

A case of neonatal lupus erythematosus with fever in an infant born from an anti-SSA/Ro and anti-SSB/Lo antibody-positive mother

Dear Editor,

Neonatal lupus erythematosus (NLE) is an uncommon syndrome, which is caused by the transplacental passage of maternal autoantibodies to anti-SSA/Ro antibody and/or anti-SSB/Lo antibody.¹

In a 21-year-old woman, infiltrative annular erythematosus regions involving the trunk, limbs, and ears appeared 1 month after the first pregnancy. A blood test showed that the ANA

titer was 1:1280, and the anti-SSB/Lo antibody titer was 1:16, whereas there was no anti-SSA/Ro antibody. There were no symptoms. The patient delivered a girl weighing 2680 g in week 38 of pregnancy.

The girl exhibited infiltrative annular erythematosus regions involving the forehead, trunk, and limbs, which appeared 20 days after birth (Figure 1A, B). Simultaneously, fever was noted (39.0°C). The

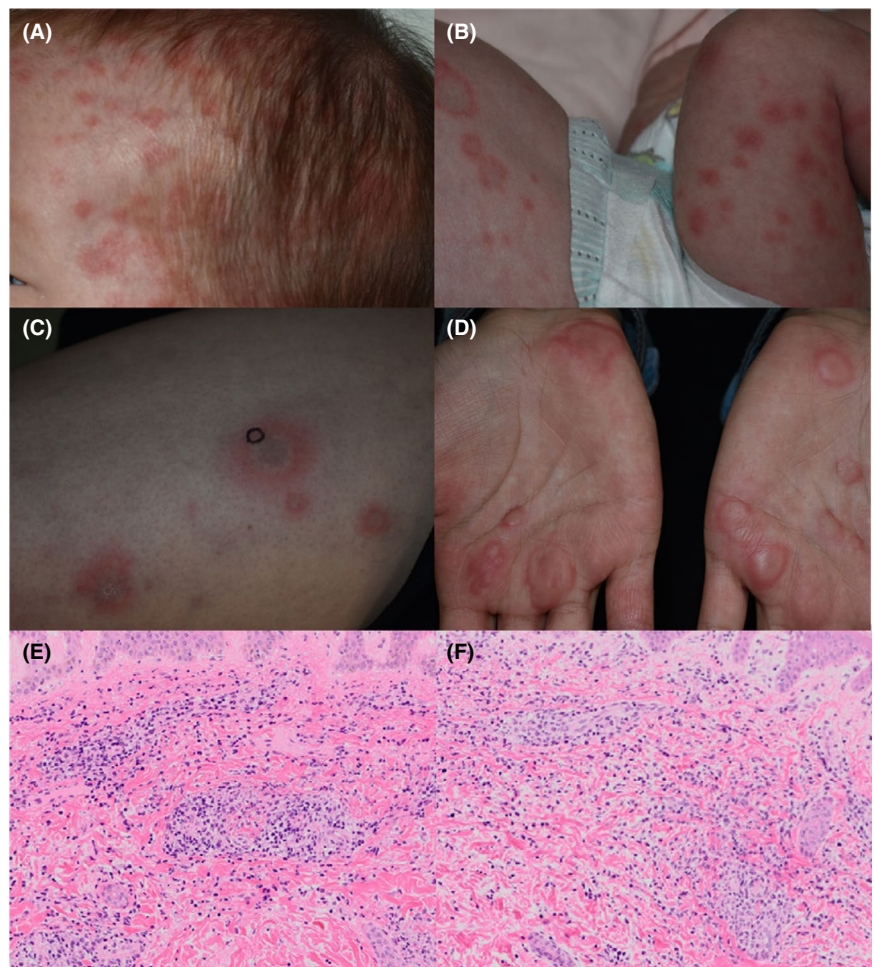


FIGURE 1 A, B, Skin rash with annular erythematosus lesions on the face, trunk, and legs of the baby. A, biopsy specimen was collected from the trunk. C, D, Infiltrative annular erythema involving the femoral region and bilateral hands. A biopsy specimen was collected from the marked point of the femoral region. E, F, Leukocytoclastic vasculitis involving the superficial to deep layers was observed in the maternal dermis (E) and in the neonatal dermis (F). (hematoxylin-eosin staining, $\times 100$)

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girl was brought to our department and referred to the Department of Pediatrics. Laboratory data showed that she was positive for anti-SSA/Ro (142 U/mL) and anti-SSB/Lo (>320 U/mL) antibodies, leading to a diagnosis of NLE. The serum aspartate aminotransferase (AST) and alanine aminotransferase (ALT) levels were 297 (reference range: 21-64 U/L) and 185 U/L (reference range: 12-50 U/L), respectively. The white blood cell (WBC) count and C-reactive protein (CRP) level were 6940/ μ L (4800-18 500/ μ L) and 0.27 mg/dL (<0.3 mg/dL), respectively.

In the mother, exanthema involved the limbs and trunk (Figure 1C, D). A blood test was positive for anti-SSA/Ro (>240 U/mL) and anti-SSB/Lo (>320 U/mL) antibodies. Skin biopsy was performed from annular erythema specimens of the patient's femoral region and baby's trunk (Figure 1B, C).

The results of the biopsy showed leukocytoclastic vasculitis (Figure 1E, F). On direct immunofluorescence, there was no deposition of immunoglobulin G/A/M, C1q, or C2 in either.

The baby was admitted to the Department of Pediatrics. To reduce the fever, the drip infusion of an antibiotic and acyclovir was started. A blood test 3 days after admission showed that the WBC count, AST level, and ALT level were 7740 μ L, 537 U/L, and 329 U/L, respectively. However, the fever subsided 5 days after admission. Blood culture was negative 7 days after admission, and hematological findings did not suggest HSV, cytomegalovirus, or Epstein-Barr virus infection.

The WBC count, AST level, and ALT level were improved, 14 days after visiting our department.

Neonatal exanthema gradually disappeared, and the anti-SSA/Ro and anti-SSB/Lo antibody titers were decreased 7 months after birth.

The incidence of an infant with potential NLE born to a mother positive for anti-SSA/Ro antibody has been reported to be 1%-2%.² NLE-related exanthema was reported to disappear after 9 months, when transitional antibodies may disappear.³ In the present case, the course was also similar, but fever of the baby was relatively infrequent. An association between cutaneous vasculitis and the presence of anti-Ro (SSA)/La (SSB) antibodies was reported.⁴ The fever in this case may have been caused by cutaneous vasculitis. Further accumulation of cases is needed to clarify the involvement of the antibodies in the pathogenesis.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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