SPINAL ANGIOMA PRESENTING DURING PREGNANCY*

CAPTAIN RICHARD E. NEWQUIST, (MC) USAF,† AND FRANK H. MAYFIELD, M.D.‡

(Received for publication February 19, 1959)

Spinal angioma, with symptoms occurring during pregnancy and with remission beginning prior to delivery, has not been reported previously. For this reason the following case is presented.

CASE REPORT

N.S., an 18-year-old white female, noted tingling in her right leg when she was in the 4th month of her 1st pregnancy. In the 5th month similar paresthesias developed in the left leg. By the 6th month weakness of the right lower extremity was observed. She was admitted to the Good Samaritan Hospital, Cincinnati, on July 2, 1957. In addition to the above symptoms, she complained of aching pain in the lower cervical region.

In retrospect, the patient recalled that periodically during her teens she had had unusual cramping in both legs.

Examination. The patient was a well-developed, well-nourished young white female, whose uterus was enlarged to just above the umbilicus. Fetal heart tones were heard.

There was tenderness to pressure over the spinous processes of C7 and T1.

There was complete paralysis of the right leg with marked weakness of all muscle groups in the left leg. Patellar and ankle reflexes were symmetrically hyperactive. Ankle clonus was present bilaterally. There were bilateral pathological toe signs. Below T3 there was poor appreciation of all sensory modalities bilaterally, and marked hyperesthesia, dysesthesia and reversal of temperature distinction were found in the right lower extremity. Dissociation of sensation was not present.

Rectal sphincter tone was poor, and there was urinary retention.

Diagnostic Procedures. Roentgenograms of the spine revealed a small area of translucency in the pedicle of T4 (Fig. 1).

Spinal puncture yielded clear, colorless fluid under pressure of 120 mm. cerebrospinal fluid and with sluggish rise and fall on bilateral jugular compression. Total spinal fluid protein was 48 mg. per cent.

Myelography demonstrated a partial obstruction to the flow of dye between C7 and T4, with the typical worm-like defects of a vascular anomaly at this level (Fig. 2).

Operation. On July 3, 1957 laminectomy was performed in the sitting position under general anesthesia. The laminae of C7 through T4 were removed, during which procedure venous and arterial bleeding was profuse. The extradural fat was replaced by tough, exceedingly vascular fibrous tissue, a portion of which was excised for biopsy. The dura mater was not pulsating and therefore was incised throughout the length of the exposure. Upon opening the dura mater, a pulsating mass of dilated, tortuous vessels was seen on the posterior surface of the cord and dipping into the posterior longitudinal sulcus of the cord. It appeared to consist of both arteries and veins. The arachnoid was opened to afford complete decompression, but no attempt was made to obliterate or excise the angioma. The dura mater was left widely open; the rest of the wound was closed in layers with black silk.

Pathology. The epidural tissue removed for biopsy was described by Dr. Louis Z. Gordon, pathologist, as follows:

“Vascular adipose tissue interlaced by fibrous tissue septa. The vascular tissue shows a

* The contents of this article are the personal views of the authors and are not to be construed as statement of official Air Force policy nor as Air Force indorsement of any commercial product described.
† Address after July 1, 1960: USAF Hospital Carswell, Carswell Air Force Base, Texas.
‡ Address: 506 Oak Street, Cincinnati 19, Ohio.
significant change in the form of degeneration of the walls. A number of vessels show only basophilic degeneration of the adventitia and muscularis. In other areas the entire wall of the vessel is involved in such a degenerative process. At one end of the specimen the vascular channels can be identified as such only because of their characteristic shape and are arranged in a noncommunicating network. Both arterial and venous channels appear to be involved in this process. Impression: Arterial angioma."

Postoperative Course. The patient's course was complicated. On the 2nd postoperative day she had three grand mal convulsions. She had not been in shock and was not toxic. Postictally neurological findings were unchanged. She had no further seizures. One was led to wonder whether she might not also have an intracranial vascular anomaly.

The wound continued to drain spinal fluid, which eventually grew *Pseudomonas aeruginosa* on culture. The organism was sensitive *in vitro* only to dihydrostreptomycin and Polymixin. The patient was placed on penicillin and dihydrostreptomycin, and by July 15, 1957 the wound was clean enough to permit secondary closure to seal off the spinal fluid leak.

On July 23, 1957, 20 days after operation, the patient could move the right leg slightly, and strength in the left leg had improved. The sensory level had fallen to T9 on the right and L1 on the left. An indwelling urethral catheter was still necessary. From that time on she continued to improve and when discharged from the hospital on Aug. 17, 1957 could stand unassisted.

She was readmitted on Aug. 24, 1957 because of a large collection of fluid beneath the incision, with fever. Fluid containing *Pseudomonas aeruginosa*, sensitive only to Polymixin, was aspirated.

At the time of this admission she was still further improved neurologically. She was able to
walk unassisted, and the only sensory deficit present was mild hypalgesia below L1 on the left side. Patellar and ankle reflexes were hyperactive, and there was a bilateral Babinski's sign. The wound was aspirated daily and a solution of Polymixin was instilled.

On Sept. 10, 1957 a normal premature female infant was delivered by Caesarian section by Dr. J. O. Porter. This mode of delivery was decided upon in order to prevent possible engorgement of the angioma during straining of natural childbirth.

Production of fluid in the wound eventually disappeared, culture became negative, and the patient was discharged, walking, on Oct. 2, 1957.

On her latest follow-up visit, July 19, 1958, the patient was walking without a limp and showed only slight spasticity, hyperreflexia and Babinski's sign in the right lower extremity.

Fig. 2. Myelogram showing partial obstruction and tortuous vascular channels.

DISCUSSION

So far as can be determined from reviewing the literature, decompressive laminectomy during pregnancy for paraparesis secondary to spinal angioma has been reported previously in 1 instance. This case was described by Guthkelch in 1948.10 A 34-year-old woman had paraplegia in the 30th week of her 3rd pregnancy. At laminectomy a vertebral and epidural hemangioma at the level of the 4th thoracic vertebra was exposed. Because of the profuse bleeding no biopsy could be made. The patient delivered spontaneously 5 days later, but died of acute dilatation of the stomach in the early postpartum period, at which time she was still paraplegic. This case was later included in Newman's report.13

Other patients have been subjected to laminectomy for symptoms recurring at some time after delivery. These include Newman's first case,13 in which an extra-
dural angioma was removed with complete recovery from paraplegia occurring 3½ years after the last delivery of a woman who had had symptoms during 2 pregnancies with remission after each delivery.

Delmas-Marsalet recorded the removal of an intradural angioma for relief of paraplegia occurring 3 years after the last pregnancy of a woman who had had symptoms of a transverse spinal cord lesion during her 2 pregnancies, with remission after delivery.

Lam et al. reported the case of a 30-year-old woman who became paraplegic in the 3rd trimester of her 9th pregnancy, with prompt improvement immediately after delivery, and with subsequent exacerbation of symptoms about 1 month after delivery. An extradural hemangioma at T3 was removed, following which neurological deficit disappeared.

Laminectomy with verification of an intramedullary vascular malformation in a woman whose first, transient symptoms had occurred during pregnancy 2 years earlier, was reported by Brion et al.

The conclusion stated by Newman that “spontaneous remission does not occur except post-partum” thus seems well substantiated. Balado and Morea had a patient with an extradural angioma who had spastic paraparesis in 7 out of 8 pregnancies with spontaneous recovery after all except the last delivery, when fatality occurred secondary to heart failure. The lesion was verified at necropsy.

Newman’s third case was of a 35-year-old woman in whom paraplegia developed during her 3rd pregnancy and recovery began spontaneously 4 months after delivery. Two years later, when she was not pregnant, symptoms recurred and myelography disclosed an intradural angioma at T11. Radiotherapy was followed by temporary improvement, but with subsequent paraparesis.

Fields and Jones reported the case of a 30-year-old woman who, after Caesarian section, partially recovered from paraparesis which began in the 6th month of pregnancy. One month after delivery an epidural hemangioma at T6 was resected with subsequent complete recovery.

A similar history of spontaneous improvement after delivery has been reported by Glaser and Michon et al.

The occurrence of prompt improvement prior to delivery in our case would seem to indicate that decompressive laminectomy had a beneficial effect; albeit from the reports above one might have expected a spontaneous remission after delivery. Nevertheless, since only a decompression was accomplished, support is added to the theory that symptoms are caused by vascular engorgement from pressure of the enlarging uterus. This theory has been well presented by Lam et al. and Newman.

That the explanation is not so simple, however, is apparent from reviewing other evidence. Delmas-Marsalet’s patient had onset of symptoms in the 2nd month of pregnancy, before any great degree of vascular engorgement would be expected to occur. The fact that symptoms have been reported to occur during menstrual periods lends support to the theory that hormones of pregnancy have an adverse effect on the walls of the vessels within an angioma. The appearance of spider nevi and palmar erythema during pregnancy, presumably on a hormonal basis, offers an analogy.

An explanation has not been offered for the absence of symptoms during pregnancies preceding, intervening between, or following pregnancies in which symptoms have occurred. Also, the occurrence of symptoms, in the absence of pregnancy,
in patients who had had reversible lesions during previous pregnancies\textsuperscript{1,5,11,13} is difficult to reconcile with any theory relating pregnancy to the onset of symptoms.

The occurrence of symptoms of other neurological disorders, in particular intracranial meningioma,\textsuperscript{2} during pregnancy would suggest that there is nothing specific about the lesion itself.

The role of thrombosis and recanalization within the angioma has been considered as an explanation for the progression and remission of neurological symptoms in nonpregnant patients.\textsuperscript{7,8} What effect pregnancy may have on this pathological process is uncertain.

SUMMARY

1. A case of spinal angioma, with onset of paraparesis in the 6th month of pregnancy and with recovery beginning after decompressive laminectomy and prior to delivery, is described.

2. Various explanations, in particular venous engorgement and hormonal influence on smooth muscle, are considered.

REFERENCES


