Metameric Spinal Cord and Skin Hemangiomas

Case Report

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Hemangiomas of the spinal cord, both intramedullary and leptomeningeal, associated with cutaneous hemangiomas of the same metamere are rare. Thirteen cases have been reported in the literature.

The following case is an example of an intramedullary spinal hemangioma with a segmentally related angiomatous lesion.

Case Report

A 38-year-old white man was transferred from another hospital to the Presbyterian-University Hospital, Pittsburgh, Pennsylvania, on November 5, 1965. He complained of chest pain, marked weakness in the left leg and numbness in the right leg. His difficulty had begun 3 months before admission with the appearance of upper thoracic pain which radiated anteriorly to just below the left nipple. Shortly thereafter, he developed weakness of the left leg and numbness in the right foot. These symptoms progressed in severity to the time of admission. Examination of the spinal fluid at another hospital had revealed a protein of 100 mg% and no cells.

The past history was significant in that he had had removal of hemangiomas from the right axilla and right lateral chest wall in 1953, 1957, 1961, and 1962.

Examination. Blood pressure was 120/80, pulse was 80 and regular. There were several well-healed surgical scars and a 6 × 4 cm non-tender mass in the upper right lateral chest wall. There was no discoloration of the skin overlying the mass. However, the patient stated that the skin at the sites of the previous surgery showed irregular red areas. The remainder of the general physical examination was normal.

There was marked weakness and atrophy of all muscle groups in the left leg. Strength was mildly decreased in the right leg. Deep tendon reflexes were abnormally hyperactive, but equal in the legs. The plantar response was extensor bilaterally and ankle clonus was present on both sides, but more marked on the left. Sensory examination revealed hypalgesia, hypesthesia, and thermohypesthesia below D-4 on the right. Vibratory sensation was decreased at the ankles, and position sense was diminished in the toes bilaterally. Hemogram, urinalysis, fasting blood sugar, blood urea nitrogen, and electrocardiogram were normal. Plain films of the dorsal spine revealed old compression fractures of the D-5 and D-6 vertebral bodies. A myelogram showed a partial block at D2–3. The Pantopaque column flowed very slowly around the right at this level. None of the contrast material passed on the left.

Operation. On November 11, 1965, a dorsal laminectomy was performed at D-2 to D-4. The subcutaneous fat was about 2 to 3 times its normal thickness and somewhat indurated. The bone appeared normal, and there was no evidence of extradural pathology.

After the dura was opened, the cord was seen to be swollen and the dorsum avascular and yellow, with a small gray-blue area on the right side. Aspiration of this region yielded only small amounts of old blood. A vertical midline incision was made in the avascular area of the cord, exposing a dark purple mass. This was largely removed except for small scattered areas, which fused with the cord.

Pathology. Histological examination of the subcutaneous fat showed fibroadipose tissue with large vessels containing fibrin and polymorphonuclear leucocytes. Microscopic examination of the mass in the spinal cord showed numerous thin vascular channels with prominent endothelial lining, walls made up of connective tissue, and, rarely, a few smooth muscle cells. There were areas

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of recent hemorrhage with many fresh red blood cells, as well as numerous phagocytes containing dark brown granular hemosiderin pigment (Fig. 1 left).

A biopsy from the soft tissue mass in the right lateral chest wall was obtained on December 6 under local anesthesia. The histopathological features included numerous dilated vascular channels resembling venules and capillaries. Some of these vessels contained smooth muscle suggestive of a thin, incomplete, tunica media. The diagnosis was vascular malformation or hemangioma (Fig. 1 right).

**Postoperative Course.** Postoperatively, the sensory level to pinprick extended up to D-2 on the right, and, in addition, there was a segmental hypalgesia from D-8 to D-2 on the left. Motor power in the left leg gradually improved over the preoperative status. By December 11, the patient was able to walk well with a left foot drop brace and a walkerette. The level of perception of pinprick had dropped from D-2 to D-8 on the right, and there was now sacral sparing. The segmental hypalgesia on the left side decreased to involve an area from D-4 to D-8. Vibratory and position sense remained the same as they were preoperatively.

In January, 1967, a little more than a year postoperatively, the patient was reexamined. Neurological findings were essentially the same as in December, 1965.

**Discussion**

Fine\(^4\) believes that these spinal vascular lesions, Sturge-Weber syndrome, and von Hippel-Lindau syndrome, represent a related group of disorders in which vascular hematomas of the central nervous system coexist with other neural or extraneural lesions of a similar dysplastic nature. Cobb\(^2\) concludes that the lesions could occur in any organ innervated by filaments from the same neuromere. Subarachnoid hemorrhage has occurred as a presenting symptom in patients with segmentally related skin angiomas.\(^6\) Spinal bruit has also been reported.\(^4,19\)

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![Fig. 1. Left: Section of intramedullary spinal mass showing abnormal vessels, recent hemorrhage, and hemosiderin laden phagocytes. Right: Skin biopsy with abnormal blood vessels of hemangioma. H. & E., × 300.](image-url)