

# Multiple Cavernomatosis Presenting during Pregnancy

Gilad Yahalom MD<sup>1,2</sup>, Arie Yagoda MD<sup>5</sup>, Chen Hoffmann MD<sup>3</sup>, Orit Dollberg MD<sup>4</sup> and Natan Gadoth MD<sup>1,6</sup>

<sup>1</sup>Neurology Unit, Maayanei Hayeshua Medical Center, Bnei Brak, Israel

<sup>2</sup>Departments of Neurology, <sup>3</sup>Radiology and <sup>4</sup>Anesthesiology, Sheba Medical Center, Tel Hashomer, Israel

<sup>5</sup>IVF Unit, Herzliya Medical Center, Herzliya on Sea, Israel

<sup>6</sup>Sackler Faculty of Medicine, Tel Aviv University, Ramat Aviv, Israel

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Cavernous angiomas of the brain are quite rare. Their frequency, based on magnetic resonance imaging reports, is approximately 0.4% [1]. Multiple CA may be present in 6%–33% of patients with CA [2] and may be either clinically silent or present with a variety of neurological symptoms and signs. The utilization of magnetic resonance imaging with gradient echo sequence enables detection of multiple and even tiny CA due to the sensitivity of this sequence to the magnetic susceptibility induced by hemosiderin [3].

Symptomatic CA presenting first during pregnancy are rare. We reviewed the English-language medical literature and found only four articles, each describing a single symptomatic CA that was diagnosed during pregnancy. To the best of our knowledge, MCA first presenting during pregnancy have not been documented to date.

## PATIENT DESCRIPTION

A 30 year old previously healthy woman – gravida 3, para 1, abortion 1 – was admitted to the department of high risk pregnancies due to premature uterine contractures at 33+2 weeks of an otherwise uneventful twin pregnancy.

CA = cavernous angiomas  
MCA = multiple cavernous angiomas

Her first pregnancy was medically terminated because of anencephalus and the second normal pregnancy resulted in a healthy girl.

On the third hospital day she experienced vomiting, vertical diplopia and left hemifacial paresthesias without headaches or seizures. Neurological examination demonstrated that she was relaxed, alert and oriented to time, place and situation. She kept covering her left eye to prevent double vision. Cognition was normal and the neck was supple. Vertical diplopia was reported on left upward gaze. Ocular movements were full. Fundoscopic examination was unremarkable. Brisk reflexes with a mild increase in muscle tone and an equivocal plantar response were present on the left side. Cerebellar functions were normal. She reported left-face hypoesthesia and a change in the quality of taste, which was attributed to the magnesium she had received for premature contractions. Cranial MRI with GRE sequences showed at least 39 CA that were located in the cerebral hemispheres, brainstem and left middle cerebellar peduncle. Two bleeding CA were seen, one in the right midbrain and the other in the right thalamus, which was slightly impinging the wall of the third ventricle [Figure]. Small old hemorrhagic foci were seen in the parietal and frontal lobes, bilaterally. Routine complete blood counts, urinalysis, blood chemistry, coagulation function tests, chest X-rays and electrocardiogram were unremarkable.

Her clinical condition stabilized and during the 34th week of gestation she underwent an uncomplicated cesar-

GRE = gradient echo

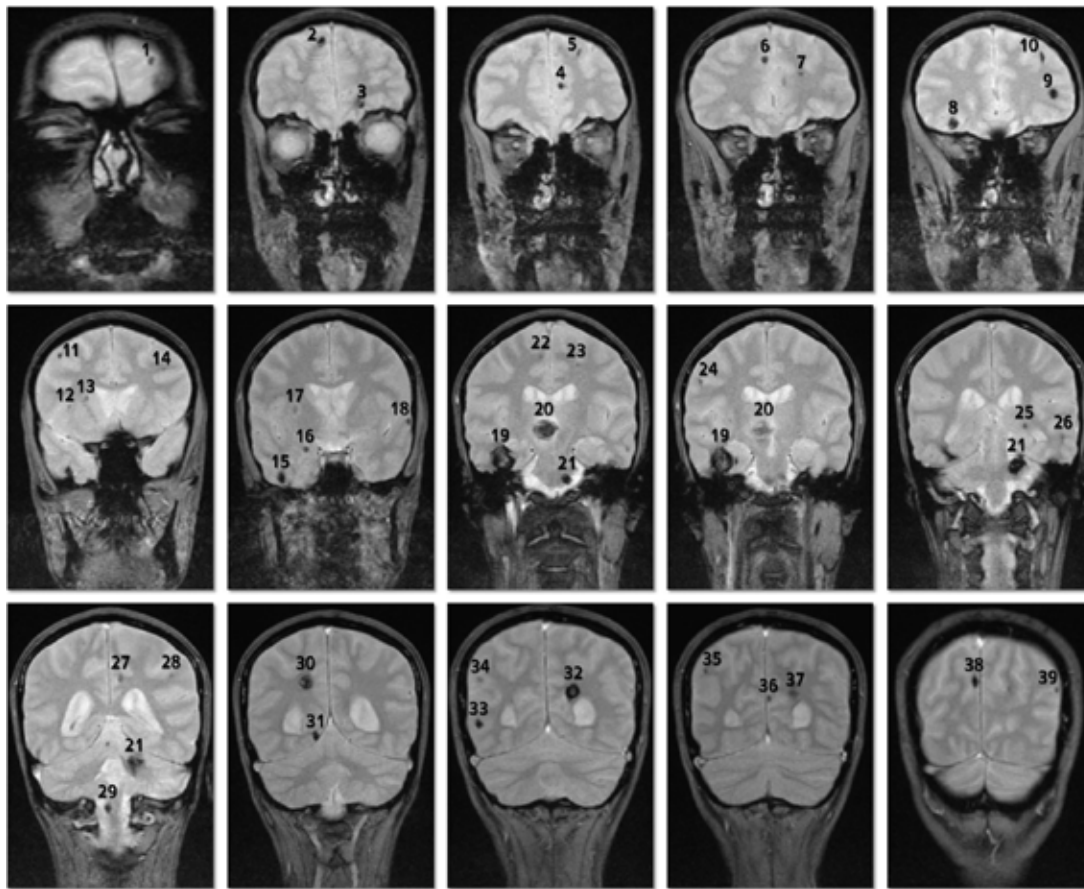
ean section under general anesthesia due to breech presentation. One of the twins, who weighed 1850 g, received mechanical ventilation for 24 hours and was discharged after 2 weeks. The other weighed 1500 g, had an uneventful postnatal course and was discharged after 3 weeks. The mother was well. The diplopia persisted for the following 3 months. At present she is asymptomatic and the twins are developing normally.

## COMMENT

CA first manifesting during pregnancy is a rarity. Pregnancy is a physiological state with a hyperdynamic circulation due to increased fluid volume and cardiac output. This state enhances turbulent blood flow. CA, which are congenital vascular malformations, have a sinusoid wall that is thinner than usual, hence the tendency of those malformations to bleed. Considering the above, it is surprising that CA that become symptomatic during pregnancy are so rare.

A single CA occurring during pregnancy was previously described in the form of single case reports in four women, all of whom presented with seizures. In contrast, our patient had neither seizures nor headaches but suffered from vomiting, vertical diplopia, mild pyramidal involvement of the left side and left facial paresthesias. Those signs and symptoms could be partially explained by the right midbrain bleeding CA.

As mentioned above, our patient is unique as there are no reports of women with MCA presenting during pregnancy. Another unique feature of this case is the



Brain MRI: A coronal T2 weighted GRE image (T2\*) showing at least 39 cavernous angiomas. Of those, the most prominent CA were in the right thalamus, the right temporal node and the left pons

total number of CA in a single patient. Zhao et al. [4] retrospectively studied 273 patients with radiological proof of CA and found 30 patients (19 males and 11 females) in whom a total of 79 CA had occurred. The highest number of CA detected in a single subject was 6, while our patient had at least 39.

The sensitivity of MRI performed with GRE sequences is well demonstrated in the present case. With routine T2 and FLAIR sequences only a small portion of the total number of CA were seen, while with GRE sequences the complete number of CA was revealed.

The described pregnancy was indeed high risk, with twins in breech presentation and a mother with MCA, two of which were bleeding, one of them located in the right thalamus, slightly impinging the third ventricle. All those variables were indeed an obstetric management challenge. Although a cesarean

section would probably have been performed for the twins' breech presentation regardless of the CA, the choice of a cesarean section could also decrease the possibility of raised intracranial pressure, which might be present during a traditional spontaneous delivery.

Since CA may exist also in the spinal cord, spinal MRI should be performed in a pregnant woman with brain CA, when epidural anesthesia is planned. This mode of anesthesia during labor is preferred when aiming to reduce the chance of induced increased intracranial pressure during natural delivery.

Those considerations should be kept in mind when a reproductive woman is diagnosed with CA. Such women should be carefully followed and as soon as they become pregnant a brain MRI including GRE sequences should be obtained. If MCA are detected the management of pregnancy and deliv-

ery should follow the above mentioned precautions.

**Corresponding author:**

**Dr. N. Gadoth**

Maayenei Hayeshua Medical Center, Bnei Brak 51544, Israel

**Phone:** (972-3) 577-5455

**Fax:** (972-30) 577-5454

**email:** gadoth@post.tau.ac.il

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