Esophageal Sarcomatoid Carcinoma Presenting as a Fever With Elevated Serum Leukocytes

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The study presented a case of esophageal cancer presenting as intermittent fever with markedly elevated serum leukocyte and C-reactive protein. The patient’s symptoms had not improved with antibiotic treatment. However, after thoracic esophagectomy, the fever faded and leukocyte serum levels rapidly normalized.

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Esophageal sarcomatoid carcinoma is a rare malignant neoplasm. Typical clinical manifestations include progressive dysphagia. A few cases presented with cancerous fever [1, 2]. These cases were mentioned in case reports. However, the serum leukocyte levels are still normal. Cases presenting as intermittent fever with markedly elevated serum leukocyte levels are rarely seen, and none has ever been reported in detail. Herein we present a case of esophageal sarcomatoid carcinoma presenting as intermittent fever with markedly elevated serum leukocyte and C-reactive protein (CRP).

A 56-year-old man presented with a 1-month history of progressive dysphagia, anorexia, and a 3-kg weight loss. He also reported a sudden onset of intermittent fever during the previous 6 days with the highest temperature of 38.6°C. He intermittently took a few tablets of nonsteroidal antiinflammatory drugs to control the fever. He said that he had experienced no hoarseness, choking, or productive cough. He had no significant medical or family history of esophageal cancer. Physical examination of the chest and neck revealed no mass, lymphadenopathy, or tenderness on palpation and no lung rales on auscultation. Analysis of serum leukocyte levels was elevated at 19.44 × 10^9/L, and the neutrophils were elevated at 85%. C-reactive protein was also high at 9.75 mg/dL. Anteroposterior plain radiographs did not reveal any abnormalities. Computed tomography of the chest showed a circumferential mass in the middle thoracic esophagus spanning 10 cm without any abscesses around the mass (Fig 1A). Upper endoscopy identified a large mass located 29 cm from the upper incisors that was almost completely obstructive. The endoscopy or nutrient canal was unable to traverse the esophageal mass (Fig 1B). A biopsy specimen taken from the tumor was found to be poorly differentiated squamous cell carcinoma (Fig 1C). After admission, nutritional support and empirical antibiotics using third-generation cephalosporin were administered for 3 weeks. However, the patient continued to experience intermittent fever without any improvement. Several blood analyses showed peripheral blood leukocyte levels to fluctuate from 16.24 × 10^9/L to 24.14 × 10^9/L. The relative number of neutrophils varied from 13.25 × 10^9/L to 22.76 × 10^9/L. Sputum, blood, and urine bacterial cultures showed no pathogenic bacteria, and the serum immune index was normal. Bone marrow aspiration showed reduced erythroid hyperplasia, although the percentage of neutrophilic myelocytes increased significantly, and the diseases of the circulatory system were excluded. Radical treatment for esophageal carcinoma was performed with a right thoracotomy. The neoesophagus was reconstructed with a gastric conduit, and intraoperative examination showed that the tumor did not invade adjacent organs (Fig 2A). Mediastinal and epigastric nodes were not pathologically involved. The tumor was bisected, revealing a mass about 9.0 × 2.5 × 2.5 cm in the esophageal lumen with only a thin pedicel connecting it to the mucous membrane of...
the esophagus (Fig 2B). Final pathologic examination confirmed the tumor as an esophageal sarcomatoid carcinoma, which had invaded the lamina muscularis mucosae without metastasis to the resected lymph nodes. Histopathologic examination showed two tumor components: squamous cell carcinoma and malignant spindle-shaped cells (Fig 2C). Immunohistochemical analysis showed that the cells are vimentin, pan-cytokeratin (CK-P), low molecular weight cytokeratin (CK-L), high molecular weight cytokeratin (CK-H) positive. No bacterial flora or abscesses were found around the tumor. The patient recovered without incident after the operation. Temperature spikes began to subside from the third day after surgery. Serum leukocyte levels also dropped to 8.58 × 10^9/L, and CRP levels dropped to 3.82 mg/dL (Fig 3). The patient began a liquid diet on postoperative day 7 and a semiliquid diet on postoperative day 9, and was discharged from the hospital on postoperative day 11. One month after the operation, the patient’s serum leukocyte and CRP had returned to normal.

Comment
Esophageal sarcomatoid carcinoma is a rare malignant neoplasm with both carcinomatous and sarcomatous components. Its incidence has been reported to range from 0.26% to 1.46% of all esophageal neoplasms [3]. Tumors are mainly located in the middle or lower esophagus with polypoid and mushroomlike projections to the esophageal cavity. Esophagectomy, endoscopic resection, and chemoradiotherapy can improve prognosis [4–6]. The typical clinical manifestation is progressive dysphagia. A few cases present as fever, but serum leukocyte levels usually are still normal [1, 2]. Esophageal cancer presenting as intermittent fever with markedly elevated serum leukocyte is seldom seen. To our knowledge, this has never been reported. Esophageal cancer can lead to respiratory tract aspiration as a result of malignant obstruction of the esophagus. This can lead to aspiration pneumonia. The cause of fever and markedly elevated serum leukocyte levels observed in this case may have been mistakenly attributed to bacterial infection. However, despite the use of antibiotics for 3 weeks, the fevers persisted. The patient became frailer because of the significant dysphagia and the intermittent fever. After radical resection of the esophageal tumor, the fluctuating fevers subsided and leukocyte and CRP serum levels normalized. For esophageal cancer patients whose fever is accompanied by markedly elevated serum leukocyte levels, it is necessary to determine whether the elevation is caused by a paraneoplastic phenomena, which this case seems to indicate, or a bacterial infection because the treatments for these two conditions are quite different. If a bacterial infection is suspected, effective preoperative antimicrobial measures should be implemented to prevent the spread of infection. For paraneoplastic phenomenon, timely operation is an effective method of treatment. Delay aggravates the disease and can even cause the patient to miss the window for a successful operation.

Although acute bacterial infections are often accompanied by a notable elevation of serum CRP (>10 mg/dL) within 8 to 12 hours, long-term, moderate elevation of serum CRP (<10 mg/dL) is observed in cancers [7]. In this case, the patient’s CRP serum levels fluctuated from 5.8 to 9.75 mg/dL.
In conclusion, we report a rare case of a patient with esophageal carcinosarcoma who presented with fever and markedly elevated serum leukocyte levels. When patients show no signs of local infection and no response to antimicrobial treatment, cancerous fever should be taken into consideration. Persistent but moderate serum CRP elevation (<10 mg/dL) further supports a diagnosis of cancerous fever, and radical treatment for esophageal carcinoma should be performed in a timely manner.

References