

Neonatal Genital Herpes Simplex Virus Type 1 Infection After Jewish Ritual Circumcision: Modern Medicine and Religious Tradition

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ABSTRACT. *Objective.* Genital neonatal herpes simplex virus type 1 (HSV-1) infection was observed in a series of neonates after traditional Jewish ritual circumcision. The objective of this study was to describe neonate genital HSV-1 infection after ritual circumcision and investigate the association between genital HSV-1 after circumcision and the practice of the traditional circumcision.

Methods. Eight neonates with genital HSV-1 infection after ritual circumcision were identified.

Results. The average interval from circumcision to clinical manifestations was 7.25 ± 2.5 days. In all cases, the traditional circumciser (the *mohel*) had performed the ancient custom of orally suctioning the blood after cutting the foreskin (oral *metzitzah*), which is currently practiced by only a minority of *mohels*. Six infants received intravenous acyclovir therapy. Four infants had recurrent episodes of genital HSV infection, and 1 developed HSV encephalitis with neurologic sequelae. All four *mohels* tested for HSV antibodies were seropositive.

Conclusion. Ritual Jewish circumcision that includes *metzitzah* with direct oral-genital contact carries a serious risk for transmission of HSV from *mohels* to neonates, which can be complicated by protracted or severe infection. Oral *metzitzah* after ritual circumcision may be hazardous to the neonate. *Pediatrics* 2004;114:e259–e263. URL: <http://www.pediatrics.org/cgi/content/full/114/2/e259>; ritual circumcision, herpes simplex infection, infection of the newborn, Jewish tradition.

ABBREVIATIONS. HSV, herpes simplex virus.

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Biblical sources dictate routine ritual circumcision at 8 days of age for Jewish boys. This procedure is widely accepted, and 60% to 90% of newborn boys of the Jewish population in the United States undergo this procedure,^{1,2} which also has an important cultural and historical role. The medically beneficial versus harmful consequences have long been debated.³ Circumcision has been reported to reduce the incidence of urinary tract infections in infants,⁴ young boys under the age of 2 years,⁵ and preschool boys.⁶ Pathologic phimosis and paraphimosis is precluded by the absence of a foreskin, and balanitis and posthitis (inflammation of the prepuce) primarily affect uncircumcised male individuals. Virtually all sexually transmitted diseases,⁷ including human immunodeficiency virus infection,^{6,8} are reported to be more common in uncircumcised men.

Both immediate and long-term complications of ritual circumcision are rare, probably because of the specific and meticulous precautions required by Jewish law. Only an experienced and qualified circumciser, the *mohel*, is allowed to perform circumcision. Historically, Jewish ritual circumcision consists of 3 parts: 1) the excision of the outer part of the prepuce (*milah*), 2) slitting of the foreskin's inner lining to facilitate the total uncovering of the glans (*peri'ah*), and 3) the sucking of the blood from the wound. Formerly, the *mohel* took some wine in his mouth and applied his lips to the part involved in the operation and exerted suction, after which he expelled the mixture of wine and blood into a receptacle provided for this purpose; this procedure was repeated several times until bleeding stopped (*metzitzah*). The first 2 parts are the act of circumcision, whereas the removal of the blood was done for medical reasons of wound care. However, the ancient procedure of *metzitzah* also carries a risk of infection, and currently most *mohels* use an appropriate suction device, such as a mucus extractor.

The incidence of neonatal herpes simplex virus (HSV) infections ranges from 1 to 6 per 20 000 live births. Most neonatal HSV infections result from exposure to infectious maternal genital secretions at delivery. Postnatal transmission usually results from nongenital infection of a caregiver, including parent or nursery personnel with oral lesions.⁹ Nosocomial transmission in nurseries has been documented.⁹ We present 8 infants who developed neonatal HSV-1

infection after oral *metzitzah* following ritual circumcision, most probably as a consequence of transmission by the *mohel's* saliva.

METHODS

Neonates who developed genital HSV-1 infection after circumcision from the following medical centers were evaluated: Soroka University Medical Center (Beer Sheva, Israel); Safra Children's Hospital, Sheba Medical Center (Tel Aviv, Israel); Hadassah Hospitals at Mt. Scopus and Ein Kerem (Jerusalem, Israel); and the Hospital for Sick Children (Toronto, Ontario, Canada). There is no official registry in the United States or in Israel to document the incidence of medical complications after ritual circumcision. The percentage of *metzitzah* performed by oral versus instrumental suction could not be evaluated statistically. The 8 cases were collected from personal communication and the experience of the authors from 1997 to 2003. Clinical data from all patients were collected, and follow-up was conducted during hospitalization and after the discharge of the infants from the hospital.

Identification of herpesvirus from lesions was performed by microscopic examination of Tzanck preparations and electron microscopy of specimens directly from the lesions. The virus was determined to be HSV-1 by immunofluorescence microscopy, isolation in cell culture, or polymerase chain reaction. HSV serostatus and seroconversion were determined by complement fixation or enzyme immunoassays. In all cases, the details of the circumcision procedures were reviewed. When possible, the *mohel* was tested for virus shedding in saliva and for HSV serostatus. Tests were performed separately in the diagnostic virology laboratory routinely used by each hospital.

RESULTS

Eight neonates with documented genital HSV-1 infections were identified (Table 1). In all cases, the *mohel* had performed the ancient custom of oral *metzitzah*. On 2 occasions, the same *mohel* performed the circumcision in 2 different infants: patients 1 and 4 (an interval of 5 years) and patients 7 and 8 (an interval of 5 weeks).

The mean birth weight was 3220 g (standard deviation: ± 696). The circumcision of patient 1 was delayed because he was born prematurely, and the circumcision of patient 3 was delayed because of suspected sepsis. All other infants were circumcised on day 8 of life. The mean interval from circumcision to HSV clinical infection was 7.25 ± 2.5 days. On admission, all infants were reported to have poor appetite. Mean temperature was $37.8 \pm 0.8^\circ\text{C}$. Vesicular rash over the scrotum and penis was noted in all patients (Fig 1). In 5 patients, Tzanck preparation was performed and showed cellular changes consistent with herpesvirus; in 1 case, herpes group virus was detected by negative contrast electron microscopy. HSV was isolated from all patients. None of the mothers had clinical evidence of oral or vaginal herpes. Most infants and their mothers were seronegative for HSV; only patient 7 and his mother were seropositive with a titer of 1:16. In patient 7, a cerebrospinal fluid sample was positive for HSV-1 by polymerase chain reaction, consistent with the clinical course and radiologic diagnosis of HSV encephalitis.

Six infants were admitted to the hospital and received intravenous acyclovir therapy. Four infants had recurrent episodes of genital herpes, and 1 infant presented with severe encephalitis followed by long-term neurologic sequelae, including seizures. Only four *mohels* could be tested, and they were found to

be seropositive for HSV; the mouth cultures obtained from *mohels* all were negative for HSV.

DISCUSSION

In the 8 infants presented here, the association between genital HSV-1 infection and the performance of the ancient procedure of oral *metzitzah* during the circumcision is strongly suggested on the basis of the following criteria: exclusive genital distribution of the lesions, timing of their appearance (4–11 days after circumcision), isolation of HSV-1, absence of HSV exposure in mothers (based on both clinical observation and negative serology in most of the mothers), and absence of clinical signs and symptoms consistent with HSV infections among family members. Furthermore, although the oral *metzitzah* is performed by only a minority of *mohels*, all infants described here underwent this procedure.

On 2 occasions, patients 1 and 4 and patients 7 and 8, the circumcision was performed in different infants by the same *mohel*. On 1 occasion, the interval between the 2 cases was 5 weeks; in the other, 5 years. In previous reports, HSV-1 genital infection occurred in 2 infants who were circumcised by the same *mohel* 10 years apart.^{10,11} Because HSV-1 can be secreted intermittently in saliva for several days to weeks, it is likely that other infants were infected. We suspect, therefore, that this entity is underreported for cultural reasons and that the studies described here are only the "tip of the iceberg" of the true incidence of the disease.

Because in every case the *mohel* had removed the blood by mouth after cutting the foreskin, it was most likely that the infection was transmitted directly from this oral or salivary contact. All of the *mohels* who consented to be tested were seropositive. The likelihood of other sources for an HSV-1 infection in the area of the glans penis in the region of the wound of the circumcision is minimal.¹² Because shedding of HSV-1 in the saliva of both symptomatic and asymptomatic individuals has been documented repeatedly, the act of *metzitzah* represents a potential source of orogenital transmission to the nonimmune infant whose skin integrity was disrupted by circumcision, especially if the infant is seronegative for HSV.

The genital infection in 7 patients remained localized, but patient 7 developed HSV encephalitis followed by long-term brain damage manifested by seizures and infantile spasms. Four infants experienced recurrent episodes of genital herpes simplex and received long-term prophylaxis with oral acyclovir.

In the past, reports of HSV genital infections after circumcision have been relatively rare, not withstanding the high frequency of active herpes labialis among the population, which would include the *mohels*.¹⁰ This may be accounted for in part by the observation that all but 1 of these cases were seronegative for antibody to HSV, which suggests that seropositive infants might be protected to some degree. Likewise, the practice of oral *metzitzah* is limited to only a small subset of ritual circumcisions. There is, however, the possibility that some previous

TABLE 1. Clinical and Laboratory Findings of 8 Neonates With Genital HSV-1 Infection After Ritual Circumcision Followed by Oral *Metzitzah*

	Patient							
	1997	1994	2000	2002	2001	1999	2002	2001
Admitting hospital	Hadassah Mt. Scopus Jerusalem, Israel	Hospital for Sick Children, Toronto, Canada	Hadassah Ein Karem, Jerusalem, Israel	Soroka University Medical Center, Beer Sheva, Israel	Soroka University Medical Center, Beer Sheva, Israel	Hadassah Ein Karem, Jerusalem, Israel	Sheba Medical Center, Tel Aviv, Israel	Sheba Medical Center, Tel Aviv, Israel
Birth weight (gestational age)	1880 g (35 wk)	3980 g (41 wk)	3170 g (40 wk)	3475 g (40 wk)	3200 g (40 wk)	4100 g (42 wk)	3155 g (40 wk)	2800 g (40 wk)
Age (d) at circumcision	31	8	16	8	8	8	8	8
Days from circumcision at presentation	4	9	4	9	6	11	8	7
Temperature	38.0°C	38.1°C	38.2°C	38.5°C	36.1°C	37.0°C	38.0°C	38.4°C
Serology of mother (infant)	Negative (negative)	Negative (negative)	Negative (negative)	Negative (negative)	Negative (negative)	Negative (negative)	Positive 1:16 (positive 1:16)	Negative (negative)
Serology <i>molitel</i> Initial treatment (acyclovir in mg/kg/day)	N/A† IV acyclovir × 8 d	Positive (>1:64) IV acyclovir × 26 d × 10 d	N/A IV acyclovir × 10 d + PO acyclovir × 14 d	N/A† IV acyclovir × 8 d + PO acyclovir × 2 d	N/A No treatment	Positive No treatment	Positive (1:64)* IV acyclovir × 21 d	Positive (1:64)* IV acyclovir × 14 d
Course and treatment	4 recurrences over 8 mo; treated with PO acyclovir × 4 d	Discharged after 26 d IV acyclovir, no neurologic deficit; treated with local acyclovir ointment	5 episodes over 20 mo, treated with local acyclovir ointment	3 episodes over 7 mo, treated with local acyclovir ointment	Spontaneous recovery	Spontaneous recovery	4 d in intensive care unit as a result of seizures (same dosage of acyclovir)	5 more episodes over 11 mo with positive HSV cultures from lesions; treated with PO-acyclovir × 7–14 d
Long-term outcome	Prophylactic treatment with acyclovir for 1 y, normal development	N/A	Delayed psychomotor development	N/A	>2 mo N/A	> 3 mo N/A	Infantile spasm, hirsutism	Prophylactic treatment with acyclovir, normal development

IV indicates intravenous; PO, oral; N/A, not available.

* Same *molitel* for patients 7 and 8 with an interval of ± 5 weeks.

† Same *molitel* for patients 1 and 4 with an interval of 5 years.



Fig 1. Genital HSV-1 infection after ritual circumcision (from patient 6).

cases were not reported for cultural reasons. To clarify this statement, the cultural background requires elaboration.

According to Biblical law, a male infant should be circumcised at the age of 8 days as a sign of the eternal covenant between God and the Jewish people (Genesis 17:10–14; Leviticus 12:3). According to classical rabbinical interpretation, performance of this religious ritual offers medical advantages, a view upheld by many modern medical authorities, as noted earlier. The Babylonian Talmud (Sabbath 133b), the main rabbinical literature completed in the fifth century of the common era, states that for the sake of the infant, the *mohel* is obliged to perform the *metzitzah*“ so as not to bring on risk.” The nature of the risk was not specified. It specifically states that “this procedure is performed for the sake of the infant’s safety and if a *mohel* does not perform the suction [of the wound], this is deemed dangerous and he is to be dismissed.” To prevent medical complications, the Talmud permits only an experienced and responsible *mohel* to perform the ritual circumcision. The Talmud (Tossefta Sabbath 15:8) was aware of potential medical problems that could arise from ritual circumcision¹³ and in fact provided the first description of hemophilia in the history of medicine, manifested as a familial bleeding disorder that required circumcision to be postponed.¹⁴

In the 19th century, Ignaz Philipp Semmelweis (1818–1865) established the principles of hygiene and disease transmission,¹⁵ after neonatal tuberculosis was documented after circumcision by an infected *mohel*.¹⁶ Since then, most rabbinical authorities modified their approach in response to these findings. Because the Talmudic injunction to perform *metzitzah* did not explicitly stipulate oral suction, >160 years ago, Rabbi Moses Schreiber (Pressburg, 1762–1839), a leading rabbinical authority, ruled that *metzitzah* could be conducted by instrumental suction,¹⁶ a ruling quickly adopted by most rabbinical authorities.¹⁷ Consequently, the great majority of ritual circumcisions are performed today with a sterile device and not by oral suction by the *mohel*. However, some orthodox rabbis have felt threatened by criticism of the old religious customs and strongly

resist any change in the traditional custom of oral *metzitzah*. The cultural process of replacing ancient customs by modern wound care has to be encouraged by a heightened awareness of this potentially life-threatening medical complication.¹⁶

On the basis of our observations, the medicolegal impact of neonatal infection by the *mohel* has to be redefined. Our findings provide evidence that ritual Jewish circumcision with oral *metzitzah* may cause oral–genital transmission of HSV infection, resulting in clinical disease including involvement of the skin, mucous membranes, and HSV encephalitis. Furthermore, oral suction may not only endanger the child but also may expose the *mohel* to human immunodeficiency virus or hepatitis B from infected infants. The same consideration that led the Talmudic sages once to establish the custom of the *metzitzah* for the sake of the infant could now be applied to persuade the *mohel* to use instrumental suction.

Indeed, after our first cases, the Chief Rabbinate of Israel pronounced in 2002 the legitimacy of using instrumental suction in cases in which there is a risk of contagious disease. We support ritual circumcision but without oral *metzitzah*, which might endanger the newborns and is not part of the religious procedure.

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