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Lyme Chondritis Presenting as Painless Ear Erythema

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KEY WORDS

Lyme disease, chondritis, otolaryngologic manifestations, ENT

ABBREVIATIONS

EM—erythema migrans

Ig—immunoglobulin

Dr Srinivasalu was involved in the care of the patient, conceptualized the study, and drafted the initial manuscript; Dr Brescia was involved in the care of the patient, conceptualized the study, and critically reviewed and revised the manuscript; Dr Rose supervised the preparation of the manuscript, and critically reviewed and revised the manuscript; and all authors approved the final manuscript as submitted.

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abstract

Erythema of the ear lobe in the context of Lyme disease is caused by either borrelial lymphocytoma or localized erythema migrans. Here we present a case of chondritis limited to the ear cartilage caused by Lyme disease. The patient was treated with ceftriaxone with complete resolution of symptoms. *Pediatrics* 2013;131:e1977–e1981

Lyme disease is endemic in northeastern states and the upper Midwest of the United States¹ and is caused by the tick-borne spirochete *Borrelia burgdorferi*. The clinical manifestations range from erythema migrans in early infection to disseminated disease involving the nervous, cardiovascular, and musculoskeletal systems. The otolaryngologic manifestations of Lyme disease include Bell's palsy, sensorineural deafness, cervical adenopathy, vocal cord involvement, and stiff neck.² Although *Borrelia burgdorferi* causes arthritis by involving the synovial lining and subsequent cartilage damage from inflammation, direct involvement of cartilage has not been reported.

CASE PRESENTATION

We present the case of a 12-year-old white girl with a history of painless right ear swelling and redness of 1 month's duration that started in early June. The erythema, swelling, and warmth initially started over the helical rim of the right ear and expanded to involve the entire right pinna and surrounding ipsilateral face and jaw with distinct borders over 1 to 2 weeks. There was no central clearing. There was no history of pain, itching, oozing, or purulent discharge from the lesion. Cellulitis was suspected initially and she was treated with 3 courses of antibiotics over 6 weeks (cephalexin 250 mg 3 times a day for 2 weeks followed by cephalexin 500 mg twice a day for 2 weeks, and ciprofloxacin 500 mg twice a day for 2 weeks) during which time there appeared to be resolution of swelling and erythema of ipsilateral surrounding face and jaw with the exception of the cartilaginous portion of the right pinna. Her past medical history was unremarkable; specifically, it was negative for any history of trauma or recent ear piercing, symptoms of relapsing polychondritis, including previous ear involvement, involvement

of the nasal bridge, hoarseness, tinnitus, dizziness, or history of painful ocular redness. Notably, a tick was removed from the occipital region of the scalp a few weeks previous. Review of systems was otherwise unremarkable. Physical examination revealed swelling of the superior half of the right pinna along with dusky appearance and minimal tenderness over the helical rim for about 7 mm. The pinna was deformed and floppy (Fig 1A). There was an enlarged, nontender jugulodigastric node on the right. Laboratory studies were significant for sedimentation rate of 36 mm per hour, normal complete blood count, negative antinuclear antibody, negative anti-neutrophil cytoplasmic antibody, and negative rheumatoid factor. Lyme serology showed enzyme immunoassay: >5.0 U/mL (normal <0.9); Western blot 10/10 immunoglobulin G bands and 3/3 immunoglobulin M bands. Ultrasound showed thickening of the soft tissues with increased echogenicity and hyperemia compatible with inflammation of the right auricular cartilage

(Fig 2). No focal fluid collection was identified. The patient was treated with ceftriaxone (2 g daily) intravenously for 4 weeks. She had remarkable improvement of the ear swelling and erythema with treatment (Fig 1B).

DISCUSSION

We report herein the case of a 12-year-old girl with auricular chondritis and positive Lyme serology with subsequent resolution of the inflammation with antibiotic treatment for Lyme disease. Auricular chondritis caused by Lyme disease has not been reported in the medical literature.

The possibility of a bacterial superinfection from the tick bite leading to perichondritis cannot be completely excluded. Further, perichondritis and chondritis are indistinguishable by ultrasound. Lyme disease was considered more likely in this patient because of the location of the tick bite, temporal association of the tick bite to the onset of symptoms, and positive serology for Lyme disease.

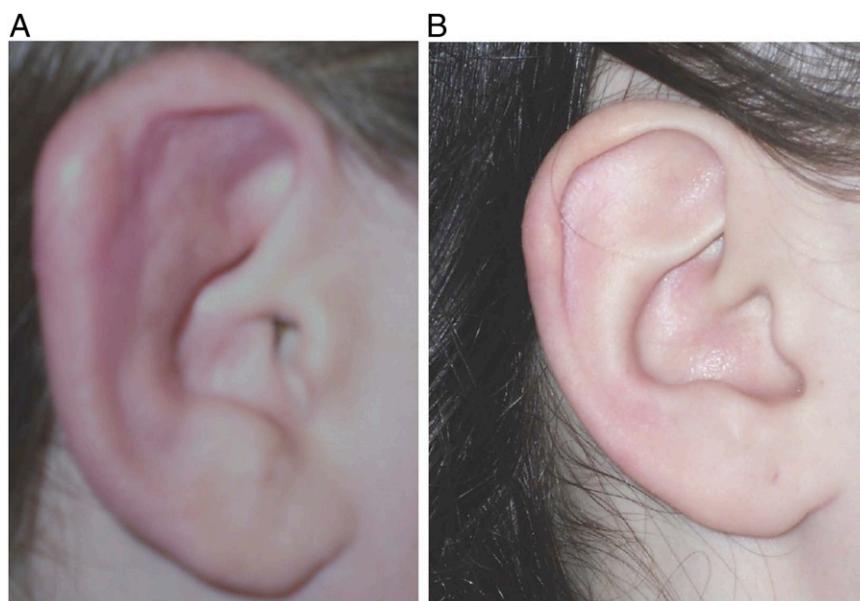


FIGURE 1
A, Photograph of the right ear taken 1 month after onset of symptoms. B, Repeat photograph of right ear taken after patient received treatment for Lyme disease. Note the erythema and distortion of the cartilaginous portion of right ear before the treatment of Lyme disease (A).

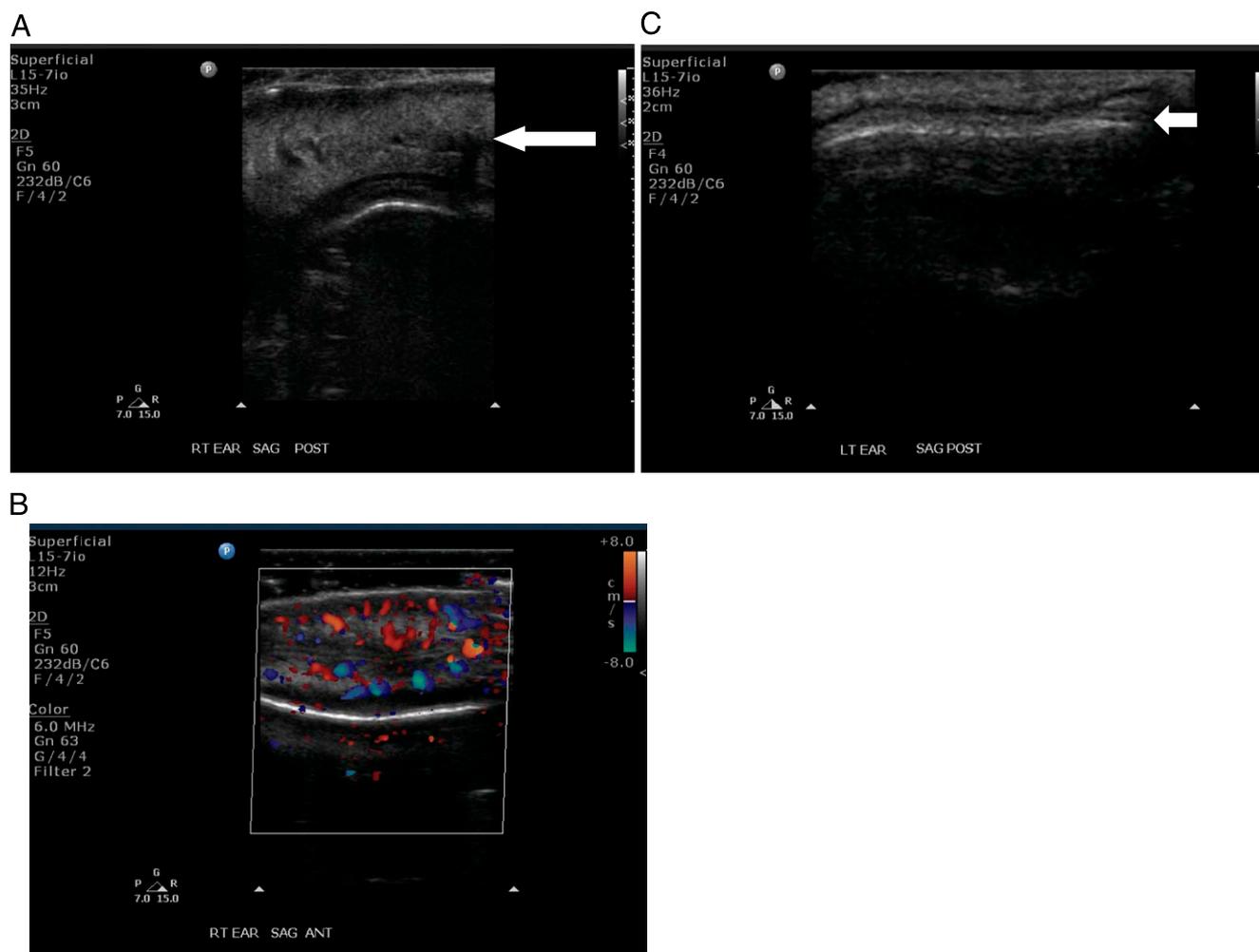


FIGURE 2

Ultrasound of bilateral ear showing thickening and increased echogenicity of the subcutaneous soft tissues (long white arrow) (A) along with hyperemia on color flow Doppler of the right ear (B). Contrast this with the hypoechoic well-defined linear cartilage on the unaffected ear of the same patient (short white arrow) (C).

The most common presentation of erythema migrans (EM) in North America is homogeneous erythema (59%); central clearing is noted in only 9%. Further, based on cleavage lines, the EM lesion may not be annular in certain body locations.³ EM lesions typically appear in or around a large joint, but can be located anywhere on the body except the palms and soles. In children, the head and neck region is the most common location.⁵ EM was misdiagnosed as cellulitis in this patient, which resulted in delay in diagnosis of early Lyme disease and led to the subsequent chondritis of the right ear cartilage. EM lesions cause local itching or

burning in 50% of cases. In contrast, cellulitis is associated with warmth, pain, tenderness, and possibly discharge. Tender lymphadenopathy is more common with cellulitis.⁴ Erysipelas may sometimes be confused with EM but is generally associated with fever and chills.⁵ The clinical presentation in our patient was similar to relapsing polychondritis with respect to the chronicity of the auricular cartilage involvement, along with deformity of the pinna. However, there were no previous episodes of chondritis and no other features on history suggestive of this diagnosis. Borrelial lymphocytoma, a rare manifestation of

Lyme disease⁶ caused by B-cell lymphocytic infiltration of the dermis and subcutis can produce ear discoloration and affect the lobular portion of the ear. A biopsy was not performed in our patient because of the entailed risks of trauma and scarring; further, at the time, significant modification to the treatment plan based on biopsy was not expected. The distribution of the erythema, with characteristic sparing of the lobule and thickening of the cartilaginous portion of the ear, support the diagnosis of acute chondritis. *Borrelia burgdorferi* spreads locally in the skin after being inoculated through tick salivary glands.⁷ Homing of the

spirochete to specific sites and dissemination occurs when the organism attaches to extracellular proteins, matrix glycosaminoglycans, and host integrins.⁸ Binding of the organism to platelets facilitates in hematogenous spread.⁹ The spirochete is presumed to reach the synovium through the bloodstream. The organism indirectly binds to collagen fibers via binding to decorin, a proteoglycan on collagen fibers through borrelia decorin-binding proteins A and B.¹⁰

Various atypical manifestations of Lyme disease, such as Horner syndrome,¹¹ amyopathic dermatomyositis,¹² recurrent nerve palsy,¹³ uveitis, and other ocular manifestations,¹⁴ have been well documented in literature. Affinity of *Borrelia burgdorferi* for tissues of mesenchymal origin in the form of myositis and osteomyelitis is also well recognized.^{15,16} We posit that there was local invasion from the superficial skin infection into the auricular cartilage in addition to becoming blood borne. This is theoretically possible because of the close proximity of the skin to the

underlying cartilage in this anatomic location.

Although the rash appeared to improve while the patient received cephalexin and ciprofloxacin, this may perhaps have been due to the natural history of EM. Neither cephalexin nor ciprofloxacin are effective in treatment of EM. According to the Infectious Diseases Society of America guidelines for treatment of EM, amoxicillin at a dosage of 50 mg/kg per day (maximum 500 mg/dose) or cefuroxime at 30 mg/kg per day (maximum 500 mg/dose) is recommended for children younger than 8 years for 14 to 21 days; doxycycline is recommended at a dosage of 4 mg/kg per day (maximum 100 mg/dose) for children older than 8 years for 10 to 21 days. If clinicians cannot effectively differentiate between cellulitis and EM, such as in this case, treatment with either cefuroxime axetil or amoxicillin/clavulanic acid (Augmentin) is recommended, as they are effective against either condition.¹⁷ Oral antibiotics are the recommended first-line treatment of Lyme arthritis. However, to prevent

cosmetic deformity from the chondritis and because of the superior tissue penetrance, intravenous ceftriaxone was chosen for treatment in our patient. Cartilaginous infections can be difficult to treat because of limited blood supply. Prompt diagnosis and treatment are essential in auricular chondritis to prevent cosmetic deformity of the ear lobe from long-standing inflammation.

This case report emphasizes that Lyme disease should be considered in the differential diagnosis of erythematous lesions during the spring/summer months in endemic areas. Further, EM can be confused with cellulitis, and Lyme disease should be considered in the differential diagnosis of cellulitis. Finally, auricular chondritis should be added to the list of rare complications from untreated Lyme disease.

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