

## IATROGENIC LUMBAR INTRASPINAL EPIDERMOID TUMOR IN ADULTS: 2 CASE REPORTS AND LITERATURE REVIEW

### ERİŞKİN İATROJENİK LOMBER İNTRASPİNAL EPİDERMOİD TÜMÖR: 2 OLGU SUNUMU VE LİTERATÜR GÖZDEN GEÇİRME

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#### SUMMARY:

Iatrogenic lumbar tumors are usually occur in childhood, through implantation of epidermal cells to the intraspinal space during repetitive lumbar puncture. Cells iatrogenically implanted into the spinal canal during lumbar puncture can slowly grow until symptomatic. Diagnosis can be difficult and is often delayed. MRI appears to offer some advantages in diagnosis provided that gadolinium is used. They are rarely seen in adults. We present successful surgical treatment of two lumbar epidermoid tumor cases one 64, and the other 34 year old whom had repetitive LP application in their medical history.

**Key words:** Iatrogenic, epidermoid tumor, lumbar puncture,

**Level of evidence:** Case report, Level IV

#### ÖZET:

İyatrojenik lomber epidermoid tümörler, genellikle çocukluk çağında, tekrarlayıcı lomber ponksiyonlar sırasında epidermoid hücrelerin intraspinal alana implantasyonu ile oluşur ve yetişkinlerde nadiren görülürler. Lomber ponksiyon sırasında spinal kanala iyatrojenik olarak implante olan hücreler semptom verene kadar yavaşça çoğalırlar. Teşhisi oldukça zordur ve sıklıkla gecikir. Kontrastlı manyetik rezonans görüntüleme, tanı maksadıyla kullanılan en önemli görüntüleme yöntemidir. Bu makalede, medikal hikâyelerinde tekrarlayan lomber ponksiyonlar olan ve cerrahi olarak başarı ile tedavi edilmiş, biri 64 diğeri 34 yaşlarında iki vaka sunulmuştur.

**Anahtar kelimeler:** İyatrojenik, epidermoid tümör, lomber ponksiyon

**Kanıt düzeyi:** Olgu sunumu, düzey IV

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## INTRODUCTION:

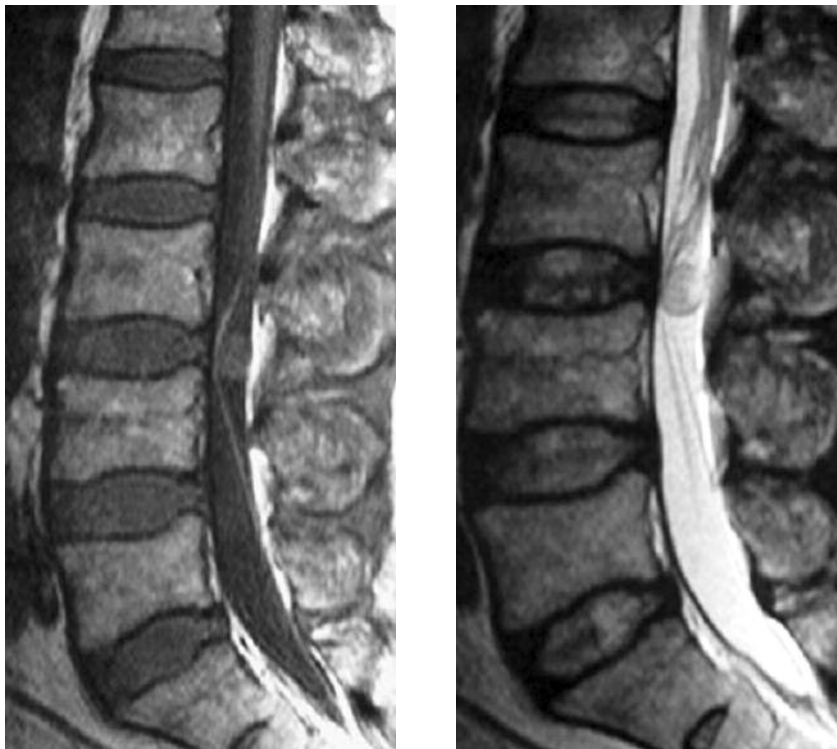
Epidermoid tumors arise from epidermal cells implanted intradurally. This implantation may be either congenital or iatrogenic. The congenital form is associated with other spinal abnormalities. Lumbar puncture is a well-recognized iatrogenic cause of implantation of epidermal cells inside the dural sac.<sup>(1,6,11,16)</sup> Intraspidal epidermoid tumors occurring several years after lumbar puncture are rare. We describe two cases of iatrogenic lumbar epidermoid tumor in 64-year-old male and 34-year old female patients, one of which presented with bilateral leg pain without neurologic findings and one of which presented with low back pain.

## CASE-1:

A 64 year old male patient complaining for back pain and pain in his both legs (more in his

right leg) was admitted to our clinic. He told that he had difficulties in walking, could not walk a long way that he had pain particularly in his right hip and the pain reflected from the hip towards the posterior region of the thigh. The physical examination was normal, while bilateral leg extension strength tests were positive and no sphincter failure was found. No abnormal findings were found in the lumbar Xray.

Conservative treatment was performed to the patient. Since his complaints did not subside in ten days, a magnetic resonance imaging was performed and an intradural mass was found at the level of third and fourth lumbar vertebra localization. The mass was found as hypo/isointense in T1 weighted imaging and hyperintense in T2 weighted imaging and was operated with pre-diagnosis of schwannoma (Figure-1). The mass was totally removed through L3 total laminectomy. It was found during



**Figure-1.** T1-weighted sagittal image demonstrating a 1 cm sized hypo-signal intensity intradural extramedullary mass at the level of third and fourth lumbar vertebra (a), T2-weighted sagittal image demonstrating a high signal intensity mass (b). (Case-1)

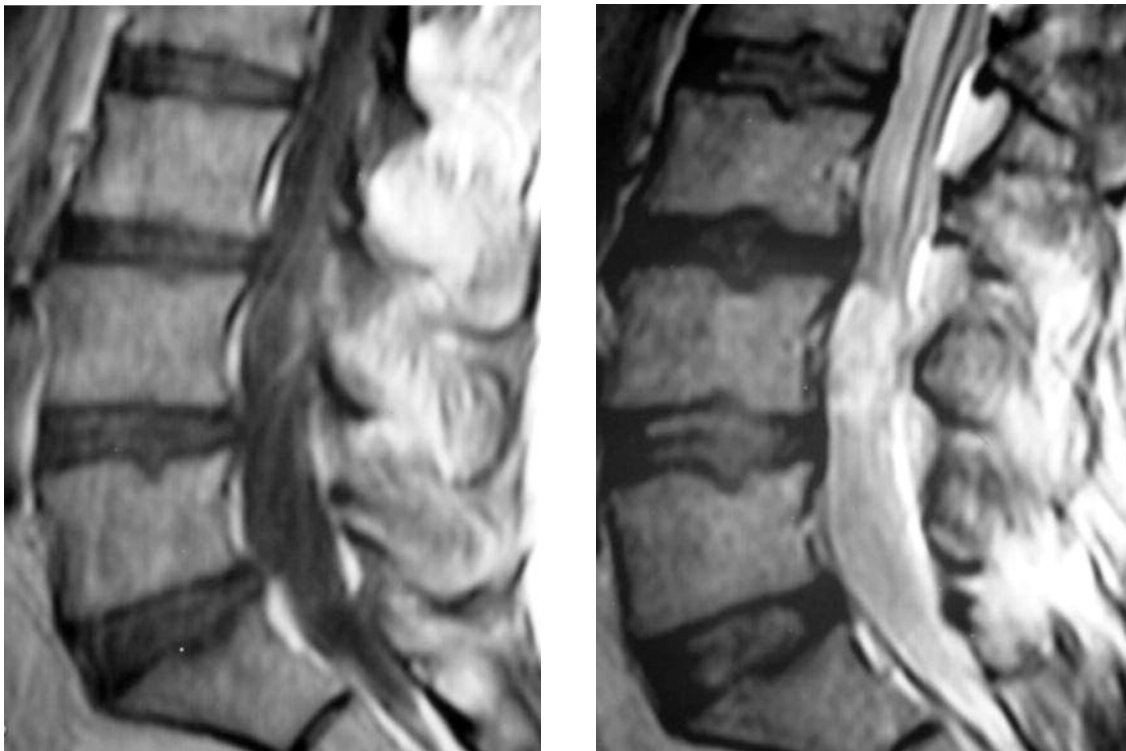
the operation that the mass was localized among the cauda fibers, however did not invade any nerve fiber. The pathological examination of the mass was reported as an epidermoid tumor. It was found following the detailed medical history that the patient was hospitalized and treated for a purulent meningitis 2 years ago and he had had repetitive lumbar punctures during treatment.

#### **CASE-2:**

A 34-year-old female patient complaining for back pain and pain in his both legs (more in his left leg) was referred. Her clinical history revealed that she had had her complaints since 3 years, and intensified during the last 2-3 months, that she did not have any benefit from the medical treatment, and her medical history also revealed that she was referred to the neurology clinic for headache 5 years ago, was

treated for pseudomotor cerebri pre-diagnosis, was hospitalized in a neurology clinic and a draining lumbar puncture was applied every other day. The patient also told that she had draining lumbar puncture once in a week during 5 months after being released from the hospital and back pain starting after the procedure intensified from time in time and sometimes reduced.

In her neurological examination, plain leg extension test was normal in both legs, lower extremity power was full, deep tendon reflexes were normoactive, TCR gave bilaterally plantar response, and sensorial examination of the extremities was normal bilaterally. There was no muscular atrophy, neural claudication and incontinence. Her lumbar MRI revealed a nodular lesion with a cystic component localized in L4 corpus level, 12 mm sized and showing slight contrast. (Figure-2) The patient



**Figure-2.** T1-weighted sagittal image demonstrating a hypo-signal intensity intradural extramedullary mass at the level of fourth lumbar vertebra (a), T2-weighted sagittal image demonstrating a high signal intensity mass (b). (Case-2)

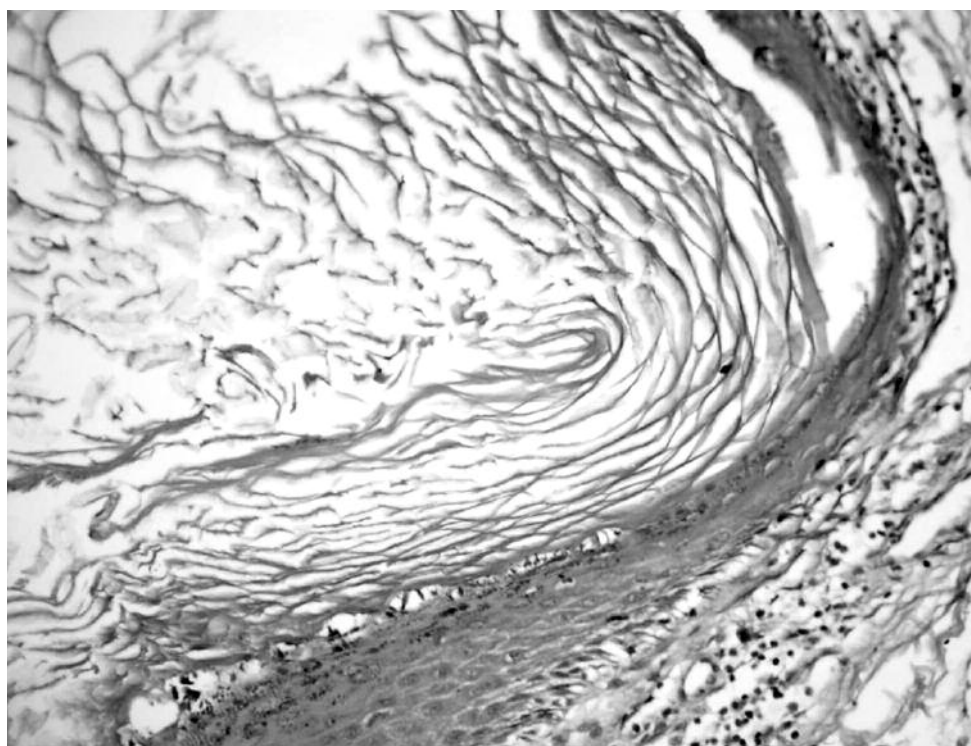
was operated and the intradurally localized mass was totally removed through L4 total laminectomy. The patient did not develop any additional deficit during the postoperative period and was released with full recovery.

#### DISCUSSION:

Epidermoid tumors are benign lesions originating from ectodermal cells and consist less than 1 % of intraspinal tumors.<sup>(6,10,13)</sup> They can be congenital or acquired. The congenital type occurs within the 2-7th weeks of the embryonic life, during the closure of the neural tube, by abnormal implantation of ectodermal cells to the spinal canal and is more frequent than the acquired type. Congenital epidermoid tumors are usually associated with other neural tube defects, such as dermal sinus.<sup>(3,8)</sup>

Iatrogenic epidermoid tumors must be differentiated from congenital epidermoid tumors, which occur from inclusion of epithelial elements during closure of the neural groove; and dermoid tumors, which are congenital and also result from a similar inclusion of tissue during embryonic development.<sup>(9)</sup>

Acquired lumbar epidermoid tumors are iatrogenic lesions frequently showing extramedullary localization, rarely intramedullary.<sup>(5)</sup> They occur by transferring epidermal cells to the intraspinal canal through invasive procedures such as lumbar punctures, spinal anesthesia, myelography and pneumoencephalography.<sup>(2,11)</sup> Iatrogenic lumbar epidermoid tumor was firstly described by Choremis et al. in 1956.<sup>(15)</sup> This theory was proved by finding an epidermoid tumor in the lumbar intraspinal region of patients, who



**Figure-3.** Pathologic examination: Light microscopic photograph showing desquamated cells containing keratohyalin encased within a laminated capsule of well-differentiated stratified squamous epithelium.



previously had normal myelography. It was hypothesized and has been experimentally demonstrated that during skin puncture with a hollow needle a core of epidermis can be transported within the needle bore. (4,7,12)

Iatrogenic lumbar tumors are more frequently seen during the childhood. The reason for that is the fact that diagnostic lumbar puncture is frequently used during the neonatal period and puncture needle with no introducers are preferred in this period. However, iatrogenic lumbar epidermoid tumors are less frequently seen in our times with using disposable, small needles, with sharp blades and introducer. In contrary to this theory, however, spinal needs used in lumbar punctures in our cases presented were with introducers. The implanted ectodermal cells continue to grow slowly, until neural pressure starts and the symptoms occur. This growing may take sometimes long years, and sometimes only few months. There are several times in the literature, ranging between 18 months and 40 years. Symptoms averagely emerge 2-6 years after the lumbar puncture (11). Since the pressure has a slow course, the symptoms are noticed late, and thus chronic pressure findings, such as abnormal walking, disability in the legs and muscular atrophy occur, and usually positive leg extension test, leg and hip pain and sensorial loss accompany this picture. There is a negative correlation between the emerging time of the symptoms and the neurological findings. Extension of the time increases the occurrence of the neurological findings. Both in our two cases, the time between the lumbar puncture and occurrence of the symptoms was less than 24 months and the complaint of the patients was pain spreading to both legs.

Direct X-ray examination has no place in the diagnosis of intradural epidermoid tumors. However, they may have a guiding role for

diagnosing cases developing scoliosis resulting from the pain in childhood tumors. Lumbar myelography was the most frequently used diagnosis method before MRI. In our time, however, MRI should be imaging method of choice in epidermoid tumors, as it is in other intraspinal pathologies. (5) Epidermoid tumors in MRI are typically seen as hypo/isointense in T1 weighted examination and hyperintense in T2 weighted imaging, and usually without contrast. A slight homogen contrasting was found in our first case, while very slight circumferential contrasting was present in our second case.

Surgical removal is the absolute treatment of the epidermoid tumors. The tumor tissue is within the neural tissue, is usually localized among the caudal fibers in the lumbar region, and can be completely distinguished from the intact nerve fibers if dissected carefully. This is the most important difference distinguishing it from schwannoma. The point that should be considered in surgery is that the probability of damage in the dura caused by the repetitive LP is high and that dura closure should be performed very carefully after tumor removal. In case that dura can not be closed in a waterproof way, dura grafts and tissue sealants should be used, otherwise CSF fistula is inevitable. In our second case, the patient had to be revised twice because of CSF fistula, and lastly subarachnoidal and operation lodges were drained separately and waited for the wound healing, and the patient could be treated only by this approach and was released without a problem.

The absence of congenital defects of the spinal column in our patient, and a history of difficult LP supports iatrogenic implantation as a possible cause. The diagnosis of the disease can be possible through taking a good clinical history. The use of lumbar needles without stylettes seems to elevate the incidence of epidermoid tumor after LP. As a matter of

cause, stylettes should be used in LP needles. The present cases emphasizes the fact that epidermoid tumor can occur in adults after LP. Although its incidence may be low, we should be aware of potential adverse complications such as epidermoid tumors, after LP.

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