Percutaneous Embolisation of a Giant Placental Chorioangioma with n-Butyl-2-Cyanoacrylate: a Case Report and Review of the Literature

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Abstract

Giantplacental chorioangiomas (CA) are benign tumors usually associated with unfavorable fetal outcome. The management of symptomatic tumors depends mainly on gestational age and fetal symptoms. Intervention aims to block the vascular supply to the tumor. Various methods have been described such as fetoscopic or interstitial laser coagulation, intratumoral alcohol injection, endoscopic ligation, and embolisation. We present a case of a large CA treated by tissue glue (n-Butyl-2-Cyanoacrylate) injection into the feeding vessel. In a 31 yearold woman at 22 weeks of gestation, routine ultrasound examination showed a solid, giant hypoechogenic tumor arising from the right side of an anterior placenta, and protruding into the amniotic cavity. The tumor was highly vascularized, and supplied by a large feeding artery and vein with pulsatile blood flow on Doppler investigation. There was polyhydramnion with a deep vertical pocket of 10.4 cm. No signs of fetal anemia or hydrops were observed. The embolisation was performed under ultrasound guidance. A 20-gauge needle was inserted into the arterial feeding vessel, flushed with 5% glucose solution, followed by the injection of a mixture of n-Butyl-2-Cyanoacrylate diluted with lipiodol. Immediate interruption of the vascular supply was observed. Subsequently, the patient was discharged without any complications and delivered spontaneously a healthy newborn at 37 2/7 weeks of gestation.

Our case emphasizes the potential of percutaneous embolisation of CA as a minimal invasive procedure compared to laser coagulation and a further option in cases of CA with large feeding vessels.

Keywords: Chorioangioma; n-Butyl-2-Cyanoacrylate; Embolization; Laser; Placenta
1. Introduction

Placental Chorioangioma (CA) is a benign tumor that represents, with an incidence of 1%, the most common tumor of the placenta. In the majority of cases it is less than 4 cm in size and is located close to the cord insertion of the placenta (Gajewska et al., 2010). These small CA have no or minor hemodynamic implications for the fetus. In rare cases, the CA can grow rapidly, causing fetal hemodynamic and circulatory alterations due to the presence of arteriovenous shunts within the tumor. This is particularly true for tumors greater than 4 cm (García-Díaz et al., 2012). We present a case of a successful prenatal percutaneous embolisation of a large placental CA using cyanoacrylate tissue glue.

2. Case Report

A 31-year-old woman was referred at 22 weeks of gestation for fetoscopic laser treatment of a large CA. Ultrasound examination showed a solid, hypoechogenic tumor of 62x44x56mm (80mL) arising from the right side of the placenta, located on the anterior wall, and protruding into the amniotic cavity. The tumor was highly vascularized, supplied by a large feeding artery (5mm diameter) and a vein (6.5mm diameter) with pulsatile blood flow (Figure 1).

The umbilical cord insertion (UCI) was next to the tumor. There was a polyhydramnion with a 10.4 cm deep amniotic fluid pocket. The highest peak systolic velocity (PSV) of the middle cerebral artery (MCA) was 33.6cm/s (<1.5 MoM) and no signs of fetal heart failure was present. Due to the large feeding artery of the CA and after extensive counseling a percutaneous embolisation of the tumor was proposed. The procedure was performed under regional anesthesia and ultrasound guidance (Voluson 730 – GE Medical Systems). A 20-gauge Chiba needle (15 cm in length, Kimberly-Clark, Zaventem, Belgium) was inserted into the arterial feeding vessel and the inner stylet was
removed and flushed with a 5% glucose solution, followed by two subsequent injections of n-Butyl-2-Cyanoacrylate (Histoacryl, Braun Melsungen AG, Melsungen, Germany) 1:4, diluted with lipiodol (Lipiodol Ultra Fluid, Guerbet, Roissy, France). The total volume of the injected mixture was 2.9 ml. Immediate interruption of the vascular supply was observed.

Pulsatility index of the ductus venosus increased rapidly soon after embolisation. MCA PSV increased from 33.6 cm/s (1.2 MoM) to 54.8 cm/s (1.96 MoM). However, no cordocentesis for diagnosis of fetal anemia was performed as the MCA PSV returned spontaneously to values below 1.5 MoM within a few days (Figure 2).

**Fig. 2.** Measurement of fetal peak systolic velocity of the middle cerebral artery before and after the injection of n-Butyl-2-Cyanoacrylate. Abbreviations: MCA PSV= Peak Systolic Velocity in the Middle Cerebral Artery.

Both volume and appearance of the tumor changed after the procedure (Figure 3 and 4).
The patient was discharged on day 2 after procedure. Pregnancy was uneventful and induction of labor was performed at 37 2/7 weeks gestation for gestational hypertension. The patient delivered a healthy newborn of 2590g (18th percentile) with a 5’ Apgar score of 9. Pathological examination of the placenta (600 g) confirmed the presence of a 6cm CA with necrotic changes.

In table 1 we summarized a literature review of chorioangiomas treated by ultrasound guided transcaneous embolisation during pregnancy.
Table 1 Literature review of chorioangiomas treated by ultrasound guided transcutaneous embolisation during pregnancy.

<table>
<thead>
<tr>
<th>Autor, year</th>
<th>Cases (n)</th>
<th>Material used</th>
<th>Gestational age at surgery (weeks)</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nicolini, 1999</td>
<td>2</td>
<td>Alcohol</td>
<td>27; 25</td>
<td>Survived</td>
</tr>
<tr>
<td>Jauniaux and Ogle, 2000</td>
<td>1</td>
<td>Alcohol</td>
<td>32</td>
<td>Neonatal death</td>
</tr>
<tr>
<td>Wanapirak, 2002</td>
<td>1</td>
<td>Alcohol</td>
<td>27</td>
<td>Survived</td>
</tr>
<tr>
<td>Lau, 2003</td>
<td>1</td>
<td>Microcoil</td>
<td>25</td>
<td>Neonatal death</td>
</tr>
<tr>
<td>Sepúlveda, 2003</td>
<td>1</td>
<td>Alcohol</td>
<td>26</td>
<td>Fetal death</td>
</tr>
<tr>
<td>Perrotin, 2004</td>
<td>1</td>
<td>Tissue glue (n-Butyl-2-Cyanoacrylate)</td>
<td>27</td>
<td>Survived</td>
</tr>
<tr>
<td>Lau, 2005</td>
<td>1</td>
<td>Tissue glue (Enbucrilate)</td>
<td>24</td>
<td>Neonatal death</td>
</tr>
<tr>
<td>Deren, 2007</td>
<td>1</td>
<td>Alcohol</td>
<td>26</td>
<td>Survived</td>
</tr>
<tr>
<td>Gajewska, 2010</td>
<td>1</td>
<td>Tissue glue (n-Butyl-2-Cyanoacrylate)</td>
<td>23</td>
<td>Survived</td>
</tr>
<tr>
<td>Haddad, 2010</td>
<td>2</td>
<td>Tissue glue (n-Butyl-2-Cyanoacrylate)</td>
<td>27; 29</td>
<td>Survived</td>
</tr>
<tr>
<td>Babic, 2012</td>
<td>1</td>
<td>Tissue glue (n-Butyl-2-Cyanoacrylate)</td>
<td>22</td>
<td>Survived</td>
</tr>
<tr>
<td>Present report</td>
<td>1</td>
<td>Tissue glue (n-Butyl-2-Cyanoacrylate)</td>
<td>22</td>
<td>Survived</td>
</tr>
</tbody>
</table>

We searched literature published in PubMed between 1978 and 2014. The terms searched without language restriction were: chorioangioma, embolisation, placental chorioangioma, giant chorioangioma, and management of chorioangiomas, and we focused on studies/cases showing the outcome of pregnancy after embolisation. Each article was carefully evaluated and any pertinent references from the manuscripts where obtained and reviewed. A total of 14 pregnancies were treated with either alcohol (n=6), tissue glue (n=7) or microcoil (n=1), resulting in 10 (71%) neonatal survival.

3. Discussion

In 50% of the cases of large placental CA severe fetal complications may be observed, such as hydrops due to congestive heart failure, anemia and other hematologic disorders (e.g. thrombocytopenia), polyhydramnios, intrauterine growth restriction, preterm delivery and fetal death (García-Díaz et al., 2012). However, since the first prenatally diagnosed case by Asokan et al. (1978), only limited interventions have been proposed and performed for large, clinically significant placental CA. For example, poor results were observed after amniolal and fetal death. Invasive procedures such as endoscopic suture of the vessels (Mendez-Figueroa et al., 2009), fetoscopic laser coagulation (Bermudez et al., 2007), interstitial laser coagulation (Zanardini et al., 2010), alcoholic ablation
(Nicolini et al., 1999; Jauniaux et al., 2000; Wanapirak et al., 2002; Sepulveda et al., 2003; Deren et al., 2007), microcoil (Lau et al., 2003) and tissue glue injection (Gajewska et al., 2010; Perrotin et al., 2004; Lau et al., 2005; Haddad et al., 2010; Babic et al., 2012) were performed. However, perinatal mortality after the above-reported procedures remains high.

Here, we report another successful prenatal percutaneous embolisation of a placental CA using cyanoacrylate tissue glue. This technique, compared with the above-described endoscopic methods, is a less invasive procedure that can be performed under local anesthesia. After identification of the feeding vessel by ultrasound, a needle is inserted into the vessel and the injected glue causes devascularisation of the CA. This substance is widely used by interventional radiologists for embolisation with good results. We report the fourth successfully antenatal treated case by using cyanoacrylate tissue glue. As reported in table 1 all patients that underwent a percutaneous embolisation using n-Butyl-2-Cyanoacrylate presented successful results with good perinatal outcomes. Also in our case we opted for percutaneous embolisation with n-Butyl-2-Cyanoacrylate, as this technique is easier, less invasive, and less dangerous for the pregnancy. In addition, a laser procedure would have been more challenging, because the CA was adjacent to the umbilical cord insertion, the placenta was anterior, and the risk of bleeding from the large feeding vessel during coagulation is higher. Immediately after treatment, we observed a temporary increase of the MCA PSV that resolved spontaneously. This can be explained by a temporal redistribution of fetal-placental blood volume.

4. Conclusion

Our case emphasizes the potential of percutaneous embolisation of CA as a minimal invasive procedure. It can be an alternative to laser in particular in cases where the localization of CA is unfavorable and large feeding vessels are present.

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