

Anterior Tibial Compartment Syndrome in a Child with Osteogenesis Imperfecta: An Unusual Complication of Spine Surgery.

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ABSTRACT

Objectives. To report the case of a child with osteogenesis imperfecta (type I) who developed bilateral anterior tibial compartment syndrome following posterior spinal instrumentation and fusion.

Summary of Background Data. No previous study has reported anterior tibial compartment syndrome after spinal instrumented fusion in prone position in a patient with osteogenesis imperfecta.

Methods. A 16 year old female with osteogenesis imperfecta (type I) and progressive scoliosis underwent posterior thoracic instrumented fusion. Surgery was performed with the patient placed in prone position and continuously monitored with somatosensory evoked potentials that showed no neurologic changes. The patient was transferred to the Paediatric High Dependency Unit and immediately after her recovery from general anaesthesia she complained of severe cramping in both lower legs anteriorly. Clinical examination revealed decreased dorsiflexion of both tibia (grade 2/5 on the right and 3/5 on the left leg). Intramuscular pressure was found to be 75 mmHg and she was taken for bilateral fasciotomy. After decompression pain improved in both legs but there was still a decrease of right dorsiflexion (grade 2/5).

Results. At the follow up 3 months after the operation the patient had improved right dorsiflexion (grade 4/5) and normal power in the left leg.

Conclusion. This is a very rare case of anterior tibial compartment syndrome following spine surgery in prone position. This position does not predispose to tibial compartment syndrome as other special positioning (tuck, kneeling) does. The continuous nerve stimulation during the evoked potential monitoring may have contributed to the development of the compartment syndrome. High suspicion for compartment syndrome because of unexplained leg pain, early diagnosis and urgent intervention (fasciotomy) is required for good results.

Keywords: Compartment Syndrome; Intramuscular Pressure; Scoliosis; Osteogenesis Imperfecta; Patient Positioning; Spine Surgery

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Although compartment syndromes of the tibia have been well described, this is an unusual complication after spinal surgery. Similar spinal surgery complications have been reported sporadically, such as one case of medial compartment syndrome of the foot after posterior instrumented fusion for idiopathic scoliosis¹, 2 cases of compartment syndrome of the thigh² and the lower leg³ respectively in knee-chest position and 2 cases of anterior tibial compartment syndrome in prone-sitting position⁴. In only one case compartment syndrome of the thigh has been described after femoral fracture in a patient with osteogenesis imperfecta⁵.

Case report.

A 16 years old female with osteogenesis imperfecta (type I), conductive hearing loss secondary to otosclerosis, multiple lower limb fractures (10 fractures) in the past, and no dentogenesis imperfecta, presented with progressive scoliosis. Health status was generally good with no recent fractures. The patient's weight was 31.1 kg, and her height was 139.9 cm, both progressing under 3rd centile. She was going through puberty-breast stage IV, pubic hair stage III, axillary hair stage III, and was expected to start periods within the next year. She had a lot of back pain. She was neurologically and vascularly intact distally. Her mother has osteogenesis imperfecta and had a Harrington rod inserted for scoliosis.

Radiographic evaluation revealed a right thoracic scoliosis of 87° and lumbar curve of 64°.

After informed consent, the patient was taken to surgery for posterior spinal instrumentation and fusion. She had isoflurane, fentanyl and vecuronium on induction followed by intrathecal morphine and clonidine. Maintenance of anesthesia was O₂/N₂O/ isoflurane based and she did not receive any additional muscle relaxant for the remainder of the procedure.

Patient was placed in prone position on well padded Montral mattress. Systolic pressure averaged 84 mm Hg (range 70-95), and diastolic pressure averaged 50 mm Hg (range 40-60). During the surgery the patient was monitored using somatosensory (SSEPs) evoked potentials by placing stimulating

nerve electrodes at T2 and percutaneously over the bilateral posterior tibial nerves. The patient was intermittently stimulated for 0.2-ms durations at 40 mA at a rate of 11 impulses per second. Acquisition frames comprised 350 stimulations during key portions of the procedure. The SSEP were normal throughout the procedure.

The fusion extended from T4 to L2 using pedicle screws and rods (**Fig. 1**). The dura was breached (it was exceptionally friable) during an effort to place a sublaminar device to help with reduction of the curve and to reinforce the fixation. Finally this idea was abandoned and meniscal repair performed to stop the cerebrospinal fluid leak. Blood loss during the surgery was 1600 ml.

Postoperative analgesia was provided from the previously given intrathecal morphine and clonidine for the next 18 hours. Additional pain control was achieved with patient-controlled analgesia delivered morphine (30 mg Morphine in 50 mls saline 1 ml (600mcg) bolus with 5 mins lockout).

In the Paediatric High Dependency Unit the patient complained of pain down front in both lower legs. Sensation and muscular power were intact. There was some tenderness in the extensors of both lower legs but no clinical evidence of fracture. The Doppler performed showed absent dorsalis pedis pulse and good posterior tibial pulse in both legs. Intramuscular pressure was measured using a Stryker pressure monitor (Stryker, Kalamazoo, MI) and was found to be 75 mmHg in the left anterior, 18 mmHg in the left posterior, 29 mmHg in the right anterior and 34 mmHg in the right posterior compartment.

The patient was taken urgently to the operating room and four compartment fasciotomy of both legs was performed. Legs pain resolved after fasciotomy. Next day sensation of the right foot was decreased, right dorsiflexion was grade 2 and left dorsiflexion was grade 3 of 5. The following days her recovery got complicated by ileus which resolved conservatively. She had a gradual improvement of sensation and muscular power. Ten days after her operation she was well mobilised and discharged home.

At the follow up 3 months after the operation the patient had improved right dorsiflexion (grade 5 of 5), normal muscular power of the left leg and

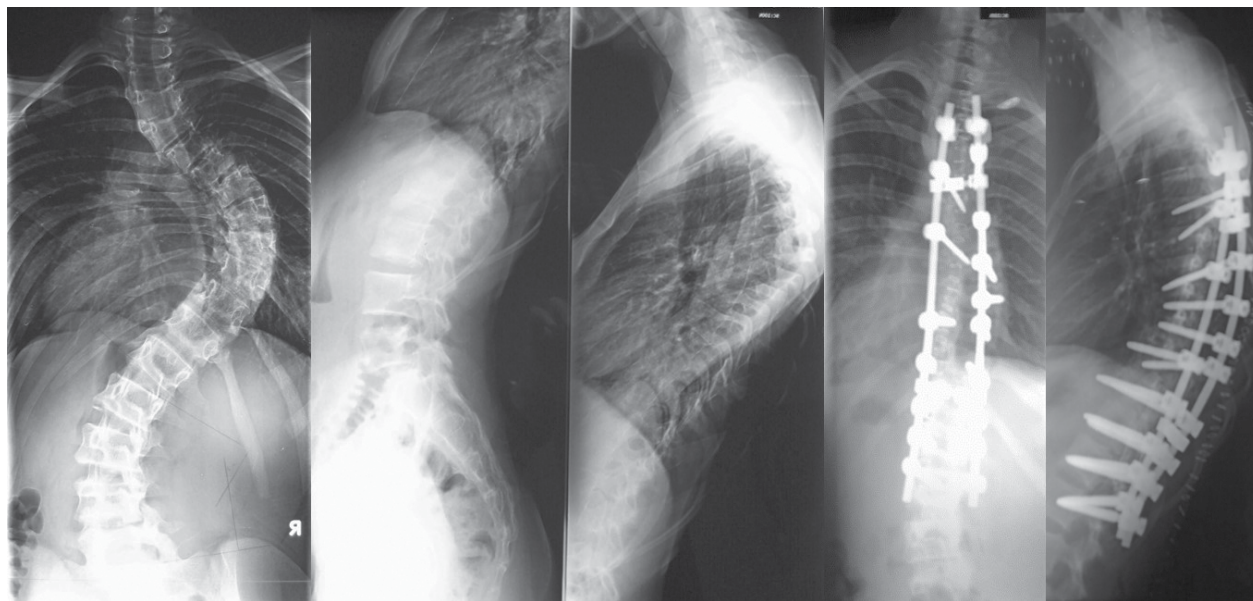


Figure 1. On the left, preoperative x rays of the spine of a 16 y.o. girl with scoliosis and osteogenesis imperfecta. On the right, the postoperative x-rays of the spine after posterior instrumented fusion.

normal sensation in both legs, however the loss of sensation on the dorsal area of the right foot persisted. Because of the poor bone quality, brace was required for 3 months. Clinically satisfactory spine correction was achieved. Radiography showed a right thoracic curve of 20° and a left lumbar curve of 15°. Metalwork was in place and there was evidence of bone fusion.

Discussion

Osteogenesis imperfecta (OI) is a rare heterogeneous group of inherited disorders that affect 1 in 5000 to 10000 individuals⁶. This disorder is caused by quantitative and qualitative defects in the synthesis of collagen I⁷. Major clinical characteristics of OI include generalized osteoporosis, age at the onset of fractures, bowing of the long bones, dentogenesis imperfecta, blue sclera, joint laxity, various degrees of short stature, basilar invagination and spinal deformities⁸. The incidence of scoliosis in OI varies between 39% and 100% in large retrospective series^{9,10,11}. Progression of the scoliosis appears very likely in curves of greater than 20°, in patients with the congenital form of the disease, in patients who have had more than 10 fractures, and in patient with severe long bone deformity¹². The

presence of six or more biconcave vertebral bodies before puberty has also been correlated with high likelihood of development of scoliosis greater than 50°¹³. The natural history of scoliosis is usually one of progression, which continues beyond skeletal maturity¹⁰. Continuous progression of the scoliosis leads to deterioration in the pulmonary and motor function of patients with OI, thereby affecting mortal and social prognosis¹⁴. Given the chest wall and rib fragility in patients with OI, bracing often fails to restrain progressive scoliosis¹⁵.

Posterior instrumented fusion is the treatment of choice, but is correlated with high risk of complications. Yong-Hing and MacEwen¹⁵ reported the results of arthrodesis of the spine in sixty patients. An average correction of 36% was obtained. One third of the patients had some complication; pseudarthrosis developed in five patients, nine patients lost more than 2.5 liters of blood, fourteen patients had problems related to the Harrington rods or hooks. A case report refers death because of intraoperative haemorrhage during spinal fusion surgery for osteogenesis imperfecta¹⁶. Despite the high rate of complications in patients with OI no previous study, in those patients, has reported anterior compartment syndrome after spine surgery in prone position.

Compartment syndromes have been reported in special positioning of the patient such as knee-chest or prone sitting position following spine surgery¹⁷. Only one case has been reported with compartment syndrome of the thigh in a patient with OI after femoral fracture².

We are uncertain of the etiology of the complication in our patient. We were careful positioning the patient and padding pressure points during surgery to ensure that nothing rested on her legs, and no areas of pressure developed. At the end of the operation there were no overlying skin marks on the patient. We suppose that this patient with osteogenesis imperfecta had a predisposition to develop a compartment syndrome. Another factor that could have contributed to the development of compartment syndrome is the continuous nerve stimulation during the spinal cord monitoring. Evoked potentials can increase the muscular activity within the anterior compartment. This activity theoretically can increase the pressure within the compartment to such an extent that the perfusion of the muscle can be compromised, despite a maintained diastolic average pressure of over 50 mm Hg throughout the case. This explanation has also been suggested in a reported case of medial compartment syndrome of the foot after spine surgery¹.


The postoperative analgesia by intrathecal morphine and clonidine given preoperatively plus

intravenous morphine delivered by patient controlled analgesia postoperatively did not mask this patient's symptoms as leg pain was present immediately after surgery¹⁸.

Early diagnosis and prompt fasciotomy resulted in good recovery of the patient. At the follow up 3 months after the surgery the patient had full recovery of the muscular power of both her legs and a persistent loss of sensation on the dorsal area of the right foot.

We recommend high suspicion of compartment syndrome in all patients with unresolved localized leg pain after spinal operation.

Key Points

- Legs should be carefully placed, padded and monitored during spine surgery.
- A sufficient average diastolic pressure should be maintained throughout the spinal operation to ensure adequate leg perfusion.
- Spinal surgeons should have a high suspicion for compartment syndrome in patients with unresolved leg pain.
- Postoperative analgesic techniques should not mask the symptoms of the syndrome.
- Early diagnosis and prompt fasciotomy is required for good results. 

Conflict of interest:

The authors declared no conflicts of interest.

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READY - MADE
CITATION

Sekouris N, Wilson McDonald J, Rogers R. Anterior Tibial Compartment Syndrome in a Child with Osteogenesis Imperfecta: An Unusual Complication of Spine Surgery. *Acta Orthop Trauma Hell* 2019; 70(3): 108-112.