Case Report

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Lower-limb progressive paraparesis management and diagnosis overview in a pregnant woman with vertebral haemangioma

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Abstract

Vertebral haemangioma is a benign vascular tumour mostly seen in the thoracic region of the spine. Spinal haemangiomas are usually asymptomatic and are discovered incidentally. In a few patients, however, aggressive vertebral haemangiomas can cause local pain, radicular pain or neurologic deficits, which result from neural compression. The aetiology of the origin is unclear and is probably multifactorial. Hormonal and biological changes in pregnant women can lead to accelerated vascular growth of haemangioma. In our report, we present the case of a pregnant patient who was diagnosed with an aggressive vertebral haemangioma that further led to progressive paraparesis. We had to take the fact that she was pregnant into account in the diagnostic procedure, in the choice of examination method and also in the method of therapy. The goal of this case report is threefold: (1) provide an overview of the possible methods of management, specifically imaging, which will aid in diagnosis and based on that, (2) determining the appropriate therapy and (3) review the risks and benefits of each will be presented when choosing individual approaches.

Keywords

bleeding, magnetic resonance imaging, paraparesis in pregnant woman, vertebral haemangioma

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Introduction

Vertebral haemangiomas are usually asymptomatic and discovered incidentally.[1,2] The possibility of a haemangioma increasing in size during pregnancy is well known and is related to altered progesterone and oestrogen levels and/or obstruction of paravertebral veins draining into the inferior vena cava by the gravid uterus.[3] The enlarged haemangioma then produces symptoms in the form of local bone pain, radicular pain or compressive myelopathy.[4] The type and timing of treatment for symptomatic haemangiomas during pregnancy are challenging.[5] Our goal is to provide an overview of the possible methods of diagnosis and therapy of vertebral haemangiomas in a female patient at 23 weeks of pregnancy and the potential risks that should be taken into account when choosing individual treatment approaches.

Case report

A 26-year-old patient, in the 23rd week of pregnancy, was acutely admitted to the neurological department on 21 January 2021, with a 1-month progression of bilateral

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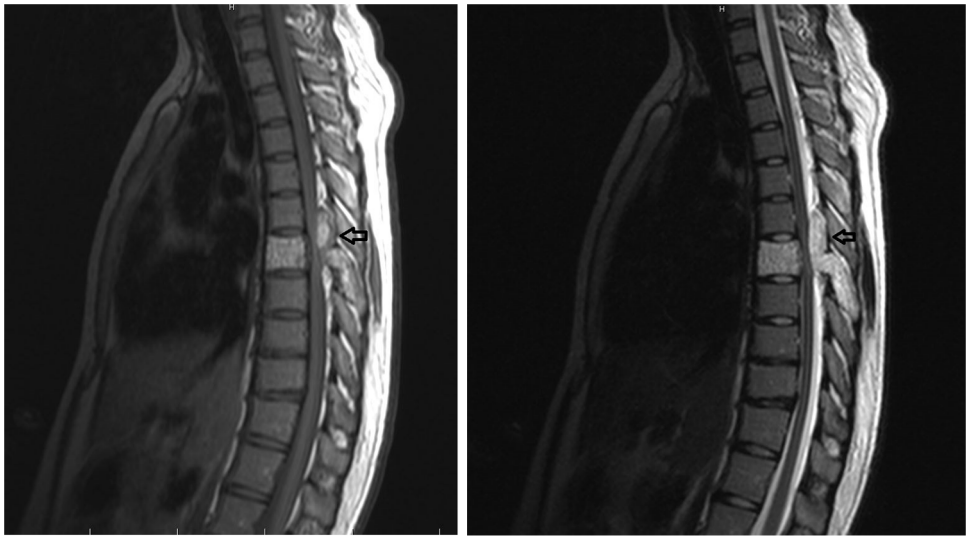

Figure 1. (a) MRI T1-weighted image. Sagittal projection. The lesion affects the entire vertebral body, arches and processes and propagates from the bone structures to the surroundings, invading the spinal canal with canal stenosis, compression of the structures of the dural sac and thus also the spinal cord. The lesion is hyperintense in the T1, T2 and T2 fat-suppressed weighted image. (b) MRI T2-weighted image. Sagittal projection. The lesion affects the entire vertebral body, arches and processes and propagates from the bone structures to the surroundings, invading the spinal canal with canal stenosis, compression of the structures of the dural sac and thus also the spinal cord. The lesion is hyperintense in the T1, T2 and T2 fat-suppressed weighted image.

lower extremities weakness. The patient was without a significant medical history. According to anamnestic data, the walking deficit first appeared at Christmas 2020. The weakness was more pronounced on the right, but gradually increased on the left lower limb, too. Tingling and numbness were present in both legs, and sensation was reduced from thoracic (Th) 11–12 dermatome downwards. A week before admission she did not consent to hospitalization due to the Covid-19 pandemic, but the day before admission she was no longer able to walk on her own. The patient had not previously suffered any injuries or taken any medication during pregnancy. In the ninth week of pregnancy, she experienced vaginal bleeding, and as a result, she took sick leave for the remainder of her pregnancy.

Physical exam

At admission, a neurological examination revealed severe central paraparesis more pronounced on the right side (bilateral positive Babinski sign, increased muscle tone in the patient, with weakness of both lower extremities, patient was only able to perform mild plantar flexion), and distal hypoesthesia. History and remainder of the physical exam revealed a positive straight leg raise test in the end position and mild oedema of the lower extremities; her spine was not painful to the touch; urgent urination corresponded to pregnancy and no other sphincter problems were present.

Laboratory parameters tests

Laboratory tests were without pathology: blood sugar 4.7 mmol/L (reference range: 4.1–5.9 mmL/L), Hb 13.30 g/dL (13–18 g/dL), erythrocytes 4.19×10^{12}/L ($4.4–5.9 \times 10^{12}$/L), white blood cells 9.17×10^{9}/L ($4–10 \times 10^{9}$/L) and platelets 147×10^{9}/L ($150–400 \times 10^{9}$).

Imaging technique

Since the presumed location of the pathology was in the thoracic region of the spinal cord, magnetic resonance imaging (MRI) was performed. We decided on MRI in a 1.5 Tesla (T) device due to the possible risk of overheating during examination in a 3.0 T device.[6] MRI showed the presence of a soft-tissue tumour of the Th seventh vertebra. The lesion affected the entire vertebral body, arches and processes and extended to the spinal canal causing spinal cord compression. The spinal cord in the compressed segment showed signs of myelopathy. The lesion was hyperintense in the T1, T2 and T2 fat-suppressed weighted image. The finding was evaluated by a radiologist as a vascular tumour in the Th7 vertebra region, resembling a

benign haemangioma. However, the possibility of another type of vascular soft-tissue tumour, including a rare Masson tumour, was not ruled out. (Figure 1(a) and (b)).

Procedures

A neurosurgeon, an interventional radiologist and a gynaecologist were consulted simultaneously. After multidisciplinary consensus, the options of further action and their potential risks were explained to the patient by the neurosurgeon. One option was to watch and wait approach, which means to observe the haemangioma for any changes that may require immediate intervention and if there are none, to determine an appropriate intervention following birth. With this option, the patient reversible spinal cord injury with subsequent paraparesis was highly likely. Another option was initial CT angiography with embolization of the supplying segmental arteries to reduce postoperative blood loss, but with the risk of endangering the foetus. The third treatment option, ultimately chosen by the patient, was to perform acute decompression of the spinal cord, while taking into account the risk of high blood loss and therefore placental hypoperfusion and abortion.

Endovascular embolization can be used in less severe haemangiomas as a stand-alone method to reduce compression, or in more severe conditions preoperatively to reduce the risk of bleeding complications. Percutaneous vertebroplasty (which reduces tumour vascularity), alcohol ablation, and radiation therapy are used for milder lesions alone or in various combinations with these methods.[7]

Before surgery

A gynaecological examination was performed to examine the health and development of the foetus and the potential risk of foetal complication with surgery. There were no signs of endangering the foetus, and the foetus was normally developed. The gynaecologist recommended the preparation of foetal lung maturity due to the risk of impending preterm birth at the diagnosis. Therefore, 4 g of magnesium sulfuricum intravenously (i.v.) up to 500 mL of sodium chloride physiological solution every 24 h and 6 mg of dexamethasone every 12 h applied intramuscularly up to a total dose of 24 mg were administered to the patient.

Surgery

The patient was operated on the next day. Surgery was performed in the left flank position, and a Th6–Th8 laminectomy and tumour extirpation from the spinal canal were performed for histological examination. The operation resulted in extreme bleeding, the patient was transferred to the Department of Anaesthesiology, and the viability of the foetus was repeatedly confirmed by gynaecological examination. Based on the gynaecologist's recommendation, tocolytic treatment was given (4 g of magnesium sulfuricum i.v. up to 500 mL sodium chloride physiological solution every 12 h) and corticoid preparation of lung maturity was also continued in case of the need to terminate the pregnancy early (4 mg of dexamethasone every 6 h i.v. up to a total dose of 24 mg). Blood loss was compensated for using erymass. Histological examination confirmed the diagnosis of vertebral haemangioma. The patient was discharged to outpatient care 1 week following surgery. At discharge, the neurological status showed improvement; the patient elevated legs above the mat with mild imbalance, performed plantar and dorsal flexion and walked using a walker.

Delivery

Before the expected date of delivery at the 39th week of gestation, the patient was hospitalized in the Department of Gynaecology and Obstetrics and an elective termination of the pregnancy by Caesarean section was performed, as vaginal delivery would have posed a great risk to both mother and foetus. At that time, the patient was already walking without the assistance of the walker. The patient gave birth to a healthy baby by caesarean section without any complications (girl: 4030 g/50 cm, Apgar score: 9/1'min, 10/5'min, 10/10'min). After 96 h, mother and baby were released from the hospital. No significant histological findings were found in placental tissues (fibrosis, etc.) that could confirm uteroplacental hypoxia during neurosurgery at 23 weeks of gestation.

Follow-up neurological examination

At the follow-up neurological examination, 8 months after surgery, an occasional tingling sensation in the right lower leg was present, without motor deficit. At the same time, an MRI control was performed and showed that the haemangioma filled the whole Th7 vertebral body and its residual arch, but the spinal canal was without stenosis and the spinal cord was not compressed. MRI showed signs of spinal myelopathy ranging from the Th6/7 vertebra to the Th7/8 vertebra. (Figure 2(a) and (b)) The patient was referred for oncology consultation regarding further action.

Discussion

Vertebral haemangioma is a benign vascular tumour that results from abnormal proliferation of blood vessels. It is most commonly seen in the thoracic region of the spine, less often in the lumbar region, and rarely in the cervical or cruciate region.[1,3] Spinal haemangiomas are usually asymptomatic and are discovered incidentally.[1,2] The aetiology of the origin is unclear and is probably multifactorial.[1] Hormonal and biological changes in pregnant women can lead to

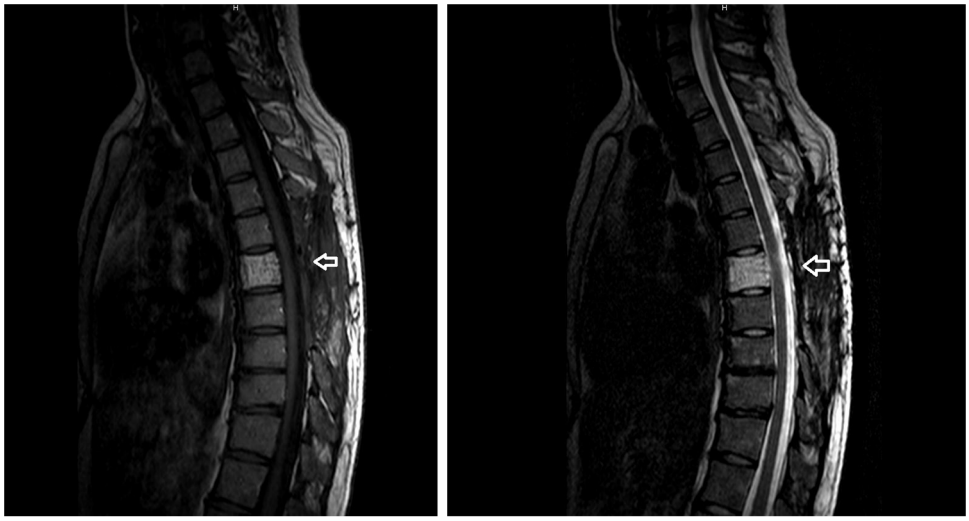

Figure 2. (a) Control MRI after surgery. MRI T1-weighted image. Sagittal projection. Haemangioma fills the whole Th7 vertebral body and its residual arch, but the spinal canal is without stenosis and the spinal cord is not compressed. MRI shows signs of spinal myelopathy ranging from the Th6/7 vertebra to the Th7/8 vertebra. (b) Control MRI after surgery. MRI T2-weighted image. Sagittal projection. Haemangioma fills the whole Th7 vertebral body and its residual arch, but the spinal canal is without stenosis and the spinal cord is not compressed. MRI shows signs of spinal myelopathy ranging from the Th6/7 vertebra to the Th7/8 vertebra.

accelerated vascular growth and distension until the entire vertebral body is affected, and nerve structures (spinal cord or nerve roots) are compressed by the extradural growth.[3,4]

There are few cases of pregnancy-related vertebral haemangiomas in the literature. The majority of them presented during the third trimester and followed by the second trimester.[4,8] Symptoms may include local back pain, leg paresthesia, spastic paresis and incontinence. Spinal cord or root symptoms appear during aggressive tumour growth into the epidural space, with subsequent compression of the spinal cord and/or nerve roots.[1,3,9]

Symptomatic, aggressive haemangiomas of the vertebrae require treatment. To date, there is not enough sufficient evidence to determine the optimal option, thus the options remain controversial. Modalities, such as radiotherapy and embolization, are not suitable for pregnant women, and surgery during pregnancy has a risk of preterm labour. Possible treatment options include surgical resection, decompressive laminectomy, vertebroplasty, endovascular embolization and radiotherapy.[3,10] When deciding on treatment, it is also necessary to consider the benefit/risk ratio of the examinations that precede the treatment itself.

Imaging method

MRI plays a key role in diagnosis and thus far appears to be safe, though its possible side effects are still not ruled out.[11,12] A study from Ontario, Canada,[13] revealed that exposure to MRI in the first trimester compared with non-exposure was not associated with an increased risk of harm to the foetus. The theoretical risks of undergoing MRI are biological effects of static and a time-varying magnetic field, the thermal effects associated with radiofrequency exposure and the acoustic noise.

The effect of static and a time-varying magnetic field: Studies performed on chicken embryos confirmed a higher incidence of abnormal embryos and their damage after exposure to a strong static and switched magnetic field gradient in the first 48 h of life. Although these studies are not applicable to humans, they still provide sufficient cause for concern.[14] Therefore, MRI in the first trimester of pregnancy should be performed only in cases of clinical need. There are currently no studies demonstrating adverse effects when a pregnant patient is scanned at 1.5 or 3 T in any trimester, but depending on the circumstances, an MRI at 1.5 T or an MRI at 3 T may be recommended.[12]

Thermal effect: Overheating may increase the risk of birth defects. Tissue heating from MRI procedures is greatest at the surface of the mother's body, while at the centre of the body it is negligible; therefore, it is unlikely that thermal damage to the foetus would pose a serious risk.[5,15]

Acoustic noise caused by switching on and off the gradient coils during scanning as a potential risk of acoustic damage has not yet been confirmed in studies using 1.5 and 3 T MRI during the second and third trimesters of pregnancy. There is no evidence of damage to the foetal auditory organs during the first trimester; if clinically

necessary, MRI in the first trimester at a field strength of 1.5 T is generally considered safe.[6,12]

Contrast medium

Gadolinium contrast media are contraindicated in pregnant women. Free gadolinium is toxic and therefore is administered only in chelated (bound) form. Gadolinium-based contrast agent (GBCA) is water soluble and can cross the placenta into foetal circulation.[15] GBCA is excreted through the foetal renal system into the amniotic fluid, which the foetus regularly swallows, so it is absorbed through the foetal gastrointestinal tract back into the foetal blood stream. The longer gadolinium-based products remain in the amniotic fluid, the greater is the potential for dissociation from the chelate and, thus, the risk of causing harm to the foetus.[16,17] While in a retrospective review of a Canadian provincial database of births, exposure to GBCAs was associated with an increased risk to a child at any time during pregnancy, further analysis confirmed this association only in the first trimester.[18]

We decided on thoracal spinal cord MRI without the use of a contrast medium. Due to pregnancy and the proximity of the examined area to the foetus, we decided to use a 1.5 T device. Before selecting a magnetic resonance with a magnetic field strength of 3 and 1.5 T, it is necessary to evaluate whether the 1.5 T intensity field will answer our clinical question and whether the result will affect patient management. No adverse effects were demonstrated in the studies for 1.5 T, while for 3 T MRI we still do not have enough studies for pregnant women; therefore, the use of a 1.5 T device is preferred if both options are available, especially in the first trimester of pregnancy.[12,19]

Treatment

After receiving the results from the MRI, we faced a decision on the next steps. Due to the significant worsening of paraparesis over the previous 24 h, we consulted a neurosurgeon regarding the acute decompression. An interventional radiologist and a gynaecologist were consulted, as well.

In the treatment of vertebral haemangiomas with progressive neurological deficiency, prompt surgical decompression with total or partial resection of the tumour is preferred, with histological identification of the lesion.[1] This procedure is associated with a high risk of intraoperative bleeding and the development of postoperative epidural haematoma, which should be considered in advance. The rapid progression of the symptoms could have been associated with a recurrence of the lesion, and therefore postoperative radiotherapy is recommended to reduce the risk of recurrence.[5] The best treatment of aggressive vertebral haemangioma in pregnancy is still not clear, but, from our experience and a review of the literature, the suggestion should be as radical as possible. We cannot forget that communication with the patient and her decision is also very important.

Conclusion

Vertebral haematoma is a rare cause of progressive paraparesis and when it occurs in a female patient during pregnancy means deciding between risk of permanent paraplegia and the life of the foetus. A multidisciplinary approach involving neurologists, neurosurgeons, gynaecologists and interventional radiologists is essential in decision-making, but discussion with the patient and her family and their opinions must also be taken considered. The role of imaging is critical in the evaluation of a haemangioma, and we discussed the benefits and risks of different imaging procedures. Pregnancy-related symptomatic vertebral haemangiomas with progressive or severe neurological deficit due to spinal cord compression are candidates for immediate surgical decompression.

Author contribution(s)

Lívia Ridzoňová: Conceptualization; Investigation; Methodology; Project administration; Writing – original draft.
Miriam Fedičová: Conceptualization; Investigation; Project administration; Supervision; Writing – review & editing.
Tomáš Andráš: Data curation; Investigation; Visualization.
Peter Urdzík: Data curation; Investigation; Visualization.
Zuzana Gdovinova: Conceptualization; Formal analysis; Methodology; Project administration; Resources; Supervision; Writing – review & editing.

Declaration of conflicting interests

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Statement of ethics

No Statement of Ethical Approval from the ethics committee was required due to the nature of this case report. The patient declared written consent to the publication of her case, including the publication of images.

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