CASE REPORT AND TECHNICAL NOTES

Ependymoma of Spinal Cord: Subtotal Intramedullary Removal from Syringomyelic Cavity

CASE REPORT

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The following case is reported primarily to illustrate the value of intramedullary exploration of the spinal cord, particularly where the primary diagnosis is difficult to establish. (For several months prior to operation the case was considered as an example of conversion hysteria.) It is also of interest that a syringomyelic cavity was associated with the tumor; and that after operation the patient suffered “tract” pain of spinal cord origin.

HISTORY

Chief Complaint. Weakness of legs and sensation of pressure in girdle distribution following herniorrhaphy under spinal anesthesia. Symptoms were of approximately 2 years’ duration.

Present Illness. This 35-year-old soldier first noted an uncomfortable pressure sensation in July 1943, immediately after the repair of a right inguinal hernia under spinal anesthesia. The entire abdominal wall was affected, from the level of the 12th ribs to the groins bilaterally. In addition he complained of vague numbness and weakness in the legs. These symptoms lasted about 2 weeks and did not recur until February 1945. At the time of recurrence the patient was overseas and his outfit was alerted for a move forward. No definite neurological signs being found, he was thought to be suffering from conversion hysteria.

Prior to herniorrhaphy he had noted pain of an entirely different character from that of his present illness. There were no known complications during the spinal anesthesia.

Past History. Civilian occupation: Boilermaker. Tropical service: None. Habits: Very moderate use of tobacco, alcohol; no drugs. Illnesses: Usual childhood diseases with no known sequelae. Venereal disease denied. Injuries: Struck in occiput early in 1943, after which he was unconscious for about 10 minutes. The only residual symptoms were occasional mild dizziness and slight blurring of vision on reading. Operations: Herniorrhaphy, inguinal, spinal anesthesia, July 1943.

Family History. No known mental or nervous diseases.

Course of Present Illness. In February 1945 the patient awoke one morning unable to turn in bed because of pain in the lower thoracic spine, accompanied by numbness and weakness of the legs. The back pain was accentuated by attempting to turn, and was accompanied by the same girdle-like sense of uncomfortable pressure that he had experienced nearly 2 years previously. Coughing or straining never aggravated his symptoms, though change of position, particularly forward bending, did. The initial acuity of these complaints soon abated, though the symptoms persisted to the time of operation, along with a tendency to staggering gait, and occasional “dancing” (clonus) of the feet. Tingling sensations of glove distribution began to affect the hands and forearms in February 1945.

Because of these complaints he was admitted to a General Hospital overseas, where the following notes were made: 13 March 1945, normal reflexes of lower extremities; no Babinski. Lumbar puncture: Initial pressure ~15 mm. CSF; no block; total protein ~27 mg. per cent. X-rays of spine showed minimal osteoarthritic changes in the lower dorsal and upper lumbar vertebrae. 27 March, tests with a Richter neurodendrometer showed no sensory loss, which

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together with the history and vague neurological findings led to a tentative diagnosis of conversion hysteria.

On 28 March 1945 the tendon reflexes of the left arm were found slightly more active than those of the right; there was a positive left Hoffmann reflex; the lower abdominal reflexes were sluggish; and there was hypalgesia from the nipple line downwards, with equivocal impairment of position sense and reduced vibratory perception in the toes of both feet. He walked with a rather wide base. On 1 April the sensory level was found to be at the umbilicus. The diagnosis of pernicious anemia was considered until the gastric analysis and blood counts proved to be normal. By 15 April there was a suggestive left Babinski and bilateral ankle clonus.

On 10 July 1945 he was admitted to a General Hospital in the United States with the same complaints. Initial neurological examination disclosed fibrillations of the muscles of the shoulder girdle; some spasticity of the lower extremities; a wide base, some staggering of gait, and a slightly positive Romberg test. The tendon reflexes of all four extremities were hyperactive and rated as 3; the upper abdominals were listed as 2; the lower abdominals 0; cremasterics 2; Babinski positive bilaterally; Hoffmann negative.

Sensation: Glove distribution of hyposthesia almost up to the elbows; variable and vague bilateral sensory level in region of umbilicus downwards for pain, with preservation of touch. Vibratory and thermal perception reduced from knees downwards; position sense

Fig. 1. Photographs of (A) the spinal cord, Th. 8–Th. 10, before removal of tumor, and (B) the tumor tissue (ependymoma), immediately after intramedullary removal, placed on the cottonoid strip at the right. The operative opening in the dorsal part of the cord appears at the left.
reported intact. There was no evidence of neurofibromatosis. Lumbar puncture: "Slightly xanthochromic" cerebrospinal fluid; total protein 25 mg. per cent. Grant-Cone test: Slightly positive. Laboratory work-up: Within normal limits. Serology: Negative. Tentative diagnoses: Conversion hysteria; or amyotrophic lateral sclerosis secondary to spinal anesthesia.

Myelography. This revealed an almost complete central block at the level of the 10th thoracic vertebra. The patient was transferred to Neurosurgery for operation.

Laminectomy, Th. 8–Th. 10 inclusive. Anesthesia: Intratracheal gas-oxygen-ether. The spinal cord was swollen for a distance of 2.5 cm., almost completely filling the intrathecal space. Its dorsal surface appeared somewhat irregular and blanched, and palpation suggested the presence of an intramedullary cyst, confirmed by midline aspiration of the distended cord with a fine hypodermic needle, which yielded a few drops of yellow fluid. The dorsal aspect of the cord was, therefore, split along the midline for 2.0 cm., disclosing a soft mass of irregular reddish-blue tissue 2.0 cm. in length and 1.0 cm. in diameter. The bulk of this tumor was carefully shelled out of a smooth lined cystic cavity in the center of the cord (Fig. 1). At the ventral aspect of the cyst, fine strands of tumor tissue extended into the substance of the cord. These were not mosted lest viable cord be damaged. It was noted that the cystic cavity, cephalic end, was funnel-shaped, suggesting the prolongation of a syringomyelic cyst rather than a degenerative area caused by pressure ischemia.

Microscopical Diagnosis. Ependymoma.

Postoperative Course. Eight hours after operation the patient was able to move both legs to some extent. There was almost complete anesthesia for all modalities from Th.10 downwards, and urininary retention. The patient complained of a continuous tingling burning pain that seemed to affect both lower extremities in diffuse fashion, and was different from any sensation he had noted prior to operation. This sensation gradually disappeared as his control of bladder and legs improved. Three weeks after operation, he began to walk and had regained approximately 75 per cent pain and position sense in the left leg, and 50 per cent pain perception on the right but no position or vibratory sensation. The level of hypesthesia was Th. 8 on the right and Th. 10 on the left, with approximately 50 per cent sparing in the saddle area. Voluntary bladder function had returned to normal. The numbness of hands and forearms completely vanished after the operation.

On the 21st postoperative day, the patient was discharged to another hospital for deep x-ray therapy, in excellent condition, with strength and sensation improving steadily.

COMMENT

With regard to the incidence and distribution of spinal cord ependymomas, Shenkin and Alpers 1 state that 10–20 per cent of all spinal cord tumors are intramedullary gliomas, of which ependymomas are the predominant type, and that the highest incidence of ependymomas is within the thoracic segments. In 7 of their 27 reported cases there were associated cystic cavities of the cord. Associated sensory disturbances suggesting "tract pain" are characterized by diffuse painful sensations of burning or boring character, distant from the tumor level, and neither of radicular nor peripheral distribution. Intramedullary removal of ependymomas is not a novel procedure.1,2

Differential diagnosis, according to most writers on the subject of intramedullary cord neoplasms, constitutes an acknowledged difficulty. For example, Lichtenstein3 speaks of remissions reminiscent of multiple sclerosis, and emphasizes the fact that multiple intramedullary tumors of the cord sometimes occur, making diagnosis still more difficult.

The present case report illustrates many of the features common to reports in the literature; e.g. (1) the difficulty of diagnosis, illustrated by the fact that conversion hysteria, pernicious anemia and amyotrophic lateral sclerosis were seriously considered; (2) the thoracic site of the tumor; (3) the associated syrinx; and (4) the possibility of a multiple process, suggested by the fact that the "glove" hypesthesia and paresthesia vanished after operation. Perhaps a syringomyelic cavity of the cervical cord may have been drained through the central canal when the cord was opened at the thoracic level.
METHOD OF VENTRICULAR FLUID REPLACEMENT

The difficulty of early diagnosis in this and similar cases should carry a note of warning, especially since remissions may occur during the development of neurological symptoms and signs which suggest demyelinating diseases of the spinal cord.

As to prognosis, ependymomas are usually slowly growing tumors, and do not tend to metastasize. They are, however, resistant to x-ray therapy, and hence the ultimate prognosis should be guarded.

The painful dysesthesias noted by this patient and others mentioned in the literature are apparently of spinal cord origin. This is worthy of note since there seems to be some controversy as to whether such a phenomenon exists. However, the pain experienced postoperatively in the present instance is similar to the burning dysesthesias felt "in the legs" of some paraplegic patients with traumatically divided spinal cords, and suggests abnormal stimulation of sensory pathways either by a pathological or physiological process, acting within the cord itself. Since effective pathological, physiological or operative interruption of the spinothalamic pathways too often fails to relieve this type of pain, such dysesthesia may well be due to abnormal neural volleys reaching the thalamus via other sensory pathways; i.e., via the intact portions of the dorsal columns immediately above the site of the cord lesion. These tracts may be stimulated pathologically by cicatricial traction or by changes in the local fluid or vascular milieu; or neurogenically, by disturbed sensory circuits.

SUMMARY

1. A case of intramedullary spinal cord tumor (ependymoma) at Th. 8–Th. 10 is reported.
2. Syringomyelic cavitation (perhaps multiple) was associated with the tumor.
3. Subtotal surgical removal of the tumor was accomplished.
4. Pitfalls of diagnosis are briefly discussed.
5. The significance of pain of intramedullary spinal cord origin is mentioned.

REFERENCES


METHOD OF VENTRICULAR FLUID REPLACEMENT FOLLOWING VENTRICULOGRAPHY

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A description of a method designed to replace ventricular fluid following ventriculography is presented. Replacement of ventricular fluid is often desirable in hydrocephalus of congenital origin, but more especially in those cases due to a periaqueductal stenosis. Attempts to lessen reactions, for example, by the use of oxygen instead of ordinary air, during and following ventriculography, are well recognized. Other methods for replacement of ventricular fluid also have been utilized.