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Case Report

Recurrent Gastric Cancer Initially Presenting with Dysphagia induced by Idiopathic Inflammatory Myositis: A Case Report

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Abstract

We present the case of a 51-year-old female patient who was diagnosed with gastric cancer and subsequently underwent surgical resection, followed by adjuvant chemotherapy. Despite these interventions, the disease recurred, manifesting as laryngeal weakness and clinical features indicative of an autoimmune disorder. Further diagnostic evaluation confirmed the presence of a paraneoplastic autoimmune syndrome, specifically dermatomyositis with concomitant features of Sjögren's syndrome. Notably, the patient also had dysphagia, a relatively rare manifestation in this clinical context. The optimal therapeutic approach for managing this paraneoplastic autoimmune condition remains to be investigated.

Keywords: Dermatomyositis, dysphagia, gastric cancer, paraneoplastic syndrome

Introduction

Idiopathic inflammatory myositis, which includes dermatomyositis and polymyositis, is primarily characterized by muscle weakness, elevated creatine kinase levels, and distinctive cutaneous manifestations. An epidemiological cohort study in Taiwan found that patients with dermatomyositis and polymyositis had a significantly higher risk of developing cancer compared to the general population.[1] In addition, a comprehensive meta-analysis of 52 observational studies reported that the relative risk of comorbid gastric cancer in patients with idiopathic inflammatory myositis was 3.69.[2] Notably, approximately two-thirds of these patients were diagnosed with myositis before the detection of malignancy. Furthermore, comorbid patients have been reported to have a poorer prognosis compared to those with autoimmune disease alone.[3]

In this report, we present a case of gastric cancer complicated by a paraneoplastic syndrome manifesting as an autoimmune disease with laryngeal weakness. While dysphagia in patients with recurrent gastric cancer is often attributed to mechanical obstruction, this case underscores the importance of considering a rare differential diagnosis:

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laryngeal weakness secondary to idiopathic inflammatory myositis. This unusual presentation highlights the need for clinicians to maintain a broad differential when evaluating patients with a history of gastric cancer presenting with atypical symptoms.

Case Report

A 51-year-old woman initially presented with a 1-month history of watery diarrhea, without accompanying symptoms such as nausea, vomiting, weight loss, or weakness. Colonoscopy and esophagogastroduodenoscopy revealed a polypoid tumor at the esophagogastric junction, and an endoscopic biopsy confirmed tubulovillous adenoma with focal invasive adenocarcinoma. A computed tomography (CT) scan showed abnormal eccentric wall thickening of the upper gastric body, leading to a diagnosis of gastric cancer. The patient underwent laparoscopic proximal gastrectomy with D1 lymph node dissection. Pathological staging classified the cancer as pT1bN2M0, Stage IIA. Immunohistochemistry demonstrated preserved mismatch repair and HER-2/neu expression at 3+/3+. She received adjuvant chemotherapy with oxaliplatin and TS-1 for two cycles, followed by TS-1 alone for 1 year. During treatment, she experienced adverse effects including Grade II diarrhea, Grade I hand–foot rash, and left lower quadrant mesenteric panniculitis.

Two months after completing adjuvant chemotherapy, a CT scan revealed a recurrence of para-aortic lymphadenopathy. She was then treated with trastuzumab, cisplatin, and fluorouracil plus leucovorin. However, she developed progressive dysphagia that was severe enough to prevent her from tolerating solid or liquid food, along with general weakness, particularly of the proximal muscles. Confluent erythematous-to-brownish patches also developed on her neck, upper back, arms, lateral thighs, and forehead, with mild excoriation [Figure 1]. Panendoscopy ruled out mechanical obstruction, revealing a 3-cm Borrmann type I recurrent lesion near the anastomotic site without outlet obstruction. A biopsy confirmed recurrent adenocarcinoma, and video pharyngoesophagography showed a mildly delayed pharyngeal phase [Figure 2]. Schirmer's test was strongly positive bilaterally, and creatine kinase levels were elevated to 522 U/L, twice the upper normal limit. Electromyography indicated myopathic changes in the left biceps brachii. A muscle biopsy was not performed due to personal reasons. Autoimmune testing revealed a positive antinuclear antibody with NuMA-like (AC-26), positive anti-extractable nuclear antigen (ENA), positive anti-SS-A, negative anti-SS-B, and negative anti-Scl-70. A comprehensive ENA panel showed positive anti-TIF1-gamma, positive anti-Ro52, and negative anti-Ro60 [Table 1]. Tc-99m sialoscintigraphy indicated moderate dysfunction in bilateral submandibular glands.

Based on the findings of muscle weakness, skin rashes, elevated creatine kinase, specific autoimmune antibodies, and positive serology tests for Sjögren's disease, a diagnosis of paraneoplastic autoimmune syndrome, dermatomyositis with Sjögren-like features, was made. Treatment with hydroxychloroquine and methylprednisolone improved her dysphagia and proximal weakness. Her anticancer treatment was resumed with the previous regimen, including anti-HER2 therapy and chemotherapy. Unfortunately, she developed prolonged neutropenia under this treatment, and follow-up imaging showed disease progression after 3 months. After discussion, the patient and her family decided to decline further anticancer treatment. She was placed in hospice care and eventually died due to sepsis 40 days after her last chemotherapy dose.

Table 1: Results of serology studies

Results	Positive	Negative
Autoantibodies	Anti-NuMA-like (AC-26)	Anti-SS-B
	Anti-ENA	Anti-Scl-70
	Anti-SS-A	Anti-Ro60
	Anti-TIF-1-gamma	
	Anti-Ro52	

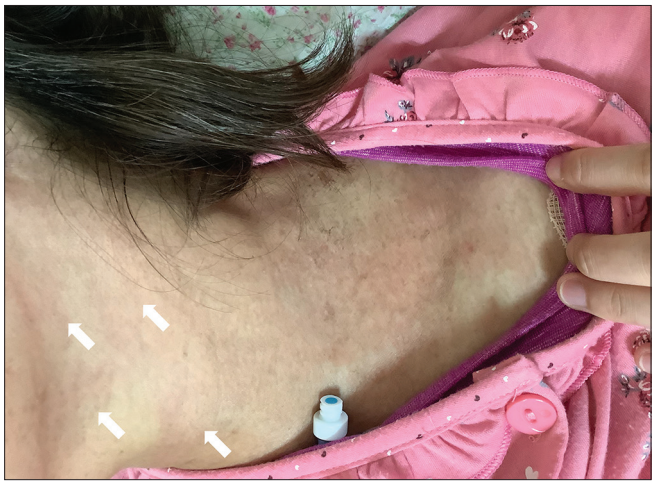

Figure 1: Confluent erythematous-to-brownish patches on her neck (arrows)

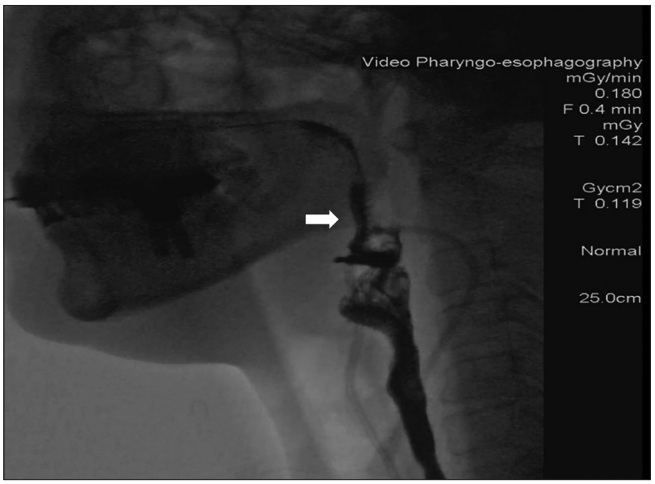

Figure 2: Video pharyngoesophagography showed barium retention at the larynx (arrow), indicating a delayed pharyngeal phase

Discussion

Recognizing the paraneoplastic autoimmune disorder in this patient was challenging due to the complexity of the disease course and its unusual presentation. The differential diagnosis of dysphagia in patients with recurrent gastric cancer includes mechanical obstruction, fatigue, decreased appetite, mood disorders, and other neurological conditions. In this case, the diagnosis was made based on the unique combination of symptoms and specific autoimmune serology tests and examinations. A previous case series of 238 patients with idiopathic inflammatory myositis in Taiwan found that those comorbid with cancer, including breast, nasopharyngeal, and rectal cancers, were more likely to present with anti-TIF1-gamma antibodies.[4]

Although our patient did not receive immunotherapy, immune checkpoint inhibitors (ICIs) are now a common treatment modality for various cancers, including gastric cancer. However, ICIs can trigger disease flares and immune-related adverse events (irAEs) in patients with preexisting autoimmune conditions.[5] Patients who develop irAEs after ICIs but do not have preexisting autoimmune diseases are usually negative for myositis-specific autoantibodies.[6] This could potentially be a way to differentiate between preexisting autoimmune disorders and irAEs. Management of Grade 3–4 irAEs typically requires intensive treatment, including high-dose corticosteroids, intravenous immunoglobulin, or plasma exchange. IL-6R inhibitors or tumor necrosis factor-alpha inhibitors may also be considered.[7,8]

There are limited data on the management of paraneoplastic autoimmune symptoms, with most patients treated with a combination of anticancer therapies and immunosuppressants. Patients with both autoimmune disease and cancer have been reported to have a higher overall mortality rate, although the risk of cancer-specific mortality has not been reported to be significantly higher across various cancer types.[9] Noncancer-related mortality is primarily due to autoimmune disease and related organ damage. Such patients are often undertreated due to poor baseline performance status, and autoimmunity may paradoxically act as a protective factor against cancer progression.[10]

In summary, we report a case of recurrent gastric adenocarcinoma initially presenting with dysphagia, which was later linked to dermatomyositis and autoimmune disorders. The presentation of paraneoplastic autoimmune symptoms can be heterogeneous, making early detection and diagnosis challenging. Further research is needed to establish optimal management strategies for patients with paraneoplastic autoimmune diseases.

Declaration of patient consent

This study was performed in accordance with and conforming to the Declaration of Helsinki. The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

Data availability statement

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

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Conflicts of interest

There are no conflicts of interest.

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