COMPRESSION OF THE LUMBOSACRAL ROOTS OF THE SPINAL CORD BY THICKENED LIGAMENTA FLAVA

By Edward Bancroft Towne, M.D.

AND

Frederick Leet Reichert, M.D.

of San Francisco, Calif.

FROM THE DEPARTMENT OF SURGERY OF STANFORD UNIVERSITY

Hypertrophy of the ligamenta flava in the lumbar region as a cause for compression of the cauda equina is a clinical entity hitherto not described. Two patients recently observed had spontaneous pain in the back and legs, progressive flaccid paralysis, fibrillary twitching of the muscles, sacral anæsthesia and absent Achilles reflexes. In addition, one had xanthochromic spinal fluid, and the other had disturbed function of the vesical and anal sphincters. In each instance a complete block in the circulation of the cerebrospinal fluid was demonstrated by lipiodol injection, and the convex or pointed lower end of the opaque shadows suggested an extradural lesion. This impression was verified at operation, which revealed thickening of several ligamenta flava, most marked at the level demonstrated by the lipiodol. Excision of the hypertrophic ligaments relieved the block in the circulation of the cerebrospinal fluid. The fact that these two cases were observed during a period of ten months would suggest that the condition is not excessively rare, but a review of the medical literature has failed to disclose another example.

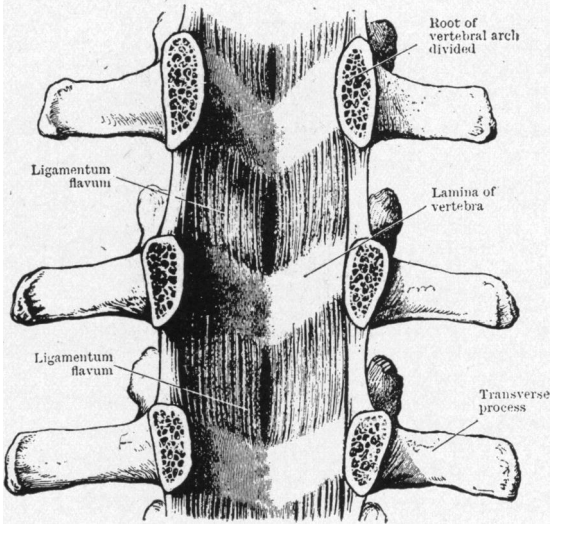

Fig. 1.—Ligamenta flava as seen from the front after removal of the bodies of the vertebræ by sawing through the roots of the vertebral arches. From Cunningham's Anatomy, Wm. Wood & Co., N. Y., 1918.

Anatomy.—The ligamenta flava or subflava stretch across the posterolateral aspect of the spinal canal between the laminæ. (Fig. 1.) The ligaments, which are made up of yellow elastic fibres, are attached to the anterior aspect of the superior laminæ and to the posterior aspect of the inferior. The two ligaments of each interlaminar space fuse in the mid-line, and extend laterally to form the posterior margin of the intervertebral foramina. (Fig.

2.) The normal ligament, as observed during a laminectomy, is one or two millimetres thick and distinctly yellow in color.

CASE I.—(E. B. Towne.) *Compression of the cauda equina by thickened ligamenta flava, causing flaccid paralysis of right lower extremity. Restoration of normal function after removal of thickened ligaments.*

S. S., a Portuguese laborer, aged fifty-three years, was admitted to the Southern Pacific General Hospital October 19, 1929, complaining of pain and weakness in the right leg.

Past History.—He was born in Portugal and had always done heavy manual labor. He had had pneumonia, malaria, gonorrhœa and syphilis, and the last had been treated by intravenous injections. There had been intermittent discharge of pus from both ears for many years, frequent head colds, occasional sore throats, and painful urination with recent difficulty in starting the stream. Otherwise, the past history was negative. He could remember no injury to the back. He used a moderate amount of wine and smoked very little. His wife and six children were alive; one boy aged sixteen was mentally

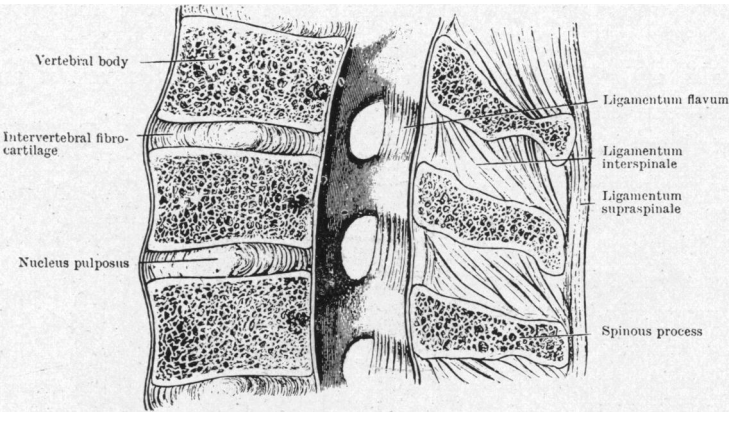

FIG. 2.—Median section through a portion of the lumbar part of the vertebral column. From Cunningham's Anatomy, Wm. Wood & Co., N. Y., 1918.

defective and was a ward of the Juvenile Court, but the other children were mentally and physically normal.

Present Illness.—About five or six weeks before admission he began to have pain in the right lumbar and sacral regions, which soon extended down the back of the right thigh and leg. The pain was always made worse by motion and relieved by rest, and it gradually became more severe. Shortly after the onset of the pain, he noticed a progressive weakness and numbness of the right leg. For a week before admission the pain had been very severe, and weakness had increased so that he could walk only with the help of a cane.

Physical Examination.—The patient was a poorly developed and poorly nourished man, 5 feet, 6 inches tall, weighing 130 pounds. Temperature was 98.6°, pulse rate 82, respiratory rate 18, blood pressure 164/90. Except for the following findings, general physical examination showed nothing abnormal. The mouth showed advanced pyorrhœa and numerous decayed teeth, and the tonsils were enlarged and hyperæmic. The tympanic membranes were perforated, and there was scanty, purulent discharge in both auditory canals.

The cranial nerves showed no abnormality. Motor power, sensation and reflexes were normal in the arms. The abdominal and cremaster reflexes were present. There was no motor loss in the abdominal muscles, and sensation was normal down to the first lumbar segment. The vesical and anal sphincters functioned normally.

All muscles of the right thigh and leg showed occasional fibrillary twitchings. Muscle tone was decreased in the right lower extremity, and there was considerable atrophy, the circumference of the right thigh and leg being about 2 centimetres less than at corresponding levels on the left. All muscle groups of the right lower extremity were weak, but he was able to walk with a cane, throwing out the leg and scuffing the toe. The loss of power progressed rapidly, so that after two weeks all motions of the right lower extremity were absent except for weak flexion of the leg and weak plantar flexion of the foot. Sensation was diminished, and in places almost totally absent, over the distribution of the third lumbar to the second sacral segments inclusive on the right side. No loss could be detected in third, fourth or fifth sacral fields on the right. No motor or sensory changes were ever found in the left lower extremity. The right patellar reflex was at first sluggish and later absent. The left patellar reflex was active. The

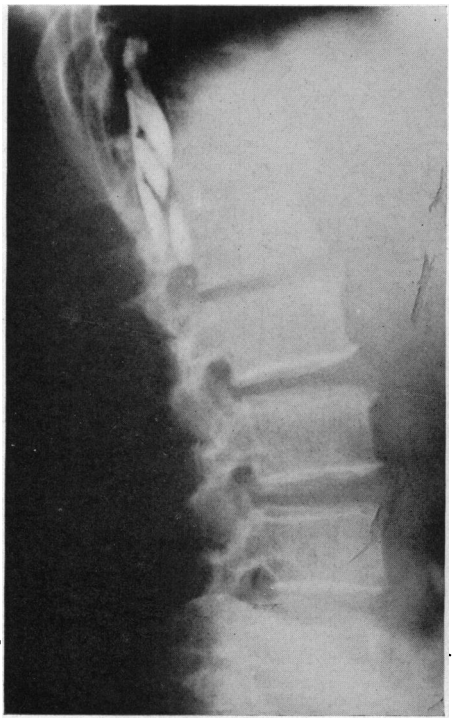

FIG. 3.—Case I. Röntgenogram showing blockade of the lipiodol just above the lower margin of the body of the second lumbar vertebra.

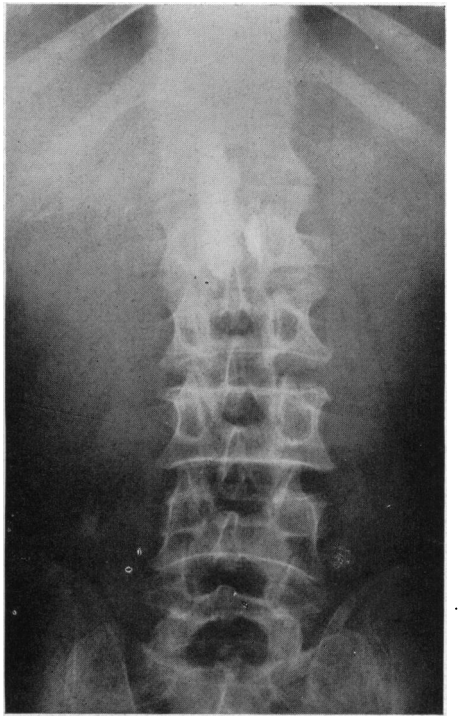

FIG. 4.—Case I. Röntgenogram showing the convexity of the lower margin of the lipiodol, suggestive of an extradural lesion.

right Achilles reflex was absent, the left active. The plantar reflex was normal on both sides.

Röntgenograms of the lumbosacral spine showed hypertrophic osteoarthritis. There were six lumbar vertebræ. Urinalysis and blood count were normal. The Wassermann reaction on the blood was negative. Spinal puncture: the fluid was xanthochromic, the pressure was not recorded, and Queckenstedt's test was not done. Wassermann reaction was negative, no leucocytes, 34 red blood-cells, globulin + + +, colloidal gold (44 54 32 22 33).

The preliminary diagnosis on the medical service was sciatica due to osteoarthritis of the spine. After the report on the spinal fluid, November 7, the patient was referred to the neurosurgical service with the diagnosis of tumor of the cauda equina. November 12, lipiodol was introduced into the subarachnoid space at the cisterna magna, and the

films showed that it was held up just above the lower margin of the body of the second lumbar vertebra (Fig. 3), and that no lipiodol had passed this point after twenty-four hours. In contrast to the concave cap usually seen above an intradural tumor, the lower margin of the lipiodol showed a convexity in the posterior projection (Fig 4), which led to a pre-operative diagnosis of possible extradural tumor.

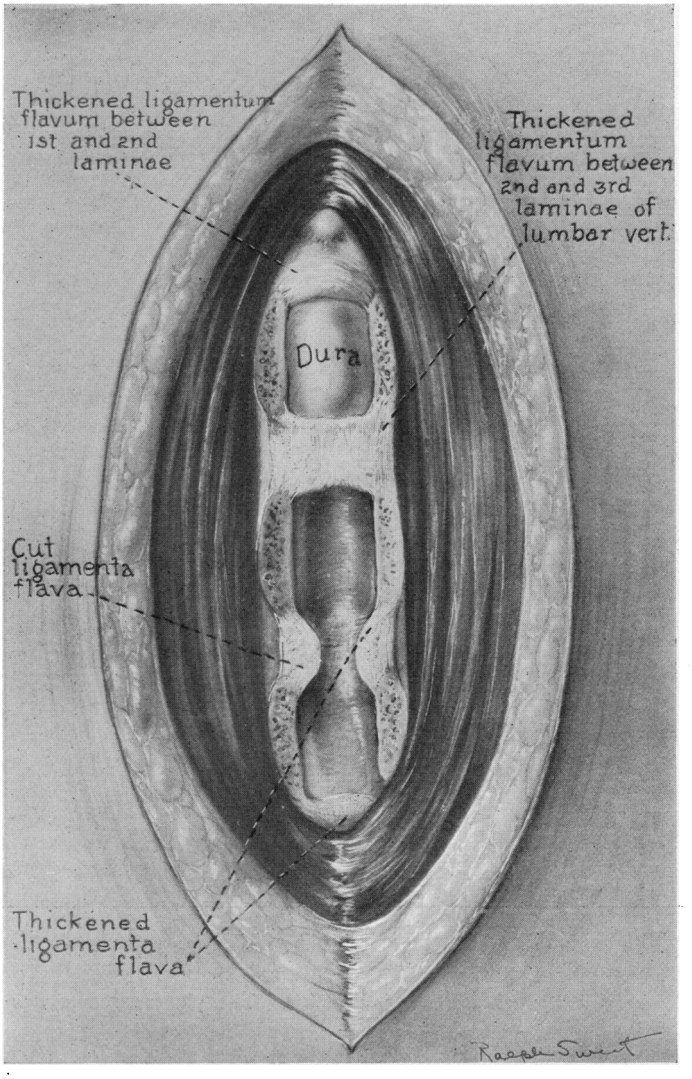

FIG. 5.—Case I. Diagrammatic sketch of the thickened ligamenta flava which compressed the roots of the spinal cord, as exposed by laminectomy.

Operation.—November 14. A laminectomy was done on second, third and fourth lumbar vertebræ. After excision of the spinous processes, dense masses of fibrous tissue bulged up between the exposed laminæ. These masses were not disturbed, though they made removal of the laminæ unusually difficult. The picture after laminectomy is shown semidiagrammatically in Fig. 5. There was no epidural fat, and the dura was so thin that it was almost transparent. The dura opposite the second lumbar laminæ was bulging

and pulsating, but that opposite third and fourth laminæ was partly collapsed, and was not pulsating. On compression of the jugular veins, the dura opposite third and fourth laminæ did not fill. Aspiration opposite the second laminæ gave clear fluid containing lipiodol, and aspiration opposite the third laminæ gave a few drops of yellow fluid, containing no lipiodol. The block was clearly due to the band of tissue which crossed the dura between the laminæ of second and third vertebræ in the normal position of the ligamenta flava. This band was divided down to the dura. It was 7 or 8 millimetres thick, and this thickening extended laterally, so that the dura was constricted to about one third or one fourth of its normal diameter. The inner aspect of the band was smooth, and it was not attached to the dura. When the tissue had been removed, fluid flowed past and filled the dura opposite the third and fourth laminæ. The cut surface of the band was yellowish white and of a very firm consistency. The thickened ligament was excised as far laterally as possible. To a less degree, there was similar thickening of the ligaments between first and second, third and fourth, and fourth and fifth laminæ, which constricted the dura at these points. These were also excised. Lipiodol was now obtained by aspiration opposite the fourth laminæ. The block in the circulation of the

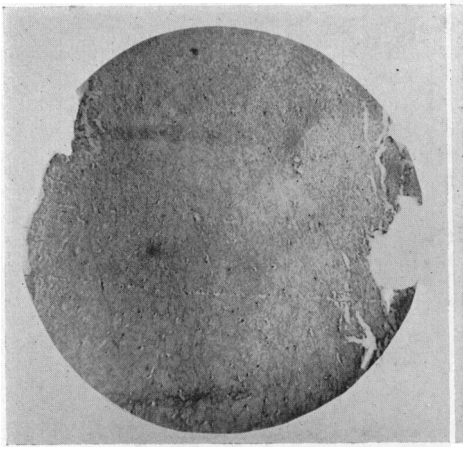

FIG. 6.—Case I. Microscopic section (hematoxylin and eosin) of hypertrophied ligamenta flava, identical in appearance with section of normal ligamenta flava. (Mag. 67 x.)

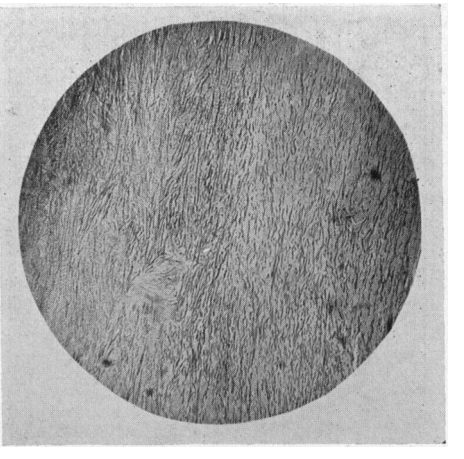

FIG. 7.—Case I. Microscopic section of thickened ligamenta flava stained by Unna's orcein method to show the elastic fibrils. (Mag. 67 x.)

cerebrospinal fluid had been removed, but it was difficult to account for the motor and sensory loss, limited to the right lower extremity, on a basis of concentric constriction of the cauda equina. To rule out a possible intradural lesion, the dura was opened. The nerve roots were slightly hyperæmic, and there was no tumor. The roots were dissected at mid-line and retracted laterally, disclosing a slight ridge, raised perhaps 2 millimetres, along the line of each exposed intervertebral disc. The dura was so fragile that sutures would not hold, so it was left open, and the soft parts were closed.

Two weeks after operation the loss of sensation in the right lower extremity had completely disappeared, and motor power was rapidly improving. December 18, 1929, in the fifth post-operative week, the muscle groups of the right thigh and leg were almost as strong as those of the left, and the patient could walk more than a mile with normal gait and little fatigue. The patellar and Achilles reflexes were active and equal. In March, 1931, sixteen months after operation, reëxamination showed no motor or sensory loss, and the reflexes were normal.

Microscopic sections, stained by hematoxylin and eosin, showed dense fibrous tissue with comparatively few cells. (Fig. 6.) In sections stained by the Van Gieson method, it was seen that only a small proportion of the intercellular substance was collagen. Most

of the fibrous tissue was made up of elastic fibrils, as demonstrated by sections stained by Unna's orcein method (Fig. 7), in which the rather broad, wavy fibrils were stained a light brown color.

CASE II.—(F. L. Reichert.) *Compression of the cauda equina by thickened and partly calcified ligamenta flava, causing flaccid paraplegia. Rapid improvement after removal of thickened ligaments.*

Mrs. C. E., an American housewife, aged thirty-two years, was admitted to Lane Hospital August 24, 1930, complaining of pain in the back and legs, and of inability to walk.

Past History.—There had been no previous illnesses except measles and mumps, acute otitis media at the age of twelve years, and headaches with nausea and vomiting

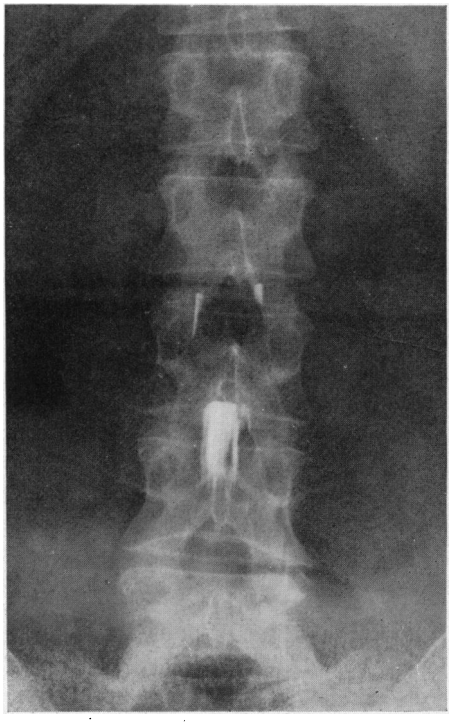

FIG. 8.

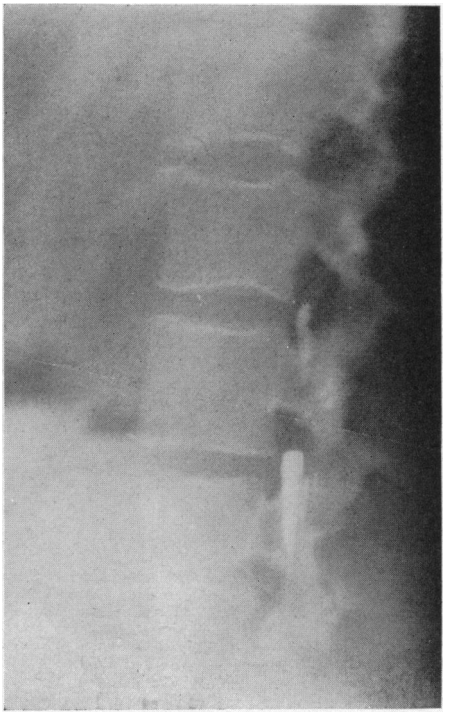

FIG. 9.

FIG. 8.—Case II. Röntgenogram showing blockade of lipiodol at the fourth lumbar vertebra.
FIG. 9.—Case II. Röntgenogram showing the tapered outline of the lipiodol at the level of the lower third of the fourth lumbar vertebra.

once or twice a month since the age of fourteen years. There was no history of injury to the back. She had been married thirteen years, and had one child, aged twelve years. There had been no further pregnancies.

Present Illness.—She had always had a "weak back," with occasional backaches. Seven years previous she had had an attack of severe pain in the lower lumbar spine, with weakness of the legs. It was difficult to arise from a chair and to walk, and she stayed in bed for a month. Three similar attacks incapacitated her, four to eight weeks at a time, during the next six years. Between attacks the legs were practically normal, but she always suffered with backache when fatigued. The fifth attack began in October, 1929, ten months before admission, with pain in the lumbar spine, followed shortly thereafter by pain down the right thigh and leg, and weakness of the right lower extrem-

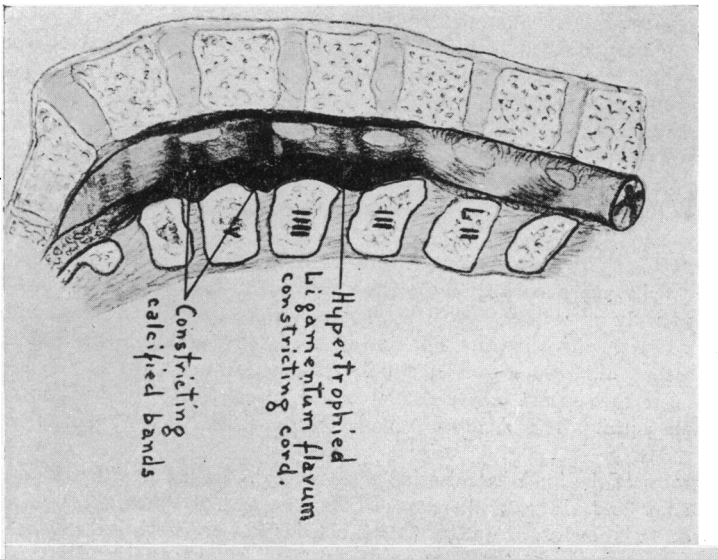

FIG. 10.—Case II. The thick, longitudinal ligamentous structure which surrounded and compressed the dura.

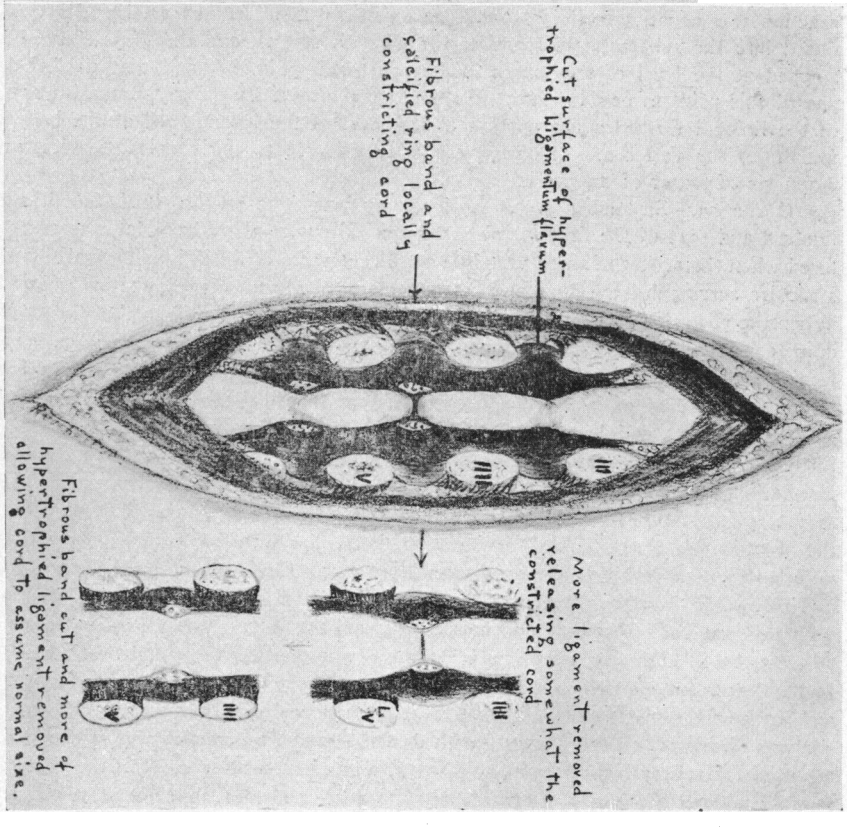

FIG. 11.—Case II. In two of the localized thickenings of the longitudinal ligamentous structure a ring of bone was incorporated. Beneath the ring of ligament and bone at the fourth interspace a fibrous band further constricted the dura.

ity. After some weeks the left lower extremity was similarly involved. In January, 1930, she became bed-ridden. In April, 1930, she noticed numbness of the feet and legs. In July, six weeks before admission, the lower extremities had lost practically all motion, and she began to have difficulty in voiding, which gradually progressed to complete retention one week before admission.

Physical Examination.—A well-developed, rather obese woman lying on her right side with hips and knees flexed. Temperature was 98.8°, pulse rate 80, respiratory rate 20, blood pressure 140/92. The cranial nerves were negative, and motor power, sensation and reflexes were normal in the upper extremities. There was pain on pressure over the lower lumbar spinous processes. The bladder was distended, and the tone of the anal sphincter was poor.

Fibrillary twitchings were seen in the muscles of the lower extremities, and there was muscular atrophy, more marked on the left. Resistance to movements of both lower extremities was hardly perceptible. All muscle groups were flaccid and weak, with very slight voluntary motion at all joints. The patient coöperated poorly in the examination of sensation, so that, although she complained of subjective numbness of the legs and feet, it was possible to map out only a partial saddle anæsthesia. The patellar reflexes were active and equal. The Achilles reflexes were absent. The plantar reflexes were normal.

Röntgenograms of the lumbosacral spine were normal. Spinal puncture in the third interspace gave no fluid. The fluid obtained from the second interspace was slightly bloody, pressure 18 centimetres of water. Queckenstedt's test was negative. Wassermann reaction was negative, colloidal gold (*22 23 21 10 00*). August 29, lipiodol was introduced into the subarachnoid space at the cisterna magna, and the films showed that it stopped at the level of the fourth lumbar vertebra, (Fig. 8), and that no lipiodol had passed this point three days later. In the lateral projection (Fig. 9), the lower margin of the lipiodol formed a sharp point at the level of the lower third of the body of the fourth lumbar vertebra. The patient was transferred to the surgical service with the diagnosis of tumor of the cauda equina.

Operation.—September 4. A laminectomy was done on the third, fourth and fifth lumbar, and part of the first sacral vertebræ. The normal epidural fat was not encountered when the arches of these vertebræ were removed. A thick, longitudinal ligamentous structure surrounded the dura and compressed it, as shown diagrammatically in Fig. 10. When the posterior portion of the ligament was removed, it was seen that the underlying dura, considerably thinned, was further constricted in the spaces between the laminæ by localized thickenings in the ligamentous structure. (Fig. 11.) In the fourth and fifth interspaces, a ring of bone was incorporated in the thickened ligament. Beneath the ring of ligament and bone at the fourth interspace, there was a fibrous band which further constricted the dura. This was the site of the block in the cerebrospinal fluid circulation, as shown by the absence of pulsation below the fourth interspace. The inner surface of the ligamentous structure was smooth and glistening and was not attached to the dura. After removal of all constricting tissue, the dural sac resumed its normal size, and pulsations appeared below the fourth interspace. Exploration above the third lumbar and below the first sacral laminæ showed that a dural sac of normal size was covered with epidural fat. The dura was not opened, and the wound was closed in layers.

On the seventh day after operation there was considerably improved motor power in the lower extremities. On the twenty-first day the patient could stand on her legs and walk with support. She began to void spontaneously on the seventeenth day. When she was dismissed, on the twenty-fourth day, the saddle anæsthesia was unchanged. She reported for reëxamination January 6, 1931, four months after operation. She was able to walk without a cane. Motions of the right ankle and toes were not as strong as those of the left. Sacral anæsthesia was still present on the right, but was now absent on the left, except in the fourth and fifth sacral segments. There was no disturbance of the

sphincters. April 14, 1931, the patient wrote that the left lower extremity was normal, and that the right was still improving.

Microscopic Examination.—The fibrous portion of the specimen was exactly like that from Case I. Fig. 12 presents the picture with hematoxylin eosin stain. The intercellular substance showed a preponderance of elastic fibres, with a few collagen fibres. Sections of the partly calcified portion showed normal bone, which was surrounded by a zone of homogeneous tissue containing cartilage cells. (Fig. 13.) Adjoining this cartilaginous tissue was the typical dense elastic tissue which made up the bulk of the specimen.

Discussion.—The pathological condition in the first case was unmistakable. The ligamenta flava were markedly thickened, and one pair of them had blocked the circulation of the cerebrospinal fluid. There was no extension of the process between adjacent ligaments, the dura being in direct contact with the laminæ. The thickened ligaments were not attached to the underlying

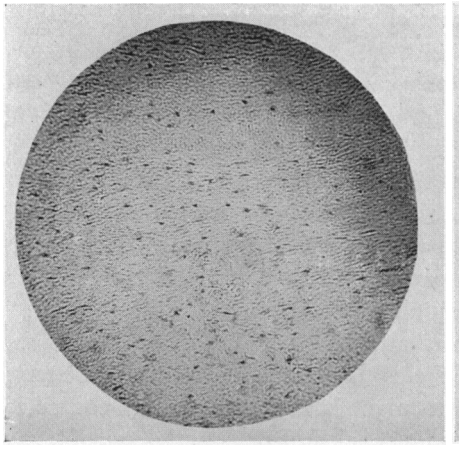

FIG. 12.

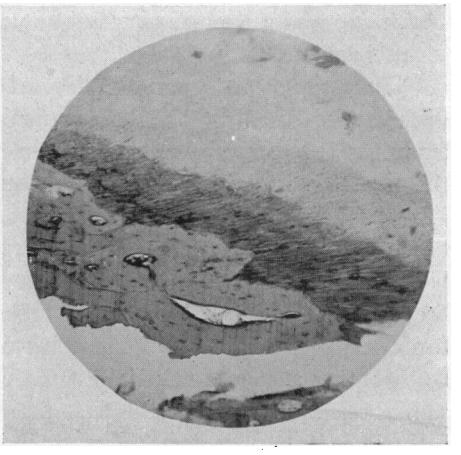

FIG. 13.

FIG. 12.—Case II. Microscopic section stained with hematoxylin and eosin. (Mag. 67 x.)
FIG. 13.—Case II. Microscopic section (hematoxylin and eosin) taken from region of the calcified ring in the ligamenta flava, showing normal bone surrounded by a zone of homogenous tissue containing cartilage cells. (Mag. 67 x.)

dura. The condition found at operation in the second case was more advanced and complicated. The greatest thickening was at the normal sites of the ligamenta flava, and here were the oldest parts of the lesion, as shown by the rings of calcification at these points. However, the proliferation of the ligamenta flava had extended upward and downward, completely ensheathing the dura, with the thinnest portions of the process under the middle of the laminæ. Although the picture, as first exposed, might have suggested an external pachymeningitis, this diagnosis was readily ruled out as the operation proceeded and showed that the extradural mass was entirely separate from the dura, and that its thickest portions were between the laminæ.

Microscopically, the specimens removed in both cases showed yellow elastic tissue, in no way distinguishable from sections of normal ligamenta flava, except that, in Case II, bone had been deposited in the thickest and

probably the oldest portions of the hypertrophied ligaments. None of the sections showed either tumor formation or inflammatory process.

The etiology of the condition is obscure. The first patient gave a history of a supposed chancre, but the Wassermann reactions on the blood and spinal fluid were negative. In the second case, there was no history of syphilis, and the Wassermann reactions were negative. The first patient had many foci of infection, in the teeth, tonsils and middle ears, and he had an advanced osteoarthritis of the spine; but the second was free from obvious foci, and the röntgenograms of her spine showed only a few hypertrophic fringes in the lumbar vertebræ. Certainly the condition here described is in no way analogous to the compression of the spinal cord and its roots by osteoarthritis, as described by Bailey and Casamajor.[1] Neither patient gave a history of trauma to the back, and although the first, being a laboring man, may well have had repeated mild injuries, the second patient at the time of onset of symptoms was a woman twenty-five years old, in whom unrecognized trauma seems most unlikely.

The diagnosis of compression of the cauda equina was made without difficulty on the history and physical findings in each case. The lipiodol examinations offered a suggestion, in the shape and position of the shadows, that the lesion might be extradural, but there was no pre-operative suspicion of the true character of the lesion. The history of the second patient was unusual in that she had passed through four transient attacks of paraplegia before the final one which did not clear up. The first of these episodes occurred seven years before she came under observation, and the lower extremities were apparently normal between attacks. It is interesting to speculate whether the elastic tissue of the thickened ligaments may explain the long remissions, and whether the first patient might have had a similar course if he had not been operated on during his first attack, before both lower extremities were involved.

Summary.—A new cause of compression of the lumbosacral roots of the spinal cord is reported, with two illustrative case histories. The condition is a proliferation of the ligamenta flava between laminæ of the lumbar spine, which eventually causes a block in the cerebrospinal fluid circulation and compression of the cauda equina. The etiology is unknown. The pathological process is a simple hypertrophy of the ligamenta flava. Extradural compression may be suspected from the position and shape of the lipiodol shadows. The symptoms of compression of the cauda equina were cured by surgical removal of the thickened ligaments.

REFERENCE

[1] Bailey, P., and Casamajor, L.: Osteoarthritis of the spine as a cause of compression of the spinal cord and its roots, with report of five cases. J. Nerv. and Ment. Dis., vol. xxxviii, p. 588, 1911.