

## Legg-calve-perthes Disease Current Concepts

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## Abstract

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Perthes is a serious but self-limited disorder. If it is a self limiting disease, then why one should think of treating it. The aim of treatment is to prevent femoral head deformity and subsequent osteoarthritis. There is still no clear guideline for the management of Perthes disease. This is because of the controversies in all most all the aspects of the disease; etiology, classification, natural course of the disease, management and its outcome. Catteral emphasized the importance of proper hip examination in Perthes of younger age group, as the femoral head still remains concentric and rounded despite an adverse looking X-ray early on. The outlook of Perthes becomes more serious in older child or early adolescent, as the femoral head lacks the regenerative capacity during this period. Whatever controversies may be there, management principle still remains the same. Before any operation in Perthes', action must be taken to unstiffen the hip. Containment procedures will fail if performed on a stiff hip and even salvage procedures are best performed on a hip that has been loosened as much as possible. Surgeons need to recall that it is better not to intervene in a Perthes child where the surgical outcome is grave and make the child a candidate for an early hip replacement. It should be remembered that most untreated Perthes' do not require intervention until the age of forty.

## Introduction

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About 100 years ago, descriptions of curious conditions affecting pediatric hips began to appear in the literature. They noted a self limiting process in which the femoral head underwent through successive stages of degeneration and varying degree of collapse, followed by regeneration and healing, albeit not necessarily with complete restitution of the femoral head. In recognition of the contribution made by Legg in the USA, by Jacques Calve in France and by Georg Perthes' in Austria at that time, the condition became known as Legg-Calve-Perthes Disease (LCPD) [1]. LCPD is a poorly understood hip disorder of unknown etiology that is associated with disturbance of the

blood supply to the capital femoral epiphysis. There is controversy about almost all its aspects, including etiology, pathogenesis, classification, management, natural history and even the out come. These factors have led to heterogeneity of different treatment modalities, ranging from the highly invasive to the fully nihilistic [2].

## Review

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### **Epidemiology:**

The disease shows variable severity and is five times more common in boys of age 3 to 8 years old [1,2,3]. The '80% rule' is useful; nearly 80% of the cases are seen in boys, 80% have unilateral involvement and 80% have at onset between 4 to 9 years of age. The incidence of the disease differ world wide, with 1: 10000 children in USA, where as the prevalence is quite high in south India which is noted to be 2.97 per 10000 children as per Benjamin's report [4]. Approximately 25% of cases show bilateral involvement. In the majority of these cases the disease does not occur synchronously in each hip. If it does, alternative causes should be considered, the most common of which would be multiple epiphyseal dysplasia.

### **Etiology:**

The exact etiology of Perthes disease is still unknown; but considered as multifactorial. Many authors consider the changes in femoral head are because of repeated episodes of ischemia to the femoral capital epiphysis. Hence it is sometimes called as 'coronary artery disease of hip'.

**Growth factor:** Several studies including the study of Benjamin in Indian population have shown that children with Perthes are of shorter stature than their peers. Their bone age trails 2 to 3 years behind their chronological age [4,5,6,7]. Some authors have found evidence of reduced growth factors, such as somatomedins, in patients with Perthes' disease. Although true bilaterality only occurs in 25%, some investigators have found abnormalities of the contra-lateral hip, which they believe represent the growth abnormalities that cause the condition.

**Genetic factor:** The role of genetic factors is

controversial. Literature reveals the genetic association with expression of HLA- A antigens in lymphocytes of LCPD patients [7].

**Environmental:** The typical geographic distributions of the disease suggest that major environmental influences are involved in the causation of LCPD [7,8]. The disease is more common in the urban areas in England, but is rare among Chinese, Negroes and in the Maori population of New Zealand. In India the disease is more prevalent in rural areas and costal plains but rare in the crowded cities [4]. A major group of LCPD attending our hospital further supports this distributive epidemiology. Most of the patients of LCPD attending our OPD services are from rural areas of Punjab, Haryana, Himachal and some areas of Uttar Pradesh and Jammu-Kashmir. One of the important observations is that the incidence is relatively higher in low socioeconomic family. Clustering in families has, however, been described on several occasions.

**Trauma:** In the developing proximal femur the major lateral epiphyseal artery must course through a narrow passage, which could make the vessel susceptible to disruption if there was trauma to this area. Despite these findings, the premise that trauma is a causative factor of the disease is difficult to substantiate because frequent mild trauma is a common part of the childhood [1,7].

**Coagulopathy:** Over the last decade, interest has been focused on abnormalities of thrombosis and fibrinolysis in the genesis of Perthes'. Some research has demonstrated a consistent relationship to a coagulopathy involving protein C and S and hypofibrinolysis [9,10,11]. Several groups have described an association between Perthes' and passive smoking [12]. Smoking has an effect on tissue plasminogen activator levels supporting the thrombotic theory, but is also potentially a cause of small stature and delayed bone age. Repeated blockage of femoral artery in experimental animals leads to Perthes like changes in the femoral head; which is otherwise called as second infarct theory. Other investigators have demonstrated increased blood viscosity in patients with Perthes' and also intra-osseous venous hypertension.

The current hypothesis that is more plausible concerning the etiology is that the child has genetic or acquired dysplasia resulting in delayed bone age and the thick preossific cartilage of the femoral head provides inadequate protection to the traversing vessels supplying the ossific epiphysis. Compression of the cartilage may reduce blood flow causing ischemia or infarction.

#### **Clinical features:**

The onset of the disease occurs in between 2 to 12 years, with most cases occurring between 4 and 9 years of age. Most common presentation of the disease is limp, with pain felt at hip, thigh and knee. The onset can be quite insidious with rather vague and intermittent symptoms during the early months [1]. This is reason why many children present late, when the disease has already been progressed in to an advanced stage. The limp and pain increase with physical activity and later part of the day. Many times the child present with history of trauma with pain in gluteal region, lateral aspect of thigh or as a referred pain in thigh or knee. The relevance of the trauma is still unclear. The unifying hypothesis correlates the history of trauma with Perthes and proposes that a hyperkinetic child sustain minor trauma after a fall which leads to injury in the capital femoral epiphysis and metaphyseal area, inducing an increased tendency to form clot. The child is deficient in anti-thrombolytic protein C and S and as result clotting occurs in the venous system of metaphysis. When the clotting propagates to femoral head, infarction occurs and it becomes radiodense, a finding typical of Perthes disease [1,7].

The clinical sign include limp that is either antalgic or trendelberg or a combination of these two. The former limp is due to short stance phase because of hip pain, where as abductor dysfunction is the cause for trendelberg gait. Wasting of gluteal and quadriceps musculature is quite obvious in few patients. Movement restriction particularly that of abduction and internal rotation is observed because of associated muscle spasm [1,3].

As the disease progresses the limp, pain and restriction of movement increases, which correlates with collapse of the femoral head. Progress there after is dictated by nature and extent of ensuing femoral head deformity. If the head remains spherical or ovoid, the hip joint still remains as a ball and socket joint and rotational movement will recover as soon as the head heals. Where as if the head remains flattened, congruency with the acetabulum will be lost and the joint will adopt the configuration of 'rolling bearing' type joint. In this type, the joint allows only flexion-extension movement but no rotation. Furthermore if the flattened head remain extruded out from the lateral lip of acetabulum, it will hinge on abduction and will cause restriction of movement even on complete healing [1].

#### **Natural course of the disease:**

The clinical course of the disease is very

unpredictable. Some cases do not present until they are well into the healing phase and some restriction of movement or an awkward gait has been noted, whereas others present with pain and stiffness before any but the most subtle radiological signs are visible. Walden-strom initially thought the disease was a form of tuberculosis and a not a distinct entity. Subsequently he described the natural course of the disease into 4 stages i.e; initial stage, fragmentation stage, healing stage and residual stage. This radiological classification is further modified by Joseph et al (modified Elizabethtown Classification) from India ( Table 1) and divided in 4 stages; sclerotic, fragmentation, healing and healed stages [13]. Deformation of femoral head occurs during the late stage of fragmentation and early stage of revascularization. Hence the surgery for containment of femoral head can be performed before the late stage of fragmentation. The study by Benjamin is very important for management of Perthes as a timely intervention is essential [14]. During initial synovitis phase the radiographic picture shows widening of the medial joint space, may be caused by synovitis or hypertrophy of the articular cartilage. However, a smaller ossific nucleus of the femoral head due to temporary cessation of endochondral ossification caused by ischemia, is an early radiological sign. The fragmentation stage is characterized radiologically by areas of radioluscencies appearing with in the normal appearing sclerotic head. The areas of calcification in the lateral aspect of the epiphysis is evidence of the extrusion of the soft, flattended head from under the cover of acetabulum, changes are frequently seen in the metaphysis where un-ossified cartilage cells rests appear as cysts. The healing phase is further heralded by the appearance of bone in the subchondral region. This often starts in the periphery, center of the head and anterior segment being the last area to re-ossify. Further remodeling with time means that the final appearance of the hip may only become apparent at the time of skeletal maturity. Radiograph made at that time may show hips that appear to be virtually normal; at the other end of spectrum, marked residual flattening of femoral head may have persisted. Further involvement of physeal growth plate will lead to decrease growth in femoral neck, and relative overgrowth of greater trochanter.

**Table 1** Modified Elizabethtown classification of natural course perthes (proposed by Benjamin Joseph from India)

Modified Elizabethtown classification	Duration
<b>I Sclerotic</b> A: no loss of height B: loss of height	220 days
<b>II Fragmentation</b> A: early B:late	240 days
<b>III Healing</b> A: peripheral B:>1/3epiphysis	255 days
<b>IV Healed</b>	

#### Prognostic factors: To whom we should treat:

In 1971 Catterall showed that 57% of untreated Perthes patients show good long term outcome. For rational management of the disease one should need to identify which patients are likely to do well if left alone and which patient may be helped by surgical intervention [15,16,17]. In other words we should know about risk signs of the disease. The extent of involvement or in other words, the severity of the disease can be classified using Catterall, Salter-Thompson or Lateral Pillar classifications. Catterall classification was widely used in the past, but because of its application difficulty and interobserver variation its acceptance has been decreased in recent days [16,17]. Because of the simplicity and better outcome assessment by lateral pillar classification, it is increasingly reported to be used by many authors [18]. Preservation of the height of lateral pillar is more important and it signifies a lesser degree of involvement with good outcome.

Catterall classification was based on the extent of epiphyseal involvement and percentage of collapse as seen in x-ray (both AP and Lateral view) [16,17]. In Group I, only the anterior portion of the epiphysis is involved. Group II, involves more of the anterior segment and central sequestrum is present. Epiphyseal height remains preserved, though affected segment may show collapse. Group III child shows most of the epiphysis is sequestered with the unaffected portion located medial or lateral to the central segment. Group IV shows sequestration of all of the epiphysis. Catterall proposed few head at risk sign to predict the prognosis, which is cited in the Table-2 [17].

**Table 2** Catterall 'head at risk sign'

Clinical	Radiographic
Progressive loss of movement	Gage sign
Abduction contracture	Lateral calcification
Flexion with abduction	Lateral subluxation
Overweight child	Metaphyseal cysts
	Horizontal growth plate

Herring classified the fragmentation stage into 3 groups. According to his classification the head is divided into three pillars; medial, central and lateral. An intact lateral pillar acts as a weight bearing support to protect the central avascular segment. When there is minimal density change in the lateral pillar and no loss of height, it is classified under group A. Group B

includes when the lateral pillar height loss is less than 50% of its original height and the collapse of the central fragment is beneath the lateral segment. Group C includes when the lateral segment height loss is more than 50% and there is no separation between lateral and central segment [18].

Salter's classification is based on subchondral fractures of femoral head due to stress on this area. When the subchondral fracture or 'caffey's crescent sign' involves less than 50% of femoral head, it is grouped under 'A' and more than this is grouped under 'B' [19].

From all these classifications, it becomes clear that a young boy with partial head involvement and without risk sign would be one of those who would have a good outcome without specific treatment. Conversely the older child with whole head involvement and signs of lateral excursions and flattening is destined for a poor outcome without treatment [27].

#### **Differential diagnosis:**

The radiological picture of Perthes simulates many conditions that cause AVN changes in the femoral head. Sick cell disease, thalassemia and leukemia are difficult to differentiate on this aspect. It is sometimes quite difficult to distinguish post-septic sequelae and developmental dysplasia of hip from Perthes disease. Gauchers disease, lymphoma are other differential diagnoses. Certain skeletal dysplasia feature abnormal ossification of cartilage and may give rise to radiological pictures similar to that of Perthes disease. The commonest of these is multiple epiphyseal dysplasias. Here the changes in hip are bilaterally symmetrical and shows familial association [1,3]. This helps in differentiating the condition from bilateral Perthes disease, where the hip involvements are sequential rather than simultaneous. Children of hypothyroidism sometimes develop appearance of fragmentation of the bony femoral head leading to flattening and confuses with perthes [1].

#### **Investigations:**

##### **Plain x-rays:**

AP and lateral x-rays of the hip is the first and most important investigation. It can tell about the severity of the disease, can identify the risk factors and hence the treatment plan can be decided

**MRI:** It is an accurate modality for the early diagnosis of LCPD. Several classifications of Perthes' disease based on MRI appearances have been proposed. At this stage, none has gained acceptance, because they have not proved definitively prognostic and the necessity for repeated imaging studies in Perthes' is

inconvenient and expensive in the context of Perthes'. However it can better evaluate the congruity of articular surfaces, femoral head containment, joint effusion and synovial hypertrophy. Its implication for better delineation of the affected sites and the degree of involvement during the early phases of the disorder can not be substantiated.

**Bone scan:** Technetium scanning is an effective means of diagnosing LCPD in its early stages before it is evident radiologically. Care must be taken, however, as early bone scan may at times suggest a more severe condition than really exists. Few authors have classified the severity of the disease on the scintigraphic finding from grade I to IV suggesting 1/4<sup>th</sup> to complete involvement. Pinhole collimation in bone scan has several advantages; as it can record the viability of lateral epiphysis and thus may predict deformity [20,21].

**Arthrography:** The arthrogram is an important investigation of the child for whom intervention is being considered. Catterall emphasizes the importance of the arthrogram being dynamic. The surgeon must certainly view how the hip moves, as medial pooling occurs both with a flattened head that moves congruently and with hinge abduction. An arthrogram may demonstrate whether the hip is containable (i.e. the area of disease can be located satisfactorily within the acetabulum) or in non-containable hips may demonstrate the position of best fit prior to valgus or valgus extension osteotomy.

#### **Treatment:**

Treatment of Perthes disease is still controversial. Benjamin's experience of early surgery in Perthes disease is good in majority of cases where as Canale writes 'we rarely recommend surgery for Perthes disease because of the complications possible after majority of hip surgery'. Perthes is a local self healing disorder and the only justification of treatment is prevention of femoral head deformity and secondary degenerative osteoarthritis. If the mechanical environment remains favorable, there is every chance that the hip will recover a normal or near normal shape and movement. This could be possible by reducing the forces acting through the hip joint, maintaining a physiologic range of motion particularly abduction and physical contact or in other term 'containment of the femoral head [1,2,3]. From the radiograph we can determine the range of movement of hip joint; at the same time it can be seen if a position of improved containment can be achieved by abducting the hip. If this is the case, an osteotomy on either femoral side or pelvic side of joint can reestablish the relationship of femoral head and acetabulum. The simple radiograph

can further say about the concentric movement of femoral head with in the acetabulum. An enlarged poorly contained head will loose ability to rotate within the acetabulum and may hinge on the lateral margin of acetabulum on attempted abduction of hip joint. This hinge phenomenon is an important prognostic factor affecting progress in the later stages of Perthes disease, and may require surgical intervention [22]. So the principles of management will remain same through out disease process though the means by which they are achieved may differ depending upon the stage of the disease.

#### **Nonoperative treatment:**

As previously described, majority of Perthes child can be treated nonoperatively. The long experience of child will remain as frustrating restriction of movement, schooling and sports activity. The job of orthopedicians and paediatricians is to explain the disease course to the children and their peers. They should appreciate that the disease progresses and resolves stage wise, which cannot be bypassed or hurried.

The force acting through the hip joint can be reduced by weight relieving and by encouragement for keeping the limb abducted. The former can be achieved by either by strict bed rest or by avoiding any physical activity or sport activity and use of crutches for walking<sup>23</sup>. Specific exercises to keep the limb abducted and swimming is recommended. One problem of keeping the limb off the ground is that it tends to encourage adduction rather than abduction. The episode of pain, limp and spasm can be relieved by simple analgesic, anti-inflammatory and intermittent home traction. The traction should be applied in flexed position of hip joint; as the volume of the joint is maximum in this attitude and hence reduces the complication because of raised intra-articular pressure [1,2,3].

Abduction of the hip maintains the containment of femoral head and reduces the lateral excursion of a deformed and enlarged femoral head. This can be achieved by abduction cast and orthoses or in the form of femoral or pelvic osteotomy. A variety of ambulatory cast or braces have been in use for years in an attempt to maintain abduction while retaining mobility. Broomstick cast or brace, Petrie cast, Synder sling are good example of these abduction braces. However their success in recent years has been questioned and patient compliance is often dubious [24,25]. Recently some surgeons have used botulinum toxin as a myoneural blocking agent to reduce spasm in abductors group of muscle.

#### **Operative treatment:**

#### **Proximal femoral varus osteotomy**

By reducing the neck shaft angle, femoral head can be better contained or may be centralized within the acetabulum. It involves resection of a bone wedge from the proximal femur and securing the divided bone with one of the variable implant. Though it appears straight forward, several disadvantages in the form of scar mark in the lateral thigh region, subsequent implant removal in a second surgery and limb shortening are inevitable. Most authors agree that the outcome of Perthes disease following varus osteotomy is no better than an untreated case after the age of 9 to 10 years. However in younger children whose femoral head is at risk but still can be contained, varus osteotomy offers a 'one off' alternative to prolonged abduction cast bracing [26].

#### **Innominate (Salter) osteotomy**

This redirection osteotomy of acetabulum provides better coverage for the extruded part of femoral head. In a young child (when the pubic syphysis is cartilaginous and mobile) division of the pelvic bone above acetabulum, from sciatic notch to antero-inferior iliac spine allows the distal fragment to rotate downwards, outwards and forward to achieve this aim. The bone graft used to maintain this position is held by two k-wires. This procedure provides the advantages in the form of a cosmetically acceptable scar and involving no additional shortening of the limb [27].

#### **Acetabular augmentation procedures**

The femoral head is out of the stage of containment is by definition when it extends beyond the confines of the acetabulum. In these cases when the head can not be contained, some benefit may be gained by extending the roof of the acetabulum anterolaterally to increase the area of weight bearing surface. This can be achieved by shelf procedure. The force per unit area on the softened head is there by decreased. It further discourages the development of hinging and in the later stages of the disease it can provide better cover for the exposed part of the femoral head following valgus osteotomy.

#### **Proximal femoral valgus osteotomy**

This appears confusing that how both varus and valgus osteotomy can be helpful for containment of the femoral head in Perthes disease. Valgus osteotomy is indicated in the healing stages of the disease when there is established deformity of the femoral head and attempted abduction of the hip results in the hinging phenomenon. In this situation, arthrography often shows that the head and acetabulum are congruous with the hip in adduction and sometimes with little flexion. In these circumstances when a wedge of bone

is removed from the lateral aspect of proximal femur, it places the limb in neutral position whilst the hip remains in adducted and congruous. Patient satisfaction following this procedure is quite high; as they regain an affective range of abduction and at the same time the limb is lengthened giving biomechanical advantage to the proximal femur with improved gait. Inevitably this procedure results in uncoverage of the femoral head and few surgeon prefer to fashion shelf procedure to cover the exposed anterolateral aspect of femoral head. It should be remembered that this is a salvage procedure designed for late cases with established deformity. Many authors suggest that the result of valgus osteotomy is not so good when it is performed earlier in the disease process at a time when the femoral head is plastic and deformable.

#### **Cheilectomy/recreation of offset:**

It is the procedure where the extruded fragment is removed surgically. As originally proposed this operation was done through an anterolateral approach with simple excision of the fragment<sup>28</sup>. However stiffness of the hip joint has been a frequent sequela and more seriously, removal of the perichondral ring, together with the fragment, has resulted in slipped upper femoral epiphysis of the remaining epiphysis. This operation had fallen into disuse until repopularised by the Bern group, who have performed it in association with trochanteric distalisation and surgical dislocation of the hip [29]. When performed on a hip with an open physis they have always stabilised the epiphysis to prevent slipped upper femoral epiphysis. Reshaping of the femoral head and recreating the femoral offset in the manner they describe is certainly a more thorough approach than "bumpectomy" and the long-term results of this are awaited.

#### **Hip Arthroscopy:**

Occasionally in Perthes', healing of the epiphysis is incomplete and patients complain of symptoms suggestive of a loose body. If this is truly the case, then hip arthroscopy can be very useful. It should be noted that sometimes the "loose body" is not loose at all and detaching it from the femoral head arthroscopically can be extremely difficult [30].

#### **Outcome evaluation:**

Though Perthes is a self limiting disease; the femoral head never regains its vascularity before it gets deformed. The natural outcome of such a deformed and incongruous joint is premature osteoarthritis and painful dysfunction hip joint.

The Stulberg and Mose classification concerns the radiographic changes of the hip at maturity [31,32]. It

shows the shape of femoral head and its relations to acetabulum. Stulberg class I and II hip develops osteoarthritis in long term; where as class III and IV develops in late adult life. Class V hip showing 'aspherical in-congruency' develops osteoarthritis early in life, before 50 years of age [31]. Children presenting before 6 years of age without head at risk signs are likely to end up with Stulberg class I and II whether they receive any treatment or not. Conversely children presenting after 9 years of age with whole head involvement are likely to develop Stulberg III and IV class irrespective of treatment given. It is likely that active intervention in the form of surgery will have greatest effect on the intermediate group of children between 6 to 9 years of age group and may bring the hip back to Stulberg I and II category. Those involved with adolescent and young adults who recovered from Perthes are often difficult to advice in regard to safe activity level and appropriate careers. Radiograph of hip at maturity and Stulberg classification are helpful in this regard. Stulberg class I and II are probably capable of supporting their owner through normal range of physical activity in both works and recreational activity for at least 30 years of adult life. Those having Stulberg III hip should be discouraged to undergo any manual labourer activity. Mc Andrew and Weinstein while studying the long term follow up of Perthes, found that nearly 40% of hip eventually require total hip replacement, mostly between 40 and 60 years [33].

## Conclusion(s)

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Perthes is a self limiting condition that may nevertheless give rise to long term sequelae. At the very least, it will involve several years of disruption of normal activity, discomfort and disability. At worst it will lead to early degenerative changes in the affected hip., limiting range of movement and nature of physical activity available to the patient and resulting ultimately in total hip arthroplasty. Supportive symptomatic treatment is applicable to all patients during the active disease process. In certain cases which can usually be identified by clinical and radiographic features, more active surgical intervention can favorably influence the long term outcome. However many cases will do well without operative intervention.

## Abbreviation(s)

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LCPD: Legg-Calve-Perthes Disease

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