

The Orthopaedic view of Transient Osteoporosis of the Hip in Pregnancy.

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ABSTRACT

Scope. The analysis of Transient Osteoporosis of the Hip, which invades the 3rd trimester of pregnancy (TOHP) and arouses the interest of the Orthopedist in its treatment. It is a rare, unknown disease that presents with localized osteoporosis in the neck and femoral head, often bilateral and resolved spontaneously after 4-8 months. The reduction of the mechanical strength of the femoral neck carries the risk of fragility fracture. The diagnosis is made with the information from the history and the objective examination and is documented with Magnetic Resonance Imaging (MRI).

Material and Methods. During the last 25 years (1998-2023), out of a total of 85,000 deliveries, 18 cases of TOHP were diagnosed (0.021%). The symptoms of the onset of the disease were pain in the affected hip with lameness that limited daily activities.

Results. Treatment was conservative and included rest, discharge of the affected hip, analgesics and anti-osteoclastic drugs after childbirth. After 6-8 months, there was a complete remission of the manifestations of the disease, and only in one woman, the diagnosis was made after a fracture of fragility of the femoral neck that was treated surgically.

Conclusions. This rare disease (TOHP) has a benign progression and requires early diagnosis, with awareness of Obstetricians Gynecologists and Orthopedic Surgeons, for the immediate initiation of therapeutic measures. Protection from a possible femoral neck fragility fracture, as well as surgical repair if it occurs, are critical issues for the successful final outcome of pregnancy.

KEYWORDS. Transient osteoporosis of the hip, Pregnancy, Orthopaedic.

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1. TOHP an unknown syndrome

1.1. Generally-Causes TOHP

The first report of the disease was made about 65 years ago, when 3 cases of pregnant women were diagnosed, with hip pain, decalcification of the femoral head radiologically and the fact was attributed to neuromuscular causes [1]. Later, the basic features of the disease were established, summarized in pain, limitation of the range of motion of the hip and subsequent claudication. It is a disease that affects middle-aged men and women during pregnancy with radiological osteoporotic image of the upper end of the femur, without narrowing of the articular space. A key feature of this rare syndrome is the automatic restoration of both clinical and radiological manifestations, after 3-12 months [2]. The etiology of this disease is unknown, and initial opinions indicated that it is a form of Südeck-Lerisch's, algodystrophy [3].

Reference is also made to the similar picture as to this, in the early stages of aseptic necrosis of the femoral head, both in the clinical and radiological picture. This has sparked debate about the common etiology of these two hip diseases, involving injury history, inflammation, use of medications such as corticosteroids, metabolic and neurological disorders, neoplasias and vascular disorders. Alcoholism, smoking, hypothyroidism, hypophosphatasia, vitamin D deficiency, low testosterone levels and certain professional activities are also implicated [4].

Relatively recently, studies involving predisposing factors compared to control cases were reported in women with the disease, who gained more weight during pregnancy, presented dental problems and reduced athletic activity in childhood and immobilization for at least 1 month during pregnancy. A causal relationship has by no means been established and all considerations are directed to the multifactorial justification for [5].

Advanced reports with literature review indicate that the etiology remains unknown and the disease is not causally associated with aseptic necrosis of the femoral head. Conditions related to pregnancy, such as reduced activity, femoral head venous posture due to the increasing size of the pregnant uterus, thyroid nerve pressure and hormone disorders

in pregnancy and lactation are implicated [6].

According to a theory about the etiology of the disease, the disorder in the synthesis of type I collagen is implicated. Since the basic solid components of the skeletal system are hydroxyapatite and collagen type I, its degradation during pregnancy, which can cause osteoporotic changes in the hip, is speculated [7]. This hypothesis is based on the case of osteogenesis imperfecta, where gene mutations associated with type I collagen, the risk of developing TOHP, is much greater.

It is known that in pregnancy, collagen I degradation changes occur to enlarge the cervical orifice, at the end of the second and third trimester of pregnancy, where body weight increases at the same time [8].

Collagenase derived from polymorphonuclei leukocytes, circulating with blood flow and settling in the bone tissue of the femur, with subsequent degradation of its mechanical strength. This theoretical view does not cover the cases of affected men and non-pregnant women, but in pregnancy the risk of fragility fracture in the hip area is clearly multiplied by weight gain and induced loads on the affected area, even with minimal strain on daily activities.

1.2. Radiological image

The radiological image is decisive in any case and is recommended for postpartum monitoring. The osteoporotic image of the femoral head and neck is characteristic, with extension in some cases to the acetabulum, as well as its restoration to normal after about 4-8 months. The preservation of interarticular space is always evident and this image resembles "ghost Joint" [9].

1.3. Magnetic Resonance Imaging (MRI)

The advent of MRI offered new possibilities for the early diagnosis of the disease, given the absence of radiation with this method and its clear applicability during pregnancy. It also can distinguish other pathological conditions, such as malignancy, septic arthritis, and stress fractures, that occur with a similar clinical picture.

MRI has proven from the beginning of its application in diagnosis, to be a sensitive, accurate and safe

diagnostic imaging test. Hip imaging gives clear evidence, where it shows reduced intensity of the bone marrow signal, corresponding to bone marrow edema in the initial stage [10]. In contrast, in aseptic necrosis of the femoral head, this edema follows as a reaction to the microfractures caused by the collapse of the femoral head in the final stage. Familiarity with the distribution of lesions in the MRI picture offers safe conclusions about the nature of the disease and is a crucial tool for diagnosis [11], [12]. There have been cases that were diagnosed early, with documentation of the radiological picture, with differential diagnosis from aseptic necrosis of the femoral head and unnecessary surgical interventions. The opportunity was also given to understand pathological lesions, by correlating the images with biopsies that had unfortunately been performed to investigate the disease, to investigate possible causes with a greater approach [13].

1.4. Laboratory tests

The hematological and biochemical investigation in this disease does not offer substantial assistance to the diagnostic process since in the cases of measurements of hematological and biochemical parameters no pathological value was found.

Relatively recently it was reported that pregnancy can cause danger to the skeleton, with fluctuations in calcium metabolism and it is possible to activate monogenic bone disorders, resulting in the appearance of a disease like this, during pregnancy where increased calcium supply is required [15].

1.5. DEXA examination

The characteristic radiological osteoporotic image of the affected pregnant women led to an investigation of the Bone Mass Density (BMD), with the Dual, Energy, X-ray, Absorptiometry (DEXA) method, where a decrease of up to 20% in the bone density of the hip was found. Restoration of BMD to normal levels occurred over a one-year period, while the same measurement in the lumbar spine decreased, even without symptoms, by up to 31%. The restoration in this area took place 2 years after the cessation of lactation [16]. This test has no place in the diagnostic process, but it can be used to as-

sess the effectiveness of the anti-osteoclastic treatment that may be administered to the patient after delivery.

1.6. Type of Delivery Recommended

Usually, this condition resolves spontaneously and no recurrence has been observed in the future, even in a subsequent pregnancy and it has been fully clarified that the delivery procedure with Normal Vaginal Delivery (NVD) is indicated and Cesarean Delivery (CD) is not required for prophylaxis the manipulations of possible musculoskeletal damage of the hip, in female sufferers during NVD. This is documented by many opinions that have been recorded in the relevant protocols by the WHO and the Academy of Gynecologists of the USA [17], [18].

1.7. Conservative treatment

The therapeutic measures proposed from the first diagnosis of this pathological entity were conservative with immediate unloading of the affected hip and rest from all physical activity. The administration of analgesic preparations mainly during pregnancy but also after delivery with additional administration of NSAIDs, aimed at relieving physical discomfort. After delivery, the treatment administered was anti-osteoclastic anti-osteoporotic preparations, with an initial application of calcitonin in which also provided an analgesic effect.

The administration of bisphosphonates after pregnancy is indicated because it did not gather the required safety guarantees for the fetus, for its administration in the first place. Supplements with calcium and vitamin D preparations complemented every medicinal effort. The administration of anticoagulation is not recommended if the patient needs immobilization for pain relief because the osteoporotic effect of these drugs has been demonstrated, which will likely worsen the already existing osteoporotic condition of the hip [6].

1.8. Surgical intervention

1.8.1. Preoperative assessment

Regarding the case where an undisplaced subcapital fracture occurs during pregnancy, immediate healing is required and in case of displacement, a

total arthroplasty of the affected hip is required [6].

In these unfortunate situations, the cooperation of the orthopedic surgeon, the patient's gynecologist and the anesthesiologist is necessary.

The preoperative gynecological and fetal assessment is a primary factor for the health of the pregnant woman and the fetus. Intraoperative monitoring of the heart function of the fetus is recommended, and if the rate drops below 100 per minute, an emergency Cesarean Delivery (CD) is recommended for fetuses older than 26 weeks. Knowledge of the changing physiological changes in physiology and anatomy in pregnancy is critical to the management of these cases.

The main ones are the following:

The relaxation of the sacroiliac joints and pubic symphysis resulting in a loss of balance and an increase in falls.

The increase in blood volume predisposes to hemorrhagic status and anemia.

The increase in the size of red blood cells and the number of white blood cells, predisposing to disseminated intravascular coagulation.

The increase in cardiac output resulting in an increase in metabolism.

The increase in the volume of blood filtered to the kidneys with consequent increase in pulse volume which increases the possibility of causing pulmonary edema.

The blood pressure is reduced, thereby camouflaging the possible early shock.

The increase in the volume of the uterus in advanced pregnancy can possibly cause hypotensive syndrome in the prone position because of the pressure of the aorta and inferior vena cava.

1.8.2. Intraoperative management

The problems faced by the orthopedic surgeon, in this rare case of TOHP fracture, concern the preparation of the operation, the surgical planning and the surgery itself with the appropriate anesthetic method and the post-operative rehabilitation care. It is important to position the patient during the operation on her left side to avoid compression of the inferior vena cava and the aorta. The choice of the surgical method must be made with the criterion

of saving time and minimal surgical intervention, with the least cost of radiation. Antibiotics to be used in the same way as outside pregnancy according to the protocols followed and prophylactic anticoagulation are considered fundamental in dealing with the increased possibility of venous thrombosis from pregnancy [19], [20].

1.8.3. Radiological assessment

Surgical planning requires the use of radiation to diagnose the nature of the fracture in order to select the appropriate method. The National Council on Radiation Protection and Measurements has established that the maximum possible safe limit of radiation that the fetus can accumulate during pregnancy should not exceed 50 mGy. However, depending on the age of the pregnancy and indeed in the last trimester, it is safely calculated that an exposure of 0.1 mGy is needed for the hip, which is in line with the protection measures. Practically in every single X-ray the use of radiation does not exceed 0.7 mGy, which ensures the protection of the fetus in every fetal period. When using the C-arm intraoperatively, the radiation source is under the operating table and there is relative protection. However, it is necessary to protect the other parts such as the pelvis and abdomen if this is possible during surgery. In general, however, it is preferable to avoid exposure of the fetus and the surgical techniques chosen do not require exposure to large amounts of radiation [21].

1.8.4. Anaesthetic intervention

The choice of anesthesia method is preferable to be made by a specialist doctor with experience in pregnancy anesthesiology, because there is familiarity with the changing physiology and pharmacokinetics of the pregnant woman and the fetus. The type of anesthesia is chosen according to the needs of the operation and there are no data documenting the influence of the type of anesthesia on the final outcome of the delivery. In any case, however, regional anesthesia is preferred, when it is possible to administer it [20].

2. Material and Methods.

During the time period 1998-2023, in 84,000 deliv-

eries that took place, 18 (0.02%) cases of pregnant women, with an average age of 33.5 years (26-41 years), were counted, who presented with pain, reduced functionality and lameness from the hip. These symptoms appeared in the 3rd trimester of pregnancy and in a single case at the end of the 1st trimester.

Bilateral location was observed in 2 (11.1%) cases, in 12 (66.6%) cases the disease was located in the left hip, while in the right hip it was located in 4 (22.2%) cases. In bilateral cases, the disease was not presented at the same stage by MRI imaging and the milder forms were asymptomatic. This fact raised the suspicion of the existence of cases with slight or non-existent symptoms, which escape diagnosis.

Regarding the gender of the newborn, boys predominated with 15 (83.3%) cases, against 3 (16.6%) cases of girls.

Phenotypically all the women were small with normal body weight and MO, BMI=22.1 and during pregnancy their weight increased by 12-16 (MO 14) kgr, a value that is considered greater than that desired by Obstetricians and Gynecologists.

In all cases, the diagnosis, treatment and follow-up were done by the same Orthopedic Surgeon in collaboration with the respective Gynecologist, who referred the patients when they presented the symptoms of the condition. The opinion they sought mainly after the diagnosis was related to the type of delivery that would follow. In all affected women, delivery was by Cesarean Delivery (CD), due to insecurity during delivery, to prevent a possible hip fracture. We note that until 2015 when the relevant WHO protocols were announced, the relevant literature did not take a clear position on the type of treatment required in case of TOHP. After 2015, CD was chosen for reasons independent of the condition.

Affected women had no history of osteogenesis imperfecta or chronic disease, no history of alcoholism, history of injury, or chronic medication, except for calcium and vitamin D supplements given at the onset of pregnancy.

Smoking was reported in 14(80%) cases and all women were not involved in sports activities, not even at a young age and had an unclear family his-

tory of postmenopausal osteoporosis.

In terms of occupation, this was sitting in private or government services, with additional standing in some cases.

The symptoms of the material patients reported were sudden onset of hip pain, worsening over the next 2-3 weeks with walking, which in some cases was difficult or impossible. The pain was located in the femoral-inguinal fold, in the abduction of the adductor muscles, in their mass, and with extension to the anterior surface of the thigh. It was combined with pain at rest or at night and with difficulty in raising the leg of the affected hip, in a supine position. In one case, the diagnosis was made after a fragility fracture of the femoral neck, which occurred at the Leto Obstetrics, in 2002 at the visit for the routine ultrasound examination at the beginning of the third trimester of pregnancy.

During the clinical examination, we found in almost all cases pain produced in the hip flexion ("log roll" test), in passive hip rotation movements, with the knee extended on the examination bed, with the patient in a supine position and in the passive abduction and adduction movements of the hip with the knee in flexion. The neurological examination did not show any pathological finding and all affected women presented a feeling of insecurity and anxiety during the first assessment.

The hematological and biochemical examination showed no abnormal findings, except for a negligible increase in the sedimentation rate and CRP in 2 cases which were considered incidental findings.

The documentation of the condition was done in all cases with an MRI check during the pregnancy and after the delivery was followed by a complementary radiological check that informed us about the recovery of the disease, with intervals every two months.

The treatment we followed during the pregnancy was the immediate unloading of the affected hip with armpit bacteria or walker, rest and in some cases, paracetamol was administered with pain at rest. Symptom resolution was gradual in all cases and required 7-10 days. Vitamin D and calcium were additionally administered in increased doses, beyond those administered by the Obstetrician dur-

ing pregnancy.

In the case of the interclavicular fracture (Garden III), reduction and union with ASNIS cannulated screws was performed immediately on the same day, followed by total arthroplasty of the affected hip, due to necrosis of the femoral head after 2 years.

After delivery, we also recommended unloading of the affected hip, calcitonin was administered in 5 cases and bisphosphonates per os and iv in the other 13 cases. calcium and vitamin D supplements were additionally administered.

3. Results

The follow-up of our material was done for up to 5 years, with clinical examination and radiological control every year, while full recovery was done in 4-8 months with an average of 6 months. No recurrence occurred even in a subsequent pregnancy that followed in 8 cases.

In the 1st case, 32-year-old woman (Picture 1a,1b,1c), pharmacist, with onset of the disease in the 9th month of pregnancy. Duration of disease 6 months. In the radiograph (Picture 1) in the left hip, femoral head is invisible ("ghost joint") from the obvious osteoporotic image. MRI lesions in the left femoral head (Picture 1b), and X-ray after 66 months (Picture 1c) reveals, full recovery.

In the 2nd case, 28-year-old woman (Pictures 2a, 2b), private employee, with sudden pain and lameness from the right hip, in the 8th month of pregnancy. The x-ray (Picture 2a) shows both hips with an osteoporotic picture. On MRI, the lesions are present in both hips (Picture 2b), with the left being lighter, without symptoms. Recovery in 8 months.

In the 3rd case, an 31-year-old woman, diagnosis of the disease, after a spontaneous fragility fracture of the left hip (figure 3a). She reported pain and lameness in the past 6 weeks. This was followed by arthrosis and after 3 years, due to necrosis of the femoral head, he underwent Total Hip Replacement (THR), as shown in figure 3b.

4. Discussion

TOHP is a rare disease that occurs in women during pregnancy and can affect both hips. The

symptoms of this condition are characteristic and Obstetricians and Gynecologists have become aware of the existence of this condition which is often confused with manifestations of back pain and pelvic pain. The physiology and anatomy of the pregnant body are subject to significant changes, which an Orthopedic Surgeon should always be aware of. In the case of our material, the awareness of fellow gynecologists resulted from informative scientific lectures and mainly from the incident of the hip fracture that happened in the Obstetrics "Leto" during the patient's visit for a standard examination of the beginning of the 3rd trimester of pregnancy. On that day, after the fracture, an x-ray check was done immediately, after being supervised by a specialist Radiophysicist and Radiologist. The surgical restoration followed immediately after the appropriate preparation in collaboration with the Obstetrician Gynecologist and the patient after about 2 months with CV, had a healthy boy weighing 2450gr. Since then there have been clinical examinations of 1-2 women every year, with similar symptoms, but no TOHP, and MRI was needed to prove the absence of the disease. Colleagues Obstetricians and Gynecologists, alarmed by the case of hip fracture, chose CV, for delivery, although they had been encouraged by the literature and Orthopedic information with special counseling lectures. TOHP, as a disease of pregnancy, needs the cooperation of many medical specialties for a successful therapeutic effect aimed at a successful outcome of childbirth as well as saving the functionality of the mother's hip. This issue is an Orthopedic challenge and we should always be prepared to deal with this peculiar case of an orthopedic patient. We are more informed and prepared today than we were 20 years ago, with the establishment of protocols by world-renowned Orthopedic Societies, such as the American Academy of Orthopedic Surgeons (AAOS). Knowledge is constantly evolving at a rapid pace and the awareness of clinicians has been achieved.

5. Conclusions


Medical Science requires constant vigilance and

daily information, even in the rare syndromes that the therapist is called upon to deal with. Prevention in any case is therapeutically superior to surgical repair in the case of TOHP hip fracture.

Basic tips in the direction of prevention are:

- Awareness of Obstetricians and Gynecologists for Orthopedic assessment when lameness is reported by a pregnant woman.
- Hip pain that persists and worsens during pregnancy, should be evaluated with great care.
- The clinical assessment should be done in

detail; the affected hip should be unloaded immediately and the possible existence of TOHP should be documented with an MRI.

- Great care is needed in the differential diagnosis in distinguishing TOHP from other serious conditions such as malignancy, osteonecrosis of the femoral head, and septic inflammation.
- Prevention of a possible fragility fracture of the femoral neck is the main therapeutic goal. 

Conflict of interest

The authors declare no conflicts of interest.

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