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SHORT CLINICAL REPORT

TITLE: Clinical case: A child dural-based occipital cavernoma with an accessory venous sinus

Cas clinique rapportant un cavernome dural occipital de l'enfant avec sinus veineux accessoire

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ABSTRACT

Background: The diagnosis of extra-axial cavernoma and surgical decision-making can be difficult on children.

Reported case: In this report we present the case of a 33-month-old child for whom the work up done for asthenia, anorexia and psychomotor regression reveals a well-defined occipital extra-axial lesion. A follow up MRI was performed two months later, revealing an increase in the size of the lesion associated with hemorrhagic changes.

The surgical decision is performed after an arteriography; it allows the total removal of the lesion contained in a duplication of the dura mater, by placing a vascular clip on an accessory venous sinus draining the lesion, respecting the integrity of the lateral sinus. Histology confirms a cavernoma. No complication resulted from the surgical procedure.

This atypical case of pediatric dural cavernoma associated with a well-defined accessory venous sinus in arteriography is to our knowledge the first description in the literature.

Conclusion: Extra-axial cavernomas have a misleading presentation. The management of these lesions is of twofold interests: to avoid a repercussion on the development of these children, and to obtain histological confirmation.

RESUME

Contexte: Le diagnostic de cavernome extra-axial et la prise de décision chirurgicale peuvent poser des difficultés chez l'enfant.

Cas rapporté: Nous présentons ici le cas d'un enfant de 33 mois à propos duquel l'exploration d'une asthénie, anorexie et régression des acquisitions met en évidence une lésion extra-axiale occipitale bien délimitée. L'IRM de contrôle à deux mois montre une augmentation de taille de la lésion, présentant des remaniements hémorragiques. La décision chirurgicale est prise après artériographie ; elle permet l'exérèse totale d'une lésion contenue dans un dédoublement de dure mère, avec pose d'un clip vasculaire sur un sinus veineux accessoire drainant la lésion, en respectant l'intégrité du sinus latéral. L'histologie est en faveur d'un cavernome. Aucune complication n'est déplorée au décours du geste chirurgical. Ce cas atypique de cavernome dural pédiatrique associé à un sinus veineux accessoire bien défini en artériographie est à notre connaissance la première description dans la littérature.

Conclusion: Les cavernomes extra-axiaux ont une présentation trompeuse. La prise en charge de ces lésions présente un double intérêt : éviter une répercussion sur le développement de ces enfants, et obtenir une confirmation histologique.

KEYWORDS

Cavernoma, cavernous hemangioma, cavernous malformation, cavernous angioma, dural cavernoma, child, pediatric neurosurgery, dura mater, accessory venous sinus

MOTS CLES

Cavernome, hémangiome caverneux, cavernome dural, enfant, neurochirurgie pédiatrique, dure-mère, sinus veineux accessoire

INTRODUCTION

Cavernous hemangiomas account for 5 to 13% of all vascular malformations of the central nervous system[1]. The natural history of pediatric cavernous malformations seems to be more aggressive than in adult patients: these lesions have higher rates of growth and hemorrhage, larger dimensions, and often atypical radiological pictures at diagnosis. Concerning location, supratentorial compartment seems to be the most frequent[2]. Surgical outcomes are related to the preoperative neurological status of the children[3].

Herein, we report a case of a pediatric patient presenting an untypical cavernoma in several aspects: unusual dura mater location between sinus's slices and a large draining vein which seems to be a duplication of transverse sinus, has been found.

CASE REPORT

A 33-month-old boy was hospitalized in our institution for asthenia, loss of acquisitions and relative anorexia.

All biological tests performed were normal, and a CT-scan revealed a right occipital extra axial lesion, heterogeneous with some bone's scalloping, sharply delineated. Brain MRI showed a right occipital lesion with T1 spontaneous high-intensity, heterogeneous, hemorrhagic and low-intensity throughout T2 gradient's echo sequence with discrete mass effect (Fig. 1). The lesion showed a peripheral enhancement, and relations between sinuses was specified: lesion seems to be in contact with right transverse sinus, possibly contained in an accessory sinus (Fig. 2). This accessory sinus or a transverse sinus's duplication seems to drain the cavernoma (Fig. 3). Fundoscopy, EEG, and PET TDM were normal.

Two month later, control MRI showing extra-axial lesion in the right occipital area, in contact with the superior part of the transversal right sinus. Lesion's size increased, hemorrhagic changes were found. Considering these different elements and mostly the growth of the lesion, after a multidisciplinary staff meeting, we decided to perform an exeresis of the lesion.

After a right occipital craniotomy extending beyond the median line and the right lateral sinus centered on the lesion by means of neuro-navigation, the opening of the dura mater is carried out under an operative microscope, with a hinge to the lateral sinus. The median incision of the dura leads to a bleeding probably caused by the section of the accessory venous sinus draining the lesion. Hemostasis is obtained by vascular clips. The lesion seems to be located in a duplication of dura mater. Section

just above the right lateral sinus is performed without any bleeding of this sinus. Resection is carried out in totality; the adhesion with the cerebral parenchyma was minimal. The closure is carried out by means of an epicranial plasty and the osseous flap is replaced. No complications can be noted in the rest of the evolution. Histology result was according to cavernoma, showing proliferative vascular structures with several calibers, some hemorrhagic changes and hemosiderin deposits. This proliferative vascular structure was well delineated.

At follow-up, 1 month later, the patient had not presented any intercurrent event. One year later, the psychomotor development of the child is normal, and the MRI does not show recurrence (Fig. 4).

DISCUSSION

The management of intracranial extra cerebral cavernomas in children is complex. Confusion is relatively frequent between meningioma and cavernoma developed at the expense of the dura, hence F. B. Meyer et al.[4], who suggests making a semantic difference since the clinical and histological characteristics of these lesions are distinct from intra-axial cavernous hemangiomas. A more appropriate term may be "sinus cavernoma" to indicate that these lesions have primarily intra sinus origin[6]. This semantic distinction is even more interesting since the group of extra-axial cavernomas developed based on the dura mater can itself be divided into two groups according to Lewis et al.[5] and Di Vitantonio et al.[6].

The majority arise from the middle fossa; they are more clinically aggressive and more difficult to resect surgically. Other cases concerns sub-tentorial locations of extra-axial dural cavernomas. Controversy still exists regarding the exact nature of these extra-axial cavernous angiomas: vascular tumor versus vascular malformation, like intra-axial cavernomas. Biondi et al. conclude that intra-cranial extra-cerebral so-called cavernous angiomas present findings suggesting that they are vascular malformations, analogous to the intra-axial cavernous angioma[7].

After a review of the literature, we found out that the present case report is the third published case of an extra-axial dural cavernoma of the sus-tentorial convexity in a child. The two previous cases were those of Joshi and al., and Saldana and al.[8][9]. To our knowledge, this is the first description in the literature of a pediatric dural cavernoma associated with an accessory venous sinus well-defined in arteriography.

CONCLUSION

Intra and extra-axial hemangiomas are pathologically the same entity but intraoperative complications and management segregate them as two different clinical diseases. Moreover, their presentation is misleading as they mimic other extra-axial tumors. The management of these lesions is of two-fold interests. First, the aim consists on avoiding a repercussion on the development of these children and histology confirmation. The relative scarcity of reported cases leads to a vagueness in the epidemiological data of extra-axial cavernomas in children. A more exhaustive

collection and analysis of cases could shed light on their location, the effectiveness of surgery and possible complications.

CONFLICTS OF INTEREST:

No conflict of interest.

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FIGURES:

Figure 1

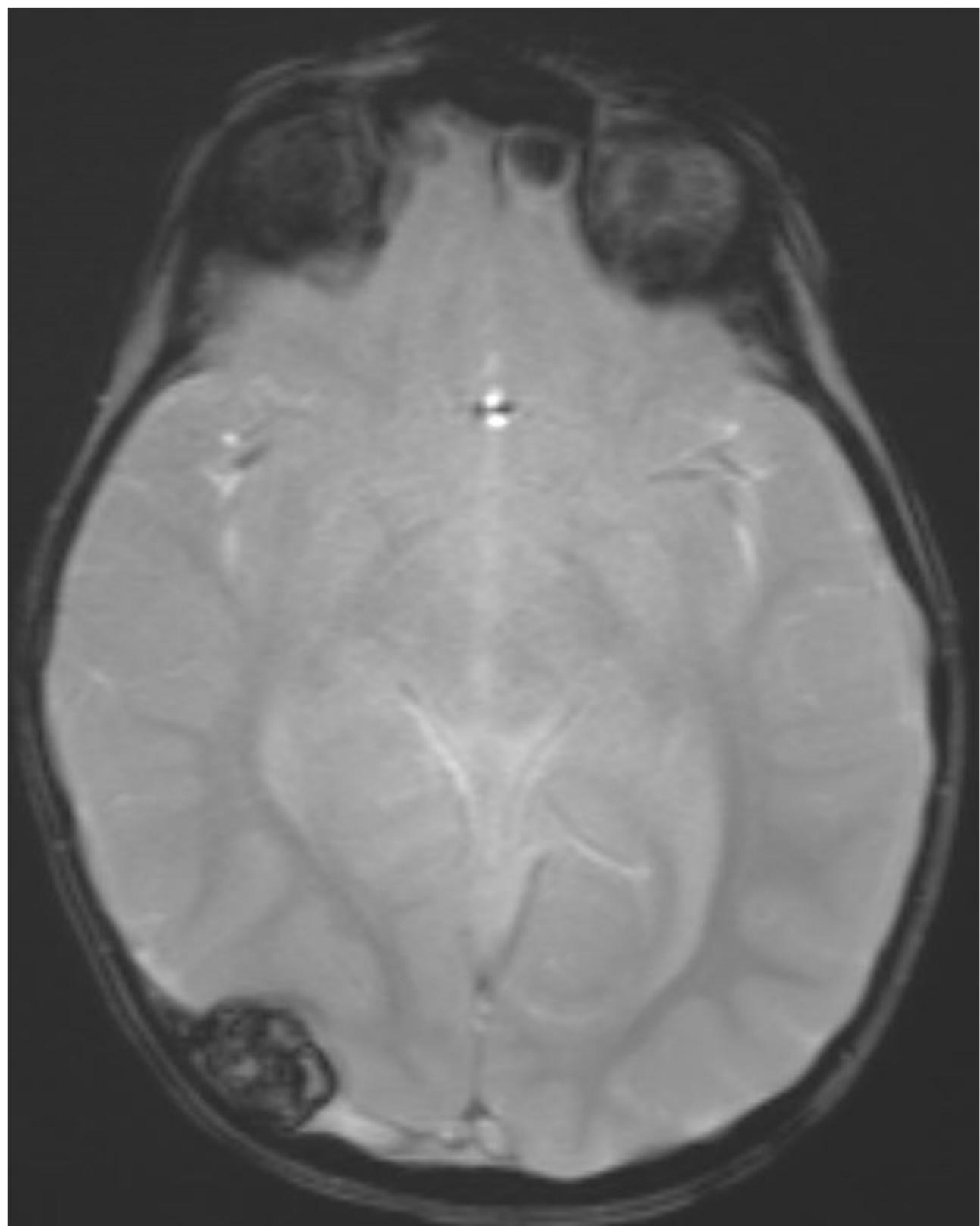


Figure 2

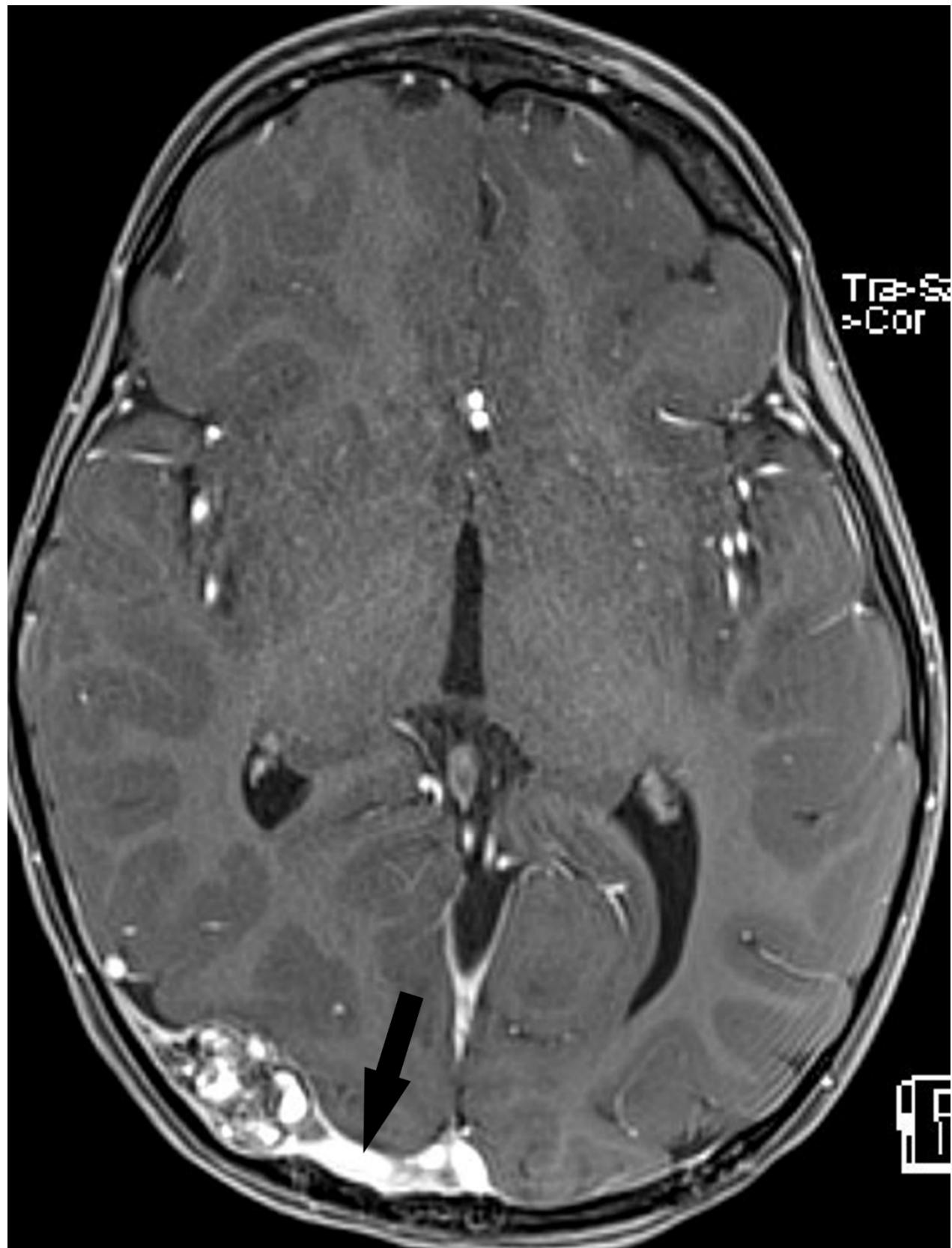


Figure 3

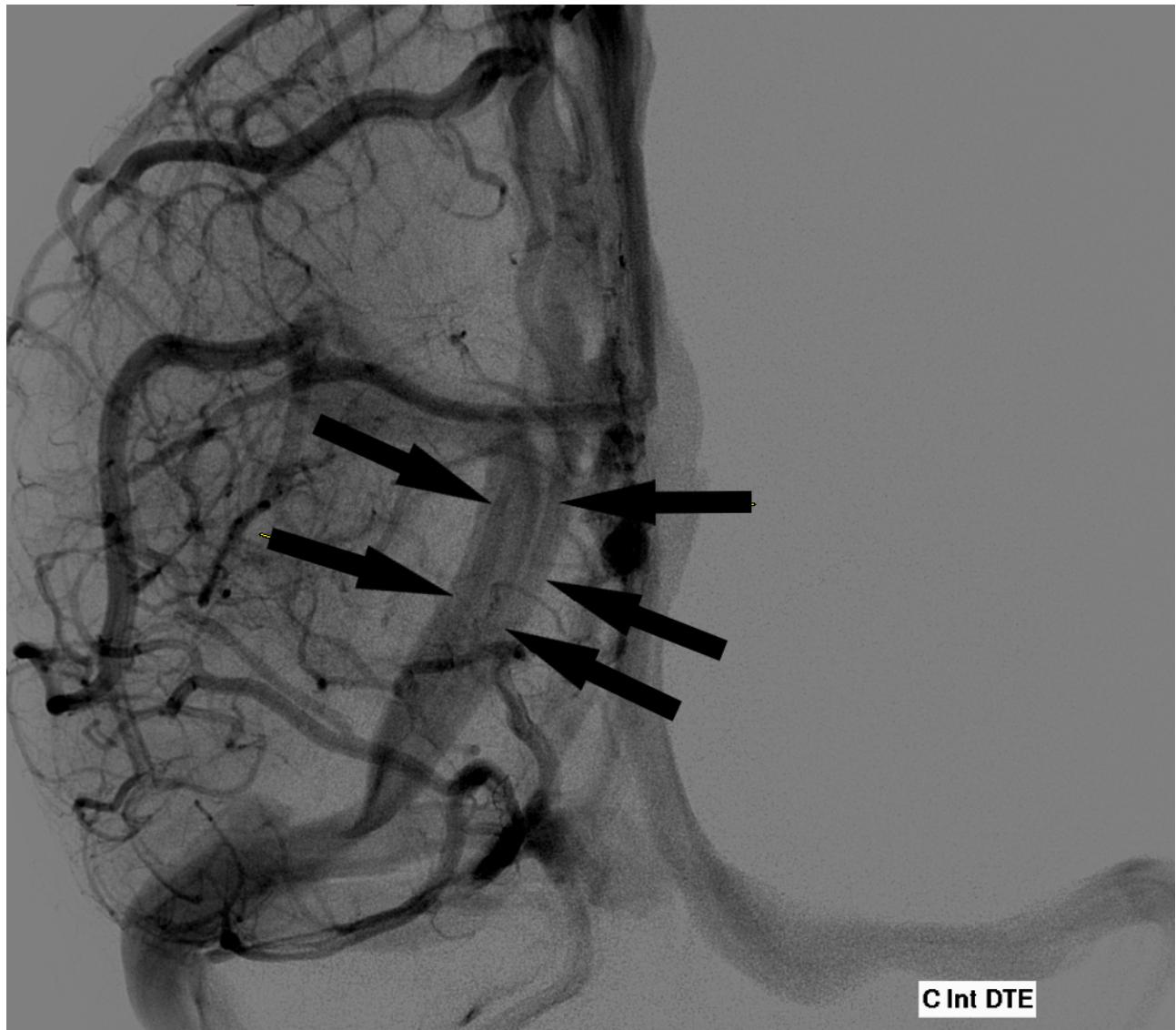
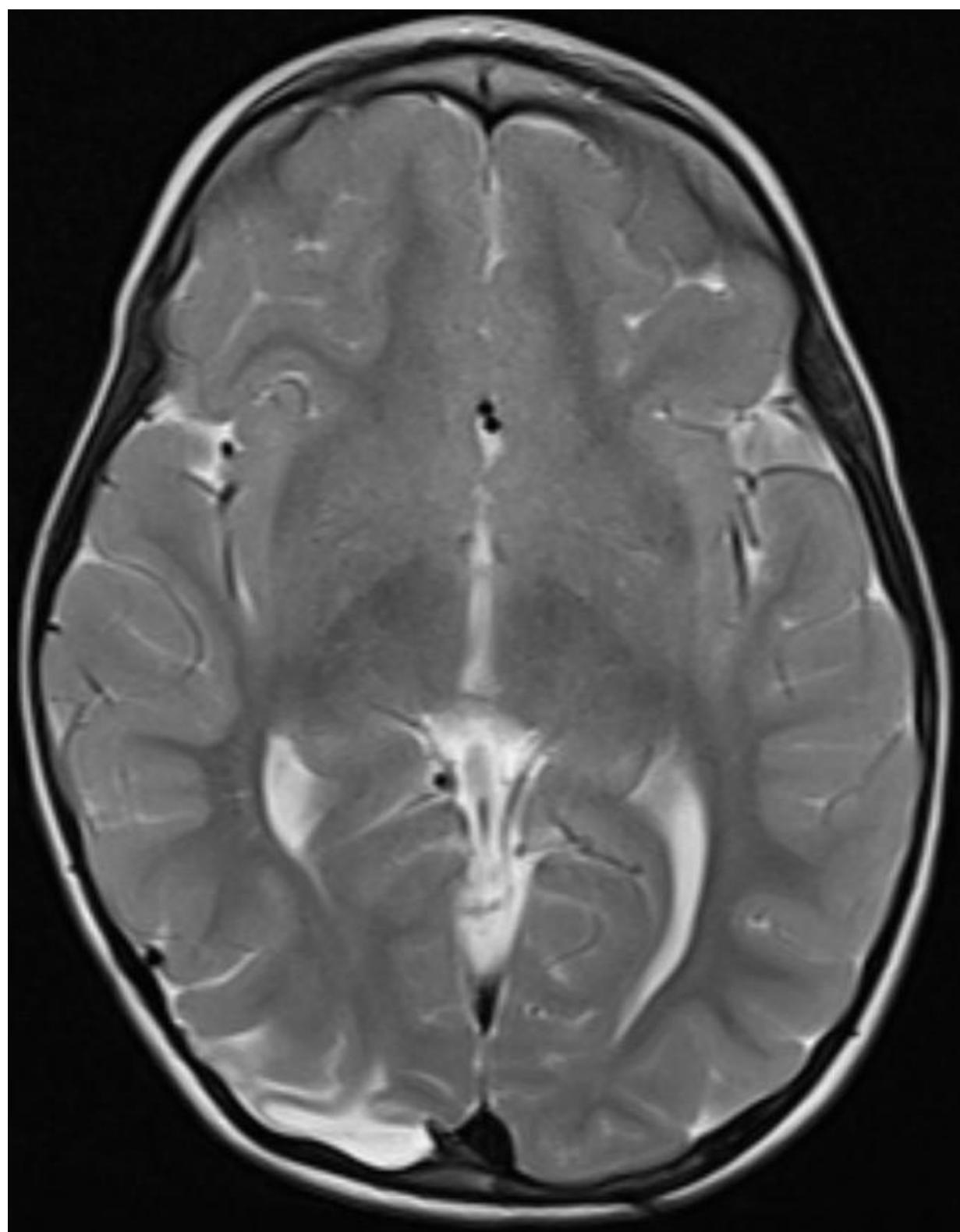


Figure 4



LEGENDS OF FIGURES :

Figure 1: MRI T2 gradient's echo: hemorrhagic and low-intensity lesion.

Figure 2: MRI T1 gadolinium: lesion seems to be contained in an accessory sinus (arrow), duplication of right transverse sinus.

Figure 3: Arteriography: accessory sinus or transverse right sinus's duplication (arrows) seems to drain the cavernoma.

Figure 4: MRI T2: one year after surgery, operative stigma without pathological character.