Dural cysts in the cervical region

Report of three cases and review of the literature

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Various types of cystic lesions are encountered in the spinal canal and are mainly congenital, inflammatory, or traumatic in origin. They are classified according to the nature of the tissue composing the cyst and its relationship to the adjacent structures. Intradural cysts are mainly arachnoid, endodermal (enterogenous), or ependymal (neuroepithelial).

Between 1992 and 1993, three patients with cystic lesions in the cervical spinal canal who presented with atypical neurological findings were admitted to our hospital. All three patients had a long history of dysesthesias and atrophic pareses in the proximal upper extremities. Intraoperatively we found cystic lesions: two were located ventrally and one dorsally to the spinal cord. All three specimens resembled dural tissue both on visual inspection and later on histological examination. The clinical, radiological, and pathological findings are described and the pertinent literature is reviewed.

Case Reports

Case 1

History. This 50-year-old woman was admitted to the neurological outpatient department of Grosshadern University Hospital for the first time in 1985. She complained of a 2-year history of neck and right shoulder pain and of weakness of the proximal left arm. The clinical history was significant for a major chemical accident in 1974, in which the patient was exposed to fluorinated and chlorinated hydrocarbons. As a result of that accident, she had to be treated for toxic lung edema for 4 weeks in an intensive care unit.

The results of cerebrospinal fluid (CSF) cytological examination and magnetic resonance (MR) imaging were normal in 1985 and subsequently in 1987; however, the weakness of the shoulder muscles and the dorsiflexors of the left foot progressed.

Examination. In 1992, the patient developed distal weakness in both legs and shoulders; the pain in her neck and left arm increased. Her joint position sensation was completely lost in the toes of both feet. Electromyography revealed generalized chronic changes that were more pronounced at C-5, bilaterally. Electromyography revealed generalized chronic changes that were more pronounced at C-5, bilaterally. Cortical stimulation was normal but tibial somatosensory evoked potentials (SSEPs) were abnormal on the right arm.

Repeated myelographic examinations showed narrowing of the subarachnoid space from C5–7. Metrizamide-enhanced computerized tomography (CT) scanning disclosed ventral dislocation of the spinal cord (Fig. 1 left) and a liquid-filled space-occupying lesion dorsal to the spinal cord, which showed delayed washout (Fig. 1 center) of contrast medium from C-2 to L-1. Additionally, a ventral liquid cyst in the thoracic region was suspected. Magnetic resonance imaging revealed only a fluid-containing space dorsal to the spinal cord from C-2 to L-1. The findings were interpreted as arachnoidal cyst.

Operation. The operation was performed using a microsurgical approach. After removing the left hemilamina from C-3 and C-4, an apparently normal dura was seen and clear CSF could be visualized within the cystic struc-
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Fig. 1. Case 1. Left: Metrizamide-enhanced CT scan obtained at C3–4 in a 50-year-old woman, showing an enlarged dorsal subarachnoidal space. Center: After 6 hours the dorsal cyst is still filled with contrast medium, whereas the subarachnoidal space is nearly clear of contrast. There is marked compression of the spinal cord. Right: After surgical communication was established between the dural cyst at C3–4 and the subarachnoidal space via hemilaminectomy, the cyst is visible only as a small margin and does not compress the cervical spinal cord.

Postoperative Course. The cyst could not be detected on postoperative MR imaging. After 14 months, the patient reported being free of pain. Her gait disturbance had been ameliorated and she claimed to have easier fine movements of the hands. On examination, the strength of her legs was almost normal; the other muscle groups were unchanged. The results of electromyelography and SSEPs remained unchanged. A control metrizamide-enhanced CT scan of the cervical spine revealed the spinal canal to be of normal size; the cyst was seen only as a small lesion without space-occupying characteristics (Fig. 1 right).

Case 2

History. This 23-year-old man, a former army officer, noticed for the first time in 1987 a minimal weakness of his left arm. This was primarily attributed to a sports accident he suffered in 1985, during which he fell on his stretched left hand. After this accident, he noticed for a short period of time a weakness in the flexor muscles of his left upper arm. He stopped his Kung-Fu activities, but remained in the service.

In 1988, a slowly progressing atrophy of the shoulder and upper arm muscles developed. In 1990, he was taken out of active service because he suffered from progressive muscle dystrophy. After he moved to the western part of Germany, diagnostic procedures were repeated, and, on confirmation of an intraspinal cyst, the patient was admitted to our department.

Examination. On admission, advanced atrophy of both shoulder and upper arm muscles was noted. The patient was not able to lift his left arm more than 30°. Although he reported tingling sensations in the left hand, disturbance of the lower extremities was absent; he had no pain or sensory and vegetative signs.

As early as 1990, myelography had shown ventral space-occupying lesions, which were suspected to be of posttraumatic nature (Fig. 2 upper left). The results of CSF cytological examination were unremarkable. On MR imaging, a low signal intensity was noted ventral to the spinal cord because of thickened ligamentous tissue. In 1993, MR imaging disclosed a cystic lesion ventral to the spinal cord, ranging from C-2 to T-3 (Fig. 2 upper right and lower left and right). Myelography also showed a ventral space-occupying lesion (Fig. 2 upper left), without early uptake of contrast medium, whereas delayed CT could detect contrast medium inside the cyst.

Operation. Hemilaminectomy of C-3 was performed. Examination of the area around the dural sac by using a nerve hook did not reveal any abnormalities. Adhesions could not be detected; the epidural veins were unremarkable. The dura was incised 1 cm paramedially and the roots as well as the denticulate ligament were exposed. There was no abnormality inside the dura, except that the ventral dura seemed to be dorsally dislocated. The cyst, containing clear CSF, was opened by puncture and incision. The wall was then fenestrated after sectioning of the denticulate ligament. Communication between the cyst and the subarachnoidal space was established and a 1-cm silicone catheter was sutured in place to keep the window open. Adhesions of the ventral dura to the posterior longitudinal ligament in the exposed level could not be detected.

Postoperative Course. Six months postoperatively the patient reported having more strength in his arms and that the tingling sensations had ceased. He was able to lift
his arms more than 90˚, although the muscle atrophy remained unchanged.

Case 3

History. This 51-year-old man had a 12-year history of intermittent head and neck pain radiating into both shoulders. After manipulation of the cervical spine 10 years ago, he developed progressive headache. A CT scan of the head revealed a left-sided chronic subdural hematoma. In 1983, the patient had been treated by burr-hole trepanation. A typical hematoma membrane was found, and the hematoma was evacuated. There was no visceral membrane. The patient's postoperative course was uneventful and he went back to work as an author. Two and a half years before the second admission, he noticed a slowly progressive weakness of the upper extremities, which was slightly more pronounced on the left side. Sensory dysfunction and signs of the long spinal pathways were absent. A cervical space-occupying lesion was diagnosed and the patient was referred to our hospital.

On admission, marked atrophy of both shoulder muscles was evident. Electromyographic examination of the upper extremities showed no spontaneous discharges, but marked chronic neurogenic changes in the biceps, triceps, and deltoid muscles on both sides. The results of electromyographic examination of the lower extremities were normal.

Plain cervical CT scanning revealed marked degenerative changes in the lower spine, most pronounced at C5–6 and C6–7. Magnetic resonance imaging could not detect a disc protrusion, but revealed a dorsal displacement of the spinal cord by a liquid mass extending from C-2 to the upper thoracic level. Myelography demonstrated bone spurs compressing the ventral arachnoid space at C5–6 and C6–7 and an increased distance between the ventral dura and the dorsal longitudinal ligament. This proved to be a CSF-containing cyst as revealed by slow contrast medium ingress and washout on sequential postmyelographic CT scanning (Fig. 3).

Operation. Laminectomy of C-5 and C-6 was performed to achieve dorsal decompression and gain access to the cystic lesion. After the paramedian opening of the dura, the fascicles of the root appeared normal. The inner aspect of the ventral dural sac seemed to be dorsally dislocated. After dissection of the denticulate ligament, the ventral dura was fenestrated. The dural cyst was opened; clear CSF was exuded and a 1-cm silicone catheter was sutured in place.

Postoperative Course. The patient reported slow recovery of his weakness in both arms; muscular atrophy was not significantly altered after 1 year.

Histological Findings. On histological examination in all three cases, the “duralike” structure consisted of dense fibrous tissue with irregular texture, dense areas beneath the reticular structures, and small areas of dystrophic calcification (Fig. 4). The parallel arrangements of collagen fibers to the secondary structure of ligaments was seen only in some areas. Elastic fibers were also present in the specimen. No epithelial or arachnoid lining or neural tissue (for example, nerve fibers) could be detected. In all three cases, the diagnosis was a cyst formation of the dura mater.

Discussion

Spinal cysts are benign lesions that are classified as arachnoid, endodermal, or ependymal cysts. 4,6,9,11,12

Arachnoid cysts and diverticula account for approximately 50% of all intradural cysts and are predominantly located at the thoracic level in a posterolateral position. The mean age of the patients in the studies reviewed was 34.8 years; there was no gender preponderance. Histologically, the lesions are mainly composed of arachnoid tissue.

Endodermal (neurenteric, enterogenous) cysts comprise approximately 40% of all intradural cysts and are located predominantly at the cervical level in an anterior position and at the conus–cauda level in a posterior position. The mean age of the patients reported was 27 years. There was a 2:1 male preponderance. Microscopically, the cyst is lined by stratified or cuboidal partially ciliated epithelial cells that rest on a basement membrane.

Ependymal (neuroepithelial) cysts account for approximately 10% of all intradural cysts and are located predominantly at the thoracicolumbar level in an anterolateral position. The mean age of the patients reported was 42.4 years. There was a 2:1 female preponderance. Histologically, these cysts show an ependymal (simple cuboidal) lining without a basement membrane.

In their reviews, Fortuna and Mercuri 2 and Goyal et al., 4 do not describe a cystic lesion originating from the dura itself. To the best of our knowledge, only seven patients with similar dural cysts have been reported in the literature, and these are summarized in Table 1. 2,5,8,10 A similar dural cyst of the cerebellum has been reported in a 9-month-old boy. 7

Whereas the intracranial dura mater can be separated into two distinct layers with a potential epidural space, the spinal dura represents a downward extension of the inner layer of the cerebral dura. 2,8 In accordance with the observation made by Leaver and Kempe 8 and Done, et al., 7 we

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![Fig. 3. Case 3. Computerized tomography scan obtained at the level of C-6 in a 51-year-old man. Twelve hours after instillation of metrizamide, the contrast medium is still present within the ventrally located intraspinal cyst, which proved to be of dural origin.](image-url)
consider dural cysts to be a developmental duplication of a localized area of spinal dura mater, which slowly evolves into a space-occupying lesion as CSF accumulates within it. In contrast to Leaver and Kempe,8 we did not observe enlarged veins in the periphery of the cavity. The arachnoid membrane itself did not contribute to the cystic lesions and, thus, an arachnoid cyst was excluded. Furthermore, an epithelial or neural lining, consistent with an endodermal, or ependymal, or perineurial cyst, could not be detected. Therefore, because of their completely dural composition, these lesions are correctly classified as dural cysts.

We consider that dural cysts originate from the splitting of the two leaves of the spinal dura mater at locations where the adherence of the two leaves is significantly reduced due to unknown mechanisms, which might be congenital. We assume this is a long-term process during which CSF diffuses continuously and slowly into the cavity and accumulates until the size of the cavity produces problems in the spinal cord. Although one of our patients (Case 2) had a history of major trauma and another (Case 3) a history of manipulation of the cervical spine that led to a chronic subdural hematoma, we found no evidence of trauma in Case 1. Therefore, we do not think that trauma is responsible for this lesion. Additional immunohistochemical investigations into the distribution of connective tissue proteins (such as laminin, fibronectin, collagen, and heparan-sulfate proteoglycans) might provide new insights into the understanding of these rare entities.

In all three cases, cystoarachnoidal shunting proved to be effective. The wall between the arachnoidal space and the cyst could be fenestrated via a limited microsurgical approach at the level of the largest diameter of the cystic lesion and kept open by a small silicone catheter. Thus, no recurrence of the cyst has been observed so far. Clinically, the progression of the muscular atrophy could be halted and motor function was improved to a certain degree. Complete restitution was most probably not possible because of the long preoperative course. On MR imaging, these cysts presented an intraluminal signal identical to that of CSF; only in Case 1 could the cystic wall be detected (Fig. 2 upper right). Confirmation of the diagnosis could only be obtained by late postmetrizamide CT scanning.

Conclusions

Because these cysts were undiagnosed in our patients for such a long period of time, it was not possible for the patients to recover completely. If MR imaging presents long-range dislocation of the spinal cord, myelography and late CT scanning are recommended. Cystoarachnoidal shunt placement can stop the progression of motor deficits.

References


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