

Antepartum surgical management of Pott's paraplegia along with maintenance of pregnancy during second trimester

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Abstract



Introduction The existing literature is limited and inconclusive regarding management of spinal tuberculosis with neurological deficit during advanced pregnancy. None of the previously published case series concerning this problem during the second trimester of pregnancy have explored the option of simultaneous surgical intervention for it along with maintenance of pregnancy.

Case report A 22-year-old woman with 26 weeks of pregnancy (2nd trimester) presented with upper back pain for the past 2 months, inability to move both lower limbs for the last 1 week, bladder and bowel dysfunction for the

past 5 days (Frankel Grade B). Patient subsequently underwent MRI scan dorsal spine and the image findings were suggestive of spinal tuberculosis T2 level. After obstetric evaluation and opinion of the expectant mother, in view of extensive neurological deficit which progressed rapidly, decision was taken for surgical intervention along with maintenance of pregnancy. Patient was positioned in right lateral position after giving general anesthesia using double lumen endotracheal tube with lung isolation technique. Exposure was done using transthoracic third rib excision approach. Decompression was achieved by radical debridement at T2 vertebrae level followed by multiple rib strut grafts and stabilization with screw and rod construct between T1 and T3 vertebrae. Intra-operative measures including type of anesthesia, prevention of maternal hypotension, hypoxemia and hypothermia, and fetal monitoring by attending obstetrician were undertaken to maintain fetomaternal safety. Postoperative ultrasonography evaluation of the fetus revealed a normal study. Post-surgery histopathological evaluation of the surgical specimen confirmed tuberculosis infection and the patient continued anti-tubercular drug therapy for 9 months. She delivered a healthy girl child at 36 weeks of gestation by cesarean section. After about 14 months of postoperative follow-up, patient has completely recovered motor power with mild persistent sensory symptoms. She is self-voiding with mild constipation requiring occasional intermittent laxative use. Radiological improvements in comparison to the previous reports were also seen at the last follow-up.

Conclusion Although this is only a single case but being the first to our knowledge, the good results highlight the point that both surgical management and maintenance of pregnancy during second trimester complicated by Pott's paraplegia are possible, involving a multi-disciplinary team approach for optimal maternal and fetal outcome.

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Keywords Second trimester pregnancy · Pott's paraplegia · Anti-tubercular drugs · Surgical decompression

Case presentation

A 22-year female with 26 weeks of pregnancy (second trimester) presented with upper back pain for the past 2 months, inability to move both lower limbs for the past 1 week, bladder and bowel dysfunction for the past 5 days. On examination there was significant tenderness along T2–T3 level with marked paravertebral spasm. Neurological evaluation revealed spastic paraplegia with upper motor neuron signs and sensory hypoesthesia below T4 level. Perianal sensations were decreased but present, and voluntary anal contraction was absent (Frankel Grade B). Laboratory investigations showed normal blood counts, increased ESR (Westergreen method: 60 mm/1st hour) and normal liver function tests. MRI dorsal spine demonstrated collapsed T2 vertebra with altered signal intensity, iso-hypointense on T1 sagittal and hyperintense on T2 sagittal and TIRM (Turbo inversion recovery magnitude) images. Abnormal pre-paravertebral and epidural collection compressing the cord were seen (Fig. 1). Following clinical, MRI and laboratory investigation evaluation, our presumptive differential diagnosis included tubercular spondylitis, pyogenic spondylitis, brucellar spondylitis, hemangioma, giant cell tumor, aneurysmal bone cyst and hydatid disease of spine [1]. With a high prevalence of tuberculosis in our country which includes about quarter of the world's tuberculosis population and on basis of the available above mentioned data regarding present case, we felt the diagnosis to be most likely of tubercular spondylitis [2].

In view of extensive neurological deficit which progressed rapidly, decision was taken for surgical decompression and stabilization. Obstetric evaluation revealed a healthy fetus and it was planned to maintain pregnancy after consultation with the attending obstetrician and opinion of the expectant mother. The patient was taken up for the surgery the day following admission.

Historical review of the condition, epidemiology, diagnosis, pathology, differential diagnosis

From the era of Hippocrates up to the nineteenth century, pregnancy was considered beneficial to tuberculosis [3]. By the early twentieth century opinion changed concerning deleterious effects of pregnancy on tuberculosis, to the extent that abortion was recommended [4]. Immediately before the chemotherapy era, Hedvall demonstrated no net benefit or adverse effect of pregnancy on the progression of

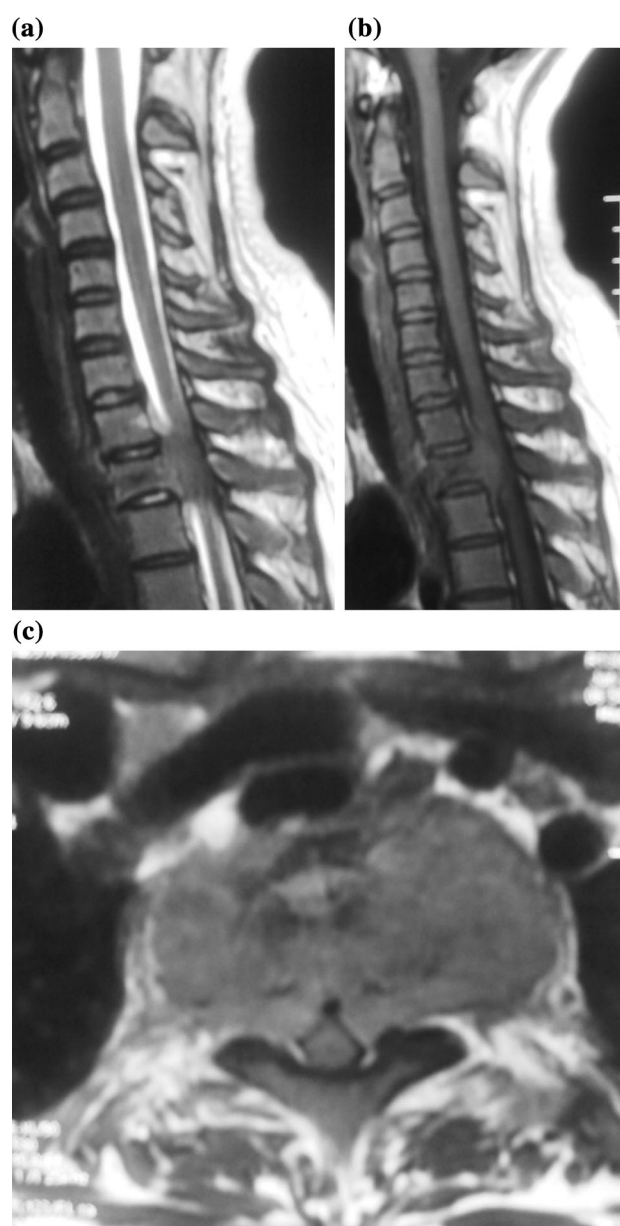


Fig. 1 Pre-operative MRI shows pathological lesion of T2 vertebrae. **a** Sagittal T2-weighted image; **b** Sagittal T1 image; **c** axial T2-weighted image

tuberculosis [5]. However, recent studies have challenged this opinion and suggest that biological changes during pregnancy and postpartum influence tuberculosis epidemiology [6].

Tuberculosis contributes significantly to maternal mortality, being among the three leading causes of death among women aged 15–44 years [7]. The data regarding exact incidence of tuberculosis during pregnancy are not readily available in many countries due to various confounding factors. A recent study has shown the prevalence of active pulmonary tuberculosis in pregnant and postpartum women from high-burden countries to be >60 cases

per 100,000 population per year and <20 cases per 100,000 population per year in low-burden countries [6]. In general, musculoskeletal involvement is noted in overall 4 % of all cases of tuberculosis, 50 % of which involve spine. Incidence of neurological complication in TB spine is about 10–20 % in highly developed nations and 20–41 % in developing countries [8]. There exists limited literature regarding tuberculosis of spine during pregnancy complicated by neurological deficit, either as small case series or single case reports [9–16].

The absence of systemic symptoms and reluctance to perform spinal radiography including computed tomography (CT scan) guided biopsy in pregnant women often delays the diagnosis, leading to serious morbidity like early onset paraplegia [17]. Factors prevalent in developing countries like poverty, malnutrition, suboptimal living conditions, human immunodeficiency virus infection, and multidrug-resistant tuberculosis further complicate the scenario [11]. Regarding safety of MRI during pregnancy, it has shown not be associated with any known adverse fetal effects as it lacks ionizing radiation. It is the recommended diagnostic modality for spinal problems in pregnant women with no contradiction for it during any stage of pregnancy [18]. Diagnosis of spinal tuberculosis during pregnancy is based primarily on the ground of clinical manifestations and radiography. Obtaining tissue specimen for etiological confirmation is avoided in view of radiation exposure involved during fluoroscopic or CT scan guided biopsy [11].

Considering use of molecular techniques such as polymerase chain reaction (PCR) as diagnostic tool for extrapulmonary tuberculosis, it is more reliable to conduct them directly on tissue sample rather than on blood, though the latter option may be utilized in situations wherein retrieving a tissue sample is technically demanding [19]. Although peripheral blood based PCR has proven to be a good option for identifying patients with active pulmonary tuberculosis, it has shown low sensitivity for extrapulmonary tuberculosis, especially with regard to musculoskeletal tuberculosis [20].

Recent immunologic diagnostic tools like interferon gamma release assay (IGRA tests) may prove helpful during the diagnostic workup for tubercular infection. It seems advisable to perform this test in any patient suspected of having tubercular vertebral osteomyelitis because of their high negative predictive value, regardless of the prevalence of tuberculosis. A negative test makes the diagnosis of spinal tuberculosis unlikely and requires other causes of vertebral osteomyelitis to be ruled out [21]. IGRA tests which measure the T-helper 1 (Th 1) mediated immune response may pose a challenge during pregnancy, in which Th 1 immunity is altered [22]. However, in a recent meta-analysis by Fan et al. regarding IGRA tests for

the diagnosis of extrapulmonary tuberculosis (EPTB), authors have stated that the immunosuppressive status of patients does not affect the diagnostic accuracy of these tests for EPTB [23].

Physiological changes associated with pregnancy including suppression of T-helper 1 proinflammatory response and high serum steroid levels may lead to a relatively immunocompromised state. This may lead to an aggressive behavior of skeletal tuberculosis including that of spinal involvement, leading to rapid and marked vertebral destruction, and even early neurological involvement [6, 11].

Rationale for treatment and review of the literature

The effects of extrapulmonary tuberculosis infection during pregnancy depend on the site of involvement, the severity and duration of disease, and the occurrence of pregnancy-related complications [17]. Maternal and fetal outcomes following extrapulmonary tuberculosis, other than tuberculous lymphadenitis infection during pregnancy, have shown an increased maternal disability resulting in frequent hospitalization, fetal growth retardation, low birth weight, and lower Apgar scores at birth [17].

For spinal tuberculosis during pregnancy, not complicated by neurological deficit or significant vertebral body destruction, conservative treatment with first line of anti-tubercular drugs is considered appropriate [11]. Regarding first line drugs rifampicin, isoniazid, and ethambutol, none of the available data have shown any significant adverse maternal–fetal effects or requirement of dose adjustment during pregnancy and postpartum [6]. Although there exist insufficient data regarding safety of pyrazinamide during pregnancy, it is recommended for routine use in pregnant women and postpartum from high burden countries by WHO (World Health Organization) and IUALTD (International Union against Tuberculosis and Lung Disease) [24]. Many experts recommend 9–12 months of anti-tubercular drug treatment for skeletal tuberculosis, as was followed in our case [25].

Progressive neurological deficit in an acute tubercular spondylitis requires early surgical intervention, but the prevalent literature is inconclusive regarding the time and nature of surgical intervention for spinal tuberculosis with neurological deficit in advanced pregnancy [11, 26]. As per general guidelines for spinal surgery in pregnant women with progressive neurological deficit, delivery should be induced or caesarean section is performed before spinal surgery at 34–36 weeks of gestation or later. In cases earlier than 34–36 weeks of gestation, prepartum spinal surgery should be performed [18]. Existing case series with regard to management of spinal tuberculosis with

Table 1 Literature review regarding management of spinal tuberculosis with neurological deficit during 2nd trimester of pregnancy

References	No. of cases	Age (year)	GA (week)	Vertebra level	Management
Govender et al. [9]	1	38	24	T8	Normal term vaginal delivery followed by surgical decompression
Vaidya et al. [10]	1	30	24	C6	Induced delivery followed by laminectomy
Han et al. [18]	1	27	16	T3–T5	Pre-operative therapeutic abortion followed by corpectomy with fusion
Our case	1	22	26	T2	Ventral debridement, fusion and instrumented stabilization, along with maintenance of pregnancy. Normal term delivery following cesarean section

GA gestational age

neurological deficit during second trimester pregnancy show variability in the measures taken (Table 1). In view of avoiding associated morbidity to the fetus if delivered premature, maintenance of pregnancy was considered. Studies have shown that survival of infants born between 23 and 25 weeks of gestation increases with each additional week. However, the overall neonatal survival rate of infants born during this early gestational period remains less than 40 %. Of those who survive, approximately 40 % have moderate to serious disabilities and many have neurobehavioral dysfunction and poor school performance [27]. Similarly, the EPIPAGE study (Etude Epidemiologique sur les Petits Ages Gestationnels) results showed highest cognitive and neuromotor impairment in children at 5 years of age, who were born at 24–28 completed week of gestation [28]. Therapeutic abortion of pregnancy may not be an appropriate option during advanced pregnancy, as was decided in our case [11]. To the author's best knowledge, this is the first case to report simultaneous surgical decompression and stabilization of spinal tuberculosis with neurological deficit along with maintenance of pregnancy during second trimester.

For performing spine surgery during second trimester pregnancy left lateral position is usually recommended to avoid aortocaval compression by gravid uterus [18]. Han et al. used lateral decubitus position for spine surgeries in both right and left directions along with maintenance of pregnancy during second trimester, depending upon the direction of the lesion [18]. Similarly, for approaching the concerned pathology anteriorly at T2 vertebrae level using transthoracic third rib excision approach in our case, we placed the patient in right lateral decubitus position [29]. With regard to performing thoracotomy or thoracoscopic procedures during advanced pregnancy, mainly to treat spontaneous thorax, studies have reported successful maternal and fetal outcomes [30].

According to MRC (Medical Research Council Working party on Tuberculosis of the Spine) guidelines regarding surgical management of tubercular spondylitis, radical

ventral debridement, fusion and reconstruction of the vertebral column remain the gold standard [31]. In the present case decompression was achieved by radical debridement at T2 vertebrae level followed by multiple rib strut grafts and stabilization with screw and rod construct between T1 and T3 vertebrae. In order to avoid morbidity following harvesting of the iliac bone autograft in our patient with advanced pregnancy, we decided to use rib autograft which was readily available following transthoracic approach. Although clinical results regarding use of rib autograft in adults with infective pathology have been mixed, it was further supplemented by anterior instrumentation (titanium pedicle screws), the use of which has been successfully described in the literature [31–33]. Minimal use of image intensifier was done for assessing proper implant position, taking adequate precaution by covering patient's abdomen with double lead apron wrapped in a sterile sheet.

Surgery and anesthesia during advanced pregnancy are associated with increased risk of spontaneous abortion, preterm labor, prematurity and low birth weight [11]. In our case, we avoided nitrous oxide during anesthesia as animal studies have shown it to be teratogenic affecting DNA synthesis [34]. Generous intravenous fluid administration matching surgical blood loss to maintain maternal arterial pressure and supplementary oxygen were given to prevent maternal hypoxemia which can lead to fetal hypoxemia, acidosis and death [35]. Hypothermia which can lead to fetal bradycardia was prevented by using warm air blanket [36]. DVT pneumatic pump for thromboprophylaxis and elastic stockings were used in view of hypercoagulable state which starts during late first trimester and attains a plateau level during second trimester onwards [37]. Isoxsuprine infusion was given to prevent premature labor [38]. Fetal monitoring was performed by attending obstetrician, as per recommendation in the given literature to be done after 23 weeks of gestation for fetal safeguard [18]. Overall, these intraoperative measures taken to maintain fetomaternal safety might have contributed to the successful outcome in our case.

Paraplegia during pregnancy is associated with an increased risk of urinary tract infection, decubitus ulcers, preterm labor, and autonomic hyperreflexia—a potentially life threatening complication [9]. We encountered no significant problems in our case, might be because of the improved neurological outcome following timely surgical intervention.

Procedure

Patient was positioned in right lateral position after giving general anesthesia using double lumen endotracheal tube with lung isolation technique. During patient positioning care was taken to pad all bony prominences and a soft roll was placed just distal to the axilla. Ipsilateral arm was kept in abduction by placing pillows underneath it and elbows were kept in slight flexed position. Isoflurane was given as inhalation agent along with supplementary oxygen. Nitrous oxide was avoided. Intra-operative isoxsuprine infusion was given to maintain uterine relaxation. Warm air blanket and pneumatic DVT pump along with elastic stockings were used. Exposure was done using transthoracic third rib excision approach. After retracting the scapula proximally following division of adjacent muscle layers, ribs were counted from below and 3rd rib resection was done. After deflating the ipsilateral lung, under direct vision, decompression was done by radical debridement at T2 vertebrae level followed by multiple rib strut grafts and stabilization with titanium pedicle screws and rod construct between T1 and T3 vertebrae. Minimal fluoroscopic exposures were taken after covering the patient's abdomen with double lead apron wrapped in a sterile sheet to assess implant position. Intraoperatively, generous intravenous fluid administration matching surgical blood loss was given. Proper oxygen saturation was maintained throughout procedure and arterial blood gas

evaluation showed normocapnia. Fetal monitoring was done by the attending obstetrician. Finally, thorough wound irrigation was done followed by layered closure of wound over a suction drain and a chest tube was placed through a different port.

Outcome and follow-up

In the immediate postoperative period patient started showing neurological improvement, and gradually kept on improving. Ultrasound evaluation of fetus done on first postoperative day revealed normal parameters. Histopathological evaluation of the surgical specimen confirmed tuberculosis, and patient continued with anti-tubercular drug therapy which was started during immediate postoperative period after she was allowed orally. She took first line of anti-tubercular drugs (Rifampicin, Isoniazid, Pyrazinamide and Ethambutol) for 3 months followed by two drugs (Rifampicin and Isoniazid) during the subsequent 6 months. Baseline liver function tests were assessed before initiating anti-tubercular drug therapy and were subsequently done after every 4–6 weeks along with ESR (erythrocyte sedimentation rate). The further course postsurgery was uneventful. The patient delivered a healthy girl child with a birth weight of 3.4 kg at 36 weeks of gestation by cesarean section as she had difficulty in perceiving contractions during labor. At the most recent follow-up, 14 months after the surgery, patient has completely recovered motor power with mild persistent sensory symptoms. She is self-voiding with mild constipation requiring intermittent laxative use. Computed tomography of dorsal spine done during this time showed stable implant with complete fusion, and MRI dorsal spine showed lack of any neural element compression (Fig. 2). Her child was also healthy and showed no ill effects of the treatment received by the mother.

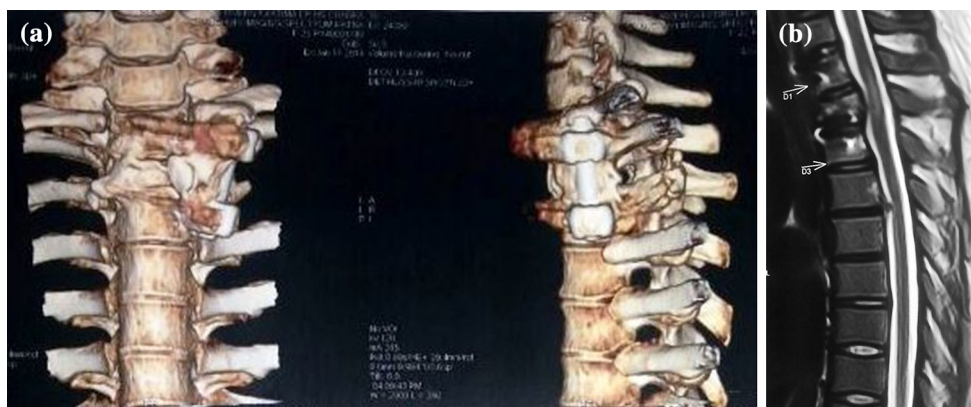


Fig. 2 3-D reconstruction CT scan (a) and MRI Sagittal T2 weighted image (b) at 14 months follow up

Conflict of interest None of the authors has any potential conflict of interest.

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