Case Report

A case of stillbirth caused by rupture of an intrahepatic hemangioma and the wish for litigation

U. Indraccolo^{1,2,*}, S.R. Indraccolo³, P. Greco⁴, P. Fedeli⁵

¹Maternal-Infantile Department. Section of Obstetrics and Gynecology. Sant'Anna University Hospital of Ferrara – Cona (Italy)

²Complex Operative Unit of Obstetrics and Gynecology. "Alto Tevere" Hospital of Città di Castello. ASL 1 Umbria (Italy)

³Department of Gynecological, Obstetrical, and Urological Sciences. "Sapienza" University of Rome (Italy)

⁴Department of Morphology, Surgery and Experimental Medicine, Section of Obstetric and Gynaecology, University of Ferrara, Ferrara (Italy)

⁵School of Law, Legal medicine. University of Camerino (Italy)

Summary

A very uncommon case of an intrahepatic hemangioma rupture as a cause of a stillborn baby is presented. It seems that the event could not be detected. However, the idea of litigation pushed parents to ascertain hypothetical liability of physicians 10 years after the stillbirth. The reason that parents do not comprehend that such a rare event could occur and opt for litigation was investigated. The conclusion seems that some patients' beliefs do not agree with physician counseling and cause the failure of the medical goals for the best care; thus, an improvement in counseling would still be ineffective. Rather, lowering parents' expectations from medical assessments and care seems the right way to reduce the incidence of litigation.

Key words: Fetal intrahepatic hemangioma; Stillbirth; Litigation.

Introduction

Obstetric-related uncertainty in the management of rare diseases is still a concern for obstetricians. Unfavorable outcomes after pregnancy and birth as reported by the parents trigger litigation against obstetricians because unfavorable outcomes are inconceivable in non-medical people's beliefs.

The following situation described in this case report was faced by two of the authors because medico-legal consultation was requested of them by the parents with the aim of evaluating the likelihood of claiming litigation for malpractice.

Case

The case events were reconstructed by reading medical charts as provided by the parents 10 years after the birth of their child. No signs of labor were found. In the medical chart, it was reported that the second trimester second level ultrasound was normal. The first trimester ultrasound was appropriate for the date (basing on crown-rump length measurements) with a nuchal translucency of 2.5 mm at 10 weeks and six days of gestational age. The combined test for Down syndrome screening resulted in a risk of 1: 1891 (with nuchal translucency 2.2 mm at 12 weeks and one day of gestation). The pregnancy was uneventful up to that point. However, the third trimester ultrasound detected an IUGR of three weeks. On the obstetric ultrasound performed at 35 weeks and five days of gestation, it was reported that the growth was 34/35 weeks old. Femoral length

was 65 mm (5° percentile), a finding that confirmed mild IUGR. Cardiac activity was regular, and kidneys, cerebral ventricles, bladder, and stomach were displayed and appeared normal. No pathological findings were found. The fetal flow in umbilical artery was found to be 1.18. Seven cardiotocographic (CTG) traces appeared normal. A mild decrease in platelets without any signs of acute platelet consumption was found.

The patients was discharged with a diagnosis of mild IUGR. A check-up for the 38th week was scheduled.

The patient was re-admitted about one month after the previous discharge. A diagnosis of symmetrical IUGR at 40 weeks and two days of gestation was made. The ultrasound performed upon admission confirmed the presence of an IUGR, and a diagnosis of oligohydramnios was also made. The standard CTG was normal and satisfied the Dawes-Redman criteria. No signs of entering labor were detected. Labor induction was initiated by applying the dinoprostone 10 mg vaginal delivery system. A cardiotocographic check was cautiously and continuously performed for many hours after labor induction, and evidence of fetal heart activity was detected. The last CTG record depicted mild reduction in variability. It was stopped and repeated several hours later. However, some hours after the end of the last cardiotocographic monitoring, the ultrasound examination did not detect fetal heart activity.

Labor began via induction and ended with the delivery of a female baby, who weighed 2670 g in which a wide hematoma of the cord (10 cm in length) was detected. It

originated from the abdomen across the navel.

The rupture of a hepatic angioma caused severe hemorrhaging and was determined as the cause of death after pathological examination. A cavernous angioma of the right lobe of the fetal liver in the anterior and inferior portion was detected. The estimated blood during the glissonian approach was about 200 mL, and it extended to the umbilical cord as previously described. A peritoneal serous and hematic ascites was also found. Malformations or abnormal patterns on the placenta were not found.

From a medico-legal point of view and to answer question: "Was it appropriate to induce labor at all or to induce labor at that time?" The answer seems to be that it was appropriate even in 2008. We currently know that mild IUGR should be managed conservatively with the aim of avoiding unnecessary cesarean sections by performing labor induction [1]. Labor induction was also correct in light of the oligohydramnios. Basing on the final weight of the baby (2670 g), it could be confirmed that the IUGR was very mild.

To answer the question: "Could fetal death be prevented with an immediate intervention?" The answer seems to be yes. Immediate intervention in the case of the pathological CTG trace prevented in-utero loss of the baby in the case reported by Morimura [2]. However, in the case presented here, the last CTG record did not imply immediate delivery as seen in the case of Morimura et al. [2].

To answer the question: "Could the hepatic angioma be detected before rupture?" The answer seems to be yes, but detecting a fetal hepatic angioma is not necessarily an indication to change the method of labor management. Usually, hepatic angiomas are followed-up because of the rapid growth and the shunt effect they can cause [2, 3]. Additionally, in Italy, some obstetricians switched from the second level second trimester echograph to a sonographic scan, which is more accurate than the one used as screening for fetal malformations and performed in agreement with Italian guidelines for sonographic examinations during pregnancy. Therefore, at the time of the second trimester sonographic scan, no malformations were detected. Moreover, viewing the images of abdominal circumferences collected at 35 weeks and five days, no sign of hepatic angioma appeared. Therefore, the onset of the baby's hepatic angioma would have occurred few days before the end of the gestation.

To answer the question: "Could an immediate intervention for the hepatic angioma have been successful?" The answer seems to be no. The literature does not seem to support the immediate operation on the hepatic angioma aiming to treat successfully the disease [2, 4, 5]. Moreover, even in case of the delivery of a live fetus, emergency pediatric surgery could not be performed in the hospital in which the case occurred because that particular hospital did not have a pediatric surgery unit.

Discussion

By digitizing the PubMed search engine, "hepatic" AND "fetal" AND "angioma" AND "hemorrhage" (September 24, 2018), we found six items. Among them, one item did not address fetal intrauterine diagnosis. The remaining five items [2, 4–7] related epidemiology [6], pediatric surgeries [4, 5], intrauterine diagnosis [7], and peripartum management of an intra-tumoral hemorrhage [2]. The latter case [2] has had an unfavorable prognosis because after obstetricians diagnosed an angioma, they were also able detect a pathological CTG and then proceeded with a cesarean section. However, due to disseminated intravascular coagulation, the baby could not be treated surgically and died several days after the delivery. Even if surgery could be performed (removing the hepatic tumor) in newborns, uncertain outcomes were reported by the Authors [4].

The liability of obstetricians and gynecologists in the case presented in this paper could not be determined by assessing the medical charts. In previous reports [8, 9], even from a medico-legal point of view, we had already discussed the expectations of parents after ultrasound fetal examinations by speculating on the difficulties involved in counseling in cases of obstetricians' uncertainty [9]. In light of the rising litigation rates, the lack of communication between obstetricians and parents on the advantages of fetal echography cannot be the only reason for disappointment. Parents do not understand why an echograph does not guarantee favorable outcomes. In Italy, a scan is provided for free, but it is expensive when someone wants to refer to the experts.

Conclusion

To the best of our knowledge, the case presented here seems to be the first one of a stillbirth caused by acute hemorrhage secondary to the rupture of a fetal hepatic hemangioma. Angioma was difficult to diagnose in utero. Additionally, it seems that in the event of in utero rupture, it would still have been very difficult to be detected in a timely manner in order to prevent fetal death. Moreover, treatment was impossible. Despite those conditions, such a rare event has generated the desire for litigation since 2008. The wish for litigation could not have been prevented by just an improvement in counseling. Patients' beliefs do not always agree with physician counseling and can causes the failure of the medical goals of the best care [10, 11]. Concerning the sonographic screening of fetal malformations, it seems mandatory to lower the parents' expectations of sonographic fetal examinations rather than improving counseling on the advantages of sonographic fetal examinations.

This paper provides an English edited version of an already published article of same case, reported by same authors [12]. English editing was done for the best of intelligibility.

Ethics Approval and Consent to Participate

Authors have been received consent for reporting the case.

Conflict of Interest

The authors declare no competing interests.

Submitted: November 09, 2018 Accepted: November 29, 2018 Published: June 15, 2020

References

- www.sigo.it/wp-content/uploads/2016/03/
 Induzione-al-Travaglio-di-Parto.pdf (24 Sep 2018).
- [2] Morimura Y., Fujimori K., Ishida T., Ito A., Momura Y., Sato A.: "Fetal hepatic hemangioma representing non-reassuring pattern in fetal heart rate monitoring". J. Obstet. Gynaecol. Res., 2003, 29, 347
- [3] Gembruch U., Baschat A.A., Gloeckner-Hoffmann K., Gortner L., Germer U.: "Prenatal diagnosis and management of fetuses with liver hemangiomata". *Ultrasound. Obstet. Gynecol.*, 2002, 19, 454.
- [4] Walton J.M., Rubin S.Z., Soucy P., Benzie R., Ash K., Nimrod C.: "Fetal tumors associated with hydrops: the role of the pediatric surgeon". J. Pediatr. Surg., 1993, 28, 1151.
- [5] Tawes R.L. Jr., Nelson J.A., Hyde J.A. Jr.: "Hepatic hemangioma: successful resection in a neonate". *Surgery.*, 1971, 70, 782.

- [6] Isaacs. H. Jr.: "Fetal and neonatal hepatic tumors". J. Pediatr. Surg., 2007, 42, 1797.
- [7] Aslan H., Dural O., Yildirim G., Acar D.K.: "Prenatal diagnosis of a liver cavernous hemangioma". Fetal. Pediatr. Pathol., 2013, 32, 341
- [8] Indraccolo U., Indraccolo S.R., Fedeli P.: "Another Case of de novo 3q26.33q27.3 microdeletion and its medicolegal sequel". Case. Rep. Obstet. Gynecol., 2018, 2018, 1909056.
- [9] Indraccolo U., Scutiero G., Greco P.: "Correction of the sensitivity reported in the 2015 Italian Society of Obstetrical and Gynecological Ecography (SIEOG) guidelines for the ecographic screening of fetal malformations: a meta-analytic approach". Clin. Exp. Obstet. Gynecol., 2018, 45, 651.
- [10] Indraccolo U., Scutiero G., Matteo M., Indraccolo S.R., Greco P.: "Cesarean section on maternal request: should it be formally prohibited in Italy?" *Ann. Ist. Sup. Sanita.*, 2015, 51, 162.
- [11] Indraccolo U.: "Punches and knocks to the physicians: choosing wisely or self-protection?" *Recenti. Prog. Med.*, 2016, 107, 607.
- [12] Indraccolo U., Indraccolo S.R., Greco P., Fedeli P.: "A case of still-birth for rupture of intrahepatic hemangioma and the wish of litigation". Clin. Exp. Obstet. Gynecol., 2020, 47, 86.

Corresponding Author: UGO INDRACCOLO, M.D., PhD Via P. Veronese 2/c 06024 Gubbio (PG) (Italy) e-mail: ugo.indraccolo@libero.it