Acute inflammatory edema in the setting of bilateral lung transplantation

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INTRODUCTION

Acute inflammatory edema (AIE) is a non-infectious pseudocellulitis that was initially described by Marchione et al[1] in a case series of 15 patients in 2019. AIE typically presents as bilateral blanchable, erythematous, and edematous plaques in predisposed patients. AIE tends to be localized to the thighs, lower abdomen, and flanks in a dependent, pressure-sparing distribution.[2] Given the paucity of cases reported in the literature, little is known about the risk factors and clinical presentation of AIE. Herein, we present a rare case of AIE in a non-obese patient who had undergone bilateral lung transplantation.

Abbreviations used:

AIE: acute inflammatory edema
BMI: body mass index
LDS: lipodermatosclerosis

CASE PRESENTATION

A 55-year-old man with a history of Crohn's disease and end-stage interstitial lung disease was admitted for bilateral lung transplantation. His immediate postoperative course was complicated by thoracic compartment syndrome and allograft dysfunction requiring veno-venous extracorporeal membrane oxygenation support and tracheostomy with continuous mechanical ventilation. The patient developed multiorgan complications including multidrug resistant pneumonia, pneumomediastinum, sacral osteomyelitis, and severe volume overload requiring continuous veno-venous hemodialysis. During the hospitalization, the patient had developed erythematous plaques involving the bilateral thighs. History could not be obtained from the patient due to his altered mentation. There was no apparent trigger to the development of this eruption and no new medications.

On exam, the patient was intubated, mechanically ventilated and frail appearing with a body mass index (BMI) of 16.4 kg/m^2. His blood pressure was 108/78 mmHg, and heart rate was 84 beats per minute. Skin examination was significant for erythematous, edematous plaques involving the bilateral thighs with extension to the abdominal pannus. These indurated plaques spared the inguinal and infra-abdominal folds and were firm to palpation without tenderness (Fig 1). Laboratory studies were notable for hypoalbuminemia of 3.4 g/dL (Normal: 3.5-5.2 g/dL) and leukopenia of 2600 cells/mm^3 (Normal: 4-10,000 cells/mm^3). The differential diagnosis included AIE, lipodermatosclerosis (LDS), and stasis dermatitis. A punch biopsy demonstrated superficial dermal edema along with a perivascular and interstitial lymphohistiocytic infiltrate (Fig 2). These findings along with the patient's clinical presentation were consistent with AIE. Recommendations for frequent repositioning, volume status correction, and increased mobility were made. Over the next few days, the patient continued to receive intermittent dialysis, and nursing staff worked on repositioning and mobility with the patient.

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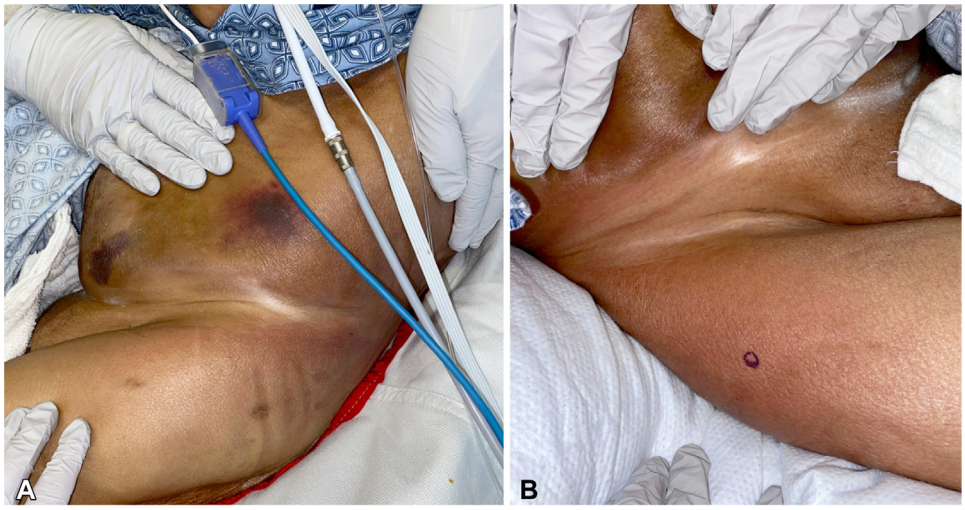

Fig 1. A and **B,** Erythematous indurated plaques involving the lower abdomen and anterior thighs but sparing the inguinal folds in a patient with acute inflammatory edema.

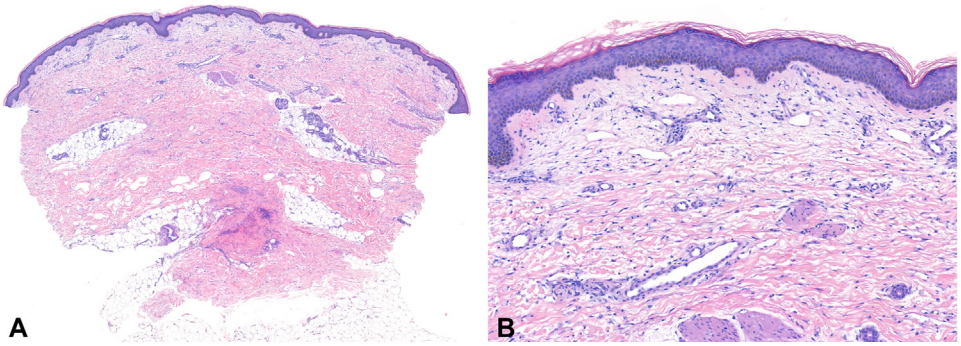

Fig 2. A, Nonspecific but consistent histopathologic findings of acute inflammatory edema include papillary dermal edema and dilated capillaries and venules in the superficial dermis (hematoxylin & eosin, 30× magnification). **B,** Papillary dermal edema, dilated capillaries and venules, and a sparse dermal infiltrate with mast cells (hematoxylin & eosin, 150× magnification).

DISCUSSION

AIE is associated with the development of erythematous and edematous plaques, typically presenting in dependent areas in patients who are critically ill.[1] Predisposing risk factors include fluid overload, renal dysfunction, an elevated BMI, and hypoalbuminemia.[2,3] The precise pathogenetic mechanism is unclear; however, prevailing theories suggest that AIE develops secondary to volume overload in patients who are critically ill with impaired lymphatic drainage.[1,3] This in turn leads to dermal edema and tissue microtears, with subsequent activation of the inflammatory cascade. AIE has been predominantly reported to occur in patients with an elevated BMI likely due to the association of impaired lymphatic drainage and obesity.[2] Our patient, however, was malnourished and a had a BMI of 16.4 kg/m^2, suggesting that alternate mechanisms may have contributed to its development. The patient's prolonged hospitalization was further complicated by profound sepsis along with development of a hypervolemic state due to acute renal failure, leading to increased hydrostatic pressure and vascular permeability. Severe malnutrition and consequential hypoalbuminemia resulted in low oncotic pressures, likely contributing to fluid extravasation and the development of AIE. The drastic improvement following correction of fluid status supports this proposed hypothesis.

AIE is primarily a clinical diagnosis.[1] Typical findings include erythematous and edematous plaques localized to fluid-dependent areas, such as the

lower abdomen and proximal lower extremities.[2] In all 15 patients reported by Marchionne et al,[1] the bilateral thighs were involved, similar to what was observed in this patient. Classic sparing of areas of pressure such as sites of contact with external objects and the inguinal folds is typically seen in patients with AIE. Elevation of inflammatory markers and leukocytosis are variably present, reflecting an underlying inflammatory rather than infectious process. Skin biopsy may prove helpful in distinguishing AIE from cellulitis and other infectious dermatoses for which antimicrobials would be indicated, especially since this condition is found to be associated with-patients who are critically ill.[2,3]

AIE is clinically differentiated from cellulitis, which is usually unilateral, by its bilateral distribution that spares areas of pressure.[1] On histopathology, cellulitis demonstrates neutrophilic infiltration, capillary dilatation, and bacterial invasion of tissue. Another finding that that differentiates AIE from cellulitis is the abnormal dilation of lymphatic channels.[4] LDS and stasis dermatitis can share a common distribution with AIE. These conditions can present on the thighs and pannus but typically manifest on the distal lower extremities and fail to spare skin folds. Additionally, the course of LDS and stasis dermatitis is usually chronic rather than acute, as seen in AIE. Histopathology of AIE typically shows findings of papillary and upper reticular dermal edema. An inflammatory infiltrate composed of scattered neutrophils, lymphocytes, and histiocytes may be present.[1,4] LDS demonstrates a lobular panniculitis with fat necrosis, lipomembranous change, and septal fibrosis. In stasis dermatitis, there are an increased number of thickened capillaries, red blood cell extravasation, and siderophages.

Management of AIE involves a supportive approach centered around optimization of volume status. Fluid restriction, diuresis, frequent repositioning, and increased mobility are the mainstays of therapy.[2,3] AIE may be misdiagnosed as cellulitis in patients who are critically ill, underscoring the importance of early dermatology consultation in improving patient outcomes and excluding clinical mimickers.[5]

Conflicts of interest

None disclosed.

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