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

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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 asymptomatically through direct orogenital suction performed during circumcision in the newborn period.

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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. 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. 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 asymptomatically 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, mirroring the circumcision wound and the im-

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 kg⁻¹ 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. 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.

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. 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. 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.

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
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. A report from Israel described 8 cases, 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 seronegative. Recently, a series of 11 cases from New York City was reported. 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.

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, 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. 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. 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. 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). Recurrent skin lesions are common, including after HSV-1 infections, appearing at the site of the primary lesions. 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. Upon questioning, the patients 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.

REFERENCES