Neonatal Genital HSV-I After Jewish Circumcision

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Introduction

Neonatal herpes simplex virus (HSV) infection incidence has increased in the past decade to 8 to 60 per 100 000 live births in the United States.¹ In Israel, the current incidence is estimated to be 8.4 per 100 000 live births². In 85% of the cases, the infection is transmitted vertically during the passage in the birth canal, the minority of the cases are acquired intrauterine, and in 10% of the cases, the infection is acquired in the postnatal period, from oral HSV shedding of a caregiver or postcircumcision.³

Ritual circumcision is a common custom in many cultures. In the Jewish tradition, there is an ancient custom of orogenital suctioning of the penile blood (*metzitza*), which is currently done by a minority of traditional circumcisers (*mohels*). Part of the ultraorthodox population adheres to this custom because it is considered glorified, without knowing the hazardous potential of it.

The aim of this study is to describe a cluster of cases of postcircumcision neonatal genital HSV infection in a single medical center serving an ultraorthodox population.

Cases

Five infants with genital herpes simplex virus type 1 (HSV-1) infection after Jewish ritual circumcision were admitted to our hospital for a genital vesicular rash between January 1, 2011 and December 31, 2013. All had a history of direct orogenital suctioning of penile blood (*metzitza*) after cutting the foreskin, and all of them were circumcised by 5 different traditional circumcisers (*mohels*). None of the infants' caregivers had a history of oral or genital herpetic lesions.

Clinical and laboratory data of the infants are shown on Table 1.

The average interval between circumcision and onset of symptoms was 8.5 ± 1.9 days, with a range of 6 to 10 days. In 2 cases, the delivery was performed by caesarian section. In all cases, HSV-1 was identified by polymerase chain reaction (PCR) performed on fluid from the peripheral lesions. In 3 cases, the cerebral spinal fluid (CSF) white blood cells (WBC) count exceeded the normal limits, and in 2 cases, PCR was positive for HSV-1 in the CSF. All infants were treated with intravenous acyclovir for 14 to 21 days as recommended, and were discharged at good clinical condition. Three of the patients received long-term treatment with oral acyclovir for 6 months. At 1-year follow up, all infants were normally developed with no recurrences of herpetic lesions; one of the children suffers from febrile seizures.

Discussion

Herpes simplex infection in newborns is a life-threatening infection with high rates of morbidity and mortality. The US Centers for Disease Control and Prevention reported 11 cases of HSV-1 infection after Jewish ritual circumcision between November 2000 and December 2011; and 2 of the infants died.⁴ The estimated risk of herpes infection after orogenital suction was estimated to be 1 to 4098, which is 3.4 times greater than the risk among male infants not exposed to direct orogenital suction.⁴

A recent study from Israel reported 7 cases of neonatal HSV-1 infection associated with ritual circumcision over a 7-year period. These cases accounted for 32% of all neonatal HSV infections.² A former report from Israel described 8 cases over a 7-year period in 5 large hospitals.⁵

We present a cluster of 5 cases of postcircumcision genital HSV-1 infection in a single medical center, who were admitted over less than a year. No other neonatal HSV infection was seen in our medical center during this time frame. Possible explanations for our cluster can be a rise in the performance of the oral suctioning

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Patient	Gestational Age (Weeks)	Delivery	Birth Weight (kg)	Age at Diagnosis (Days)	Time Interval From Circumcision to Infection (Days)	CSF PCR	CSF WBC/µL	Additional Findings	Treatment
I	41	Vaginal	3.000	17	Few days	Negative	55	_	IV acyclovir × 21 d, oral acyclovir × 6 mo
2	37	Cesarean section	3.572	20	10	Positive	60	Fever, elevated LFT	IV acyclovir × 21 d, oral acyclovir × 6 mo
3	39	Vaginal	3.650	16	8	Negative	0	Fever	IV acyclovir × 14 d
4	41	Cesarean section	2.800	14	6	Positive	20	_	IV acyclovir × 21 d
5	38	Vaginal	3.344	18	10	Negative	7	_	IV acyclovir × 14 d, oral acyclovir × 6 mo

Table 1. Clinical characteristics of infants with post-circumcision neonatal HSV-1 infection.

Abbreviations: CSF, cerebrospinal fluid; PCR, polymerase chain reaction; WBC, white blood cells; IV, intravenous; LFT, liver function tests.

custom in Jerusalem orthodox community. More than 90% of adults have antibodies to HSV-1 by the fifth decade of life and the seroprevalence is affected by socioeconomic status. Thus, another explanation for our cluster can be a higher HSV-1 oral carriage among the *mohels*.

It should be emphasized that the cause-effect relationship between the metzitza and the following herpetic infection was not fully proven in any of the cases. The only scientific method to prove this is isolation of the virus from the mohel's oral secretions and comparing the sequence of the isolate with the one of the babies. None of this was performed in any of the described cases. Serological testing of the baby, the immediate family members, and the mohel may shed some light, but in most cases will not be informative enough. Hence, one cannot exclude the possibility that the infection was not acquired either vertically or from other caregivers. Nevertheless, the fact that the infection was observed only in the genital area of male infants undergoing ritual metzitza including 2 who were born by Caesarian section, the well-defined interval from circumcision to clinical signs, and that the isolate in all 5 cases was HSV type 1, strongly advocate the metzitza as the source of infection.

Untreated neonatal herpes infection undergoes dissemination or develops to central nervous system (CNS) infection in more than 70% of cases. The mortality rate is 65% of untreated neonatal herpes infections. Only 20% of CNS infection will develop normally.

Surprisingly, all 5 cases, including the children with CNS involvement, had benign course of disease and no long-term severe sequelae. This can be due to increased awareness and early diagnosis or possibly due to lower viral load compared with the viral load in birth canal transmission. This assumption can be supported by a previous report of 2 neonates with cutaneous genital infection that recovered without any antiviral treatment.⁵

None of the infants had cutaneous recurrences at 1-year follow-up. This is in contrast to a previous report in which 4 out of 8 children with neonatal post circumcision HSV-1 infection had cutaneous recurrences.⁵ It is unknown whether early diagnosis and treatment reduces the cutaneous recurrences, and if this can be the explanation in our cases. Another explanation for the low rate of cutaneous recurrences can be the administration of oral acyclovir for 6 months in 3 of our infants as recently recommended by Kimberlin et al.⁶ One of the infants with CNS involvement developed febrile seizures after the age of 6 months. It is unknown whether these are related to the perinatal herpes CNS infection.

In summary, neonatal HSV infection carries a risk for substantial morbidity and mortality, and can be completely prevented by avoiding oral suctioning during circumcision (eg, using a glass tube). The general population should be aware of the disease and the simple way to prevent it.

Author Contributions

ST-B designed the study and drafted the initial manuscript. MB-M and YS reviewed and revised the manuscript. OM supervised data collection, designed the study, reviewed and revised the manuscript. All authors treated the patients presented in the manuscript.

Declaration of Conflicting Interests

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References

- Corey L, Wald A. Maternal and neonatal herpes simplex virus infection. N Engl J Med. 2009;361:1376-1385.
- Koren A, Tasher D, Stein M, Yossepowitch O, Somekh E. Neonatal herpes simplex virus infection in Israel. *Pediatr Infect Dis J.* 2013;32:120-123.
- Fidler KJ, Pierce CM, Cubitt WD, Novelli V, Peters MJ. Neonatal disseminated herpes simplex virus infections be treated earlier? *J Infect*. 2004;49:141-146.
- Centers for Disease Control and Prevention. Neonatal herpes simplex virus infection following Jewish ritual circumcisions that included direct orogenital suction—New York City, 2000-2011. *MMWR Morb Mortal Wkly Rep.* 2012;61:405-409.
- Gesundheit B, Grisaru-Soen G, Greenberg D, et al. Neonatal genital herpes simplex virus type 1 infection after Jewish ritual circumcision: modern medicine and religious tradition. *Pediatrics*. 2004;114:e259-e263.
- Kimberlin DW, Whitley RJ, Wan W, et al; National Institute of Allergy and Infectious Diseases Collaborative Antiviral Study Group. Oral acyclovir suppression and neurodevelopment after neonatal herpes. *N Engl J Med.* 2011;365:1284-1292.