HEMANGIOMA OF A DORSAL VERTEBRA WITH COLLAPSE AND COMPRESSION MYELOPATHY

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During the winter semester at the University of Berlin (1863), Virchow delivered a lengthy discourse on angiomata in which brief mention was made of a patient with paraplegia who, at autopsy, was shown to have tumors composed of dilated blood channels in the center of both the 10th thoracic and 3rd lumbar vertebrae. Subsequent to this there may be found sporadic reports by pathologists concerning angiomata of osseous structures. In 1917 Hitzrot recorded the first description of the roentgen appearance of hemangioma within bone. Later Perman described the roentgenographic appearance of a verified vertebral angioma. Töpfer, working in Schmorl’s clinic, investigated at autopsy 2,154 spinal columns, disclosing a 12 per cent incidence of angiomatous masses in various areas of this structure. In a comparable study of 10,000 cases Junghanns cited an incidence of 10 per cent of vertebral angiomata. These figures have recently been questioned by Lichtenstein and others as being too high because some of the cases included in this study represented so-called “blood cysts.” A review of the recent literature discloses reports of hundreds of cases of angiomata of the vertebrae; however, in only about 60 of these was there associated dysfunction of the spinal cord. Five of these 60 were well documented examples in which the vertebral body was partially collapsed; in the remainder the angioma was associated with compression myelopathy caused by narrowing of the vertebral canal by the enlarged pedicles and laminae. This latter phenomenon of thickened pedicles and laminae producing compression of the spinal cord in cases of vertebral angiomata has been reported by Bailey and Bucy.

Only true examples of massive collapse of the vertebra with concomitant dysfunction of the spinal cord are included in the present report. The 5 cases of particular interest previously mentioned were reported by Muthmann, Gold, Globus and Doshay, Sandahl, and Holta.

Muthmann’s case. A 61-year-old female had progressive paraparesis of 16 years’ duration. The clinical diagnosis was tuberculous spondylitis. No operative intervention was carried out because of complicating cardiopulmonary disease. Death resulted from pyelonephritis. Autopsy disclosed 50 per cent collapse of the body of the 6th thoracic vertebra with associated moderate kyphosis. Histologically there was a cavernous hemangioma within the involved vertebral body.
Gold's case. A male aged 23 had slow onset of paraplegia which was diagnosed preoperatively by roentgenograms as Kümml's disease. At operation a hemangioma of the epidural space was found. Death followed shortly after surgical exploration. At autopsy there were found multiple lesions of the thoracic vertebrae with compression of the body of the 6th thoracic vertebra and extension of the tumor into the epidural space at this level.

Globus and Doshay's case. A 13-year-old female had a rapidly developing paraparesis of 1 year's duration which allegedly followed trauma to the back. A laminectomy was performed, following which she died. Necropsy revealed a gross collapse of the 8th thoracic vertebra which had destroyed the centrum, pedicles, and laminae, and invaded the regional epidural space.

Sandahl's case. A female, aged 17, had progressive weakness of both legs for a period of 8 years following an injury of the back. Roentgenograms disclosed compression of the body of the 12th thoracic vertebra. However, at operation an additional lesion, namely a neurinoma of the upper cauda equina, was found and removed. Although previous reviewers of vertebral angiomata have included this case, the description given by the author was not in sufficient detail to evaluate the relative significance of the two lesions. The patient survived but follow up was not reported.

Holta's case. A 66-year-old sailor had acute cervical trauma followed by gradual development of quadriplegia. Roentgenograms showed almost complete collapse of the body of the 4th cervical vertebra with a large bony fragment of the anterior inferior aspect of the body jutting anteriorly. The vertebral column was markedly angulated dorsally at the level of the lesion. Parallel striations were demonstrated radiographically within the involved vertebra and on the basis of this finding a diagnosis of angioma of the vertebra was made. After 2 weeks of cervical traction and deep roentgen therapy he died. Necropsy revealed a cavernous hemangioma of the centrum, pedicles and laminae of the 4th cervical vertebra.

Comment. In these 5 cases there was recorded a slowly developing paraparesis or tetraparesis often progressing to paralysis. It would seem there are three modes of encroachment on the vertebral canal, resulting in compression myelopathy, namely deformation of the centrum subsequent to collapse, extension of the angioma into the regional epidural space, and hypertrophy of the vertebral arch subsequent to angiomatous invasion. In some, the pathogenesis of the implication of the spinal cord appears to invoke more than one of these mechanisms. Even though the excellency of roentgen diagnosis at the present time has aided in resolving the issues in most cases, this was not always true. Many of the early reports indicated difficulty in differentiating angioma from Kümml's disease, as well as tuberculosis.

In our clinic 4 patients with compression myelopathy secondary to vertebral angioma have been submitted to therapeutic laminectomy. No postoperative complications ensued in 3 of these. The fourth, however, had a rather rare complication, namely, collapse followed by almost complete disappearance of the centrum of the involved vertebra. The historical account of this case follows.
CASE REPORT

B.H. #134548. M.W., a 25-year-old white female, was admitted to the neurosurgical service at the Brooklyn Hospital on Oct. 5, 1948 complaining of pain in the back and weakness in both legs. She had been entirely well until March 1948 when there was pain in the left lower flank attributed to an 8½-month pregnancy. Intermittent pain in this region persisted 7 weeks. In June 1948, after a period of remission, the pain in the left flank returned, grew more intense, and tended to radiate into the left lower abdomen. Medical advice was sought and because of the distribution of pain a pyelogram was carried out. Immediately following the pyelogram there was bilateral radicular pain arising paravertebrally and projecting into the lower anterior abdominal quadrants. Coughing, sneezing or straining aggravated the discomfort. In August there were felt for the first time numbness and tingling in both legs and heaviness and coldness in both feet. Occasional hesitancy in starting of the urinary stream was present. Roentgenography in September was said to have disclosed no abnormalities of the vertebral column. On Oct. 4, 1948, disabling weakness of both lower extremities appeared rather precipitously, and on the following day she entered the hospital.

Examination. The head and cranial nerves were within normal limits. The upper extremities and thorax disclosed no abnormalities. The upper abdominal reflexes were equal and active, whereas the lower abdominal reflexes were absent. A Beevor's sign was present. There was marked paresis of the lower extremities. Extension of the legs, with the thighs slightly flexed, was difficult to perform. There was mild spasticity of both lower extremities, slightly more evident on the right side. The sign of Babinski was present bilaterally. Appreciation of touch and pin prick was abolished below the T11 dermatome zone bilaterally, with minimal involvement of the sacral area. Appreciation of tuning fork vibration (c 128) was diminished at the level of the iliac crests, but absent at the knees and ankles bilaterally. Urinary retention developed shortly after entry.
Roentgenograms of the dorsal spine revealed moderate decalcification of the body of the 10th thoracic vertebra with "paralleling of the trabeculae" (Fig. 1). The lateral projection demonstrated a minimal loss of the substance of the involved centrum at its posterior inferior aspect. The intervertebral spaces were well preserved.

1st Operation. On Oct. 7, 1948, upon exposing the posterior spinous process of the 9th, 10th and 11th thoracic vertebrae, there was excessive bleeding from enlarged cortical foraminae of the lamina of the 10th vertebra. Laminectomy was attended by hemorrhage arising almost exclusively from the divided arch of the 10th thoracic vertebra. The left lamina of this vertebra was considerably thicker than that of the right side. The vertebral canal was obviously encroached upon by the enlarged pedicles of the implicated vertebra. The dural envelope was effectively released by the laminectomy. Hemostasis was secured, blood loss was replaced by transfusion and the wound was closed.

Histological Report. Sections of a decalcified portion of a lamina were composed of slender bony trabeculae which were widely spaced (Fig. 2). Interspersed were sinusoids of varying size lined by endothelium and supported by scanty fibrous stroma. Diagnosis: Cavernous hemangioma of bone.

Course. Postoperatively there was gradual return of muscle power in both lower extremities. Voluntary control of the urinary bladder was reestablished on the 5th postoperative day. Ambulation occurred on the 17th day. There was no residual pain in the back or flank. She was discharged Oct. 30, 1948.

At home there was continued improvement in walking. There was occasional pain in the right thigh and the right lower abdominal quadrant. About June 1949 she noticed an increase in the "sway" of her back, and associated prominence of the lower abdomen. Without medical advice, physical manipulations of the trunk were
instituted to improve muscle tone. Shortly thereafter there was noted spasm of muscles of the back and when walking a slightly flexed posture of the trunk was assumed.

In October there was pain in the lower lumbar region with occasional radiation along both iliac crests. A week later weakness in the right leg was present followed by similar weakness in the left leg and a sensation of coldness in both lower extremities. Gradually the situation deteriorated. She was readmitted to the hospital Jan. 3, 1950.

**Examination.** On entry the patient walked with a halting gait and manifested weakness of both legs, more pronounced in the right lower extremity. Appreciation of position and vibration was found impaired in the right leg and foot. Touch was not felt in either lower extremity. There was diminution of appreciation of temperature below the knee on the right, and hypalgesia below the knees bilaterally. Tendon reflexes were accentuated in the left lower extremity. The sign of Babinski was present bilaterally. The healed laminectomy scar was tender. A slight protrusion of the posterior spinous process of the 12th thoracic vertebra was present. Roentgenograms of the dorsolumbar spine showed almost complete destruction of the 10th dorsal vertebra with a portion of the remaining part extruded anteriorly (Fig. 3). The lesion appeared to be limited to the 10th thoracic vertebra.

**Course.** She was placed on a Whitman frame with bilateral Buck’s extension; however, this only served to accentuate the spinal cord dysfunction. It became evident that further surgery was necessary.

**2nd Operation.** On Feb. 10, 1950 the operative scar was excised. At the level of the 10th thoracic vertebra, the dura mater was encountered about 1 cm. beneath the skin. After exposing this structure, the laminectomy was increased by the removal of an arch at either end. This liberated the dural envelope. Upon incising the dura mater the full length of the wound, the spinal cord was found somewhat thinned at the level of the collapsed vertebra. The cord and its coverings were markedly displaced dorsally by an underlying dome-shaped mass which was not surgically investigated. Following hemostasis the wound was closed.

**Course.** Postoperatively there was immediate improvement in motor and sensory function of the lower extremities. Roentgen-ray therapy was administered. Subsequent to this a special double corset brace was provided for external stabilization. At discharge on April 4, 1950 the patient could walk with support. At this time there was impairment of appreciation of both vibration and postural movement of the lower extremities, and mild to moderate thermohypesthesia of the left leg and foot.
At home there was continued improvement in every respect. About the beginning of October 1950 she complained of the eyes feeling tired. A neurological examination disclosed bilateral moderately advanced papilledema with retinal hemorrhages. The visual fields were mildly constricted; however, visual acuity was not impaired. Small paracentral ring scotomata were demonstrated. There was no associated headache. Gradually the abnormal findings of the optic fundi receded and when examined on Feb. 26, 1951 the optic fundi were found to be normal. There was mild dysfunction of the lower extremities and rarely stress incontinence.

During the past 4 years there has been a gradual return to normal. The spinal brace has been discarded and she has been allowed to carry out all her household duties with the exception of lifting heavy objects. The most recent roentgenogram (May 27, 1954) is reproduced in Fig. 4.

**DISCUSSION**

Columniation of the centrum of a vertebra is commonly interpreted by roentgenologists as hemangioma. Seldom are such lesions of clinical significance. As stated, an impressive number of vertebral hemangiomata disclosed at autopsy have been recorded and regarded as incidental findings. Although the vast majority of these lesions are innocuous, there are a few that undergo proliferative and degenerative changes. It has been proposed by Watson\(^\text{13}\) that expansion of the tumor results from endothelial budding and subsequent canalization of the outgrowths. Cancellous bone encroached upon by these newly formed channels assumes an irregular colonnade effect or reactive trabeculae in parallelism so readily identified by roentgen examination. It has been estimated that some of the trabeculae become enlarged to as much as three times normal.

The case herein presented is rather unique because of the degree of vertebral dissolution and almost complete clinical recovery following operation. The lower dorsal region is a rather common site for the occurrence of this lesion, but it is uncommon to demonstrate anterior extrusion of a portion of a vertebral body. In light of previous experiences a complication of this character subsequent to laminectomy for compression myelopathy was not expected. The sequence of clinical events in this case serves to caution one against overestimating the strength of a vertebra involved by angioma. Fusion operations, either vertebral or rib, should be given considera-
tion. It would seem that partial collapse of a vertebral body involved by an angioma is a clear indication for a fusion operation.

The administration of roentgen therapy may help. However, it would seem unjust to recommend it preoperatively in the face of an advancing paraparesis. Certainly a judicious use of radiation for this form of lesion is indicated following laminectomy. It is to be remembered that roentgen therapy may be complicated by necrosis of normal spinal cord, especially when high dosage has been employed.

In the case herein recorded, the cause of the transitory papilledema remains obscure. Intracranial angiomatous lesions may be present although there are no other clinical features to support such an opinion.

CONCLUSIONS

Dissolution of a vertebra implicated by hemangiomatous process is a relatively rare occurrence. In the presence of clinical evidence of compression myelopathy associated with this disease, laminectomy is clearly indicated. After such an operation, roentgen studies of the involved area should be performed at intervals, particularly if there are complaints suggesting advancement of the disease within the vertebra. Furthermore, if by roentgen examination there is disclosed beginning collapse of the vertebra, a fusion operation should be performed.

REFERENCES