

CLINICAL VIGNETTE

Milian's Ear Sign: Bilateral Ear Redness, Facial Rash, and Fever

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Case

A 59-year-old male, former smoker, with Hashimoto's thyroiditis, testicular hypogonadism, HLD, HTN, and OSA presented to his Primary Care Physician with rapid heart rate, fever and red erythematous ears. He noted a nodule behind the left ear for two days with worsening, left-sided neck pain and associated headache and photophobia. He had traveled to Hawaii 2 weeks prior with no infectious ocean water exposure. The PCP escorted the patient to the emergency department because of hemodynamic instability. Physical examination revealed moderate distress with tenderness of the left neck without meningeal signs. Laboratory investigations revealed an elevated white blood cell count (WBC) of 21,190 cells/mm³ with an absolute neutrophil count (ANC) of 19.12. Ultrasonography (US) of the neck showed a hyperechoic left thyroid gland without hypervascularity to suggest thyroiditis and normal appearing bilateral cervical lymph nodes. Computed tomography (CT) of the neck with contrast to rule out deep space infection was remarkable only for a left thyroid nodule. He was discharged with lidocaine patches and instructions for follow-up.

He was seen by his primary care physician the next day after development of a painful rash over the left cheek, scalp, forehead, and left ear, with left ear swelling. Pain was described as skin soreness and burning that was painful to touch. This was associated with an ongoing fever since discharge from the emergency room. He denied hearing loss, aural fullness, ear drainage, or vertigo. Physical examination showed a temperature of 38.6°C and an erythematous, blanching, mostly confluent, raised warm rash, extending throughout the forehead and bilateral ears to the left side of the neck and chest. He was referred to the emergency room due to concern for severe infection and admitted with a differential diagnosis including relapsing polychondritis vs vasculitis or other rheumatologic process vs cellulitis vs allergic reaction. Rheumatology, ENT, and infectious diseases were consulted. Labs were notable for WBC 17.2, erythrocyte sedimentation rate (ESR) 64, C-reactive protein (CRP) 18.3, procalcitonin 2.09. Computed tomography (CT) of the chest was ordered to rule out superior vena cava (SVC) syndrome. It revealed a left upper lobe calcified granuloma and enlarged nodule on the left thyroid, but no mass. Urine and bacterial cultures were collected and he was started on intravenous (IV) piperacillin-tazobactam with clinical improvement. Testing for immunocompromised states showed no

underlying immunodeficiency. A methicillin-resistant staphylococcus aureus (MRSA) nares swab was negative and other cultures showed no growth. Antibiotics were narrowed to IV ceftriaxone with suspicion for non-purulent skin and soft tissue infection (SSTI). He continued to improve after two days of ceftriaxone and was transitioned to oral amoxicillin-clavulanic acid for 10 days. There was no underlying rheumatologic syndrome identified nor clear ENT diagnosis. His symptoms improved and the lymphocytosis resolved. He was discharged home with confirmed symptom improvement. Symptoms entirely resolved on oral antibiotics and the most likely explanation was erysipelas.

Discussion

Facial rash with ear involvement suggests erysipelas, a form of cellulitis involving the superficial skin layers. It typically affects the lower limbs or the face.² Erysipelas is often caused by beta-hemolytic streptococci, most commonly by *Streptococcus pyogenes* (group A *Streptococcus*).³ Forty percent of patients have systemic symptoms and 78% have a predisposing factor, most commonly age and immunosuppression.⁴ A large retrospective study of 1142 episodes of erysipelas in 981 patients reported patients with erysipelas were more likely to be male, around 60 years-old, with underlying diseases or predisposing conditions,⁵ as with our patient.

Other erysipelas risk factors include prior saphenous vein excision for bypass, lymphedema, lymphatic obstruction, arteriovenous fistula, status post-surgery (eg mastectomy), nephrotic syndrome, and immunocompromised state.⁶ This patient did not have specific risk factors, and immunodeficiency evaluation was negative.

Three clinical features help distinguish erysipelas from cellulitis: 1) the lesion is raised above the surrounding skin; 2) there is a clear line of demarcation; and 3) a brilliant salmon-red color of the lesions. Erysipelas is frequently preceded by 48 hours of malaise, fever, and chills, and facial erysipelas often develops after streptococcal sore throat. Lesions will often be described as burning, tender, and itchy.^{3,6} Involvement of the ear is called Milian's ear sign. This sign distinguishes erysipelas from facial cellulitis as the ear is spared in cellulitis due to the absence of deeper dermal tissue and subcutaneous fat in the ear.^{7,8} Erysipe-

las should be considered in the differential when patients have bilateral erythema of the ears, although the incidence of bilateral ear erysipelas is rare.⁸

The diagnosis of erysipelas remains clinical. Laboratory tests will show elevated WBC count with increased polymorphonuclear neutrophils (PMNs) and an elevated ESR,⁹ as was evident in our patient. Blood cultures are only positive in 5% of patients and the causative agent can be isolated by culture of the skin lesions.⁴ Additional testing and culture should be considered for immunocompromised and toxic-appearing patients.⁶ Blood cultures from our patient did not exhibit any growth.

Antibiotics are the mainstay of erysipelas management. A penicillinase-resistant semisynthetic penicillin or cephalosporin should be selected. Clindamycin, linezolid, or vancomycin may be used in penicillin-allergic patients and can be used if there is high clinical suspicion for MRSA.³ A meta-analysis showed no statistical difference between penicillin-based antibiotics versus cephalosporins, and no difference between older- and newer-generation cephalosporins regarding clinical outcomes.¹⁰

Complications include abscess formation, scarlet fever, pneumonia, meningitis, skin necrosis, hemorrhagic purpura, thrombophlebitis, and bullous formation.^{3,4} Erysipelas bullosum is when an accumulation of exudative fluid in the papillary layer causes separation of the dermis and blistering. Severe erysipelas may lead to necrosis and gangrene, called erysipelas gangrenosum.⁹

Ear redness can have varying etiologies. Differential diagnosis includes relapsing polychondritis, red ear syndrome, and erysipelas. Patients with acute fever and superficial inflammation are suspected to have erysipelas. Milian's ear sign refers to ear involvement in erysipelas. Diagnosis remains clinical and blood cultures should not be routinely requested. Antibiotics remain the mainstay of treatment for erysipelas with penicillin-based antibiotics or cephalosporins.

Frank Sun also contributed to this manuscript.

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