A 1-day-old female infant presented with red patches on the right arm that had been present since delivery. The patient was born to a healthy mother by spontaneous vaginal delivery without complications and with a good Apgar score. The newborn moved both arms and legs well and blood work was unremarkable. Her mother noted being healthy during pregnancy, and she had not taken any additional medications aside from prenatal vitamins. Examination of the infant revealed red blanchable reticulate patches in a dermatomal distribution extending from the posterior aspect of the right shoulder (top) down to the flexural aspect of the arm (bottom). There also were a few coalescing reticulate patches on the superior aspect of the right side of the chest and superior aspect of the right side of the back that resolved by 3 months of age.

WHAT’S THE DIAGNOSIS?

a. angioma serpiginosum
b. congenital unilateral nevoid telangiectasia
c. erythema ab igne
d. hemangioma
e. nevus flammeus

PLEASE TURN TO PAGE E12 FOR THE DIAGNOSIS
Two weeks later the patches were noticeably lighter (Figures 1A and 1B). She continued to be in good health, but gynecomastia was notably present on examination (Figure 1C). At 3 months of age, all patches on the right arm, superior aspect of the chest, and superior aspect of the back had resolved, along with the gynecomastia (Figure 2).

This case describes the rare condition of congenital unilateral nevoid telangiectasia (UNT). Unilateral nevoid telangiectasia is a rare cutaneous vascular condition first described by Blaschko in 1899. It is characterized by the presence of unilateral superficial telangiectases occurring most often in the cervical and upper thoracic dermatomes in a linear pattern. Females are more often affected than males (2:1 ratio), and cases of UNT are either congenital or acquired. Although most UNT cases are acquired and often found in females, approximately 15% of cases are congenital and are comprised largely by males. Acquired cases have been hypothesized to occur in association with hyperestrogenemic states such as pregnancy, puberty, oral contraceptive use and hormonal therapy, alcoholism, and liver disease including hepatitis B and C infections. There is conflicting evidence as to whether there is an absolute increase in the presence of estrogen and progesterone receptors in the skin, as many case reports show no increase. Instead, others hypothesize that the condition is actually a result of somatic mosaicism and that the cutaneous lesions are genetically predisposed to becoming visibly evident under conditions of elevated estrogen.

In our case, we hypothesize that the cause was elevated maternal estrogen levels present at higher than normal levels in the fetal circulation. The presence of gynecomastia seen in our patient supports the hypothesis that increased circulating estrogen may be present in infants with UNT.

REFERENCES