PLANCENTAL HAEMANGIOMA ASSOCIATED WITH ACUTE FETAL ANEMIA IN LABOUR

A.M.C. FRANCA-MARTINS, Z. GRAUBARD, G.A. HOLLOWAY, F.J. VAN DER MERWE
J.G. Strijdom Hospital, Department of Obstetrcs and Gynaecology, University of the Witwatersrand, Johanesburg, South Africa.

SUMMARY

A case of pregnancy complicated by a placental hemangioma is presented. This was unassociated with any of the complications commonly coexistant with this tumor, and presented with fetal anemia due to a fetomaternal transfusion.

RESUMO

Hemangioma da placenta associado a anemia fetal aguda durante o trabalho de parto

Descreve-se um caso de uma gravidez a termo, complicada por um hemangioma da placenta. As complicações mais frequentes devidas a este tipo de tumor não se observam neste caso que, se apresentou com um quadro de anemia fetal aguda devido a transfusão feto-maternal. Discute-se a incidência, tipos histológicos, etiologia e apresentação clínica deste tipo de tumor. Alerta-se para a possibilidade da frequência dos hemangiomas da placenta ser maior, escapando ao diagnóstico do obstetra bem como a transfusão feto-materna ser um facto mais comum.

INTRODUÇÃO

A 22 year old white primigravida female, blood group A Rh positive wo was 37 weeks pregnant by all parameters, was admitted to our antenatal ward with moderate elevation of her blood pressure. She had had, up to the date of admission, a pregnancy that was complicated only by sporadic elevation of her blood pressure,without proteinuria or ther features of pregnancy induced hypertention. Because of a clinical impression that mildpolyhydramnios was present, an abdominal ultrasound was performed. This revealed a large mass of 4 cm × 7 cm in diameter, attached to the placenta. The polyhydramnios was not confirmed, and a normal single fetus was seen. Noemal fetal heart beat and fetal movements were observed.

A non-stress test was performed, which was nonreactive, and therefore an oxytocin contraction test was done. This revealed a normal fetal heart baseline, borderline (+/— 5 bpm) short term variability, and regular moderate late decelerations in response to uterine contractions. This was regarded as positive by the criteria of Freeman [1].

In view of these findings, labour was induced using an infusion of oxytocin in titrated dosage starting at 1 m i.u. per minute. The fetal heart tracing altered with the onset of mild uterine contractions, and a sinusoidal fetal heart pattern developed (Hofmer and Sonnendecker) [2].

A emergency caesarean section was performed, and a 2500 gm, pale non-hydropic female infant with Apgar of 2 and 3 at 1 minute and 5 minutes respectively, was delivered. The intraoperative blood loss was minimal, and care was taken to remove the placenta by cord tranction to minimise the chance of feto-maternal transfusion. The fetal hemoglobin was 4,3 g/dl and the hematocrit 14,0%. The other hematological parameters were: MCV 131 fl MCH 41 mg; MCHC 31 g/dl: 5,9 Erythroblast per 1000 leucocytes; reticulocytes 9,1%, platelets were normal in number and morphology. This was indicative of a normoblastic anemia with no more than an early erythropoetic response. There were no morphological features in the smear to suggest an underlying hemolytic process.

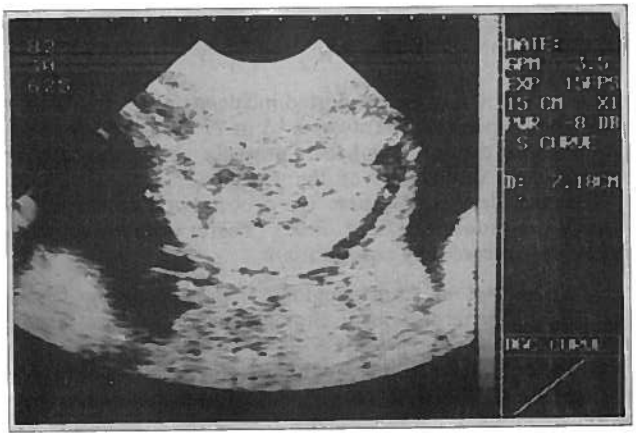

Fig. 1 — Mass seen at ultrasonography to be attached the placenta (Diasonics 100)

A Kleihauer test performed on the maternal blood immediately post-partum was strongly positive for the presence of fetal red cells.

The maternal hematological parameters were otherwise normal.

Recebido para publicação: 8 de Abril de 1987

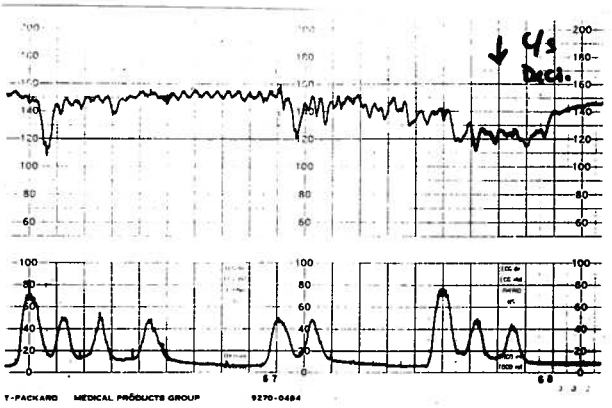

Fig. 2 — Fetal heart tracing 20 minutes before caesarean section (Doppler monitor, Hewlett Packard 8040A, paper speed 1 cm/min.).

The placenta weighed 680 gms and contained a large 8×7×4 cm mass. This was a well encapsulated angiomatous hemangioma (Chorioangioma) with numerous capillary-like vessels contained in a loosely cellular stromal background. Numerous blood vessels were markedly congested, and in one section necrosis and degenaration of the angiomatous capillaries was seen. There was no retroplacental hemorrhage or hemorrhage into the tumor, and the rest of the placenta was normal.

The infant was transfused with group O Rh negative blood in the intensive care unit and made a good recovery. There was no clinical evidence of any coagulation defect, and she was discharged to her mother on day 10.

DISCUSSION

Chorioangiomas have reported incidence of about 1% overall. Fox [3] 1 per 100, Wentworth [4] 1 in 77. The greater majority of these do not reveal their presence either clinically or on routine examination of the placenta.

The histological features have been well reviewed by Novak [5], Asadourian [6] and Fox [3]. There were three main types as observed by Asadourian:

A. Angiomatous Chorioangioma — predominantly vascular, with the overgrowth of capillary networks in a loosely supported stroma.

B. Myxomatous Chorioangioma — predominanatly consisting of a myxomatous stroma, containing abundant engorged capillaries.

C. Infiltrative — an infiltrative type representing overproliferation of the capillary network in otherwise normal chorionic villi. The thin-walled vessels are sometimes dilated, but when ectasia is less marked and the involved villi are more normal in size, distinction between tumor and telangiectasia can be made only by the increased numbers of blood vessels within a single villus.

A degenerative type was discribed by Marchetti [7]. It is thought that this could represent degeneration of the above types.

CLINICAL FEATURES

The etiology of these tumors has been the subject of speculation in papers by Fox [3], Battalgia [8], and Asadourian [6]. The weight of opinion is that they represent an excess proliferation of the undifferentiated angiobastic chorionic mesenchyme. The majority of these tumors are small, being entirely embedded in the substance of the placenta. However, a variety of complications are associated with larger chorioangiomata. These have been well reviewed by Asadourian and others [16].

Hydramnios is common. McIntroy and Kelsey (1954) postulated that the failure of the placenta to adquately clear solutes, leads to their excretion in the fetal urine. Prematury (30% cases), and premature placental separation (16-17% of cases) may occur especially in association with hydramnios.

Cardiomegaly is found in cases where the tumor exceeds 5 cm in diameter or are multiple (Wallenberg, 1971 [10]). This appears to be as a result of hyperdynamic heart failure due to the effects of the chorioangioma acting as an arteriovenous shunt. This has been well demonstrated by Reiner (1965) [11] and may also have a major role in the genesis of the fetal distress and intrauterine fetal demise that is more common, (12-16% of cases), with the larger chorioangiomata.

Congenital anomalies (5-10%) are more common than in the general population. These are often due to the presence of hemangiomata in extra placetal sites. There also appears to be an association between chorioangiomata and chromosomal anoamlies, suggesting a possible common determinant. Cases of recurring chorioangiomata in subsequent pregnancies in a woman have been reported Battalgia, 1968 [8], Berge 1966 [3]. The possibility that there may be an underlying genetic factor has been suggested Wallenberg, 1970 [10].

There have been few cases reported of severe fetal anemia associated with a chorioangioma. Hurwitz et al. 1983 [12] reported the delivery of a severely anemic infant associated with a chorioangioma. The anemia was in his case attributed to sequestration of blood within the tumor. However the nature of the anemia is not defined, nor did the authors exclude microangiopathic hemolytic anemia as a possible cause. Bauer (1978) [13], also reported an association between severe anemia and chorioangiomata.

Sims et al. (1976) [14] reported on a case of a fetal anemia, with the suggestion of massive chronic fetomaternal bledding as the main underlying cause. Another case of chronic fetomaternal hemorrhage resulting in iron deficiency anemia is reported by Cunningham and Pritchard (1985) [15].

The main features in our case are the rapid onset of severe distress against the background of a positive OCT, and the features of a fetal anemia of short duration, in the absence of hydropic changes. It is true that fetal cells were demonstrated in the maternal circulation only after delivery, and may therefore have entered during parturition. However we feel that the likelihood is strong that an antepartum fetomaternal transfusion took place, as the clinical picture was that of a relatively rapid bloodloss, and there was no evidence of hemorrhage into the tumor, or of fetal hemolysis.

It is of note that a second case like this has recently been trated in our unit. The baby, who had a hemoglobin of 3,2 g/dl died soon after delivery. Blood taken antepartum demonstrated the presence of fetal red blood cells in the maternal circulation.

CONCLUSION

It is surprising that fetomaternal transfusion has not previously been stressed as a possible complication of chorioangiomata, considering the vascularity of the tumor in the vascular setting of the placenta which is being subject to the positive intra-uterine pressure associated with labour. Attempts shoula be made to demonstrate fetomaternal transfusion in patients who present with:

1. Placental masses found on ultrasound examination.
2. Fetal distress, expecially when hydromnios co-exsts, as this may signal the presence of a chorioangioma.
3. The presence of sinusoidal fetal heart trancings in the absence of Rh disease.

This tumor remains a rare cause of pregnancy related problems, and will no doubt contonue to surprise clinicians in the future.

BIBLIOGRAFIA

1. FREEMAN R.K., GARITE T.J.: Fetal Heart Monitoring, Baltimore, Williams and Wilkins; 1981; 116.
2. HOFMEYR G.J., SONNENDECKER E.W.W.: Heart rate changes after acute fetal hemorrhage — a basis for the pathophysiology of the sinosoidal pattern. SAMJ; 1983; 64: 23-25.
3. FOX H.: Haemangiomata of the placenta. J. Clin. Path.; 1966; 19: 133-137.
4. WENTWORTH P.: The incidence and significance of hemangiomata of the placenta. J. Obstet. Gynaecol. Br. Commonw.; 1965; 72: 81-88.
5. NOVAK E.R., WOODRUFF J.D.: Gynecologic and Obstetric Pathlogy, 8th Ed., Philadelphia, Saunders; 1979; 624-625.
6. ASADOURIAN L.A., TAYLOR H.B.: The clinical significance of placental hemangiomas. Obstet. Gynecol.; 1968; 31: 551-555.
7. MARCHETTI A.A.: A consideration of certain types of benign tumors of the placenta. Surg. Gynec. Obstet.; 1939; 68: 733-743.
8. BATTALGIA F.C., WOOLEVER C.A.: Fetal and neonatal complications associated with recurrent chorioangiomas; 1968; Pediatr. 41: 62.
9. BERGE T.H.: Placental chorioangiomatosis. Acta Path. Microbiol. Scand.; 1966; 66: 465.
10. WALLENBERG H.C.S.: Chorioangiomata of the placenta. Obstet. Gynecol. Survey; 1971; 26: 411-425.
11. REINER L., FRIES E.: Chorioangioma associated with arteriovenous aneurysm. Am. J. Obstet. Gynecol.; 1965; 93: 58-64.
12. HURWITZ A., et al.: Severe foetal distress with hydramnios due to chorioangioma. Acta Obstet. Gynecol. Scand.; 1983; 62: 633-635.
13. BAUER C.R., FOCAJO R.M., BANCALARI E., FERNANDEZ-ROCHA L.: Microangiopathic hemolytic anemia and throbocytopenia in neonate associated with a large placental chorioangiomata. Pediatrics; 1978; 62: 574-577.
14. SIMS D.G., BARRON S.L., WADEHARA V., ELLIS H.A.: Massive chronic feto-maternal bleeding associated with chorioangiomas. Acta Pediatr. Scand.; 1976; 65: 271-273.

Pedido de separatas:
Dr. A.M.C. Franca-Martins
23 Francisview
Dwonham avenue 2091
Mondeor
Johannesburg
South Africa