DOUBLING OF THE SPINAL CORD*

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Variations in the gross anatomy of the spinal cord are unusual. Observation of many spinal cords, in our regular class dissections during a period of five years, tends to confirm this statement. During this period one double spinal cord was found. A careful search of the literature failed to disclose the report of a case similar to the one I describe. Although it was impossible to obtain a clinical history, the variation is so unusual as to be of more than casual interest to the clinician, the surgeon and the anatomist.

This rare variation was encountered during the regular laboratory work in the dissecting rooms at the Daniel Baugh Institute of Anatomy of the Jefferson Medical College. Unfortunately the spinal cord was not in a state of preservation to admit microscopic study of serial sections.

The specimen was obtained from an adult white male cadaver, well nourished, about 35 years of age and of unusually good physique. A careful external examination showed no abnormalities. The vertebrae were normal and there were no abnormalities of the bony canal, either ventral or dorsal to the spinal cord. The skin and overlying tissues of the lumbar and cervical region were likewise without abnormalities.

On opening the dural sac, an unusual variation of the spinal cord and inner surface of the dura was discovered. The spinal cord was divided in its caudal portion into two distinct symmetrical halves, the two entirely enclosed in a single dural sheath as shown in the illustration. Similarly, the arachnoid stretched across as a single membrane. The bifurcation began at the caudal border of the eleventh thoracic vertebra and extended to the caudal limit of the cord, forming two independent symmetrical halves. The caudal limit of each half was at the level of the cephalic border of the first sacral vertebra. The conus medullaris of each half was independent and at the level of the third sacral vertebra, they united into a single large filum terminale, the latter coursing caudally for a distance of 5 cm. to gain attachment to the dorsal surface of the first segment of the coccyx.

The cervical and thoracic portions of the cord were normal in appearance. The peripheral nerves cephalic and caudal to the bifurcation were carefully dissected and presented no variations in the formation of the different plexuses and in their normal peripheral distribution.

The inner surface of the dura at the level of the caudal border of the twelfth thoracic vertebra likewise showed an interesting and rare anomaly. Where the division of the cord was total, a distinct band of dura was found

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projecting between the two halves of the cord. The band of dura was directed ventrodorsal and had a first attachment of 1 cm. on both the ventral and dorsal ental surfaces of the dura. It was of an hour-glass shape and was placed in the sagittal plane. This band or fold was clearly a part of the dura, being of the same texture and consistency. In the unopened dura it seemingly caused no pressure on the halves of the cord. The division of the spinal cord began 0.5 cm. cephalic to dural band noted above.

There were also present, as shown in the illustration, a number of small flattened calcareous plaques adherent to the spinal arachnoid; the largest was about 7 mm. in its long diameter. Each plaque appeared to be accompanied by a small artery. The calcareous plates will be dealt with more in detail in the discussion of this paper.

The unusual features of this specimen are: first, the presence of a bifurcated spinal cord beginning at the twelfth thoracic vertebra; second, the presence of an hour-glass-shaped dural band directed in the sagittal plane and firmly attached to the ventral and dorsal ental surfaces of the dura, which entered the dorsal median fissure and completely divided the cord; third, the absence of other abnormalities that not infrequently accompany bifurcation of the spinal cord; fourth, the extreme caudal extent of the spinal cord—first sacral vertebra.

EMBRYOLOGY

Kunitomo,¹ in a careful study of a large number of human embryos, shows that in very young specimens the spinal cord reaches the extreme tip of the tail, and throughout its length is quite uniform in structure. He further calls attention to the fact that in the formation of the filum terminale in addition to the dedifferentiation of the caudal end of the medullary tube, there is also mechanical disproportion between the growth of the medullary tube and that of the vertebral column. Streeter's studies show that the filum terminale represents that portion of the spinal cord caudal to the second coccygeal segment. This strand elongates by interstitial growth in adaptation to the ascending displacement of the cord. The part of the cord that undergoes dedifferentiation into the filum terminale is caudal to the ventriculus terminalis (the fusiform dilatation of the central canal in the lower end of the conus medullaris). The caudal tip of the dural sac maintains its initial relation to the vertebra rather than to the definitive spinal cord, and remains attached to the filum terminale in the sacral region at a more or less fixed point. Early in the embryo the spinal cord and the vertebral column lie alongside of each other in a metameric manner, corresponding in position segment for segment. Owing to the disproportion in growth between the spinal cord and the vertebral column, relative displacement of their respective levels occurs. In the first twenty-five weeks there is an ascent of the ventriculus terminalis from the level of the second coccygeal vertebra to the third lumbar, and

1. Kunitomo, K.: Contributions to Embryology, Vol. 8, Publication No. 271, Carnegie Institute of Washington 8: 1918.

The lower part of the dural sac has been opened and turned aside. The arachnoid with the superimposed calcareous plaques is shown stretching across the spinal cord to the level of the eleventh thoracic spinal nerve indicated at N. The arachnoid below this level has been removed to show the dural plaque projecting between the halves of the spinal cord. Y indicates a calcareous plaque; N the eleventh thoracic nerve and X the dural plaque.

there remain but two segments before the adult position is reached. One may say that the principal part of the migration of the cord is accomplished during the first half of fetal life. It is thus evident that the adult position of the cord is the result of two factors — the dedifferentiation of the caudal portion of the neural tube into the filum terminale, and the disproportionate growth between the spinal cord and the vertebral column. Another factor may also enter into the formation of the filum terminale: The relation of the pia overlying the spinal cord is early established, likewise the caudal tip of the dural sac holds the same position in the embryo and the adult. It would seem plausible that as the cord ascends, due to the caudal growth of the vertebral column, the pia is gradually attenuated, interfering with the proper nutrition of the part of the spinal cord destined to become the filum terminale. The more caudal extent of the cord is, therefore, not due to a partial failure of the cord to ascend as is commonly thought, but is the result of a failure of a part of the embryonic undifferentiated cord to continue its retrogressive changes into the filum terminale.

Obviously, the length of the filum terminale depends on the caudal limit of the spinal cord. In cases in which the spinal cord extends more caudally than its usual limit, there is a corresponding decrease in the length of the filum terminale.

The neurenteric canal is a transitory communication between the neural tube and the primitive digestive tube. It is purely a temporary structure having normally completely disappeared by the end of the third week. Certain anococcygeal tumors are believed to arise from the neurenteric canal, as well as from the hind-gut. Such a canal is represented by a remnant passing from the rectum to the sacral region of the spinal canal. Keen, in 1906, reported a sacrococcygeal tumor associated with a spinal bifida and a sinus beginning at the skin, perforating the sacrum and the spinal canal, and terminating in the lumen of the gut.

The dural band present in this specimen seems to have been the factor that produced the division of the cord. I have been unable to explain the genesis of this variation on the basis of our known embryology of the cord and dura. The dural band appears to be due to a disturbance of the mesothelium resulting in an excessive mesothelial activity at this point with the projection of a fold of dura between the two halves of the cord.

LITERATURE AND DISCUSSION

Doubling of the spinal cord is a rare anomaly, and is the bifurcation of the medulla spinalis into two symmetrical or asymmetrical halves for a variable part of its course. The division usually occurs

in the lumbar or lower thoracic segments of the cord. Hanover describes a case of complete division of the spinal cord with a distinct central canal in each half extending to the fourth ventricle. Frequently this variation is only one of many anomalies in the same subject. Doubling of the spinal cord has been found associated with rachischisis, myelomeningocele, hydromeningocele and anencephalus. Frequently the anomaly is associated with cutaneous and bony malformations.

A complete doubling of the cord is one in which two well-marked symmetrical halves are present; at times one half is rudimentary and the other well developed, or the two halves are asymmetrical. The spinal cord substance collectively is probably decreased when the division of the cord is well marked. In a division that is not well-marked, or in an otherwise nondefective cord, the gray and white matter of both halves are equal to the normal amount of spinal intramedullary substance. The central canal may be absent or may be present as a distinct canal in each half, varying in outline. Thirty-five cases of double spinal cord were collected by Steiner;² twelve were eliminated by Bruce, McDonald and Pirie³ as being merely split cords and thirteen as artefacts, leaving ten cases of true doubling of the cord. Of the latter, seven were associated with spina bifida. In seven of Steiner's thirty-five cases the bony vertebral canal was normal. The age of Steiner's collected cases ranges from a 3 months' old fetus to an adult of 76 years. In Steiner's cases, eight occur in adults whose ages range from 19 to 76 years. In one case the doubling was limited to the cervical and upper thoracic segments; in another (Hanover's case mentioned above) the division involved the entire cord. All other cases occurred in the lumbar region. The case described by Steiner occurred in a female infant 3½ months old. A sacral swelling was present which fluctuated and when opened revealed an oval communication with the central canal. There was likewise a medial defect of the bony vertebral column consisting of a split 2 cm. long beginning on the first sacral segment and continuing through the sacrum and coccyx. The division of the cord began at the caudal border of the fifth lumbar vertebra, the cord continuing to the level of the third sacral vertebra as two independent halves. Each half resembled a normal cord.

There are cases reported showing diffusion and spreading of the gray commissure, but without division of the white matter. These are not true and complete divisions of the cord.

2. Steiner, F.: *Ueber Verdoppelung des Rückenmarks*, Königsburg, 1896.

3. Bruce, A.; McDonald, S., and Pirie, J. H. H.: A Case of Localized Doubling of the Spinal Cord, *Rev. Neurol. & Psychiat.* **111**:709, 1905. A Second Case of Partial Doubling of the Spinal Cord, *ibid.*, 1906, IV, 6-19.

Bruce, McDonald and Pirie report a case of doubling of the cord occurring in a woman, aged 31. There were no sensory or motor symptoms. In the lumbosacral region the cord divided into two distinct trunks entirely enclosed in a single dural sheath. The division was total and a fold of dura projected for a certain distance between the two halves not reaching the ventral surface of the dural sac. The cord was completely divided for about 2 inches. There were no abnormalities of the bony vertebral canal. In the case described in this paper the fold of dura projected between the halves of the cord and had a distinct attachment to the ventral and dorsal ental surfaces of the spinal dura.

Bernstein⁴ has described a case of bifurcation of the spinal cord beginning at the first lumbar segment. Examination of sections of the cord showed two seemingly complete cords united slightly along their mesial aspects. Each half of the cord presented two anterior and two posterior cornua. Caudal to the third lumbar vertebra the two halves of the cord seemed to fuse again. The filum terminale was single and large. At the level of the second lumbar segment each half exhibited a distinct central canal.

Van Gieson has called attention to the fact that artefacts are easily produced simulating a bifurcation of the spinal cord and certain cases demand the most careful study to eliminate the doubt of an artefact. This statement is confirmed by Spiller in a study of a specimen of what seemed a double spinal cord. On more careful examination and study he decided it was an artefact.

Westphal⁵ has reported an unusually interesting case of a lumbar division of the spinal cord occurring in a woman, aged 29. There were no associated abnormalities. On opening the dural sac the spinal cord was found divided in its caudal portion into two distinct halves, the left half being broader and larger than the right. The dura exhibited no abnormal variations. The left half presented in its distal portion a distinct cyst which was filled with glassy mucus. The walls of the cyst were whitish in color and spotted with yellowish red areas. The latter on close examination proved to be small vessels and small hemorrhagic spots. The cervical and thoracic segments of the spinal cord were normal in appearance. Deep in the dorsal median fissure of the spinal cord numerous smooth muscle fibers were demonstrable on microscopic examination. The strands of smooth muscle increased in number caudally, blending with a distinct swelling also composed of smooth muscle. The swelling presented in its caudal portion a lumen, cylindrical in outline and forming the walls of the cyst noted above. The mass of muscle fibers and the spinal cord were covered by the overlying pia. The right side likewise presented a swelling smaller than that of the left and similarly composed of smooth muscle. Some of the nerve fibers streamed irregularly through the cyst wall. The cyst was not a dilatation of the central canal as a distinct ependymal canal was demonstrable in each half of the cord at levels with the cyst. The walls of the cyst as noted were formed of smooth muscle and microscopic examination of serial sections revealed no nervous tissue or ganglion cells.

4. Bernstein, J. M.: Doubling of the Spinal Cord, *Trans. Path. Soc. Lond.* 7:318, 1905-1906.

5. Westphal: Ueber eine bisher auscheinend nicht beschriebene. Missbildung am Ruckenmarke, *Arch. f. Psychiat.* 46:712, 1906.

A number of swellings, myofibrolipoma in character, have been reported by Von Recklinghausen, Fischer, Von Ribbert and Marsh usually associated with lumbar spina bifida. In the case of Westphal the cyst was composed of smooth muscle fibers extending into the dorsal median fissure. The growth of smooth muscle fibers along the cord was first noticed by A. Pick. This interesting observation noted by Pick showed the presence of myomatous growths about blood vessels in the white and gray matter. Bonnet believes they represent a remnant of the hind-gut of the embryo occurring as an outgrowth from a rest of the hind-gut before the back is completely formed. Marchand and Von Recklinghausen show the relation between the neurenteric canal and the spina bifida. Middendorph comments on certain large intestinal tumors in the region of the coccyx as the persistence of the postanal opening. Muscatello has described a case of a fetal anomaly in which the lumen of the gut tract communicated by a winding canal directly with the central canal of the cord, confirming the observation of Marchand and Von Recklinghausen relative to the abnormal communication of the intestinal tract and the caudal portion of the ependymal canal. The latter is to be particularly remembered in spina bifida. Tillman states that growths in the coccygeal region may occur as rests of the medullary tube, the chorda, intestine, muscle and skeleton.

Mention was made in a description of this specimen of the occurrence of a number of calcareous plaques adherent to the spinal arachnoid. Calcareous deposits within the central nervous system are most common on each side of the superior longitudinal sinus as the result of calcium deposits in the arachnoidal granulations or villi (pacchionion bodies). The writer has observed the occurrence of macroscopic calcareous arachnoidal plaques in approximately 10 per cent. of all cadavers. The deposits, according to Cushing and Weed,⁶ are composed of true bone as is evidenced by the occurrence in them of the typical lacunar system, Haversian canals and the characteristic osteoblasts and osteoclasts. Another example of bony deposition is the occurrence of an osseous nodule in the falx cerebri. The observation of the dura and its reflections in our neuro-anatomy laboratory frequently show this example of bony deposit, in reality osteophytes and practically always confined to the falx cerebri. Calcareous deposits appear to be a normal physiologic process in two structures of the central nervous system, the corpus pineale and the plexus choroideus. Virchow called attention to the occurrence of lime salts in the cerebro-

6. Cushing, H., and Weed, L. H.: *Studies on the Cerebrospinal Fluid and Its Pathway*, No. IX. Calcareous Deposits in the Arachnoidea. *Johns Hopkins Hospital Bulletin* 26:367, 1915.

spinal axis in that type of cerebral neoplasm which he designated psammome. Weed and Cushing have noted the occurrence of a thickened arachnoid covered with plaques of true bone in a patient suffering from epilepsy. He had, in early life, been a victim of severe infection.

Weed, in a study of a series of brain tumors, found a deposition of calcium salts in 40 per cent. of all cases. They have been described in all slowly-growing cerebral neoplasms. Bony growths in the form of osteophytes arising from the meninges are of frequent occurrence and have been repeatedly recorded.

In conclusion, it is interesting to comment on the possible effect of a lumbar puncture in a patient with a cord extending caudal to third or fourth lumbar vertebra. The author is not willing to venture an opinion because of the rarity of its occurrence, yet it seems a fair assumption that injury to the cord would follow with its resulting symptoms. It is not far-fetched to state that occasional untoward effects, such as paralysis following lumbar puncture, may be due to the presence of a cord extending to levels below its normal caudal limit.⁷

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7. In addition to the references given, the following may be of interest:

Stewart, J. P., and Bernstein, J.: A Case of Partial Doubling of the Spinal Cord, *Rev. Neurol. & Psychiat.* **4**:729, 1906; Streeter, Geo. L.: Factors Involved in the Formation of the Filum Terminale, *Am. J. Anat.* **25**: No. 1.