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Retinal hemorrhage in a neonate with congenital diaphragmatic hernia

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Abstract: We present the case of birth-related retinal hemorrhage in a full-term female infant. The baby was born via cesarean section and showed left-sided congenital diaphragmatic hernia (CDH). High-frequency oscillation was initiated on the first day of life for management of pulmonary hypertension secondary to CDH. Hernia repair was performed 3 days after birth. Postoperatively, the pulmonary hypertension gradually resolved. On day 8after birth, an ophthalmoscopic examination revealed multiple oval retinal hemorrhages in the right eye. Additionally, several hemorrhages contained white-centers (Roth spots). The left eye, however, showed no abnormalities. During the 2-week follow-up period, the retinal hemorrhages gradually disappeared. We believe that the relationship between retinal hemorrhages and CDH is coincidental rather than causative in this patient.
Keywords: Retinal hemorrhage, Roth spot, congenital diaphragmatic hernia, pulmonary hypertension.

INTRODUCTION

It is well known that retinal hemorrhages occur in neonates during delivery. While the reported incidence varies from 2.6 to 50% [1-4], it is known to vary with the mode of delivery. They occur most frequently in babies delivered by vacuum extraction (75%),followed by 33% by spontaneous vaginal delivery, and 6.7% by cesarean section [2].The hemorrhage type varies including splinter-shaped, flame-shaped, dot and blot hemorrhage, and the rare sub retinal and pre retinal type[1-4].

Congenital diaphragmatic hernia (CDH) is largely due to the presence of secondary pulmonary hypoplasia and other non-pulmonary malformations and chromosomal defects [5]. Ophthalmologic findings with isolated CDH are extremely rare [5, 6]. Herein, we report aneonate with CDH who presented with a unique birth-related retinal hemorrhage.

CASE REPORT

A full-term female neonate weighing 2.94kg was referred to us for an ophthalmological examination. The baby was born via a cesarean section to a 33-year-old, gravida1 para 1, Japanese woman with negative serology following an uncomplicated pregnancy. The baby showed left-sided CD Hon prenatal ultrasonography. High-frequency oscillation (HFO) was initiated on the first day of life for management of pulmonary hypertension secondary to CDH. Hernia repair was performed 3 days after the birth. Postoperatively, the pulmonary hypertension gradually resolved. Eight days after the birth, a dilated funduscopic examination was performed and digital color photographs were taken using the Ret Cam 120 (Massie Research Laboratories, Inc., Dublin, CA). Ophthalmoscopic examination revealed multiple oval retinal hemorrhages in the right eye (Figure A). Additionally, several hemorrhages contained white-centers (Roth spots; Figure A, arrows).However, no abnormalities were found in the left eye (Figure B).There was no facial dysmorphism or obvious external eye abnormality. Family history was negative for consanguinity, eye disease, and genetic disorders. The HFO was continued for 13 days. During the 2-week follow-up period, the retinal hemorrhages gradually disappeared.

DISCUSSION

The reported incidence of retinal hemorrhage in newborns varies from 2.6 to 50% [1-4]. In the largest series of retinal hemorrhage in newborns, Li *et al.;* [4] evaluated 3,573 healthy full-term newborns that underwent eye examination. According to their report, 769 (21.5%) had retinalhemorrhages. In other prospective studies, Emerson *et al.;* [2] reported that retinal hemorrhage was found in 33.6% (50/149) of newborns. Of these, 26 (52%) showed bilateral hemorrhaging. Hughes *et al.;* [3] reported that retinal hemorrhage was found in 34% (18/53) of newborns, of which, 14 were bilateral. Furthermore, Emerson *et al.;* [2] reported that white-centered hemorrhage was commonly noted (18/76eyes; 24%). In contrast, Hughes *et al.;* [3] described that white-centered hemorrhages were present in only 5.7% (3/53) of the cases.

Roth spots were classically described as septic emboli lodged in the retina of patients with sub acute bacterial endocarditis [7]. More recent histological evidence suggests, however, that they are not foci of

bacterial abscess. Instead, they are nonspecific and may be found in other diseases as well [7]. Roth spots may result from the rupture of retinal capillaries and the extrusion of whole blood. In addition, elevated venous pressure also lead to retinal hemorrhages. Typically, elevated venous pressureis the most common predisposing factor for retinal hemorrhages in neonatal birth trauma, traumatic deliveries in mothers, battered baby syndrome, and intracranial hemorrhages. We speculate that unstable circulatory control secondary to CDH may have exacerbated the white-centered retinal hemorrhage.

In general, there is agreement in the literature that birth-related retinal hemorrhages resolve quickly. Emerson *et al.;* [2] reported that approximately 90% of intra retinal hemorrhages detected at birth resolved within 2 weeks, and none were detectable by4 weeks after birth. Consistent with previous reports, in our patient too, retinal hemorrhages gradually disappeared during the first 2 weeks.

CDH is associated with a wide range of other malformations [5]. Associated malformations may occur with trisomies 13, 18, and 21, non-trisomy chromosomal disorders, or recognizable non-chromosomal syndromes. The most common associated anomalies are cardiovascular and neural tube defects [5]. However, ophthalmologic findings with isolated CDH are extremely rare [5, 6]. Kumar *et al.;* [5] reported bilateral cataracts, retinal detachment, and vitreous hemorrhage in a newborn with CDH. In addition, asymmetrical retinopathy with intra retinal hemorrhages has been reported in patients who underwent extracorporeal membrane oxygenation for CDH [6]. However, ophthalmologic findings in our patient were not consistent with those seen in previous reports. Therefore, we believe that the association between retinal hemorrhages and CDH is coincidental rather than causative in this patient. Additional cases are necessary to further characterize birth-related retinal hemorrhage with CDH.

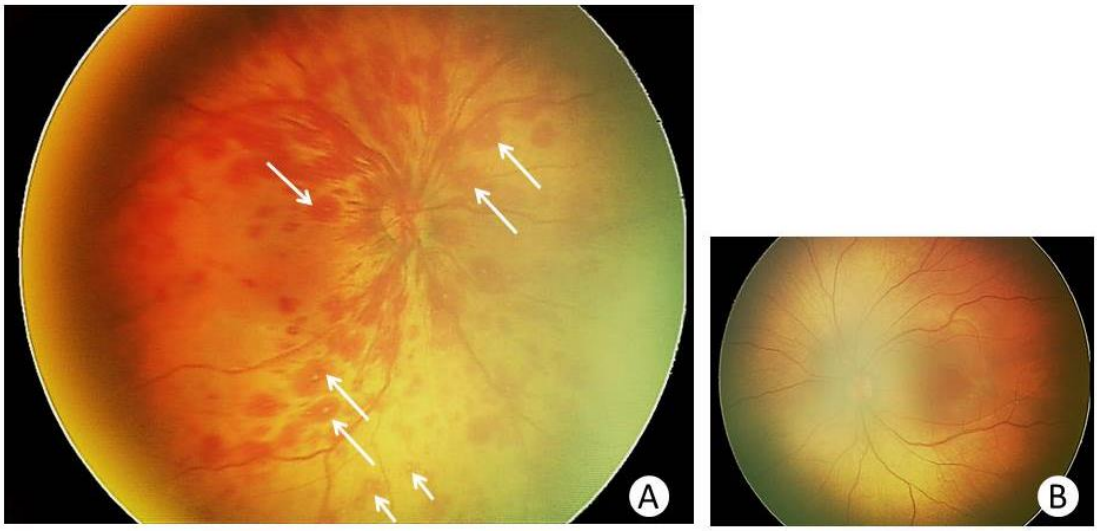

Fig.1 Funduscopic images of the right (A) and left (B) eyes

Note multiple oval retinal hemorrhages in the right eye (A). Several hemorrhages contained white-centers (arrows). No abnormal findings were detected in the left eye (B).

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