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Kaposi Sarcoma With Bilateral Chylothorax Responsive to Octreotide

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Abstract

Kaposi sarcoma (KS) is a soft tissue malignancy of the endothelial cells that can rarely invade the thoracic duct and cause bilateral chylothorax. Treatment for chylothorax includes drainage and dietary modification. However, octreotide has been reported to improve chylothorax in some pediatric and post-operative cases. We present a case in which a 9-day course of octreotide led to an improvement of non-traumatic malignant chylothorax.

Abbreviation list

AIDS: Acquired immunodeficiency syndrome CT: Computed tomography HIV: Human immunodeficiency virus KS: Kaposi sarcoma

Introduction

Kaposi sarcoma (KS) is a malignant, multifocal, highly vascularized tumor of the endothelial cells that most commonly affects the skin but may also include the lymph nodes, mucosa, and viscera (1). KS is commonly associated with human immunodeficiency virus (HIV) and can occur at any CD4 count (2). In very rare cases, Kaposi sarcoma can invade the thoracic duct and cause chylothorax (3). Chylothorax occurs when lymphatic fluid accumulates in the pleural cavity and is usually seen after damage to the thoracic duct following trauma or cardiothoracic surgery. It can also be caused by malignancy, however, bilateral chylothorax secondary to **KS** is rare. Treatment of chylothorax usually involves drainage of the effusion and initiation of a low-fat diet. Octreotide has been reported to improve traumatic chylothorax, but has only been reported in non-traumatic etiologies in a handful of cases (4). Here, we present a case of bilateral chylothorax associated with **KS**, which was successfully treated with octreotide.

Case Presentation

A 40-year-old man with a previous diagnosis of acquired immunodeficiency syndrome (AIDS) and KS presented to the emergency department due to progressive tachypnea, dyspnea, bilateral lower extremity edema, and expansion of his KS lesions onto his legs and genital region. His vital signs were significant for a respiratory rate of 25 breaths per minute and pulse of 109 beats per minute. The patient denied recent infection, trauma, or procedures. Chest X-ray showed a large left pleural effusion with midline shift and a small right pleural effusion (Figure 1).



Figure 1. Upright chest X-ray demonstrating large left pleural effusion with midline shift and small right pleural effusion.

Computed tomography (CT) scan of the chest showed large bilateral pleural effusions with collapse of the right lower lobe and partial collapse of the upper lobes bilaterally (Figure 2).



Figure 2. Representative view from computed tomography (CT) scan (axial plane) in lung windows showing bilateral pleural effusions.

The patient developed hypoxemia and underwent thoracentesis with a total of 1.5 liters of pink, milky fluid removed (Figure 3).



Figure 3. Image of pleural fluid obtained from thoracentesis demonstrating pink, milky appearance.

Bilateral PleurX catheters (PleurX; Iskus Health; London, United Kingdom) were placed for persistent drainage. Fluid studies showed a triglyceride count of 147 mg/dL on the right side and 153 mg/dL on the left side. The patient continued to self-drain when symptomatic and drained about 600 mL of light-colored opaque fluid from each side daily. Serum albumin levels decreased to about 2.0 g/dL over the next week with concurrent development of diffuse pitting edema in all four extremities and abdomen. He was started on a high-protein, low-fat diet consuming up to 6-7 nutritional protein supplements per day with little to no improvement in his clinical state or serum protein levels. Given the patient's poor response to treatment and persistence of his pleural effusions, a trial of octreotide was initiated. The patient was given octreotide 100 mg three times per day. About 3 days after initiating therapy, the patient refrained from draining his PleurX catheters for the first time and the frequency of draining decreased over the remainder of the week due to improvement in symptoms. The fluid was noted to be less opaque and clearer with each drainage. The patient's tachypnea and oxygen saturation also showed improvement. After day 9 of octreotide, the treatment was discontinued and repeat pleural fluid studies showed a triglyceride count of 69 mg/dL on the right side and 89 mg/dL on the left side. With the resolution of his chylothorax and improvement in oxygenation status as well as his edema, the patient was discharged and will follow up with Oncology for continuation of his KS treatment.

Discussion

KS is known as an AIDS-defining illness that can invade a variety of tissues in the body leading to manifestations beyond the classic

skin lesions. It can cause unusual neurologic, cardiac, orbital, laryngeal, endocrine, and gastrointestinal complications in rare cases (5). We present a case of bilateral chylothorax as another rare potential complication of KS. Other reported cases have presented similarly to our patient, such as a case presented by Pennington *et al.* (6) which also described dyspnea and hypoxemia with transient but significant improvements in ventilation with serial chest drainage as well as repeated reaccumulation of the chylothorax. In their case, however, the patient died as a result of his condition. Other cases of presumed KS-induced chylothorax have also resulted in marked nutritional deficiencies as seen in our patient (7).

Treatment of chylothorax involves therapeutic thoracentesis, a low-fat diet that is high in medium-chain triglycerides which do not pass through the thoracic duct, and surgical correction or embolization of the defect (8). Though not a standard practice, the use of octreotide has been reported to improve chylothorax in some cases. The majority of these cases have been traumatic chylothorax following cardiothoracic surgery in adults or the pediatric population, or neonates with congenital chylothorax (8). There is a paucity of literature regarding octreotide in the management of malignant and other non-traumatic causes of chylothorax in the adult population. One case has been reported by Togashi *et al.* (9) which describes chylothorax secondary to idiopathic fibrosing mediastinitis that was treated successfully with octreotide. The exact mechanism is unknown, but as a somatostatin analogue, it may involve a

decrease in splanchnic blood flow and subsequent reduction in lymphatic flow from the gastrointestinal system and through the thoracic duct (10-11). There is no standard protocol for the administration of octreotide, however, most studies report a 1-2 week course with recognizable improvements after 2-3 days of treatment, as seen in our patient (12).

Conclusion

Bilateral chylothorax is a rare manifestation of KS that can lead to respiratory failure, malnutrition, and death. We present a case of non-traumatic, malignant chylothorax that was treated successfully with octreotide, a somatostatin analogue. Further studies are necessary to elucidate the exact mechanism of its effect on chylothorax and to establish a standardized treatment protocol for the usage of octreotide in this condition.

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