Rare location of spondylitis tuberculosis; atlanto-axial, sacral and cervico-thoracic junction

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Abstract. Three cases of rare location spondylitis tuberculosis are reported, each in atlanto-axial, cervico-thoracic junction and sacral. The complaints were aweseness of motoric strength and local back pain. Patients’ thoracic x-ray was normal, there was no complaint of acough, PCR for TB was early diagnostic and positive in all three cases, HIV negative, intraoperative tissue samplings were sent for histopathology examination and the results showed the specific inflammatory process. Lesions were evaluated with computer tomography and/or MRI imaging. Preoperative TB regimens therapy were given for 2 weeks and continued for nine months. The surgical procedure was done in all cases with excellent improvement of symptoms and motoric strength. In our institution, 25 cases of total TB spondylitis were performed in 2 years, only 1 case each was found in atlanto-axial, cervico-thoracic and sacral.

1. Introduction
Atlanto-axial, sacral and cervico-thoracic junction are rare locations of spondylitis tuberculosis (Pott's disease).[1] The most frequent region of the vertebral column involved in tuberculous infection is the thoraco-lumbar.[2] Tuberculosis (TB) is a re-emerging disease worldwide, affecting about one-third of the world’s population, and second only to HIV/AIDS as the greatest killer worldwide. In 2012, 8.6 million people worldwide fell ill with TB, and 1.3 million died from TB, with over 95% of deaths occurring in low and middle-income countries. Skeletal TB constitutes 3-5% of all cases of TB, with TB of the spine being the most common form of musculoskeletal TB.[1] Atlanto-axial TB accounts for only about 0.3-1% of all cases of spinal TB.[1-5] Cervico-thoracic junction (C7-T4) tuberculosis constitutes 5% of spinal tuberculosis and isolated tuberculosis of the sacrum is rarely reported in the literature. The frequency estimated at 5%.[6-8] In a review of 63 cases of spinal tuberculosis, sacral involvement was found in just four cases, while none was involved in 107 patients.[9-12]

2. Case Report-1 (C1/C2)
A 16-year-old girl with significant complaints of upper and lower extremity weakness with 3/3 muscle strength accompanied by local neck pain, X-ray thorax in the normal range, from other physicians had PCR test positive, HIV test negative and had been treated with TB regimens for two months. Due to progressive motoric weakness, a cervical traction was performed for one week and followed by two staged procedure trans-oral debridement of granulation tissue with decompression. Posterior occipito-cervical fusion in single sitting, granulation tissue taken by surgery was sent for histopathology examination, motoric function progressively improved after surgery and patient were able to walk one
month after surgery. There were no significant complications except mild mastication pain in the temporo-mandibular joint region which disappeared two months after surgery, anti-tuberculosis drug treatment and physiotherapy were continued.

Figure 1. (A) Sagittal fragment cervical computed tomography (CT) shows invagination of odontoid tip to foramen magnum, (B) left C1 arcus destruction with odontoid dislocation to posterolateral, (C) sagittal MRI shows spinal cord compression as high as foramen magnum and retropharyngeal abscess, (D) cervical traction before surgery, (E) trans-oral decompressive procedure position, (F) trans-oral view shows visible nasopharyngeal soft tissue bulging, (G) illustration of anterior drilling C1 ring and odontoid tip location, (H) anterior C1 ring and odontoid tip drilling.

Figure 2. (I) occipito-cervical fusion procedure, (J) visible rigid occipito-cervical fusion fixation x-ray after surgery, (L;N) after surgery MRI shows achieved decompression.

3. Case Report-2 (S1/S2)
A 35-year-old male with major complaint isolated lower back pain, no history of fever and weight loss, normal lower limbs motoric and sensory examination, no sign of bowel or bladder incontinence and sacrococcygeal region pain on palpation. Lumbosacral MRI examination shows suspicious radiological expertise of a sacral chordoma, normal thoracal x-ray, positive PCR test, HIV negative, treated with TB regimens for fourteen days before surgery followed by surgical laminectomy decompression of S1/S2. Debridement and posterior L4/L5 iliac bone fusion with pedicle screw rod, granulation tissue taken by surgery were sent for histopathology examination, back pain was a relief after surgery, TB regimens was continued.
4. Case Report-3 (T1/T2)

A 17-year-old male came with the major complaint of all extremities weakness which slowly progressing for a month, muscle strength of 4/3, no defecation and micturition complaint, local neck pain, a thoracic x-ray is normal, positive PCR on TB and negative HIV. After treatment with TB regimens for fourteen days, laminectomy decompression was performed, partial sequestrectomy, debridement and cervico-thoracic posterior fusion with lateral mass/pedicle screw rod, granulation tissue taken during surgery was sent for histopathology examination, three weeks after surgery patient was able to run with no complaint of pain, TB regimens were continued. 2 years post-operation patient was in good condition with good position of the implant.

Figure 3. (M) sagittal lumbosacral MRI shows S1 and S2 bone destruction, (O;P) axial sacral MRI shows S1/S2 destruction and anterior sacral cold abscess (Q) lumbo-sacral x-ray after surgery shows L4/L5 iliac bone pedicle screw-rod fusion.

Figure 4. (R) MRI cervico-thoracic sagittal shows loss of subarachnoid space on level T1/T2 due to compression and paraspinal abscess (S) cervical sagittal CT shows T1/T2 corpus destruction, (T) laminectomy decompression of T1/T2 was performed, debridement and posterior fusion with lateral mass screw-rod C5/C6-Pedicle screw rod T3/T4, (U;V) cervico-thoracic junction x-ray after surgery shows visible rigid fixation, (W) three weeks after surgery patient was able to walk.
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, and a year after surgery evaluation with results of good implant position and
revealed by back pain without neurological deficit.

Isolated sacral tuberculosis is exceptional and often causes rapid and correct diagnos-

5. Discussion
The first paragraph consists of the burden of disease not jump to sign and symptom directly. Signs and
symptoms may be absent generally, and non-specific radiographic image enhances diagnostic
difficulty. Lesions are best with using computed tomography and/or MRI imaging. Atlanto-axial TB
imaging shows odontoid tip lateral masses of the atlas and fusion in soft tissues. Pott’s disease is able
to severely damage the odontoid apophysis and responsible for atlanto-axial dislocation and
instability, as in the case of atlanto-axial subluxation and left C1 ring destruction were found.[5]

Generally, spondylitis tuberculosis surgical procedure is performed to the relief of neural
compression, maintenance of spinal stability, obtaining tissue for thorough biopsy debridement.

Atlanto-axial TB surgical procedure can be done with single stage posterior occipitocervical fusion or
two staged procedure in single setting i.e. (transoral decompression with excision of the C1 anterior
arc and or excision dens and/or drilling clivus). In the above three cases transoral decompression
sequestrectomy of granulation tissue, anterior arc C1, and odontoid tip drilling, after surgery MRI
shows achieved decompression and patients were progressively improved clinically.[2]

Isolated sacral tuberculosis usually presents as chronic back pain in adults and discharging sinuses
or abscess formation, with or without neurological deficit. Briefly, in the literature, it was reported that
isolated sacral or coccygeal tuberculosis is revealed by back pain without neurological deficit. It is
suitable for the case above, with a complaint of local lower back pain only, without any neurological
deficit.[9,10] TB sacrum MRI usually reveals diffuse marrow edema that is hypointense on T1 and
hyperintense on T2-weighted images. The tubercular lesions may be misinterpreted with other
infectious diseases or neoplasms in MRI, like the above case the MRI imaging were amalignant
neoplasm. Surgery may be necessary due to neurological compression signs during the extensive
destruction of several vertebral bodies with aspinal deformity or to evacuate an abscess that is resistant
to medical treatment. Prognosis of sacral tuberculosis is good when the rapid and correct diagnosis is
made, and adequate treatment is provided. This pathology should always be suspected in any process
of the lytic sacrum, mainly in endemic areas of tuberculosis, to prevent or at least reduce the morbidity
of this disease, which is generally curable. Isolated sacral tuberculosis is exceptional and often causes
a diagnostic delay.[9,10]

Cervicothoracic spondylitis TB patients are more likely to experience neurologic deficits such as
tetraparesis or tetraplegia, so the choice of surgery with or without instrumentation is more preferred
than treatment with TB regimens only. The thoracic region area is known as the watershed area and
highly vulnerable to neurological disorders especially when kyphosis correction should be made. In
cases of severe kyphosis, correction is carried out through the anterior and/or posterior approach. The
anterior approach has a higher risk of bleeding due to organs and large blood vessels inside the rib
cage but the abscess and infected vertebral bodies are easy to clean up. A posterior approach may have
a smaller risk of bleeding, but lesser access to the anterior so some surgeons perform a combination of
anterior and posterior approach or vice versa.[7] In this case, we performed surgery with the only
posterior approach, and a year after surgery evaluation with results of good implant position and
ability to walk without pain were done.

In all three cases, the thoracic x-ray was normal, there was no complaint of acough, PCR for TB
was done for early diagnostic and positive in all three cases, HIV negative, intraoperative tissue samplings were sent for histopathology examination and the results showed the specific inflammatory process.

In the three cases above preoperative TB regimens therapy were given for two weeks and continued for nine months. Treatment was two phases: intensive (initial) phase and aconituous phase, according to WHO guidelines. The intensive phase consists of a two-month course of four regimens of first-line drugs: isoniazid, rifampicin, streptomycin, and pyrazinamide. In the continuous phase, isoniazid and rifampicin were given for four months. WHO recommends nine months of treatment for tuberculosis of bones or joints, whereas the American Thoracic Society recommends 6 months for spinal TB in adults and 12 months for children. The British Thoracic Society recommends six months of daily treatment with rifampicin and isoniazid, supplemented in initial two months with pyrazinamide and either ethambutol or streptomycin.[1]

6. Conclusion
Cranio-cervical junction region spondylitis TB, cervico-thoracic junction and isolated in sacral are rare cases. In our institution 25 cases of total TB, spondylitis was performed in two years, only one case was in each location of atlanto-axial, cervico-thoracic and sacral. Generally, the prognosis is good.

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