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Case report / Приказ болесника

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Ligamentum flavum hypertrophy in a patient with Pott's disease

Хипертрофија жутог лигамента код пацијента са Потовом болешћу

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SUMMARY

Introduction The spine is involved in less than 1% of all tuberculosis (TB) cases, and it is a very dangerous type of skeletal TB as it can be associated with neurologic deficit and even paraplegia due to compression of adjacent neural structures and significant spinal deformity. The spine TB is one of the most common causes for an angular kyphotic deformity of spine. Patients with 60 or more degree kyphosis at dorsolumbar spine are at great risk to develop late onset neurological deficit and paraplegia due to chronic compression and stretching of the spinal cord over bonny ridges. In small portion of cases other conditions may lead to neurological deficit in patients with long standing angular kyphosis which also alters the treatment strategy that otherwise involves prolonged and mutilant surgery.

Case outline We present a case of a 61-year-old male patient with concomitant 90-degree dorsolumbar spine kyphosis due to spinal TB and ligamentum flavum hypertrophy, which led to spinal canal stenosis with myelopathy and consequent paraplegia. The patient undergoes dorsal decompression with removal of the hypertrophic yellow ligament after which he recovered to the level of walking.

Conclusion Many authors propose guidelines for treatment of spinal TB taking into account the stage of the disease, the age of the patient, the angle of kyphosis, and other factors. We find that the best approach for each patient is personalized medical approach.

Keywords: kyphotic deformity, late onset paraplegia, TB spine, spinal canals stenosis, flavum hypertrophy
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САЖЕТАК

Увод Кичма је укључна у свега 1% случајева туберкулозе (ТБ) и овај скелетни облик ТБ може бити удружен са неуролошким дефицитом или чак са параплегијом услед компресије на нервне структуре и значаног деформитета кичме. Спинална ТБ је један од најчешћих узрока угаоног кифотичног деформитета кичменог стуба. Пацијенти са углом кифозе од преко 60 степени у грудно-слабинској регији су у великом ризику да у каснијем току болести развију неуролошки дефицит и параплегију услед дуготрајне компресије и истезања кичмене мождине преко коштаних структура. У малом броју случајева постоје други узроци неуролошког дефицита код пацијената са дуготрајном угаоном кифозом, што мења стратегију лечења, која иначе подразумева дуготрајне и мутилантне операције.

Приказ случаја Приказујемо случај мушкарца старосне доби 61 годину са израженим кифотичним деформитетом са углом од 90 степени у тораколумбалној регији насталог услед спиналне ТБ прележане у раном детињству и хипертрофијом жутог лигамента који је довео до стенозе спиналног канала удружене са мијелопатијом и последичном параплегијом. Пацијенту је урађена дорзална декомпресија у смислу уклањања хипетрофисног жутог лигемента након чега се пацијент опоравио до нивоа самосталног хода.

Закључак Многи аутори предлажу водиче за лечење спиналне ТБ узимајући у обзир фазу болести, старосну доб пацијента, угао кифозе, као и друге факторе. Ми саматрамо да је најбољи приступ сваком пацијенту персонализовани медицински приступ.

Кључне речи: кифотични деформитет, параплегија касног почетка, туберкулоза кичме, стеноза спиналног канала, хипертрофија жутог лигамента

INTRODUCTION

Tuberculosis (TB) is one of the oldest diseases affecting humans and has been found in the ancient mummies of Peru and Egypt. The disease is caused by the bacillus Mycobacterium tuberculosis, and occasionally by *Mycobacterium africanum* or *Mycobacterium bovis*. The spinal column is involved in less than 1% of all TB cases. First case was described in 1779. by Percival Pott, and since then the disease is frequently called Pott's disease. Main reasons for late onset paraplegia occurrence in patients who had Pott's disease are long standing angular kyphosis and chronic compression and stretching of the spinal cord over bonny

ridges [1]. However in small portion of cases, other conditions may simultaneously be present besides spinal deformity and result in spinal canal stenosis and neurological deficit.

We present a patient with concomitant hyperkyphosis due to spinal TB and ligamentum flavum hypertrophy.

Written consent was obtained from the patient to publish all shown material. This study was done in accordance of the institutional standards on Ethics.

CASE REPORT

We present a male, Caucasian patient, 61 years of age, presented to our clinic with long-term history of chronic back and leg pain. As a 3 year old child he was treated for spine TB in an orthopedic clinic in another country, but no medical documentation is available. It was decided not to operate at that time, and patient was recommended to use brace. Patient states that he was inconsistent in going to checkups. As a consequence of irregular treatment pronounced gibbus in thoracolumbar spinal region developed. However, he had no symptoms or difficulties of any kind. He was physically active, played football since 8 years old, finished high school and was employed. At the age of 57, pain in his back appeared. Pain was worst at night and with time his foot started stucking. He was admitted to Spinal orthopedic center in Belgrade, where first spine X-ray was performed and gibbus deformity with thoracolumbar kyphosis resulting in 90-degree angulation was observed (Figure 1). After that spine MRI was performed which showed kyphotic deformity, but with normal signs of myelon, according to radiologist's description (patient lost this MRI images), so patient was treated conservatively. He was pain free for 3 years, but in 2009 pain worsened, with right leg predominance, however, he did not receive any treatment. In 2013 patient started to have walking problems and within a few months he developed paraplegia. Spine MRI was performed and clearly demonstrated myelopathy finding at the level of Th11-Th12 with

hypertrophied yellow ligament at the same level (Figure 2). He was offered surgical treatment, but as procedure was supposed to include anterior decompression and spinal fusion, patient refused treatment at that time. After several consultations with spinal orthopedic surgeons and neurosurgeons from other institutions, patient was operated by posterior decompression via partial laminectomy and excision of hypertrophied ligamentum flavum at the level of Th11-Th12 consistent with the level of myelopathy, without other procedures. Following surgery patient gained ability of cane aided gait. Early control spine MRI showed signs of good spinal canal decompression at the level of Th11-Th12, with regression of myelopathy signs. Few months after operation he started to have lumbar pain and leg numbness with pain more pronounced in right leg, which is why he was further, evaluated in our department. Patient reported pain on visual analog scale 5/10, objectively he had bilaterally negative Lazarevic's sign (Lasègue test), normal muscle tonus, with symmetrically reduced myotonic reflexes, and normal plantar flexion response, normal muscle strength, no objective loss of sensation and antalgic cane-aided gait. Kyphosis in upright posture was evident. Control CT scan was performed and showed good decompression, without signs of spine instability or progression of kyphosis (Figure 3). Since no further neurological deficit has been developed and based on neuroradiology findings, it was concluded that no further surgical treatment was necessary, caudal epidural blockage was performed in local infiltrative anesthesia and patient was discharged in good condition with significantly reduced pain and better walking (Figure 4).

DISCUSSION

There are two basic types of spinal TB. First is the classic form or spondylodiscitis. Second one is atypical form, which is spondylitis without involvement of intervertebral disc. The basic lesion in Pott's disease is a combination of arthritis and osteomyelitis, usually

affecting more than one vertebra, and most commonly involving anterior aspect of the vertebral body. Spinal TB can include progressive bone destruction leading to kyphosis and vertebral collapse, formation of cold abscess, spinal canal narrowing by granulation tissue, abscesses, or direct dural invasion resulting in spinal cord compression and neurologic deficits [2].

Pott's disease is one of the most common cause for a angular kyphotic deformity of spine, particularly in developing countries. There is an average increase in spine kyphosis of 15 degrees in all patients treated conservatively, and a deformity greater than 60 degrees can develop in about 3% of patients [3].Children are more prone to develop greater deformity, probably due to the cartilaginous nature of their bones. Development of kyphosis occurs in two stages of disease: (phase I) during active disease and infection, (phase II) after healing of the lesion. Deformity developed in the phase II with neurologic deficit has worse prognosis than complications that occur during the phase I [4]. In our case kyphosis developed in the early stage of the disease, but paraplegia occurred at a late stage of the disease, due to hypertrophy of the yellow ligament causing stenosis of spinal canal and the consequent compression of the spinal cord.

The progression of kyphosis depends on number of vertebral involvement at the phase I of the disease, initial vertebral body loss, and segment of spine affected. There is some evidence that if patients developed 60° or more kyphosis at dorsolumbar spine they were likely to develop late onset paraplegia [5]. There are several risk factor which may indicate severe progression and patient that are at great risk of deformity progression, such as: age below ten years and loss of one or one and a half vertebral bodies (I), a pre-treatment kyphosis angle of greater than 30 degrees, especially in children (II), thoracolumbar junction lesions (III), radiological signs of "spine at risk" (IV) [6]. These signs of "spine at risk" are: separation of the facet joints (a), retropulsion of vertebras (b), lateral translation (c), and

toppling (d). This signs are manifestation of spinal instability due to dislocation of the facet joints. Each of these signs is given a score of one. Total score of three or four can predict an increase in the kyphosis by more than 30 degree and a final deformity of more than 60 degree [7]. Since our patient was inconsistent with medical checkups, disease treatment was inadequate. According to mentioned risk factors, our patient was at great risk for development of severe deformity, which happened and final manifestation was paraplegia. Analyzing radiological findings of our patient, it is most likely that he had all 4 signs of "spine at risk", finally resulting in 90 degree kyphosis. In addition, patients with pronounced kyphosis are prone to develop severe spinal canal stenosis, probably due to compensatory ligament and bone hypertrophy in the spine that is instable. Persistent deformity affects the biomechanics of all spine segments [8]. This is what happened to our patient, and which in the end turned out to be the biggest problem in our patient, since he developed myelopathy due to spinal canal stenosis.

The indications for surgery in Pott's disease are patients with neurologic deficit. Although management of patients with active disease is well defined, there is a lack of literature on the management of spinal kyphotic deformity caused by TB [9]. Combined posterior and anterior osteotomy, correction of deformity, and instrumented fusion are shown to arrest progression of kyphosis and improve neurologic symptoms. Different techniques have been used to correct the kyphotic deformity. A single-stage posterior Smith-Peterson osteotomy, pedicle subtraction osteotomy, vertebral column resection through a single-stage anterior–posterior approach (anterior decompression followed by posterior instrumentation), direct internal kyphectomy and other new approaches are mostly used techniques, but this procedures are associated with significant blood loss, major complications and high morbidity [9, 10]. In the cases of patients with severe and long standing kyphosis, who were treated for spinal TB 15 or more years ago, and with new presentation of paraplegia or upper

motor neuron spinal cord injury, anterior decompression and fusion is advocated [10, 11, 12]. Our patient was a candidate for this procedure, but after being presented with all possible complications, length of surgery and hospital stay, he refused treatment. He was than admitted to our neurosurgical department, and after additional analysis of the spine MRI, and after consultations with spinal orthopedic surgeons and neurosurgeons from other institutions, patient was offered posterior decompression with only partial laminectomy and with removal of hypertrophied yellow ligament, without kyphus correction or fusion. Our presumption that the problem was mainly due to hypertrophied yellow ligament was set on the basis that the kyphotic deformity did not change over several years, and that myelopathy sign was present at the same level where yellow ligament was thickened. Also, since our patient did not have restriction of pulmonary functions we decided that best approach was partial laminectomy with flavectomy, which proved to be a successful approach, since patient achieved neurological status improvement and good recovery.

Hyperkyphotic deformity remains the main reason for late onset paraplegia after spinal TB. However, other causes such as ligamentum flavum hypertrophy can simultaneously occur and contribute to progressive spinal canal stenosis and compression of the cord, causing a neurological deficit. Although, many authors propose guidelines for treatment of spinal TB taking account phase of the disease, patient's age, kyphosis angle and other factors, we advocate personalized approach to every patient. In addition, many complications, such as kyphosis can be avoided by early diagnosis of spine TB and by proper treatment. So, patient with cured TB must be controlled with regular checkups in order to prevent late complications.

Conflict of interest: None declared.

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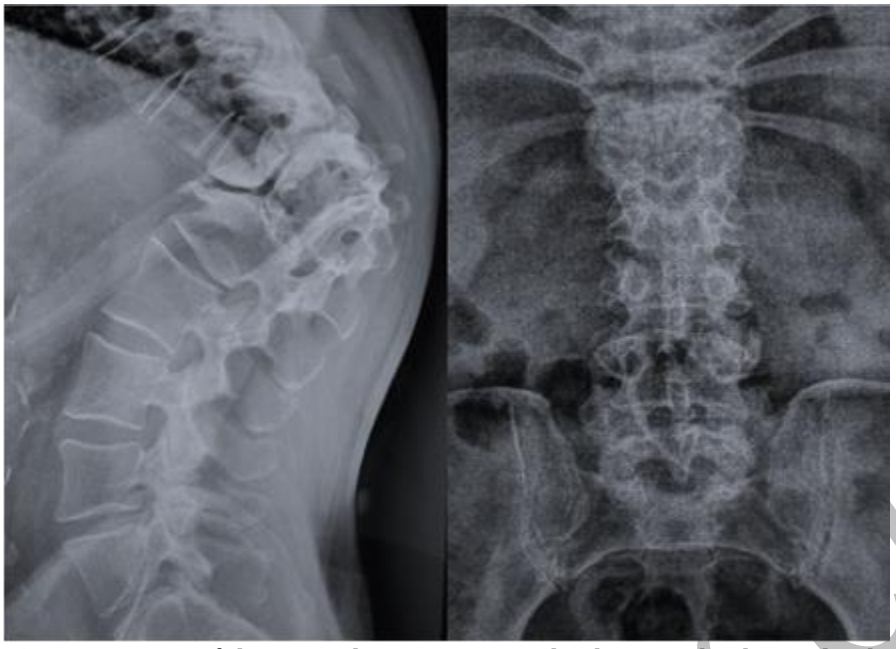

Figure 1. X-ray of the spine showing a severe kyphosis at the thoracolumbar junction

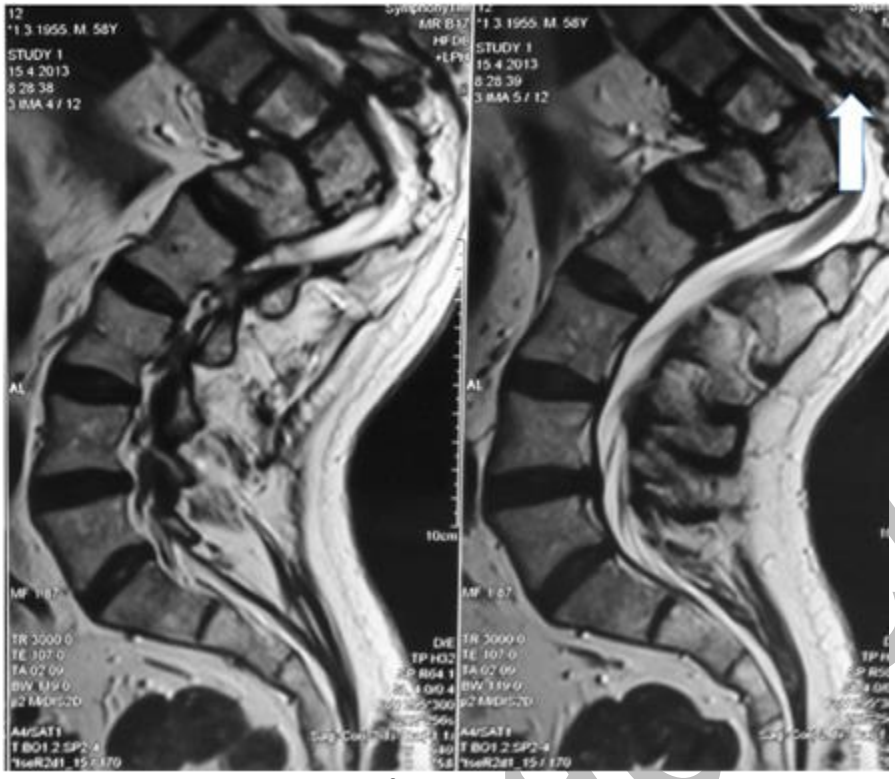

Figure 2. T2 weighted MRI of the thoracolumbar spine showing pronounced kyphosis with signs of spinal canal stenosis most pronounced at the level of Th11-Th12 with the consequent myelopathy finding, due to hypertrophied yellow ligament at the same level (arrow)

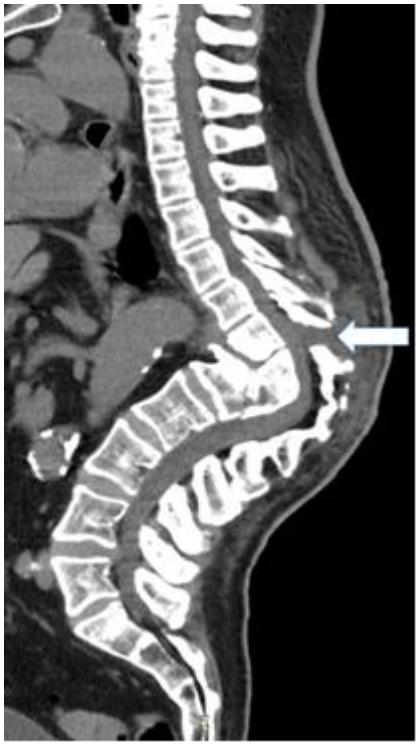

Figure 3. Control computed tomography scan of the spine showing good decompression at the level of Th11-Th12 (arrow), without signs of instability or progression of kyphosis

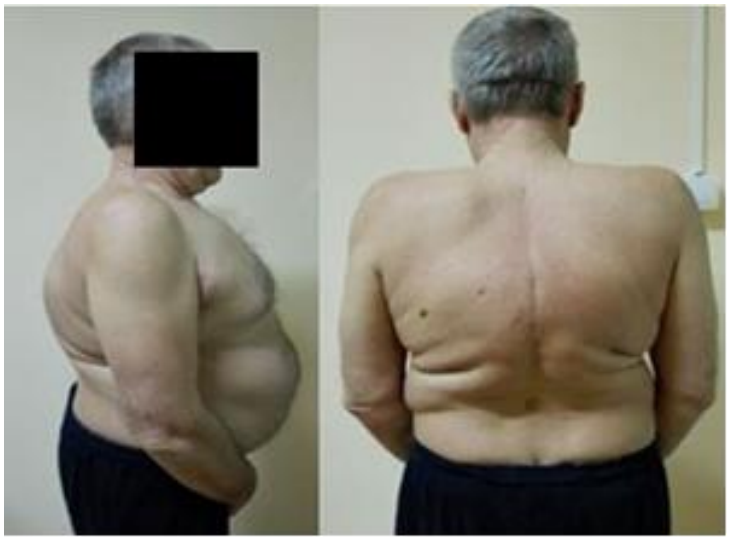

Figure 4. Patient has kyphosis in upright posture (since childhood). After operation patient can stand alone and walk with a cane. Pronounced gibbus is seen in the thoracolumbar region. On the right picture a scar from surgery in the thoracolumbar region is showed

