Ecthyma gangrenosum is an uncommon cutaneous necrotizing infection classically associated with Pseudomonas aeruginosa bacteremia. It occurs almost exclusively in immunocompromised patients, particularly in the setting of hematologic malignancy. Only one case of sinonasal ecthyma gangrenosum has been reported to date. Here, we present a second case of sinonasal ecthyma gangrenosum with unique, previously unreported features.

CASE REPORT

A 35-year-old immunocompromised woman was evaluated for acute-onset fever, left-sided facial pain and swelling, without visual changes or facial hypoesthesia. Three months earlier, she had undergone peripheral blood stem cell transplantation for the treatment of acute lymphoblastic leukemia; she had been receiving tacrolimus and prednisone since. High-resolution computed tomography revealed opacification of the left maxillary sinus, though not frankly necrotic. Nasal endoscopy revealed an eschar within the middle meatus, adjacent to an enlarged antrum. Biopsy showed no fungal elements. Due to concern for acute invasive fungal rhinosinusitis, the patient underwent sinonasal debridement via combined endoscopic endonasal and endoscopic-assisted Caldwell-Luc approach. Abundant necrotic tissue was encountered, necessitating a left maxillary sinus antrum; no intraorbital abnormalities were noted. Diagnostic nasal rhinoscopy, crusting was noted distal to the left nasal vestibule. Nasal rhinoscopy showed no evidence of necrosis or other abnormal finding. On anterior rhinoscopy, nasal mucosa and mucosa showing necrotizing inflammation (Figure 2). Partial debridement of the extensive crusting was carried out, and specimens were submitted for immediate histopathologic analysis to rule out the presence of fungal elements; however, no fungal elements were encountered.

The patient was taken to the operating room. Examination of the left nasal cavity revealed frank necrosis of the inferior turbinate and lateral nasal wall mucosa. An abnormally enlarged maxillary sinus antrum was noted. The maxillary sinus mucosa, though not frankly necrotic, was pale and poorly vascularized; there was minimal bleeding as the maxillary sinus mucosa was debrided. There was no evidence of fungal elements on Gram stain. Methenamine silver stains did not show fungi. Gram stain did not identify bacteria; however, multi-Drug-resistant Pseudomonas aeruginosa was isolated from tissue culture. Culture-directed intravenous antibiotics were administered (tobramycin and ceftazidime). The patient showed significant clinical improvement subsequently.

CONCLUSION: A diagnosis of sinonasal ecthyma gangrenosum should be considered in an immunocompromised patient with an acute necrotizing rhinosinusitis in whom biopsy fails to show fungal elements.

REFERENCES


ABSTRACT

OBJECTIVE: Ecthyma gangrenosum is an uncommon cutaneous necrotizing infection classically associated with Pseudomonas aeruginosa bacteremia. Only one case of sinonasal ecthyma gangrenosum has been reported to date. Here, we present a second case of sinonasal ecthyma gangrenosum with unique, previously unreported features.

METHODS: Retrospective review of clinical case.

RESULTS: A 35-year-old immunocompromised woman was evaluated for acute-onset fever, left-sided facial pain and swelling, without visual changes or facial hypoesthesia. Three months earlier, she had undergone peripheral blood stem cell transplantation for the treatment of acute lymphoblastic leukemia; she had been receiving tacrolimus and prednisone since. High-resolution computed tomography revealed opacification of the left maxillary sinus, though not frankly necrotic. Nasal rhinoscopy, crusting was noted distal to the left nasal vestibule. Nasal rhinoscopy showed no evidence of necrosis or other abnormal finding. On anterior rhinoscopy, nasal mucosa and mucosa showing necrotizing inflammation (Figure 2). Partial debridement of the extensive crusting was carried out, and specimens were submitted for immediate histopathologic analysis to rule out the presence of fungal elements; however, no fungal elements were encountered.

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