

# Prenatal diagnosis of thrombosis of the dural sinuses: report of six cases, review of the literature and suggested management

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**KEYWORDS:** Doppler; dural malformation; dural sinus; fetus; prenatal diagnosis; thrombosis; torcular herophili; ultrasound

## ABSTRACT

**Objectives** To describe and assess the sonographic findings, evolution and clinical implications of thrombosis of the fetal dural sinuses.

**Methods** We compiled a multicenter report of the outcomes of five cases with a prenatal diagnosis of thrombosis of the dural sinuses, and one case in which thrombosis of the dural sinus was diagnosed at necropsy after termination of pregnancy. Prognostic factors are discussed, and suggestions made for prenatal and postnatal management.

**Results** The mean (range) gestational age at diagnosis of thrombosis of the dural sinuses in the five cases in which it was made prenatally was 25.2 (22–31) weeks. In these five cases, diagnosis was made by sonography and confirmed by magnetic resonance imaging (MRI), which showed a blood clot in the region of the torcular herophili. Three of the six cases delivered vaginally with favorable sonographic findings, and normal clinical neurological development. Two pregnancies were terminated at the request of the parents. In one of these cases the prognosis was poor, with signs of fetal decompensation or cardiac failure; the pregnancy was terminated and necropsy revealed thrombosis of the occipital dural sinuses associated with a hemangioma. One infant, in whom the thrombosis developed in conjunction with a dural sinus malformation, died at 4 months of age.

**Conclusions** Thrombosis of the cerebral venous circulation can occur antenatally and is detectable by fetal

real-time and color Doppler ultrasound examination. A review of the literature supports targeted evaluation of the fetus by serial ultrasound imaging and MRI to help guide the diagnosis, and to improve the counseling and management of such cases. Partial or total regression, isolated abnormality, absence of fetal decompensation or signs of cardiac failure and favorable clinical evolution are suggestive of favorable prognosis. In such cases, non-interventional neonatal management is recommended. Copyright © 2008 ISUOG. Published by John Wiley & Sons, Ltd.

## INTRODUCTION

Thrombosis of the fetal dural sinuses is rare. The sonographic features usually mimic those of an intracranial tumor. As a result, the condition is often misdiagnosed and its frequency underestimated. There exist few reported cases and the prognosis is difficult to establish. Nevertheless, the evolution is potentially favorable, and so early detection of the features and associated signs are important in providing informed counseling.

We describe the features of five cases diagnosed on ultrasound and magnetic resonance imaging (MRI) examination, and one case in which thrombosis of the dural sinus was diagnosed on necropsy after termination of pregnancy. After a discussion and review of the literature, we suggest guidelines for antenatal and neonatal management.

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## METHODS

A multicenter report was compiled of the outcomes of five cases with a prenatal diagnosis of thrombosis of the dural sinuses, and one case in which thrombosis of the dural sinus was diagnosed after termination of pregnancy.

## RESULTS

Data on the six cases are summarized in Table 1.

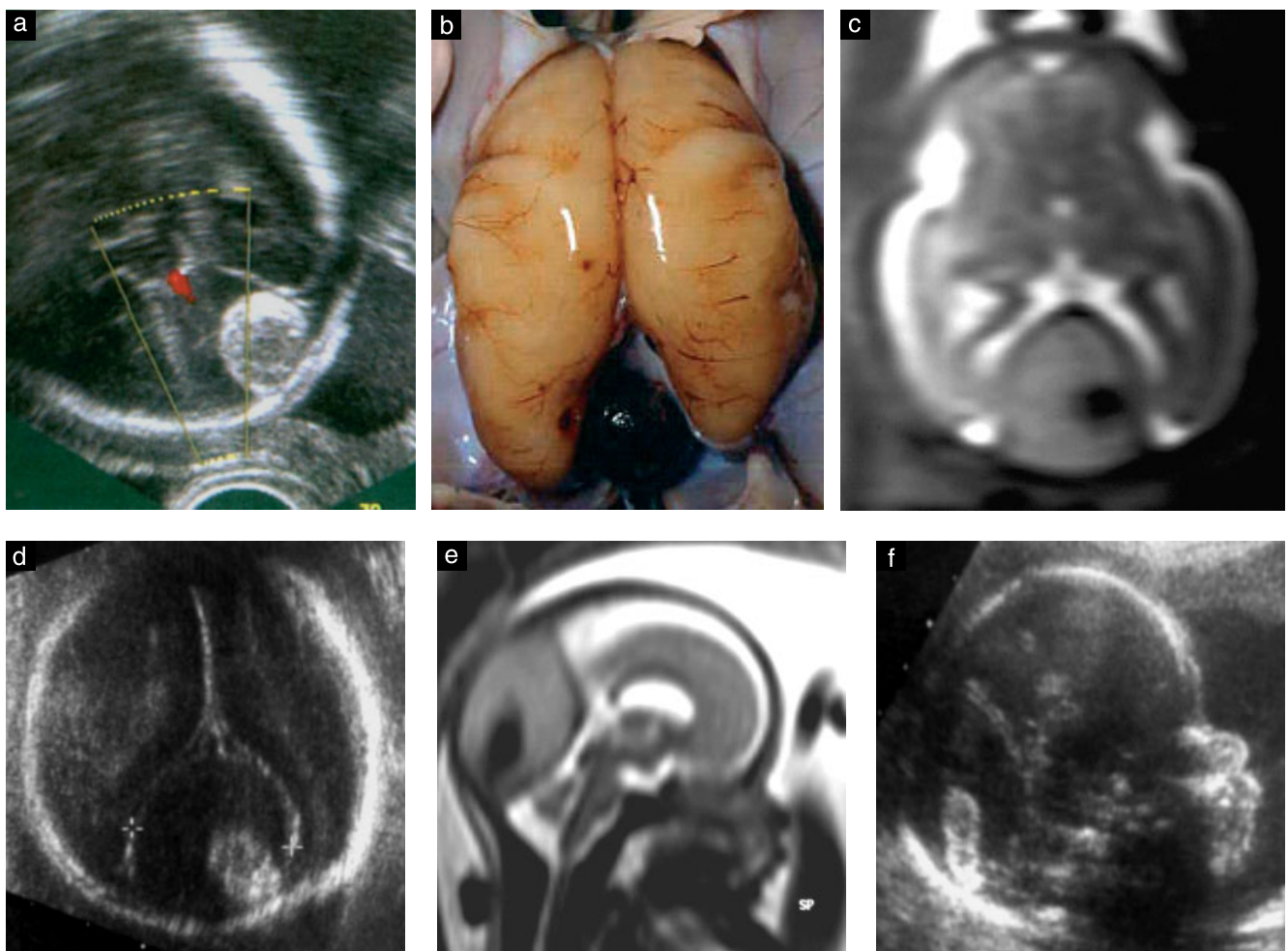
### Cases 1–3

Cases 1, 2 and 3 were similar; the patients were referred for a posterior hyperechogenic, rounded, extracerebral and intracranial mass just above the cerebellum, posterior to the occipital lobes and superior to the tentorium cerebelli, with absence of signals on color Doppler imaging (Figure 1a). The mass was surrounded by a triangular sonolucent area which was identified as dilatation of the posterior segment of the longitudinal sinus. Sinus thrombosis was suspected and confirmed by MRI, which showed an ovoid thrombus of the torcular (Figure 1c–f).

It appeared as a hypersignal on T1-weighted images and a central hyposignal on T2-weighted sequences. The earliest case in this series (1996) was terminated at the request of the parents (Figure 1b). As the condition had not been documented previously they were informed that the prognosis was uncertain. In the two other cases, both color Doppler ultrasound examination and MRI showed a normal brain, no ischemic lesion and regression of the thrombus throughout the pregnancy, which resulted in normal birth. Fetal and parental investigations failed to find any cause of the condition. At 12 months and 24 months the infants' developmental assessments were normal.

### Case 4

The patient was referred at 32 weeks, after an uneventful pregnancy, for an abnormal posterior fossa and mild polyhydramnios (largest pocket of fluid, 9 cm). Ultrasound examination revealed cephalic biometry on the 95<sup>th</sup> centile and an avascular septate and mainly anechogenic mass



**Figure 1** (a) Color Doppler image (axial plane) of the fetal head at 25 weeks showing a hyperechogenic clot surrounded by a hypoechoic area corresponding to a dilated sinus. (b) Corresponding brain at autopsy showing thrombosis of the torcular. T2-weighted axial plane magnetic resonance image (c) and corresponding ultrasound image (d) showing the dilated thrombosed superior sagittal sinus and torcular at 24 weeks. Sagittal plane magnetic resonance image (e) and corresponding ultrasound image (f) showing the clot in the torcular.

Table 1 Description of our case series of thrombosis of the dural sinuses

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6
GA at diagnosis (weeks)*	25	24	24	32	22	31
Patient's age (years)	32	31	25	23	27	25
Gravidity	2	1	1	1	2	2
Prenatal trauma or early invasive tests	No	No	No	No	No	No
Location of thrombosis	Torcular	Torcular	Torcular	Posterior sinuses Complex mass	Torcular Right transverse and sphenoparietal sinuses	Torcular straight sinus Dural sinus malformation
Initial size of clot (mm)	20 × 16 × 14	18 × 18	14 × 9	40 × 50	—	30 × 20 × 15
Associated signs	None	None	None	Mild pericardial effusion and ascites, tricuspid regurgitation, BPD on 95 <sup>th</sup> centile	Portosystemic shunt Mild transient polyhydramnios	BPD on 90 <sup>th</sup> centile EFW on 10 <sup>th</sup> centile
MRI	25 weeks Ovoid thrombus of the torcular	29 weeks Thrombus of the torcular, no ischemic lesion, normal brain	24, 27 and 30 weeks Thrombus of the torcular, no ischemic lesion, normal brain, progressive regression	32 weeks Postcerebellar mass 50 mm × 50 mm with sign of compression of cerebellum and pons	27, 31 and 35 weeks	32 + 5 weeks 52 mm × 50 mm × 38 mm, no sign of compression
Evolution	—	<i>In-utero</i> regression after slight increase in size observed at 28 weeks; clot measuring 29 mm × 28 mm × 19 mm	<i>In-utero</i> regression	—	<i>In-utero</i> regression and total disappearance at 1 month	No decrease, similar size at birth, slight decrease (1 <sup>st</sup> month) then rapid increase in size (4 <sup>th</sup> month)

\*Gestational age (GA) at which an intracranial abnormality was diagnosed.

(continued overleaf)

Table 1 (Continued)

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6
Coagulation test and thrombophilia	Not performed	Parents: no platelet antibodies, normal thrombophilia status	Fetus and parents: normal thrombophilia status	Fetus and parents: TORCH negative, normal coagulation tests and thrombophilia status	Fetus and parents: TORCH negative, normal coagulation test and thrombophilia status, no genetic mutation, no platelet antibodies	Fetus and parents: TORCH negative, no platelet antibodies, maternal coagulation test normal, no thrombophilia Neonate: coagulation tests normal
Fetal gender	Female	Female	Female	Male	Female	Male
Outcome of pregnancy	TOP at 26 weeks	Vaginal birth at 41 weeks	Vaginal birth	TOP at 33 weeks	Elective CS at 38 weeks (scarred uterus) 2850 g Apgar 8/10/10	Spontaneous vaginal birth at 36 weeks
TFS and or neonatal MRI	—	Normal; total regression of thrombus	Residual 5-mm clot and abundant flow in the surrounding region	—	Residual 13 mm × 8-mm clot at birth MRI at 10 weeks: total regression, normal brain	MRI: 5-cm echogenic mass at day 15 TFS: 30 mm mass at 3 months, then 40 mm × 20 mm at 4 months
Long-term follow-up	—	Good (24 months)	Good (12 months)	— (Normal next pregnancy)	Good (24 months)	Death at 4 months + 2 weeks
Pathology	Thrombus of the torcular surrounded by hemorrhagic subarachnoid fluid	—	—	Thrombus and hemangioma	—	Not performed

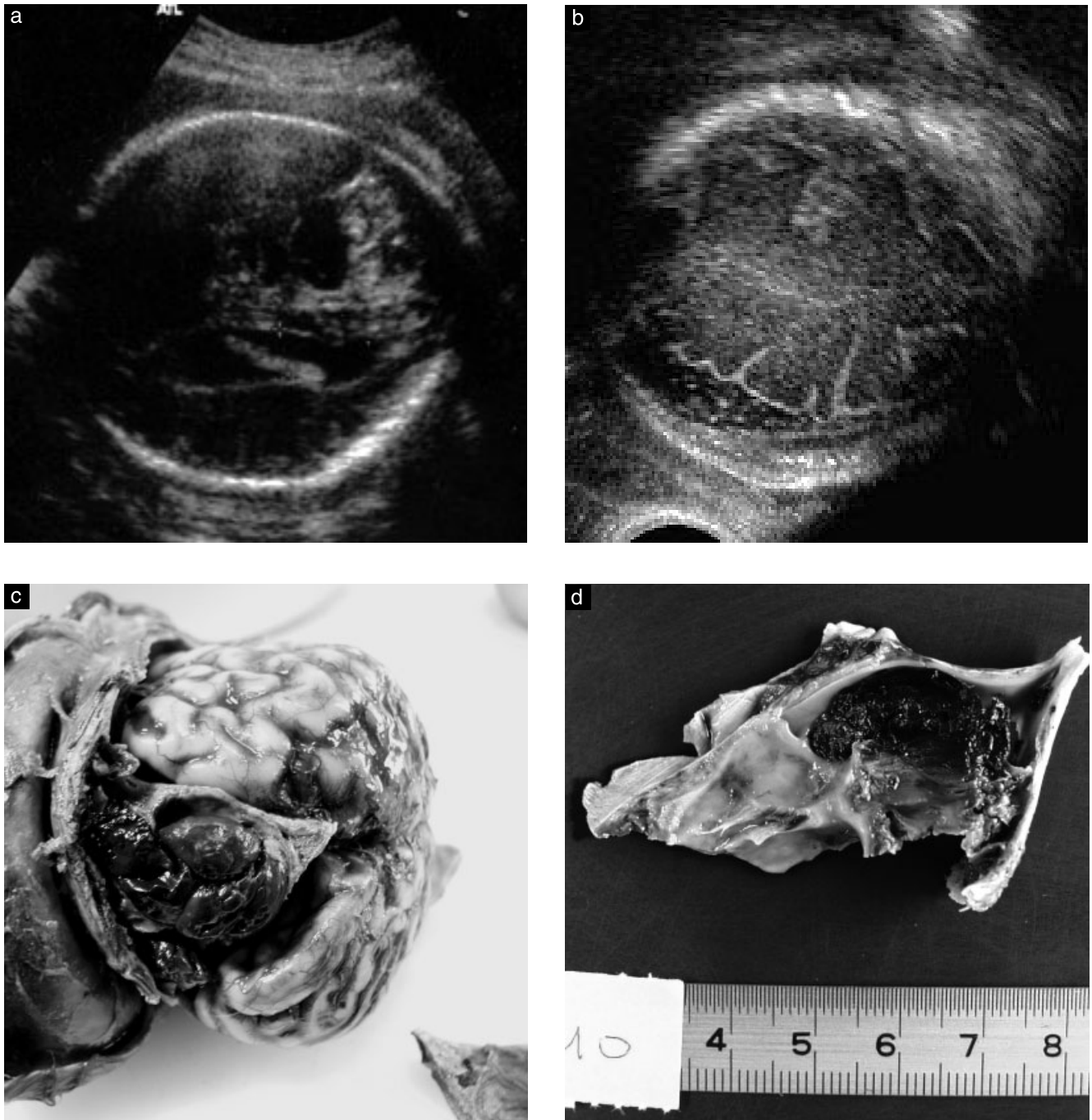
BPD, biparietal diameter; CS, Cesarean section; EFW, estimated fetal weight; MRI, magnetic resonance imaging; TFS, transfontanelle sonography; TOP, termination of pregnancy.

of the posterior fossa. An echogenic area was observed posteriorly with no color Doppler signals and a hypo-echogenic area above the mass under the calvaria. These two signs were interpreted retrospectively as the clot and the dilated superior sagittal sinus (Figure 2a and b). The cerebellum was not visible. Signs of fetal decompensation were noted. MRI detected a meningeal mass measuring 50 mm in diameter, with compression of the cerebellum and pons. In view of the poor fetal prognosis in combination with cardiac insufficiency the parents decided to terminate the pregnancy. Necropsy revealed a

thrombosis of the occipital dural sinuses associated with a hemangioma (Figure 2c and d).

#### Case 5

Case 5 presented with three concomitant thromboses and a portosystemic shunt. An abnormal fetal cerebellum was observed at 22 weeks. Ultrasound examination revealed a round, well defined complex mass just in front of the occiput (Figure 3a) and above the structurally normal cerebellum. On a sagittal view the mass was surrounded



**Figure 2** Images of Case 4. (a) Ultrasound image showing a heterogeneous mass of the posterior fossa. (b) Transverse view under the calvaria, showing dilatation of the superior sagittal sinus. (c and d) Pathological examination showing thrombosis of the dural posterior sinuses (associated with a hemangioma).

by a slightly echogenic oblong area lying just under the calvaria and identified as a dilated longitudinal sinus (Figure 3b). The latter was observed on a transverse view as an enlargement of the interhemispheric space, as described in Case 4 (Figure 2b). Color flow imaging failed to reveal any vascular flow within the lesion or in the dilated longitudinal sinus. The right transverse sinus could not be visualized but the left transverse sinus was visible (Figure 3d), showing a normal triphasic pattern. The brain was normal. A diagnosis of thrombosis of the torcular, right transverse and longitudinal sinuses was suspected. Fetal biometry and well-being were normal. Fetal hemodynamics were normal but there was a dilated umbilical vein with high-velocity flow (35 cm/s). The placental cord insertion showed a small aneurysmal dilatation and the umbilical vein seemed to directly connect to the right atrium. The ductus venosus was present, with normal flow. This feature was identified postnatally as a portosystemic shunt.

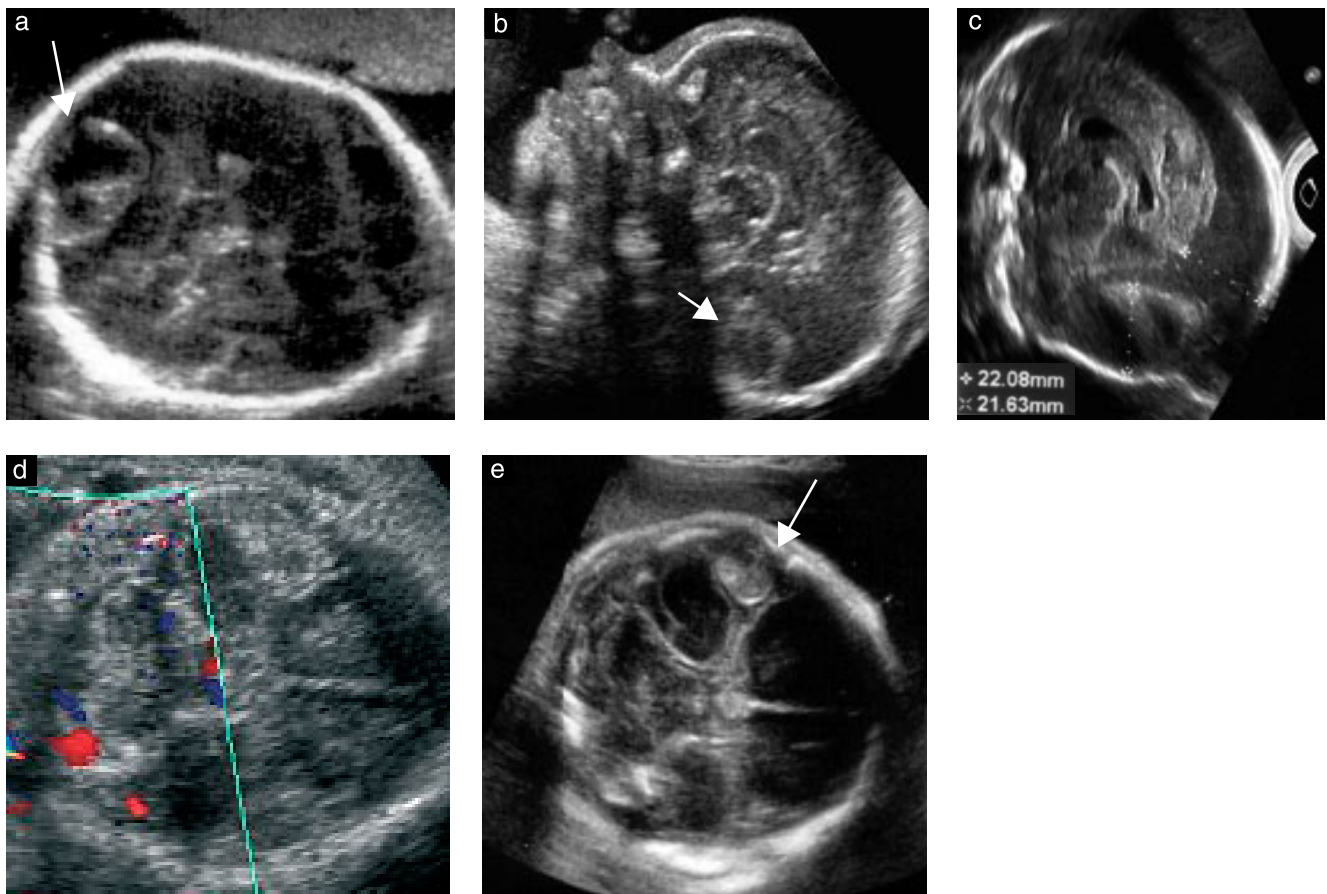
MRI confirmed thrombosis of the torcular, right transverse sinus and left sphenoparietal sinus, with a normal brain and gyration (Figure 4a–c). No deep thrombosis, ischemic lesion or arteriovenous malformation could be identified.

Sonographic follow-up showed progressive regression of the torcular thrombosis, which was no longer detected at 35 weeks (Figure 5a and b). At that time reduced flow was observed in the right transverse sinus, providing evidence of recent recanalization (Figure 5c and d).

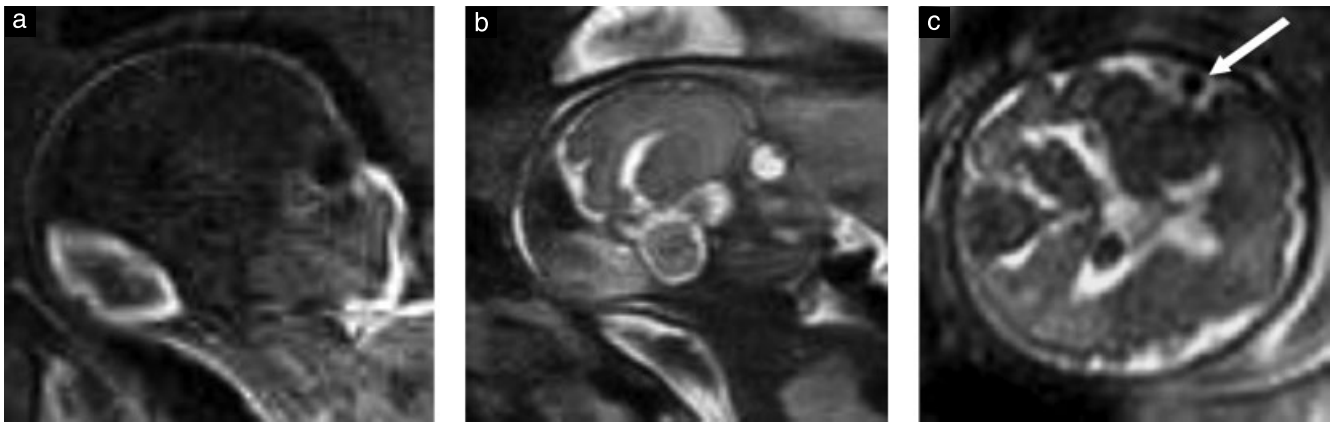
The umbilical vein remained dilated with high velocity flow, and signs of fetal cardiac overflow appeared during the last month of pregnancy (aortic flow, 1.2 L/min; pulmonary flow, 1.4 L/min) with mild right cardiac dilatation and tricuspid regurgitation (3.1 m/s). Ductus venosus hemodynamics remained normal as did fetal well-being.

Elective Cesarean section (because of a scarred uterus) was performed at 38 weeks. Postnatal transfontanelle ultrasound examination confirmed a thrombosis confined to the posterior part of the superior longitudinal sinus with normal flow in the surrounding region (Figure 5e and f).

Neonatal ultrasound examinations performed at 8, 15 and 30 days showed a persistent shunt between the right portal vein and right subhepatic vein with an unusual dilatation of the right portal vein, which appeared dysplastic. Postnatal MRI was performed at 2.5 months of age and was totally normal. At 24 months the child had normal physical and cognitive development.



**Figure 3** Ultrasound examination of Case 5 at 27 weeks. (a) Thrombus of the torcular (arrow) misdiagnosed as abnormal cerebellum at 22 weeks. (b) Mid-sagittal plane; note the normal brain and the occipital thrombus (arrow) surrounded by a hypoechoic area corresponding to the dilated superior sagittal sinus. (c) Sagittal view demonstrating a large dilatation of the superior sagittal sinus above the thrombus. Measurements of the clot (x...x) and the dilatation (+...+) are indicated. (d) Color Doppler signal in the contralateral transverse and sigmoid sinuses. Note the absence of any signal in the thrombosed transverse sinus characterized by a mild echogenic area and the normal cerebellum. (e) Axial plane showing thrombus in the left sphenoparietal sinus (arrow).



**Figure 4** Magnetic resonance images of Case 5. (a) T1-weighted image at 31 weeks demonstrating thrombus of the torcular; note the associated hypersignals and hyposignals due to different evolution of the thrombosis. Sagittal (b) and axial (c) T2-weighted images show thrombosis of the torcular, right transverse sinus and of the left sphenoparietal sinus (arrow).

### Case 6

The patient was referred at 31 weeks after an uneventful pregnancy for an isolated extracerebral mass, located just above the tentorium, measuring 30 mm × 20 mm × 15 mm. The biparietal diameter was on the 90<sup>th</sup> centile, and the abdominal circumference and femur length were on the 25<sup>th</sup> centile. There was no mass effect, no ventriculomegaly and a normal brain. At 32 + 5 weeks MRI showed an increased mass measuring 52 mm × 50 mm × 38 mm, suggestive of thrombosis of the torcular. Spontaneous vaginal birth occurred at 36 + 2 weeks. Transfontanelle ultrasound examination showed thrombosis of the torcular measuring 40 mm × 20 mm, probably associated with a dural sinus malformation. Postnatal MRI confirmed the diagnosis without signs of ischemic lesions. Serial neonatal scans showed persistent superior longitudinal sinus dilatation. At 3 months the mass had decreased slightly. At 4 months the mass increased rapidly, with a perceptible flow. The longitudinal sinus was large. Within 2 weeks the child developed respiratory failure with recurrent episodes of apnea. A painful lambdoid tumor was detectable clinically. Death occurred at the age of 4 months and 2 weeks.

### DISCUSSION

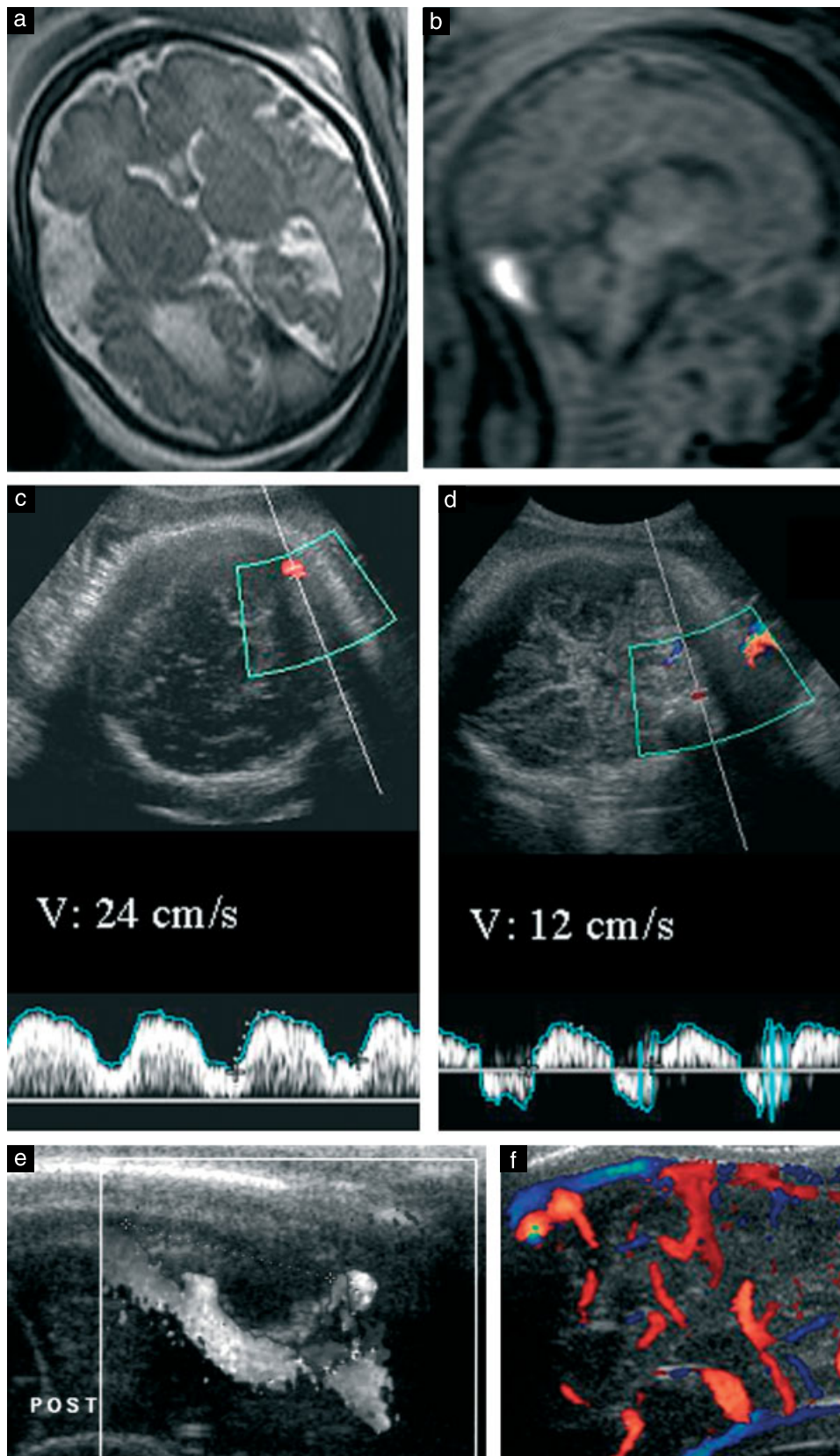
We report the detailed observation and follow-up of six cases of prenatal dural sinus thrombosis. The condition is rare and only a few cases with long-term follow-up have been reported (Table 2)<sup>1–4</sup>. There are two possible explanations for this lack of documentation. First, in the absence of any firm evidence on which to base counseling, parents prefer to terminate the pregnancy and, second, the condition is often misdiagnosed because the ultrasound features are suggestive of a tumoral disorder. Thrombosis is often recognized at a later stage on pathological examination, as in our Case 4.

The entire venous circulation of the fetal brain can be identified prenatally and studied by Doppler technology,

as we have shown previously<sup>5</sup>. Real-time ultrasound associated with color Doppler imaging is the key factor in the prenatal diagnosis as demonstrated by Visentin *et al.*<sup>2</sup>. Any tumor-like mass of the posterior fossa, particularly if it is heterogeneous, should be examined by color Doppler sonography. The association of a hyperechogenic area surrounded by a hypoechoic, possibly septate, triangular area and the absence of flow inside the mass is suggestive of thrombosis of the posterior sinuses. This tentative diagnosis can be backed up either by visualization of interruption of blood flow in the thrombosed sinus or by visualization of an enlargement of the interhemispheric space corresponding to the dilated superior longitudinal sinus, which appears slightly and homogeneously echogenic as if filled with stagnant fluid. This typical image can be observed longitudinally, as described previously, but also just under the calvaria on a transverse view. Dilatation of the superior longitudinal sinus can be great, as observed in our Case 5 (Figure 3), and is not necessarily indicative of poor prognosis.

Dilatation can also be detected by MRI, as described by Emamian *et al.*<sup>3</sup>. However, we do not totally agree with the authors' interpretation of the mixed signals as hematoma and recurrent hemorrhage. In our opinion these features could be consistent with thrombosis of the superior sagittal sinus, associated with a large dilatation of the sinus. Regarding prognosis, neither the size of the clot, the surrounding dilatation nor the number of thrombi, of which there were three in our Case 5, seem to be predictive factors of poor outcome at the initial stage of diagnosis.

Of the total of 12 cases observed in the literature and in our series, eight were live births. One infant died postnatally after surgical management and another at 4 months of age. In the six other infants neurodevelopment was good over a follow-up of 12–24 months. In all cases no other fetal abnormality was found and the brain appeared normal. These two factors seem to be essential for a favorable prognosis. Nevertheless it is particularly difficult antenatally to prove the absence of associated



**Figure 5** Antenatal and neonatal follow-up of Case 5. Transverse (a) and sagittal (b) T1-weighted magnetic resonance images showing partial recanalization of the superior sagittal sinus and torcular associated with the residual thrombosis at 35 weeks' gestation. (c and d) Pulsed Doppler images at 34 weeks showing normal left transverse sinus hemodynamics (c) and partial recanalization of the thrombosed right transverse sinus (d). (e) Transfontanelle neonatal examination; the thrombosis of the superior sagittal sinus measures  $1.3 \times 0.8$  cm. (f) Color Doppler image showing abundant flow around the thrombus. V, velocity.



Table 2 Data on previously published cases of thrombosis of the fetal dural sinuses

	Gicquel et al. <sup>1</sup> 2000	Visentin et al. <sup>2</sup> 2001	Visentin et al. <sup>2</sup> 2001	Visentin et al. <sup>2</sup> 2001	Emamian et al. <sup>3</sup> 2002	Clode et al. <sup>4</sup> 2004
Gestational age at diagnosis (weeks)	22	21	22	28	24	22
Location of thrombosis	Torcular	Torcular	Torcular	Torcular	Longitudinal superior sinus	Torcular, extended to the right transverse sinus
Associated signs	None	None	None	Borderline ventriculomegaly (13 mm) ?*	None	None
Prenatal trauma or early invasive test	Amniocentesis at 15 weeks	?*	?*		NR	NR
Coagulation test and thrombophilia	Postnatal test normal	No prenatal test performed	No prenatal test performed	No prenatal test performed	NR	NR
Gender	Female	?†	?†	Male	NR	NR
Evolution	Stable lesion (2 <sup>nd</sup> and 3 <sup>rd</sup> trimester) No thrombus observed at 38 weeks	—	—	No further evolution	Serial CT scans: slight decrease in size Angioscan: no bleeding vessel	CT scan: stable thrombus MRI at 4 days of life: clot in torcular and right transverse sinus
Outcome of pregnancy	Vaginal birth at 40 weeks	TOP	TOP	Planned CS at 38 weeks	Spontaneous vaginal birth at 36 weeks	CS at term
Long-term follow-up	Good (18 months)	—	—	Peroperative neonatal death (neurosurgery)	Good (12 months)	Good (18 months)
Pathology	—	Thrombus of torcular with normal brain	Thrombus of torcular with normal brain	Thrombosis of torcular, massive intraoperative hemorrhage	—	—

?\*, Amniocentesis was performed in two of the three cases in Visentin's series. ?†, One of these was female, the other not reported. CS, Cesarean section; CT, computed tomography; NR, not reported; TOP, termination of pregnancy.

malformation. We believe the use of MRI in association with ultrasound imaging to be of great value in ruling out brain or gyration abnormality.

In only one of the six live newborns with a favorable prognosis was an enlargement of the initial lesion seen. In four cases, serial scans showed a slow and late decrease in the size of the clot and of the stasis around the clot. In each of our three cases with favorable outcomes the lesion could not be detected at the end of the pregnancy, but a small thrombus was found on the transfontanelle neonatal scan. Color Doppler imaging identified repermeabilization of the thrombosed sinus before birth (Case 5).

The cause of prenatal thrombosis of the dural sinuses remains unknown. In neonates, infants and adults, an association with trauma has been established, but this is uncertain in the prenatal period. The pathophysiology of neonatal and infant dural thrombosis has been widely studied by Barbosa *et al.*<sup>6</sup>. These authors reported that the condition usually occurs in association with dural sinus malformations, which are rare diseases of the posterior sinuses and may develop antenatally. They correspond to a distinct entity within the group of dural arteriovenous shunts. These vascular malformations are characterized by low flows. They usually proceed either to arteriovenous shunt with unfavorable outcome or total or partial thrombosis, the regression of which (spontaneously or under

heparinotherapy) was associated with a favorable outcome in 18 of the 30 cases reported by Barbosa *et al.*<sup>6</sup>. Dural malformations with dural arteriovenous shunt lead to congestive cardiac failure and macrocrania. These features were present in two of our cases. In Case 4, signs of fetal decompensation were initially present and pathological examination disclosed a hemangioma. In Case 6, no fetal cardiac failure was observed initially but the biparietal diameter remained on the 95<sup>th</sup> centile (with fetal and neonatal weight on the 10<sup>th</sup> centile). Serial investigations failed to show a net decrease in the thrombus, which raised the possibility of dural malformation.

Little is known about genetic thrombophilia or abnormal coagulation status. In our six cases we did not observe any association. In contrast, Wu *et al.* reported four cases of thrombophilia in a series of 30 newborns with sinovenous thrombosis<sup>7</sup>. Thrombophilia in association with sinus thrombosis was also reported in an earlier study by Vielhaber *et al.*<sup>8</sup>, but the link with fetal and neonatal dural thrombosis is still a matter of debate<sup>9</sup>. We would not recommend cordocentesis as parental tests are sufficient to rule out coagulation or thrombophilic disorders.

For some authors dural sinus malformation involving the torcular is a worrying feature<sup>6</sup>. However, in our review of cases of prenatal dural thrombosis with long-term follow-up, including our series, thrombosis of the torcular was observed in all six cases with favorable outcomes (Tables 1 and 2). It is possible

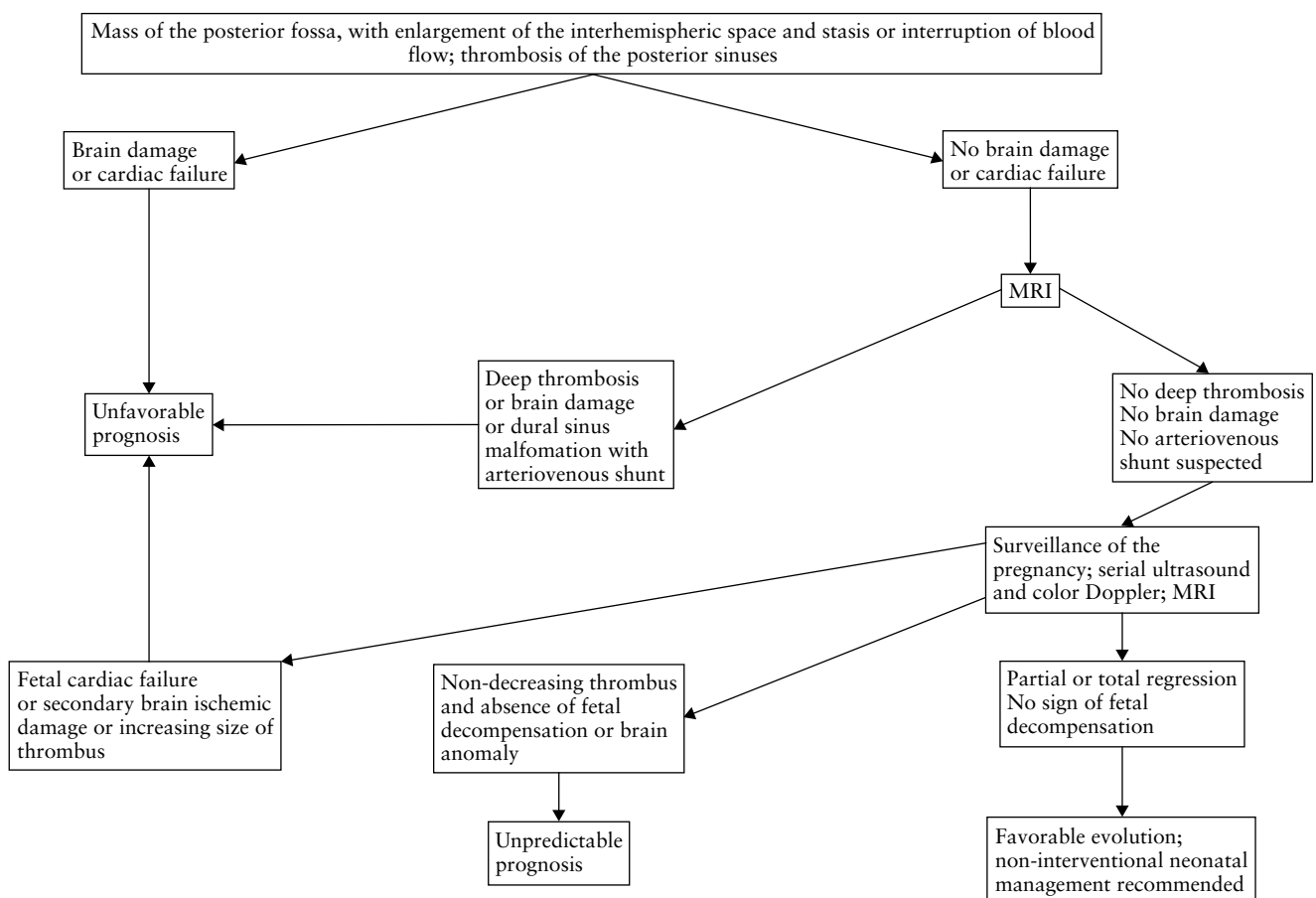


Figure 6 Counseling for management of thrombosis of the dural sinuses. MRI, magnetic resonance imaging.

that cerebral venous drainage in the fetus differs from that found postnatally. Fetal anastomoses may facilitate redirection of the venous blood flow. We know that the absence of cavernous capture in the first months of postnatal life carries an unfavorable prognosis but the cavernous sinuses are fully developed after 4 months.

Dural sinus malformations are not hereditary, but male predominance has been noted<sup>6</sup>. In our series we did not observe this predominance (four females, two males), but the prognosis seems to be worse for males.

The features of our Case 5 were suggestive of a venous dysplasia. Although follow-up at 15 months was good, the association of multiple cerebral thrombosis, portosystemic shunt and dysplasia-like hepatic veins on the abdominal ultrasound scans is consistent with this hypothesis, which needs to be confirmed by reports of similar cases. However, we agree with Visentin *et al.*<sup>2</sup> that thrombosis of the dural sinuses can occur antenatally as early as the second trimester in uncomplicated pregnancies.

Spontaneous favorable evolution appears to be possible when cerebral structures remain normal, without infarction or ventricular hemorrhage, and when there is no fetal cardiac failure suggestive of an associated shunt. MRI is essential at the initial stage to confirm the diagnosis, to accurately describe the location, size and number of the thrombi, and to rule out brain or gyration abnormality. We think that MRI should be repeated at least once, 1–2 months later, to examine for infarction or hemorrhage. Normal biparietal diameter and decreasing thrombosis size are the essential factors for a favorable prognosis, and these can be monitored by serial ultrasound and color Doppler examinations.

On the basis of findings from the literature and our experience we suggest guidelines for prenatal

management that could be helpful in prenatal counseling (Figure 6).

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