultaneous cholecystectomy. The second hypothesis is that the GERD was the primary sign of undiagnosed brain tumor which was discovered after 4 years of GERD treatment. Wong et al. in a retrospective study on children with neurological impairment who underwent laparoscopic fundoplication and within follow-up period (range from 3 months to 9 years) reported one case who died due to brain tumor [8]. To illustrate this dilemma; we recommend more investigations in this area.

Chronic reflux of gastric contents into the esophagus is the most common cause of GERD. This can result in esophageal mucosa inflammation, which can be symptomatic or asymptomatic, causing heartburn, regurgitation, or epigastric pain. Chronic GERD, especially if left untreated, can result in metaplasia of the stratified squamous epithelium into the gastric mucosa [9].

Our patient had no neurological clinical manifestations; thus, she undertook various GERD investigations. The posterior fossa is a tiny area of the brain that contains the brain stem, cerebellum, and 4th ventricle and is responsible for many essential motor skills functions like speaking, listening, moving, feeling, and swallowing. Growth of a mass in the posterior fossa happens at the expense of the region's normal structures, which may result in brain stem or cerebellar disturbance. The posterior fossa is the site of origin for less than 5% of all adult tumors, but it is the site of origin for about 50% of primary brain and spinal cord tumors in the patients under the age of 15 [10].

The clinical manifestations of posterior fossa tumors are typically characterized by intracranial hypertension and focal neurologic deficits caused by the brain stem and/or cerebellar material compromise, whereas nonspecific complaints of vague, intermittent headache, fatigue, intractable hiccups, esophageal reflux, chronic dysphagia, and personality changes may predominate early in the course of illness [10-12]. This report highlighted brain tumor as the etiology of bile reflux associated with severe GERD. Chongsrisawat et al. and Frank et al. reported similar findings in children [12,13].

5. Conclusion

This study highlighted the possibility of a brain tumor as the cause of intractable hiccups leading to chronic and unresponsive bile reflux. When encountering a patient with symptoms of chronic and not responsive bile reflux, appropriate neuroimaging studies should have been included in therapeutic strategies.

6. Acknowledgements

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7. Patient Perspective

The patient's family mentioned that they would like to thank everyone from the nurses to the physicians that helped us take care of my sister, from the day she went to the hospital to the day that she left us.

8. Informed Consent

A written informed consent was obtained from the patient's family for participation in our study.

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