CASE REPORT

Unilateral partial ossification of sacrotuberous ligament: anatomico-radiological evaluation and clinical implications

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Abstract
The present study describes the topography and morphometry of a unilateral ossified sacrotuberous ligament. It aims to discuss its anatomical, radiological and clinical implications. The pudendal nerve, internal pudendal artery, nerve to obturator internus and coccygeal branch of inferior gluteal artery are all-important structures near sacrotuberous ligament. An ossified sacrotuberous ligament may be an important etiological factor in neurovascular compression syndromes and its anatomical knowledge may help in the development of new treatment for this common clinical problem. The ossified sacrotuberous ligament in the present case was 7.6 cm in length and exhibited a characteristic anterior and posterior segment. The base was at the ischial tuberosity and the apex showed numerous small bony protuberances with deep intervening grooves. The ossified STL may be important in differential diagnosis of soft tissue pain and tenderness after trauma. It may be a challenging puzzle for the present day surgeon and radiologist in interpretation of CT-scans and MRI.

Keywords: ossification, sacrotuberous ligament, pudendal nerve, compression, radiography.

Introduction
The sacrotuberous ligament (STL) is an important architectural component of the pelvic wall since it connects the pelvic bone to the vertebral column. The STL along with the sacrospinous ligaments (SPL) stabilizes the sacrum on the pelvis. It is a strong ligament that provides support and stability to the sacroiliac region, by counterbalancing its rotation, therefore limiting the movement of the lower portion of sacrum [1]. Soft-tissue calcification is a well-known phenomenon in various pathological conditions. An ossified STL due to loss of elasticity may affect the general stability of sacropelvic articulation. The STL can act like a lobster claw with the pudendal nerve traversing the intriligamentous space where it can be crushed [2]. Ossification of STL has been reported in radiological literature. Anatomical description of the entity is however rare [3]. Due to ossification of STL, the lesser sciatic foramen which lies between STL and SPL will become a restricted anatomical space and therefore will become a potential site for compression of neurovascular structures traversing through this area. An ossified STL may therefore precipitate pudendal nerve entrapment syndrome and may be an important factor in undiagnosed chronic perineal pain. An ossified STL may be a significant factor in differential diagnosis of soft tissue pain and tenderness after trauma [4].

Material and Methods
The study was performed on dried cadaveric bone specimens obtained from the Department of Anatomy. During routine undergraduate osteological teaching program for medical students, amongst 100 specimens of human bony pelvis, a unique ossification of STL was noted in the right side of a human male pelvis. The anomalous ossified STL was studied in detail and the specimen was photographed. All the measurements of the bone specimens were recorded by using vernier calipers. A skiagram of the ossified STL was also obtained to study the radiological features. The Ethical Committee of Vardhman Mahavir Medical College & Safdarjang Hospital, New Delhi, approved the project and the reporting of dissection findings (anomalies).

Results
The ossification showed a broad based origin at the ischial tuberosity and exhibited a characteristic tapering craniomedially (Figure 1). The apex of the ossified STL was very irregular and showed multiple small bony protuberances with intervening grooves. The STL showed a distinct anterior and posterior segment. The anterior segment was 2.5 cm in length and showed a superior and an inferior surface. The anterior surface was more irregular than the superior surface. The posterior segment was 5.1 cm in length and also exhibited two surfaces, lateral and medial. The lateral surface was more irregular in the posterior segment. The total length of the ossified STL was 7.6 cm. The maximum transverse diameter of STL was 2.5 cm. The distance of the STL from the posterior inferior iliac spine was 1.5 cm. The ischial spine was 1.2 cm from the midpoint of the ossified STL. The apex of the greater sciatic notch was 5.5 cm from the apex of the ossified STL. The STL showed a characteristic spiraling along its longitudinal axis, which divided the ossified STL into anterior and posterior segments.
The posterior inferior iliac spine also showed a small-ossified segment of bone with a notched appearance. The skiagram revealed distinctly the ossification of STL (Figure 2).

The skiagram of the bone specimen was taken to depict its radiological appearance and hence ascertain the importance of these rare bony anomalies in clinical diagnosis.

The contralateral side of the male pelvis was normal and did not reveal any bony variation or ossification of STL.

**Discussion**

The STL attaches to the posterior superior iliac spine, the transverse tubercles of sacrum and the upper part of coccyx. Its oblique fibers converge and descend laterally to attach to the medial surface of ischial tuberosity. This attachment widens slightly and curves along the ramus of ischium to form the falciform process [5]. The finding of calcification of STL is rare [3]. Calcification of various soft tissues in the musculoskeletal system is a well-known phenomenon. In many cases, it may be the late sequela of a normal damage–repair process. This calcification process may affect various tissues such as synovium, muscle and cartilage [6–10]. Calcification of ligaments secondary to traumatic injury has also been reported although not a common one [4].

The first description of ossification of STL was mainly confined to its upper segment and in a cranio-caudal direction [11]. Anatomical description of STL is important because of the close relationship of the pudendal nerve and coccygeal branch of inferior gluteal artery.

Ossification of the STL has been reported in radiological literature. Radiological literature described ossification or calcification of STL as a slate pencil like formation, which begins at the ischial tuberosity, projects into the obturator foramen and has a caudo-cranial direction of growth [3]. One of the first illustrations reported that bilateral occurrence of ossified STL was rare [12]. Metaplastic ossification of ligament has been described as a normal variant [13].

A study on the diagnostic significance of radiographic abnormalities of the pelvis in patients with DISH and spondylosis deformans, showed that ossification of pelvic ligaments is a good indicator of presence of spinal DISH [14]. In a study of 101 cadavers, ossified STL was reported in eight cases (7.9%), the length of which varied between 1.1 and 7.2 cm. However, the study disapproved of any relationship between ossified STL and co-existence of DISH [3]. Interestingly, the incidence of ossified STL in the current study is much lower, one in 100 cases, and highlighting racial differences for the above. Ossification of STL has been reported mostly only in men [15]. STL ossification has never been described in children or adolescents hence proving that it is an acquired lesion and not an epigenetic variant [3].

Due to inadequate clinical data, it cannot be concluded that ossification of STL is related to particular occupation or special stress at STL. However, such a condition if present may lead to compression of neurovascular structures related closely to STL. We hypothesis that such a condition may precipitate
muscle, difficult to be diagnosed clinically. The internus may lead to consequent weakening of the ossified STL. Compression of the nerve to obturator internus, pudendal nerve, internal pudendal artery and nerve to obturator internus. The pudendal nerve (S2, S3, S4) is the principal sensory nerve of external genitalia and perineum. Its entrapment may be an important cause of chronic perineal pain. The pudendal nerve enters the pudendal canal, which is formed by duplication of the obturator fascia, inferior to the falciform process and nerve to obturator internus. The pudendal nerve is the principal sensory nerve of external genitalia and perineum. Its entrapment may also lead to fecal incontinence [17].

There are three possible sites of pudendal nerve constriction – between the STL and SPL, in the pudendal canal and by the falciform process of the STL [2]. In type 2 variety of STL described in an earlier study [18], the medial border of the falciform process of the STL descended to fuse with the lateral anococcygeal ligament, forming a continuous membrane involving three structures, the STL, the obturator fascia, and the anococcygeal ligament. The ossification of such a variety of STL would probably alter the morphology of the associated obturator fascia and anococcygeal ligament.

The STL contains the coccygeal branch of inferior gluteal artery. It passes posterior to the midportion of the SSL to pierce the STL at multiple sites [18]. The branches end by supplying the glutus maximus and the structures attached to the coccyx. Ossification of STL may lead to consequent compression of this artery and resultant ischemia in the area supplied by it [13]. Awareness of morphology of rare isolated cases of ossified STL as in the present case may be of great value to the present day surgeon operating on the pelvic floor. Ignorance of an ossified STL may pose unnecessary hindrance during sacrospinous ligament fixation procedures.

The lesser sciatic foramen which lies between STL and SSL will become a restricted anatomical space with ossification of STL and therefore will become a potential site for compression of neurovascular structures such as pudendal nerve, internal pudendal artery and nerve to obturator internus. The pudendal artery lies anterior to STL. Compression of this vessel is possible with increased transverse diameter of an ossified STL. Compression of the nerve to obturator internus may lead to consequent weakening of the muscle, difficult to be diagnosed clinically.

Study of such rare cases of ossified STL may be of great clinical and academic relevance. In the present case, partial ossification of STL can be easily made out in radiological study. Correct interpretation of such a case in MRI- and CT-scans may be beneficial for correct diagnosis of neurovascular compression syndromes of unknown etiology.

Conclusions

The anatomical and radiological knowledge of the ossification of STL may be helpful for clinicians, radiologists and surgeons dealing with neurovascular compression syndromes involving structures in the immediate vicinity of STL. Awareness of such ossification of STL may be relevant to surgeons undertaking reconstructive procedures on the pelvic floor.

References


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Received: December 29th, 2008
Accepted: August 8th, 2009