Short report

Epidural haemangiomas during pregnancy

EHUD LAVI,* DARA G JAMIESON,† MENACHEM GRANAT‡

From the Departments of Neurology* and Obstetrics and Gynecology‡, Hadassah University Hospital, Jerusalem, Israel, and Department of Neurology,† Hospital of the University of Pennsylvania, Philadelphia, Pennsylvania, USA

SUMMARY Two cases of vertebral haemangiomas are reported which presented as spinal cord syndromes during pregnancy. Eleven additional cases of epidural haemangiomas in the literature which became symptomatic during pregnancy are reviewed. In 11 out of 13 cases symptoms presented during the third trimester of pregnancy and in all but two cases the epidural lesions were in the upper six thoracic vertebrae. These features can be explained by the effect of the gravid uterus on the relatively sparse vascular supply of the upper thoracic spinal cord.

Haemangiomas of the spinal epidural space can be divided into tumours which arise primarily from within the spinal epidural space and vertebral haemangiomas which extend into the epidural space. Pure epidural haemangiomas are rare^{1,2} and almost always occur at the low thoracic level. Vertebral haemangiomas are extremely common lesions which were found in 10% of 10000 vertebral columns examined at necropsy^{1,3}; however, they rarely cause symptoms due to root or cord compression.⁴ Pregnancy has been implicated in the production of myelopathic symptoms due to vertebral haemangiomas.

This paper reports two cases of vertebral haemangiomas causing spinal cord syndromes in pregnancy and reviews the reported cases of epidural haemangiomas presenting during pregnancy. We speculate on the mechanism of symptoms produced in pregnancy especially as they relate to the vertebral blood supply and the haemodynamic and endocrinological changes of pregnancy.

Case reports

Case 1

A 25-year-old female, a primipara in her 26th week of pregnancy, was admitted to the high risk pregnancy unit of the Hadassah Hospital after ultrasound diagnosed a quadruplet pregnancy. The patient conceived during the first cycle of HMG-HCG therapy for secondary ovarian failure due to

hypogonadotropism. On admission the patient was in good general condition. After 2 weeks of bed rest and prophylactic isoxsuprine hydrochloride 60 mg/day orally she started to complain of low back pain. One week later she complained of paraesthesias in her thighs, difficulty in walking and hypaesthesia in the lower part of her body. Neurological examination at that time revealed paraparesis with brisk tendon reflexes, an extensor plantar reflex on the left and a decrease in all sensory modalities below the level of T4 with sacral sparing. She deteriorated over a period of a few hours to a state of paraplegia and urinary retention. A myelogram revealed a complete extradural block at the level of T4. The patient, in her 29th week of pregnancy, was placed in the left lateral decubitus position and a T4-T6 laminectomy was performed. A vascular tumour invading the T4 vertebra was removed which was diagnosed histologically as a cavernous haemangioma. One hour after the operation uterine contractions began and an hour later, four premature male infants, weighing between 1020 and 1200 grams, were delivered by a low segment caesarean section. The post partum course was uneventful; neurological recovery was rapid and she returned to normal within two weeks. One infant died of intracerebral haemorrhage; another was treated for secondary hydrocephalus; the two remaining infants are developing well.

Case 2

A 21-year-old female, a primipara in her 34th week of pregnancy, was admitted complaining of leg pain for the past 2 months, at first on the right, then bilaterally, and progressive bilateral leg weakness. Neurological examination on admission revealed spastic paraparesis, brisk tendon reflexes in her legs, bilateral extensor plantar responses and a decrease in all sensory modalities below the level of T6 with sacral sparing. A myelogram revealed an extradural block at T4. It was decided to perform a caesarian section prior to laminectomy to avoid risk to the foetus and to facilitate positioning of the patient during laminectomy. Immediately after a normal

*Present address and address for reprint requests: Dr Ehud Lavi: Department of Microbiology/G2, University of Pennsylvania School of Medicine, 36th and Spruce Streets, Philadelphia, Pennsylvania 19104, USA.

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weight male infant was delivered by low segment caesarian section a laminectomy at T2 to T5 was performed and an extradural, highly vascular tumour was removed. It originated from the vertebral body of T2 and extended about 8 cm into the canal. It was histologically diagnosed as a cavernous haemangioma. Her post operative course was uneventful and neurological recovery was rapid. The patient was able to walk 10 days after the operation.

Discussion

Haemangiomas of the spinal column are benign vascular tumours of cavernous or capillary structure which are thought to be due to an embryonic dysraphic disturbance which affects the differentiation of capillaries and other vessels joining arteries and veins.⁵ Vertebral haemangiomas are usually solitary and are symptomatic most often in the mid and low thoracic and lumbar levels^{6,7} and less often in the cervical region.^{8,9} The lesion can have multiple presentations: without symptoms, discovered incidentally² with symptoms of local and/or radicular pain or with spinal cord symptoms. The neurological symptoms may be due to: (1) hypertrophy of the posterior cortex of the vertebral body and/or enlargement of the lamina and facets subsequent to angiomatous invasion; (2) subperiosteal growth of the haemangioma forming an epidural mass covered with periosteum; (3) compression fracture of the involved vertebral body;^{4,10} (4) haemorrhage¹¹; (5) spinal cord ischaemia.

The table shows the 13 cases from the literature, including the two reported in this paper, of epidural haemangiomas symptomatic during pregnancy. Nine cases involved known vertebral haemangiomas. In cases 1 and 5 no mention was made of an associated vertebral lesion and case 4 showed a discrete, entirely extradural vascular tumour without mentioning a vertebral lesion. In case 10 there was a venous channel in the pedicle of T4 but no obvious vertebral haemangioma. A case (reference 18, case 3) involving a vertebral angioma at L3 but in which symptoms were due to a T11 intradural angioma was not included. In their review of the literature Fields and Jones¹² quoted extensively from a case by Delmas¹³ citing it as a "spinal epidural haemangioma" when in fact the original paper described an intradural haemangioma.

In seven out of 12 cases the symptoms presented during the first pregnancy although diagnosis was not always made at that time because of post partum remission. In 11 out of 13 cases symptoms presented during the third trimester of pregnancy, often weeks to days before term. In case 6 one episode of transitory chest dysaesthesias occurred at 6 months, then leg weakness presented at 7 to 8 months. In case 10 leg paraesthesias occurred during the 4th and 5th months and leg weakness presented during the 6th month. In

all cases but two (cases 5 and 7) the epidural lesions were in the upper six thoracic vertebrae.

It has been speculated that the presentation during pregnancy of otherwise asymptomatic spinal vascular tumours is due to mechanical and/or endocrinologic mechanisms.^{1,12,14-18} By closer examination of these possible mechanisms some of the features of epidural haemangiomas presenting during pregnancy can be explained.

The predilection for the upper thoracic levels is striking in view of the occurrence, in the non pregnant state, of epidural haemangiomas at all levels from cervical to lumbar. The region from T3 or T4 to T8 has an afferent arterial supply arising from the aorta at the level of T7 and no continuous perimedullary anastomotic pathway resulting in a poor vascular supply.¹⁹ This is in contrast to the regions above and below which are highly vascularised. Since the system of venous plexi draining the spinal cord is valveless, increased intrathoracic and intra-abdominal pressure can result in shunting of distal venous blood into the internal venous plexi then to the superior vena cava.²⁰ The upper thoracic epidural space, because of its poor arterial supply and the potential reversibility of its venous drainage, is an area in which a vascular tumour could remain quiescent until it became engorged causing spinal cord symptoms, thus explaining the location of the symptomatic haemangiomas. The vascular anatomy would also explain the relatively acute onset of symptoms in the third trimester as the rapidly enlarging uterus caused increased intra-abdominal and intrathoracic pressure and also inferior vena cava compression in the supine position.²¹ If the lesion is not treated the symptoms and signs usually completely resolve (cases 1, 2, 3, 5, 8, 9) or remit considerably (cases 4, 6) in the early post partum period. This could be explained by the rapid decrease in the pressure effect of the gravid uterus and the correction of the venous blood flow reversal.

There could be other factors contributing to epidural vascular engorgement. In pregnancy there is a 20-100% increase in plasma volume, which begins in the first trimester, accelerates during the second trimester with a slower rise in the last three months.²² The mean increase in plasma volume is greater in multigravidae than in primigravidae.²³ This may explain why myelopathic symptoms become more severe in subsequent pregnancies (cases 1, 2, 3, 5, 8, 9). Structural alterations of blood vessels may occur due to the hormonal changes of pregnancy. Increased venous distensibility may be due to increased blood levels of maternal progesterone during pregnancy. Decreased total peripheral resistance and increased cardiac output occur during the luteal phase of the normal menstrual cycle.²⁴ An increase in circulating oestrogens has been associated with the appearance in

Table Extradural haemangiomas presenting as spinal cord compression during pregnancy: review of the literature.

Author	Age at Diagnosis	Number of pregnancy (a) symptomatic (b) at diagnosis	Trimester at first Symptoms	Localisation	Pathology
1. Balado ²⁶ 1928	36	(a) 1, 2, 3, 4, 5, 7 (b) 8	3	T2-T7	epidural angiolioma
2. Michon ²⁸ 1935	34	(a) 4, 5 (b) 6	3	T4	vertebral haemangioma
3. Guthkelch ²⁷ 1948 (case 4)	34	(a) 2 (b) 3	3	T5	vertebral haemangioma
4. Lam ¹⁶ 1951	36	9	3	T3	extradural haemangioma
5. David ¹⁵ 1952	30	(a) 5, 6 (b) 7	3	T12	extradural haemangioma
6. Fields ¹² 1957	30	1	2	T6	cavernous haemangioma (vertebral)
7. Askenasy ⁶ 1957	20	—	3	T11	vertebral haemangioma
8. Acquaviva ¹⁴ 1957	40	(a) 5, 6 (b) 7	3	T3	vertebral haemangioma
9. Newman ¹⁸ 1958 (case 1)	24	(a) 1 (b) 2	3	T2	vertebral haemangioma
10. Newquist ¹ 1960	19	1	2	C7-T4	extra and intradural arterial angioma
11. Nelson ¹⁷ 1964	16	1	3	T3	cavernous haemangioma (vertebral)
12. Lavi 1985 (case 1)	25	1	3	T4	cavernous haemangioma (vertebral)
13. Lavi 1985 (case 2)	21	1	3	T2	cavernous haemangioma (vertebral)

pregnancy of spider nevi and palmar erythema and may contribute to venous shunting. Hormonal changes may be a factor in intradural haemangiomas which become symptomatic during menstruation²⁵ or early pregnancy.¹³ We were unable to find cases of epidural haemangiomas which became symptomatic during these periods. The mechanical mechanism would seem to be the major explanation for the location, the trimester of presentation and the post partum remission of epidural haemangiomas presenting during pregnancy.

According to the literature, the method of treatment of epidural spinal cord compression presenting during pregnancy is controversial. In three of the cases laminectomy was performed prior to delivery. In case 10 laminectomy was performed at 6 months gestation with a caesarian section delivery two months later. In cases 3 and 11 laminectomies were performed at 36 weeks and 7 months gestation respectively. In case 3 spontaneous delivery occurred four days after operation. The infant survived but the woman died of post-operative complications. The infant in case 11 was delivered spontaneously two days after operation and did not survive. When laminectomy was deferred because of spontaneous remission after delivery symptoms often recurred or did not completely disappear necessitating surgery at a later time. In cases 4, 5, 6, 8, 9, when surgery was done up to a few months after delivery neurologic

recovery was eventually complete. Surgical resection was the primary mode of treatment in all cases, with radiotherapy being an adjunct in some cases. Labour began immediately after laminectomy in our first case necessitating an emergency caesarian section. In our second case the caesarian section was performed first followed by laminectomy. We believe that when the foetus is immature according to conventional criteria every effort should be made to prevent premature labour. In cases of a mature foetus there is not enough data from the literature to indicate whether delivery, either vaginal or by caesarian section, should be induced prior to or following the laminectomy. However it would seem reasonable to recommend surgical resection of an epidural haemangioma at a time when, should premature labour occur post-operatively, a viable foetus could be delivered.

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