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Intradural sequestration of a L_3–L_4 herniated intervertebral disc. Case report and literature review

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The issues of etiology, pathogenesis, diagnosis and surgical treatment of intradural sequestration of herniated intervertebral discs are poorly reviewed in the world literature. This pathology occurs in 0.26–0.30 % of cases of all intervertebral disc herniations. The basis of pathogenesis is scar-adhesive changes in the spinal canal, leading to dense fusion of the dura mater with the posterior longitudinal ligament and collapse of the ventral epidural space. The main clinical manifestations are long-term pain in the lumbar spine with possible radicular symptoms. There was also a high incidence of acute caudal radicular ischemia (29 % of cases). An important factor in the differential diagnosis of intradural sequestration of intervertebral discs and intradural extramedullar neoplasms are neuroimaging phenomena (hawk-beak sign, Y-sign, ring enhancement) observed on magnetic resonance imaging in the native mode and with intravenous contrast enhancement. The correct preoperative diagnosis and, as a result, competent planning of surgical intervention are the keys to a favorable clinical outcome in this group of patients.
The article describes a clinical case of 55-year-old patient treatment who underwent surgery in 2023 to remove an intradural sequestration of a herniated intervertebral disc at the L_3–L_4 level. An overview of the world literature is also presented, and the features of this pathology important for medical practice are discussed.

Keywords: herniated intervertebral disc, intradural sequestration, microsurgical discectomy, cauda equina syndrome

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INTRODUCTION

The problems of surgical treatment of intervertebral disc herniations are the subject of many scientific papers with sufficient detailed description of tactic and surgical algorithms. At the same time, intradural sequestration of intervertebral disc herniations is extremely rare, occurring in only 0.26–0.30 % of cases [1]. The correct diagnosis at the preoperative period is established only in 25 % of cases [2]. The rare occurrence and sporadic nature of distribution explain the insufficient coverage of this problem in the world literature. Most publications are descriptions of clinical cases, there are isolated systematic reviews of the literature, and the largest series of clinical observations, published by G. Schisano et al. in 1995, includes only 9 cases [3]. That indicates the relevance of this problem, and each published clinical case is important for studying the characteristics of this pathology. This article describes a clinical case of a patient with an intradural herniated L_3–L_4 disc and also provides a review of current world literature.

CLINICAL CASE

Female patient S., *55 years old, had been suffering from lumbar pain for 5 years. According to the patient words the pain radiated along the posterolateral surface of the thigh and anterolateral surface of the lower tigh for the first time (9 points by the visual analogue scale (VAS)), numbness in the area of the 4th and 5th toes of the left foot, and the lumbar pain increased to the point of unbearable 2 months before consulting a neurosurgeon. The exacerbation of symptoms led to limited mobility of the patient as well as to decrease in the quality of life, and difficulties in self-care. The conservative therapy included nonsteroidal anti-inflammatory drugs (NSAIDs), muscle relaxants and the course of vitamin B complex injections for 3 weeks. During the treatment, the symptoms persisted, and the patient underwent magnetic resonance imaging (MRI) of the lumbar spine with contrast enhancement, which revealed an intradural space-occupying lesion in the projection of the intervertebral L_3–L_4 disc. In the T1+C TSE Fat Sat mode, the lesion had an insointensive signal with a peripheral hyperintensive rim (Fig. 1). to clarify*

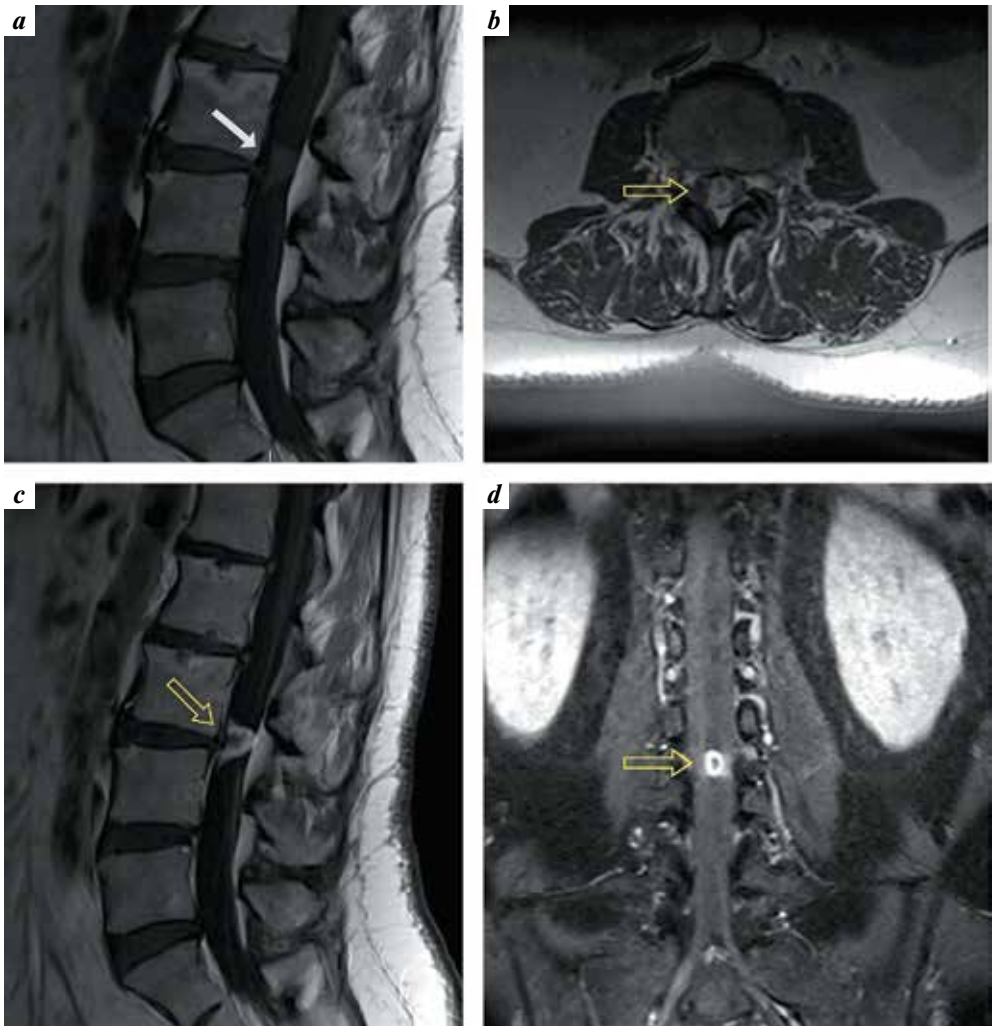

Fig. 1. *Lumbar magnetic resonance imaging of patient: a – T1 weighted image: an isointensive intradural formation is visualized (white arrow); b–d – T1+C TSE Fat Sat mode: an intradural isointensive volumetric formation with a ring-shaped accumulation of contrast along the periphery, the phenomenon of "ring enhancement" (yellow arrows) is visualized*

the diagnosis, the patient underwent computed tomography (CT) of the lumbar spine, in which the lesion looked isodense; no signs of petrification were found (Fig. 2).

The neurological status showed the depression of tendon reflexes in the left leg, hypoesthesia along the S_1 dermatome, and hyperesthesia along the L_5 dermatome on the left. When discussing the clinical features and neuroimaging data at a council of leading neurosurgeons, specialists suggested a tumor genesis of the disease (meningioma). They also did not exclude the presence of hernial sequestration. In order to relieve lumbar ischialgia syndrome resistant to drug therapy and prevent the development of irreversible neurological deficit, the patient underwent surgery.

Using a microscope and a high-speed drill, the spinal canal was opened interlaminar by means o interhemilaminectomy and medial facetectomy at the L_3–L_4 level on the left, and a tense, thinned dural sac was visualized, through the walls of which transit roots were traced, and a transmitted pulsation was determined. At the level of the L_3–L_4 disc, there was an epidural cuff-shaped constriction of the dural sac by a cloudy-white color dense structure. Medial displacement of the dural sac was impossible due to its ventral fixation to the posterior longitudinal ligament. The dura mater (DMA) was opened linearly within of 3 cm. Clear, transparent cerebrospinal fluid was obtained. A circular thickening of the arachnoid membrane of a cloudy-white color was found, embracing the roots and forming the adhesions to them with the formation of an encapsulated cavity. In the center of this cavity, a dense elastic white formation on a wide base was determined ventrally, fixed to the inner layer of the dura mater, penetrating between the roots and forming the adhesions to them (Fig. 3, a, b). The radiculolysis and mobilization of dural sac were performed. The ventral surface of the dura mater was divided into layers with total removal of the formation and the walls of the arachnoid cavity (Fig. 3, c).

As a result, dura mater defect sized 7 × 4 mm was formed on the anterolateral surface of the dural sac, which could not be sutured due to degeneration and weakness of the dura

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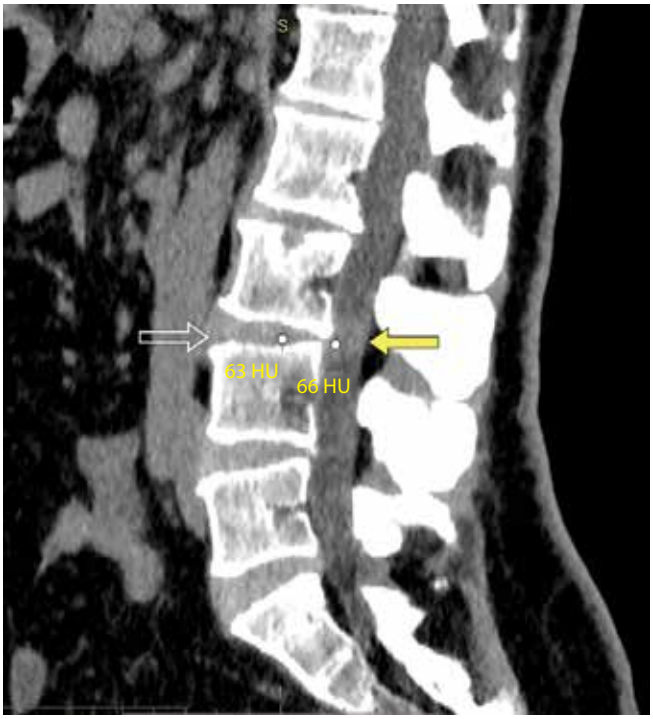

Fig. 2. *Lumbar spine CT-scan. In the projection of the intervertebral disc L_3–L_4 (white arrow), an isodense volumetric formation (66 Hounsfield units) is visualized in the spinal canal, similar in density to the tissues of the intervertebral disc (66 Hounsfield units) (yellow arrow)*

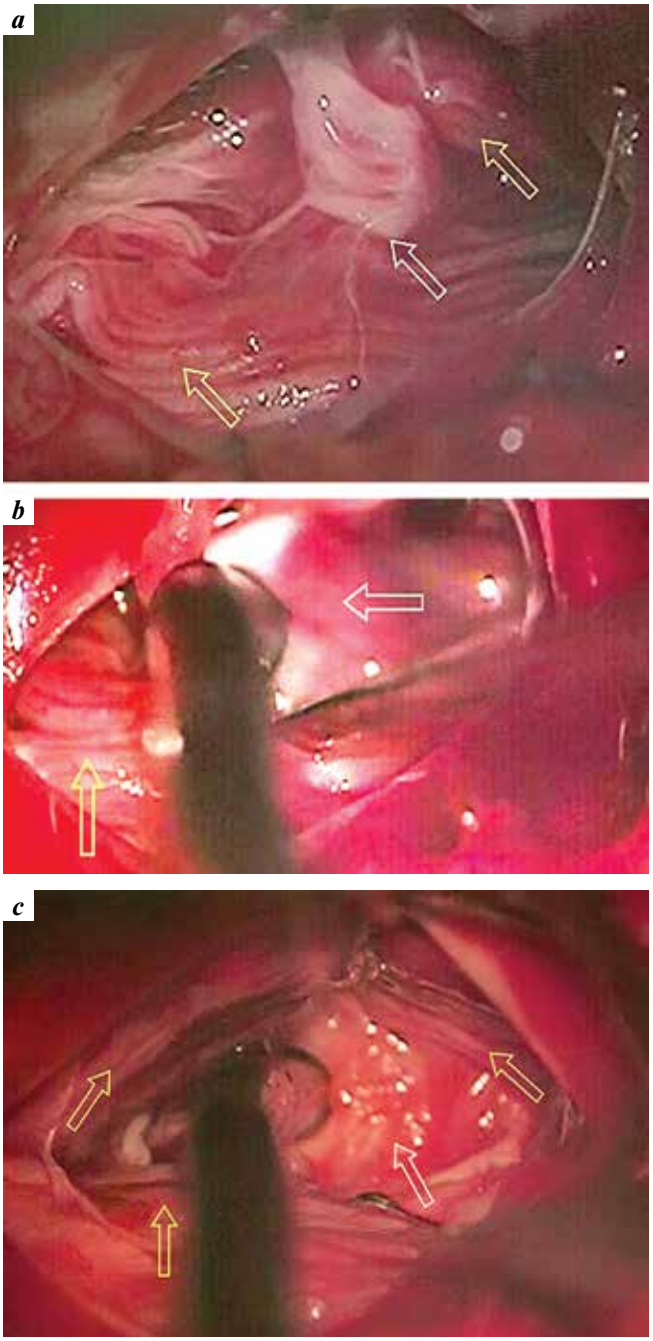

Fig. 3. *Intraoperative images: a, b – an intradural volumetric lesion adherent to the passing nerve roots (white arrows); passing nerve roots (yellow arrows); c – the cavity after removal of the intradural volumetric lesion (white arrow); passing nerve roots (yellow arrows)*

mater. The incision on the dorsal surface of the dura mater was sutured continuously with atraumatic 5/0 monofilament thread. The defect on the anterolateral surface of the dural sac was epidurally patched with a hemostatic sponge and fibrin-thrombin glue. A lumbar drainage was placed to sanitize the cerebrospinal fluid and prevent wound liquorrhea. The samples of fragments of the thickened arachnoid and the masses were sent for histological examination.

On the first postoperative day, Sol. Tramadoli 50 mg/ml – 2 ml were administered twice to relieve pain in the area of the postoperative wound. The administration of NSAIDs (no more than 2 times a day) was performed up to the fourth postoperative day. The patient was verticalized in a semi-rigid lumbar orthosis on the third postoperative day. As the cerebrospinal fluid cleared, the lumbar drainage was removed on the third day. The wound healed by primary intention.

By the time of discharge, complete relief of previously existing symptoms was noted, pain syndrome regressed (0 points by VAS), sensitivity in the left leg was completely restored.

The results of histological examination revealed the fragments of mature connective tissue in the sample, represented by loose fibrous stroma with pronounced extensive zones of fibrosis, inclusions of cartilaginous and bone tissue, vessels, and diffuse, scanty lymphocytic infiltration. No atypical cells were found (Fig. 4).

During the immunohistochemical study using monoclonal antibodies, the expression of CD34 on the membranes of the vascular endothelium, cytoplasmic staining of single reticular cells of the stroma with the CONFIRM anti-S10 antibody were determined. Stromal fibroblasts with the expression of Beta-Catenin in the cytoplasm of cells with atypical morphology and the expression of NSE, GFAP, Calretinin, Anti-EMA were not detected.

Thus, the final diagnosis was established as intervertebral L_3–L_4 disc herniation with intradural sequestration.

The patient was discharged from the hospital in satisfactory condition on the 9th day after the operation.

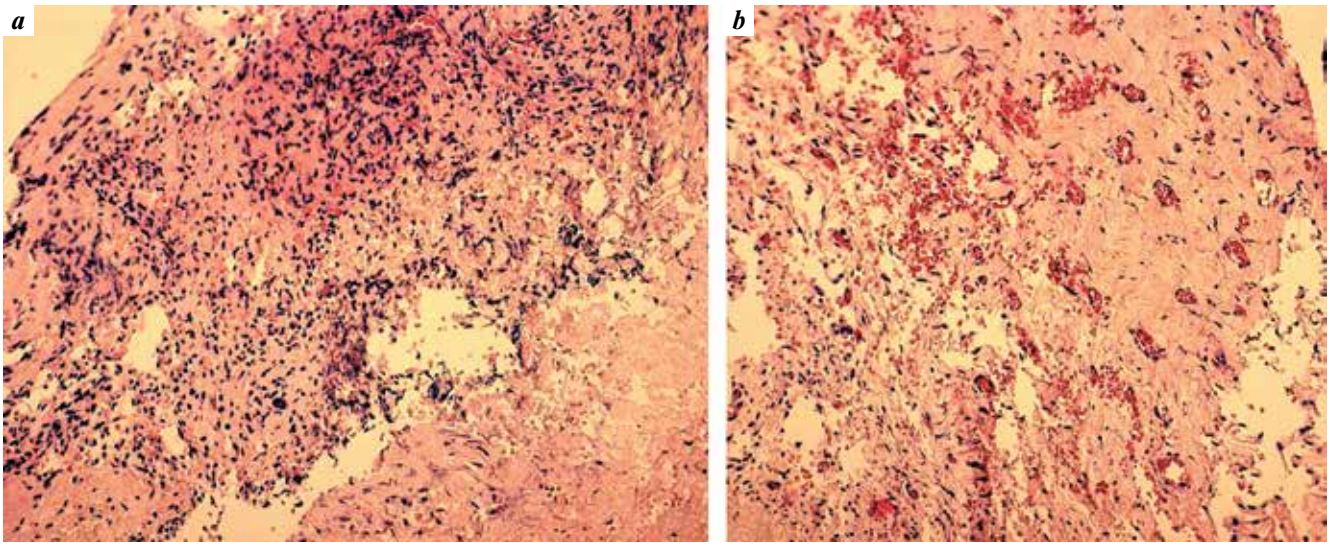

Fig. 4. *Histological examination: a – fragments of mature connective tissue represented by a loose fibrous stroma with fibrosis zones, inclusions of cartilage and bone tissue, diffuse lymphocytic infiltration; b – neovascularization in the structure of connective tissue*

DISCUSSION

Intradural sequestration is an extremely rare manifestation of spineal degenerative-dystrophic changes, occurring in 0.26–0.30 % of cases of intervertebral disc herniation [1]. In 92 % of cases, intradural intervertebral disc herniation occurs in the lumbar spine, while in the thoracic and cervical spine the incidence is 5 and 3 %, respectively [4]. The most frequently affected level is L_3–L_4 – in 55 % of cases, L_3–L_4 – in 16 % of cases, and L_5–S_1 – in 10 % of cases. At the same time, the levels L_2–L_3 and L_1–L_2 are extremely rarely involved [5].

The first clinical case of intradural sequestration at the lumbar level was described by W.E. Dandy in 1942 [6]. At present, the pathogenesis of intradural sequestration of intervertebral disc herniations has not been fully studied. The ventral epidural space usually acts as a buffer, reducing the degree of traumatic impact on the dura mater that occurs during the process of sequestration of the intervertebral disc herniation, thereby preventing the entering of the hernial fragment into the subdural space [7].

It is believed that an important pathogenetic factor is the pronounced adhesion of the ventral surface of the dura mater to the surface of the fibrous ring and the posterior longitudinal ligament, which leads to the collapse of the ventral epidural space. These changes may be the result of chronic aseptic inflammation in response to the formation of a herniated disc, previous trauma, or previous surgery at the involved level.

Thus, with an increase in intradiscal pressure as a result of various provoking factors, a breakthrough of the hernial sequestrum occurs through the fibrous ring, the posterior longitudinal ligament and the dura mater, which is tightly fused with them [8–11]. Also, a significant background may be the thinning of the dura mater in case of congenital or progressive acquired stenosis of the spinal canal [7]. As a result of intradural sequestration, the hernia can be located both in intradural and intraradicular spaces [12].

The most common clinical manifestations are long-term pain in the lumbar spine and mono-/polyradicular symptoms in the form of pain, numbness, muscle weakness in the lower extremities. In a large number of cases (29 %) against the chronic pain syndrome, the patient»s condition sharply worsens with the development of cauda equina syndrome, whereas intracanal space-occupying lesions are typically presented with a torpid development of symptoms [13].

A rare manifestation of the disease is the syndrome of intracranial hypotension due to intracanal liquorrhea, which is more often recorded when the hernia is localized in the thoracic spine [2]. An accurate diagnosis is extremely rarely established in the preoperative period; in 75 % of cases, the correct diagnosis is established intraoperatively or based on the results of a postoperative histological examination [2].

In addition to the clinical features, various neuroimaging methods are important for diagnosis. It is often difficult to differentiate intradural sequestration from intradural extramedullary neoplasms such as schwannoma, neurofibroma, meningioma, epidermoid cyst, and secondary space-occupying lesions. Lumbar spine CT can be useful for differential diagnosis. The presence of an intradural extramedullary space-occupying lesion with a density similar to that of intervertebral disc tissues, without the tumor-like accumulation of contrast suggests the presence of hernial sequestration [13].

There are also reports that the presence of gas in the spinal canal in the area of interest can be an important diagnostic sign in CT [14]. When performing MRI, a characteristic sign is the “hawk-beak sign”, which is the presence of an intracanal substrate in the projection of the

intervertebral disc that has a triangular shape with the apex directed toward the ventral surface of the dura mater and has a mass effect that is most clearly visualized on T2-weighted images [5]. Also, in the T2 mode, one can observe the "halo sign" in the form of an isointensive strip of cerebrospinal fluid, surrounding the surface of the hernial protrusion in a ring-shaped manner, and the "Y-sign" – in the form of a Y-shaped separation of the arachnoid and dura mater on sagittal MRI sections [15, 16].

Lumbar spine magnetic resonance imaging with contrast enhancement provides more additional information [7]. Intradural hernial sequestration is characterized by the presence of peripheral contrast enhancement on T1-weighted images (ring enhancement), which distinguishes it from other formations included in the differential diagnostic algorithm [2, 13, 17].

Not all of the described radiological features may be present in a particular patient. In the presented case, the key neuroimaging parameter that allowed establishing the correct diagnosis was the characteristic pattern of contrast agent accumulation by the mass during MRI (see Fig. 2).

Surgery is the primary treatment option for patients with this pathology. Careful preoperative diagnostics and surgical planning are the keys to a favorable outcome. If intradural sequestration of a herniated disc is suspected, a thorough revision of both the epidural and subdural spaces is mandatory for identifying the completely removal of all fragments of the hernial sequestration to avoid the revision surgery. Ultrasonography is a useful method of intraoperative diagnostics, allowing the operating surgeon to visualize the intradural component of the hernia before performing a durotomy [18].

An important aspect of the operation is the careful closure of the dura mater defects. The literature describes the direct suturing of the dura mater as well as the use of autofat, automuscles, and the use of adhesive compositions, artificial dura mater grafts for additional sealing [7]. In our case, in order to prevent wound liquorrhea, the lumbar drainage system was placed.

CONCLUSION

In conclusion, intervertebral disc herniation with intradural sequestration is a rare and difficult pathology to diagnose. The basis for successful treatment of this group of patients is careful preoperative diagnostics, but the correct diagnosis cannot always be made in the preoperative period. The differential diagnostics between intervertebral disc herniation and various neoplasms is important. Features of the clinical course in combination with characteristic signs according to MRI and CT of the lumbar spine in native mode and with the use of contrast enhancement allow us to assume the correct diagnosis and plan all stages of surgical treatment, which ultimately has a positive effect on the clinical outcome.

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Author's contributions
M.A. Nekrasov: development of the concept and design of the study, editing of the article, performing the surgical operation;
D.S. Glukhov: development of the concept and design of the study, obtaining data for analysis, writing and editing of the article;
V.V. Babenkov, E.F. Abdulrahman, V.M. Nekrasov: obtaining data for analysis, editing of the article.

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