

Penile Herpes Simplex Virus Type 1 Infection Presenting Two and a Half Years After Jewish Ritual Circumcision of an Infant

Orit Yossepowitch, MD,* Tamar Gottesman, MD,* Orna Schwartz, MSc,† Michal Stein, MD,‡§ Francis Serour, MD,‡§ and Michael Dan, MD*§

Abstract: The association between Jewish ritual circumcision and genital herpes simplex virus type 1 infection has been well described. We report a case of genital herpes that first presented at the age of 2½ years. We believe that the infection was acquired asymptotically through direct orogenital suction performed during circumcision in the newborn period.

The association between neonatal genital herpes simplex virus type 1 (HSV-1) infection and the Jewish ritual orthodox circumcision at the age of 8 days, which involves the practice of *mezizah b'peh*, is now well established.¹⁻⁴ In this ultraorthodox Jewish practice, the circumciser (*mohel*) places his mouth directly on the newly circumcised penis and sucks blood away from the circumcision wound (direct orogenital suction). This procedure is repeated several times until bleeding stops.¹ According to Gesundheit et al.,¹ in the typical case of circumcision-associated neonatal genital herpes, the clinical manifestations appear 7 days after the circumcision (range, 4–11 days), and most cases (75%) present with fever. Recurrences were observed in 50% of the neonates. Serology (anti-HSV-1 immunoglobulin G [IgG]) was negative most infants and their mothers.

We report herein a case of genital herpes in a boy who presented clinically for the first time at the age of 2.5 years. We believe that the infection was acquired asymptotically in the postnatal period on the occasion of ritual orthodox circumcision.

A 2.5-year-old boy was admitted to the pediatric department with a 10-day history of ulcerative eruption on the corona of the glans penis and dysuria. No improvement was observed despite treatment with local and systemic antibiotics. He was born after a normal pregnancy and vaginal vertex delivery at 39 weeks' gestation weighing 4.265 kg. The mother was a 32-year-old, gravida 2, para 2 with gestational diabetes. At birth, the infant was found to have glucose-6-phosphatodehydrogenase deficiency. At 30 days of age, a ritual circumcision was performed by a *mohel* (religious circumciser), an individual trained to perform circumcision as part of a religious ceremony (the circumcision was delayed because of the boy's prolonged jaundice).

On current admission, abnormal physical findings were limited to the genital area: several vesicles and small ulcers were observed on the corona of the glans penis, which appeared erythematous and edematous (Fig. 1). A vesicle with erythematous

borders was seen at the right base of the scrotum. There was no fever. Complete blood count showed 8300 white blood cells/ μL , with 21.5% polymorphonuclear leukocytes, 66.1% lymphocytes, 11.6% monocytes, 0.4% eosinophils, and 0.4% basophils; 10.8 g hemoglobin/dL; and 281,000 platelets/ μL . Findings of the blood chemistry (electrolytes, kidney function tests) were normal. Because of the lesions' appearance, herpetic infection was suspected. Polymerase chain reaction performed on a swab obtained from the lesions was positive for HSV type 1. Herpes simplex virus type 1 serology, performed on a blood sample obtained on the eighth hospital day, was positive for IgG and negative for IgM; no HSV-2 antibodies were detected. The patient was treated with intravenous acyclovir (30 mg $\text{kg}^{-1} \text{d}^{-1}$) for 4 days and was discharged with recommendation to continue acyclovir orally for additional 7 days. No recurrences were noted over an 11-month period. There was no maternal or paternal history of oral or genital herpetic infection. Maternal serology was positive for HSV-1 and negative for HSV-2.

When asked, the father recalled that the *mohel* who was an ultraorthodox Jew, performed *mezizah b'peh* on the bleeding penis after excising the foreskin, as part of the orthodox ritual. This specific *mohel* is known in the community to perform circumcision according to the strictest orthodox Jewish laws, including the act of *mezizah b'peh*. The infant's parents categorically opposed that we contact the *mohel* for HSV antibodies testing.

Neonatal herpes is of rare occurrence. Most (approximately 85%) infections result from exposure to infectious maternal genital secretions at delivery. Postnatally acquired neonatal infection is even rarer (10%) and usually results from orolabial infection of a caregiver, including parent or nursery personnel.⁵ Another source of postnatal acquisition that has been recently recognized is the act of *mezizah b'peh* in the Jewish traditional circumcision during which the infant's wound is in contact with oral secretions of the *mohel*.¹

At the time of the appearance of the genital herpetic lesions and the positive polymerase chain reaction for HSV-1, our patient already had a positive IgG serology for HSV-1 antibody. This pattern corresponds to a symptomatic recurrence, rather than to a primary infection.⁶ Because this was the infant's first clinical attack, we believe that the initial infection, which was acquired on the occasion of the circumcision, was asymptomatic and went unrecognized, as it often happens with initial herpes simplex infections.⁶ Acquisition of primary genital HSV-1 infection (initial episode) is characterized by initial absence of anti-HSV-1 antibody. It takes 2 to 3 months for type-specific IgG antibodies to appear. At the time of recurrent episodes, anti-HSV-1 antibody is already present, which explains the milder nature of recurrent episodes compared with the initial episode.⁶

The most plausible source of the virus in the present case was acquisition from the oral secretions of the *mohel*, mainly because the eruption involved almost exclusively the corona of the glans penis, mirroring the circumcision wound and the immediate contact with the *mohel*'s saliva. Neonatal genital HSV-1

From the *Infectious Diseases Unit, †Microbiology Laboratory, ‡Department of Pediatrics, E. Wolfson Hospital, Holon, Israel; and §Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel.

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Correspondence: Michael Dan, MD, Infectious Diseases Unit, E Wolfson Hospital, Holon 58100, Israel. E-mail: midan@post.tau.ac.il.

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Figure 1. Multiple vesicular and ulcerative lesions on the penis.

infection after Jewish traditional circumcision was reported for the first time in 2000. The authors described 2 patients who were diagnosed in 1988 and in 1998. Both infants were circumcised by the same mohel who had performed the orthodox act of *metzitzah b'peh*.² A report from Israel described 8 cases,¹ in all of which the *mohel* had performed oral *metzitzah*. Four infants had recurrent episodes of genital HSV infection, and 1 developed HSV encephalitis with neurologic sequelae. All 4 ritual circumcisers (*mohelim*) tested for HSV antibodies were seropositive. Recently, a series of 11 cases from New York City was reported.⁴ The median interval from circumcision to appearance of herpes was 8 days. The infection was caused by HSV-1 in 9 cases and by untyped HSV in 2 cases. Three had disseminated disease, and 2 had central nervous system involvement; 2 infants died. Because of the increased awareness of the infectious risk associated with this ancient procedure of *metzitzah b'peh*, many orthodox circumcisers have modified this procedure to avoid direct oral contact with the infant's blood or penis using a pipette-like device.²

The infant's mother tested positive for HSV-1 antibodies. Maternal antibodies do not guarantee protection of the infant from acquiring the infection. Indeed, in 3 of the previously described cases,^{1,2,4} penile postnatal HSV-1 developed after circumcision despite maternal HSV-1 antibodies. Nonetheless, the presence of antibodies in the mother might have played a role in the asymptomatic nature of the primary infection of the infant.

Sexually acquired genital HSV-1 is increasingly reported.⁶ After sexual acquisition, HSV-1 can be shed in the genital area with or without symptoms. According to a recent report from New York City, neonatal herpes caused by HSV-1 outnumbered cases caused by HSV-2.⁷ However, it is highly unlikely that the patient's herpes infection resulted from intrapartum transmission of the virus from his mother. Neonatal herpes is often a devastating systemic disease, with severe sequelae. Theoretically, the infant might have developed postnatally the milder skin, eye, and mouth disease, which can manifest with discrete vesicles that may be difficult to detect without careful examination.⁸ The cutaneous

rash in skin, eye, and mouth disease consists of a single lesion or clusters of vesicles, often appearing first on the presenting part of the infant (i.e., face or head in vertex deliveries and buttocks in breech deliveries).⁹ Recurrent skin lesions are common,¹⁰ including after HSV-1 infections, appearing at the site of the primary lesions.¹¹ In the present case, however, although the infant was born after vortex delivery, the herpetic lesions were limited to the site of the former circumcision wound.

We cannot exclude another form of postnatal acquisition of the virus from a caregiver with oral infection may be it the parents, other members of the family, or day care personnel.¹² Upon questioning, the parents denied any orogenital contact with the infant. Still, even if an orogenital contact with a caregiver did happen, one would expect the genital eruption to be more extensive than it was.

Although in Israel Jewish boys are universally circumcised, postcircumcision genital herpes remains uncommon, probably because most circumcisions are performed using the pipette-like device (indirect orogenital suction). Parents choosing Jewish ritual circumcision should inquire in advance whether the *mohel* performs the *metzitzah b'peh*, and direct orogenital suction should be avoided. The association with a circumcision of a genital herpes infection should not be discarded only because of its delayed clinical appearance.

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