Transient Osteoporosis of the Hip: A Mysterious Cause of Hip Pain in Adults

Abstract

Background: Transient osteoporosis of the hip (TOH) is a poorly understood and forgotten clinical entity. The diagnosis is often delayed, and inappropriate treatment is provided, due to the lack of its awareness among the clinicians. Materials and Methods: Twelve patients (11 male and one female) within the age group of 35–50 years, were identified retrospectively from the hospital records between July 2011 and June 2015 who had evidence of TOH on clinical and radiological parameters. Results: All the patients were treated conservatively by nonweight bearing mobilization, anti-inflammatory drugs, bisphosphonates, calcium, and Vitamin D supplements. None of our patients had any symptoms after 6 months of conservative management. The disease did not progress, and there was no evidence of hip joint involvement in any of the cases. Plain radiographs were not diagnostic in the early detection of TOH. Magnetic resonance imaging was found to be highly specific and sensitive in diagnosing TOH. The clinical condition of TOH is characterized by its acute onset of hip pain in middle-aged people, and its symptoms are out of proportion to the radiological findings. Conclusion: The TOH is a nondestructive and self-limiting condition of the hip, which responds well to the conservative treatment. We believe that TOH could be a subset of complex regional pain syndrome type 1, as it has many similarities in clinical presentation and management. Awareness of this entity is important to the clinicians for an early diagnosis and to avoid unnecessary treatment for other mimicking conditions.

Keywords: Avascular necrosis, reflex sympathetic dystrophy, transient osteoporosis of the hip
MeSH terms: Osteoporosis, sudeck dystrophy, avascular necrosis of bone, magnetic resonance imaging

Introduction

The transient osteoporosis of hip (TOH) is a poorly understood and forgotten clinical entity. TOH was reported first by Ravault (1947) followed by Curtiss and Kincaid in 1959. It has been described with different names such as bone marrow edema (BME) syndrome, transient demineralization, complex regional pain syndrome (CRPS) type 1, migratory osteolysis, and algodystrophy of the hip. It is an idiopathic and self-limiting disorder which is characterized by unexplained hip pain. The diagnosis is often delayed, and inappropriate treatment is provided, due to the lack of its awareness among the clinicians. It is followed by spontaneous recovery, within a period of 2–9 months. The TOH is associated with reduced mobility of the hip, nonspecific laboratory findings, and mostly uncertain radiographic findings.

We present our retrospective analysis of such cases.

Materials and Methods

14 hips (n = 12) who were diagnosed as TOH between July 2011 and June 2015 comprised this study. The study was undertaken after obtaining ethical clearance, and written explained consent from all the patients, included in the study. The study group comprised 11 male and 1 female, with an average age of 41 years (range 35–50 years). They presented with the complaints of moderate or severe pain in thigh or groin. The right hip was affected in 8 cases and left hip in 4 cases; two patients had the bilateral affection of hip. A complete record of clinical examination and investigations including complete hemogram, erythrocyte sedimentation rate, and the rheumatoid profile was evaluated. History about excessive intake of alcohol, prolonged use of steroids, trauma or any other major or small joint involvement was also obtained from all the patients. All the patients underwent radiological examination including plain radiographs of the hips, magnetic resonance imaging (MRI), and bone scan. The MRI scans were performed...
on 3.0 Tesla (Philips® Achieva®) machine. The protocol for MRI included T1 (longitudinal relaxation time) weighted, T2 (transverse relaxation time) weighted and short tau inversion recovery (STIR) images. The T1- and T2-weighted images were obtained in coronal and axial planes, whereas the STIR images were obtained in coronal and sagittal planes. Clinical assessment was performed using Harris Hip Score.

All patients underwent conservative management in the form of protected weight bearing using crutches, nonsteroidal anti-inflammatory drugs (NSAIDs), bisphosphonates, calcium, and Vitamin D supplements. All the patients were accessed weekly for initial 2 weeks, then 4-week interval for 3 months and at 3 months interval until 1 year of followup. During followup, patients underwent plain radiographs of the pelvis and MRI, at 3, 6, and 12 months. The patient was declared treated with remission of initial presenting complaints and regaining full movement of the affected hip joint.

**Results**

The mean duration of the period from the onset of symptoms and treatment taken was 3.2 months (range 1–6 months) [Table 1]. The majority of these cases had presented with a sudden onset of pain in groin area, with no associated history of trauma or any other associated constitutional symptoms such as fever, weight loss, and polyarthralgia. The nocturnal pain was present in 10 of 14 patients. The pain was aggravated by weight bearing or exercise and was partially relieved by rest. The local examination revealed painful and moderate restriction of range of movement of the affected hip. Of 14 hips studied, 5 hips showed localized osteopenia extending from femoral head to intertrochanteric region. All patients underwent MRI and revealed a uniform, and diffuse hypointensity on T1-weighted sequences and hyperintense when associated with joint effusion on T2-weighted sequences [Figure 1]. Two cases reported with a nonuniform signal with alternate areas of intensity. Clinical and radiological improvement were analyzed.

None of the patients progressed to femoral head collapse, arthritic changes, or avascular necrosis of the head [Figures 2 and 3]. The average period for complete resolution of symptoms was 17.1 weeks (range 13-25 weeks). All the patients could join their work back after conservative management, with no recurrence reported in an average followup 1.3 years (range 1–2 years and 6 months).

**Discussion**

The TOH has been reported more frequently in healthy middle-aged males with a male:female ratio of 3:1.7,8

| Case number | Name | Age/sex     | Side | Duration of symptoms (days) | Pain         | X-ray features | MRI                          | Treatment   | Time for resolution (weeks) |
|-------------|------|-------------|------|----------------------------|--------------|----------------|------------------------------|-------------|-----------------------------|
| 1           | AC   | 35/male     | Right| 28                         | Sudden onset | Normal         | Uniform and diffuse signal intensity | Conservative | 18                          |
| 2           | RM   | 42/male     | Right| 35                         | Progressive  | Normal         | Uniform and diffuse signal intensity with effusion | Conservative | 16                          |
| 3           | YS   | 37/male     | Left | 30                         | Sudden onset | Localized Osteopenia | Uniform and diffuse signal intensity | Conservative | 14                          |
| 4           | VK   | 50/male     | BL   | 28                         | Progressive  | Localized Osteopenia | Uniform and diffuse signal intensity with effusion | Conservative | 23                          |
| 5           | SH   | 38/male     | Right| 33                         | Sudden onset | Normal         | Nonuniform signal with alternate intensity | Conservative | 15                          |
| 6           | MK   | 35/male     | Left | 32                         | Sudden onset | Localized Osteopenia | Uniform and diffuse signal intensity | Conservative | 17                          |
| 7           | SS   | 41/male     | Left | 27                         | Progressive  | Localized Osteopenia | Uniform and diffuse signal intensity | Conservative | 16                          |
| 8           | AD   | 45/male     | Left | 35                         | Progressive  | Normal         | Nonuniform signal with alternate intensity | Conservative | 13                          |
| 9           | SK   | 36/male     | Right| 30                         | Sudden onset | Normal         | Positive | Conservative | 14                          |
| 10          | RM   | 35/female   | BL   | 33                         | Progressive  | Normal         | Uniform and diffuse signal intensity with effusion | Conservative | 25                          |
| 11          | VG   | 43/male     | Right| 27                         | Sudden onset | Localized Osteopenia | Uniform and diffuse signal intensity with effusion | Conservative | 18                          |
| 12          | AA   | 41/male     | Right| 31                         | Sudden onset | Normal         | Uniform and diffuse signal intensity | Conservative | 16                          |

BL=Bilateral, MRI=Magnetic resonance imaging
Although lesser in females it is more commonly seen in pregnant women in their third trimester, has also been reported in nonpregnant women without any previous history of infection or trauma.\(^9\)

The exact etiology of this condition is still unknown, and some theories have been proposed, explaining this condition. The etiopathogenesis of TOH may include microvascular injury, nontraumatic reflex sympathetic dystrophy, metabolic, viral infection, neurological, and endocrine factors.\(^{10}\) Curtiss and Kincaid advocated a neurogenic hypothesis, child’s head compressing mother’s obturator nerve, as one of the contributory factors of TOH. Viral infection simulating an increased osteoclastic bone resorption, resulting in hip pain because of stress fractures in femoral head was proposed as a possible etiological factor.\(^{11}\) BME surrounding the femoral hip has led to believe in the theory of ischemia as one of the underlying causes. According to some researchers, this is due to a disturbance of the venous drainage.\(^{12}\) This theory is also supported by angiographic and scintigraphic studies, showing dilated nutrient vessels of the femoral head resulting an increased perfusion in the affected area, suggesting ischemia as a causative factor.\(^{13}\) Due to these associated findings, various acronyms of TOH have been proposed including “bone marrow edema syndrome” or “transient osteoporosis of the hip.”\(^{14}\) A transient ischemic episode leading to a contained area of cell necrosis was thought to be the cause of TOH.\(^{15}\) It was described as an early reversible phase of other mimicking pathology, i.e., avascular osteonecrosis of the hip.\(^{16}\) Some authors believe TOH to be a nontraumatic type of CRPS, because of its striking resemblance in various clinical, radiological findings of both these pathologies. However, TOH lacks specific cutaneous changes that are characteristic of reflex sympathetic dystrophy,\(^{17}\) which may be due to the deep location of the hip joint. We believe that TOH could be a subset of CRPS, as it has many similarities with it, namely, an unknown cause, characteristic pain (which is usually out of proportion to the physical and radiological findings), vasomotor dysfunction of the extremity and it develops in the absence of an identifiable precipitating event. TOH need to be differentiated from other similar clinical conditions such as avascular necrosis [Table 2], insufficiency fracture, infective, and inflammatory arthritis.

The onset of pain in TOH is sudden and more severe than osteonecrosis. Clinically, it presents as a dull aching pain in the groin region, buttocks, or anterior aspect of the thigh. The pain is worse in the night and on weight bearing. Functional disability is often disproportionate to the symptoms as described by Lequesne,\(^2\) with preservation of a range of movement. Occasionally, there may be some limitation of abduction and rotation. The TOH usually affects a single, but in sporadic cases, bilateral involvement has also been reported, similar to 2/12 cases in this series. Xyda et al.\(^{18}\) reported bilateral TOH in 3 cases in postpartum females.

Three distinct phases of TOH are known.\(^{19}\) The initial phase is defined by a sudden onset of pain associated with functional limitation lasting for 1 month. It is followed by a phase of which sign and symptoms follows a consistent trend with no further aggravation of pain and lasts for about 1–2 months. During this stage, osteopenia is a specific finding on routine radiographs. The final phase is characterized by spontaneous regression of clinical sign and symptoms and the bone density returning to normalcy; this period is usually as long as 4 months.

The radiographic features often lag behind clinical symptoms by 1–2 months. Initial X-rays show focal osteopenia involving femoral head and neck region. As the disease progresses, there might be complete effacement of the subchondral cortex of femoral head, and sometimes, a near absence of the osseous architecture, thereby creating
an optical void known as phantom appearance’ of the femoral head. Characteristically, during the whole course of the disease, there is the preservation of joint space with no osseous erosion or subchondral collapse, similar to our experience in this study. A bone scan is sensitive, but a nonspecific test. However, it can be a valuable screening tool for an early diagnosis of TOH. There is an increased uptake with radioisotope bone scanning even before radiographic changes are visible, thereby helping in early diagnosis of the pathology. Bone scintigram is characterized by an increased uptake in all the three phases indicating a focal area of hyperemia and increased capillary permeability with an increase in osteoblastic activity. Knees, or the shoulders. Radionuclide scanning provides an efficient screening tool for the entire body, and it may also reveal the asymptomatic involvement of other body parts such as the contralateral hip. An MRI is a very sensitive test to diagnose TOH and was described first in the radiology literature by Bloem.\textsuperscript{20} T1-weighted images demonstrate low signal intensity, and the T2-weighted images reveal matching high intensity involving femoral head to the intertrochanteric region, usually associated with effusion. MRI also helps in ruling out other pathologies such as avascular necrosis, insufficiency fractures, infection, and neoplasm which mimic TOH [Table 3].

TOH is a self limiting disease, a symptomatic and supportive treatment is recommended, which includes the use of NSAIDs with protected weight bearing and graduated physiotherapy regime.\textsuperscript{21,22} In an acute phase, intermittent traction helps in preventing and simultaneous correction

Table 2: Radiological differentiation between transient osteoporosis of hip and avascular necrosis

| Radiological investigation | TOH | AVN |
|---------------------------|-----|-----|
| **Plain radiograph**      |     |     |
| Subchondral osteoporosis/cortical loss involving femoral head and neck: Virtually pathognomonic | Early disease | Minor osteopenia, followed by variable density |
| Profound osteopenia involving femoral head and neck region, sometimes periacetabular osteopenia | Serpiginous area in femoral head with distinct sclerotic margins |
| Joint effusion may be present | Late disease | Micro fractures of the subchondral bone (followed by articular surface collapse, demonstrating a crescent sign) |
| Joint space is preserved | Absence of diffuse bone oedema (in most cases) | Collapse or fragmentation of femoral head leading to secondary arthritic changes |

| **MRI** | Bone marrow oedema extending from the femoral head to intertrochanteric region (may be appreciated within 48 h of symptoms) | Absence of diffuse bone oedema (in most cases) |
| T1: Poorly defined region of homogenously decreased signal intensity with loss of normal fatty marrow signal compared with normal bone marrow of the proximal femur | Reactive interface line is a low signal serpentine line surrounding a fatty centre (most consistent and early sign) |
| With or without joint effusion | Double line sign: Serpiginous peripheral/outer dark (sclerosis) and inner bright (granulation tissue) is a characteristic of T2-weighted image |
| T2FS/STIR: Increased signal, often heterogeneous, and may be striking | Rim sign: Characterised by osteochondral fragmentation |
| Maintained joint surface | Secondary arthritic change |

| Tc-99 methylene diphosphonate bone scan | Markedly increased homogeneous uptake in the femoral head; seen well before osteopenia is seen on plain films | Early disease: Likely vascular occlusion, reflected by a cold area |
| | Early disease: “Doughnut sign” (surrounding hyperaemia and adjacent synovitis) | Late disease |

MRI=Magnetic resonance imaging, AVN=Avascular necrosis, TOH=Transient osteoporosis of hip, STIR=Short tau inversion recovery
of deformity associated with a joint effusion. Although nonweight bearing is advantageous on the affected joint in long duration, this may lead to disuse demineralization. Trevisan\textsuperscript{23} advocated densitometric analysis in the conservative treatment of TOH. Physiotherapy regime must include abductor strengthening exercise and preservation of a range of hip movements. In this study, the average time for resolution of symptoms was 17.1 weeks. Of the 12 cases reported, 2 cases had a period of complete resolution of >20 weeks (23 and 25 weeks). The possible reason could be bilateral involvement, which led to delayed mobilization and rehabilitation protocol.
Several authors have discussed the beneficial role of various therapeutic agents such as bisphosphonate (both oral and intravenous), as effective means of treatment and may also speed up the recovery.\textsuperscript{24} Calcitonin in the form of a nasal spray (200 IU daily) has also been described,\textsuperscript{25} to reduce the bone loss during the acute phase of the disease. Iloprost (a prostacyclin analog) has been used recently with the satisfactory outcome with painful BME of the knee joint.\textsuperscript{26} Pain management and rapid regression of pathology are due to prostacyclin properties, dilating blood vessels and also reducing the permeability of small vessels (capillaries). Its effectiveness in TOH is still under consideration. The various studies which have described the role of various pharmacological agents such as calcitonin and bisphosphonates are based on small case series and sample size.\textsuperscript{24,26} The absence of a series with a large sample size can be attributed to the rare incidence of the disease along with delay in diagnosis of the disease, due to it closely mimicking avascular necrosis. To assess the role of these pharmacological agents in TOH, large-scale studies with control groups are needed.

**Conclusion**

We believe that the TOH is not an uncommon condition in clinical practice of an orthopedic surgeon and rheumatologist. However, its diagnosis is often difficult in the early stages, due to lack of awareness and suspicion by the treating clinicians. We also believe that TOH could be a subset of CRPS, as it has many similarities with it, namely, unknown cause, characteristic pain (which is usually out of proportion to the physical and radiological findings), vasomotor dysfunction of the extremity and it develops in the absence of an identifiable precipitating event. An early and accurate diagnosis of TOH can lead to early resolution of symptoms of the patients and avoid unnecessary investigations and treatment for other mimicking conditions. This condition can usually be managed by conservative means, and the surgical intervention is often not recommended.

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**Conflicts of interest**

There are no conflicts of interest.

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