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Case Report

Madelung's Disease as the First Presentation of an Occult Gastric Adenocarcinoma

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Madelung's disease is a rare disorder defined as cervical benign symmetric lipomatosis. Its association to alcoholism, pulmonary pathology, and neck cancer has been reported. We first describe the occurrence of this syndrome during the course of a gastric cancer. A 65-year-old white Latino male patient was referred to our service for investigation of dyspeptic symptoms, weight loss, and small bilateral symmetric and painless cervical masses. Upper endoscopy revealed a pyloric ulcer, treated with a 3-month regimen of antibiotics and omeprazole. A new endoscopy revealed persistence of the gastric ulcer, and biopsy pathology report showed chronic inflammatory changes but no malignant cells. Two months after the biopsy, the patient was admitted to the Emergency unit for gastric obstruction. Gastric wall enlargement and distention were found at the operation. Intraoperative frozen section gastric biopsy was inconclusive. Resection was not performed and a gastroenteric anastomosis was constructed. Definitive pathology report revealed gastric adenocarcinoma with signet-ring cells. Thus, the patient underwent subtotal gastrectomy. Definitive pathology report confirmed diffuse signet-ring cell gastric cancer with duodenal invasion; 1 lymph node was positive for cancer. Postoperative course was uneventful, and adjuvant chemotherapy was performed. Gastric cancer recurred and the patient died 2 years after the operation. Madelung's disease did not show any progress during the disease-free period. Madelung's disease has been associated with head and neck cancers and now with gastric adenocarcinoma. We suggest that patients presenting with cervical lipomatosis also should be screened for occult gastric cancer and abdominal malignancy.

Key words: Madelung's disease – Gastric cancer – Cervical lipomatosis – Paraneoplastic syndrome – Cervical tumor

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Madelung's disease, also known as cervical benign symmetric lipomatosis, is a rare syndrome defined as multiple symmetric lipomatous nodules. The pathogenesis of this syndrome is poorly understood but may involve abnormal lipogenesis induced by catecholamines. Madelung's disease has been rarely associated with the presence of an occult malignancy, mainly head and neck cancers.[1] We report the occurrence of cervical lipomatosis as the index presentation of an occult gastric cancer.

Case Report

A 65-year-old white Latino male was referred to our service for investigation of dyspeptic symptoms, weight loss, and small bilateral symmetric and painless cervical masses (Fig. 1). He had quit alcohol and cigarette consumption 6 years ago. Upper endoscopy revealed a pyloric ulcer that was treated with a 3-month regimen of omeprazole, amoxicillin, and clarithromycin without improvement of dyspeptic symptoms. A new upper endoscopy was performed and revealed the persistence of the gastric ulcer, and biopsy pathology report showed chronic inflammatory changes but no malignant cells. One month later, he noticed enlargement of the cervical masses (Fig. 2). Biopsy of the cervical lesions showed lipoid mass, revealing the diagnosis of Madelung's disease.

Two months after the biopsy, the patient was admitted to the hospital Emergency Unit with the complaints of nausea and vomiting. Clinical examination revealed abdominal distention. Laparotomy was indicated with the presumptive diagnosis of gastric obstruction. Gastric wall enlargement and distention were found at the operation. Frozen section of the gastric biopsy was not conclusive. Regarding the benign macroscopic aspect the ulcer, resection was not carried and a gastroenteric anastomosis was performed. Definitive pathology report revealed a diffuse gastric adenocarcinoma with signet-ring cells. Thus, the patient underwent reoperation, undergoing subtotal gastrectomy with Roux-en-Y reconstruction. Definitive pathology report was confirmatory for diffuse gastric cancer with duodenal invasion; 1 lymph node was positive for cancer. Postoperative course was uneventful and the patient was discharged home 20 days after the first operation. Adjuvant chemotherapy was performed. The patient died of gastric cancer recurrence 2 years after the first operation. Madelung's disease did not show any progress during the disease-free period.

Fig. 1 Anterior view of the patient with Madelung's disease, showing large symmetric subcutaneous masses in the anterior and lateral cervical regions.

Fig. 2 Lateral view of the patient with Madelung's disease, showing the areas of fat deposition.

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Discussion

Benign cervical lipomatosis was first reported by Brodie in 1846.[2] It is a syndrome of unknown etiology that mostly affects middle-aged males and is strongly associated to chronic alcohol consumption.

Chan *et al* reported 8 cases of Madelung's disease associated with head and neck cancers.[1] They postulated that alcohol and tobacco use may have a primary role in the development of benign cervical lipomatosis.

Although cervical lipomatosis has been reported to occur in association with esophageal cancer and metastatic intra-abdominal malignancies, this is the first report of the association of Madelung's disease and gastric cancer.[1,3,4] Cervical lipomatosis here was associated with dyspeptic symptoms, prompting clinical investigation that revealed an occult gastric cancer.

Considering that Madelung's disease has been associated with esophageal adenocarcinoma, metastatic abdominal cancer, and now with gastric adenocarcinoma, we suggest that every patient presenting with cervical lipomatosis should be promptly investigated with upper esophagogastroduodenoscopy and probably with abdominal imaging (computed tomography or ultrasound) to rule out an occult gastrointestinal malignancy.

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