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Review Article

Developmental Dysplasia of the Hip-A Current Systematic Review

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Abstract

Developmental dysplasia of the hip (DDH) is one of the most serious and challenging pathology of Pediatric Orthopedics, consisting of a wide spectrum of clinical changes, which present in different ways and at different ages. These changes include joint instability in reduced but easily luxable hips; joint subluxation, in which there is contact between articular surfaces, but eccentrically; complete dislocation of the joint - without any point of contact between articular surfaces; and femoroacetabular dysplasia- morphological alteration of the bone anatomy, which prevents perfect joint congruence between femur and acetabulum. It is important to differentiate these various entities, as treatment and clinical evolution differ between them, as well as the final prognosis [1-3].

Keywords: Developmental Dysplasia; Hip; Anatomy

Introduction

The classic term used was congenital hip dislocation, however, it was progressively abandoned to the detriment of DDH, since it may be not present at birth and sometimes appearing during the child's development and growth [4].

Diagnosis is clinical and supported by imaging - ultrasound up to 6 months of life and pelvic radiography after 6 months. Early detection with consequent therapeutic onset is the most important factor in the final result, being the most favorable prognosis at younger ages [4,5].

Anatomy, pathophysiology, and etiological factors

The hip joint, formed by the acetabulum and proximal femur, has its stability based on bone joint congruence and surrounding soft tissues (labrum and joint capsule, capsular ligaments, round and transverse and pelvic musculature) [1].

In the newborn it is, however, a relatively unstable joint, since the surrounding musculature is not developed and the joint capsule and ligaments peri-articular not strong enough to stabilize it. Thus, small changes in these elements lead to joint instability compatible with AD [5].

In turn, this initial joint instability may be responsible for the development of acetabular and femoral dysplasia, because the normal development of the acetabular cavity requires a concentric movement of the femoral head inside. In the absence of joint contact, as occurs in dislocation, or in cases of subluxation, in which there is contact, but asymmetric, can occur progressive morphologic changes of the acetabular ring, adopting a shallow and enlarged shape, and sometimes flattening the femoral head itself [7].

Excessive anteversion, valgism, delay in the appearance of the ossification nucleus and shortened femoral neck are other anatomical variation associated with the dysplastic proximal femur [8].

The surrounding soft tissues may also undergo modifications that in turn hinder the concentric reduction of the femoral head in the acetabular cavity, aggravating instability- the labrum can become inverted, the round and transverse ligament became hypertrophic, the joint capsule constricted by the shortened iliac psoas tendon and the contracted adductor muscles [1,9].

Several theories and risk factors have been proposed for DDH. [10] Pelvic presentation in childbirth is pointed out as an important risk factor, since the fetal position adopted in forced hyperflexion of the hip with adduction can lead to an excessive stretch and laxity of the articular capsule [10]. Females are another important risk factor, corresponding to 80% of cases, and is often the only one present during diagnosis [10,11].

Family history for the disease is also well documented in the literature, with a predisposition to DDH, especially in first-degree relatives. A comparative study between risk of DDH in children with a family history reported a relative risk increased by 12 times for first-degree family members and only 1.7 times for second-degree relatives [12].

Although pelvic presentation, family history and female gender are considered by many authors as the main risk factors for DDH, others have been described: first pregnancy, situations that decrease intrauterine mobility such arthrogryposis, laterality (left hip is responsible for 60% of cases, right 20%, and bilateral in 20%), and congenital postural deformities such as congenital torticollis and more controversial, clubfoot and metatarsus varus, should also make suspect and exclude DDA [6,10].

Prematurity and hormonal theory in which increased levels of progesterone, relaxin and beta-estradiol in the mother would be related to DDH were not corroborated by recent studies [13].

As a protective factor, it has been demonstrated that societies that have a cultural habit to transport the baby with the hips in flexion and abduction, the prevalence is lower [14].

Natural history

The incidence of DDH varies according to the methodology used, with related occurrences of 10:1000 in series whose criteria are strictly clinical, and 25/50:1000, when considered ultrasound diagnostic criteria [6,15,16].

However, 88% of newborns with mild instability at birth resolve spontaneously within the first 8 weeks of life [16]. Resolution after 6 months of life is uncommon. Persistent cases, if untreated, lead to permanent dysplasia, modifying the biomechanics of the hip, and may be responsible for an accelerated wear of the articular cartilage and thus early originates osteoarthrosis (OA) of the hip. It is estimated that DDH is responsible for 2.6% to 9% of all Total Hip Arthroplasties (THA), and for 21% to 29% in young populations, being the main cause for THA in this age group [17,18].

If it persists during adolescence and adulthood, AD may cause functional changes, such as gait alteration, lower limbs dysmetry, hip contracture in flexion-adduction, and also compensatory changes in other joints such as ipsilateral knee valgism and postural scoliosis [19].

Diagnosis

Early detection is essential to treat dysplasia and achieve a good outcome. However, it is not always easy to make an early diagnose, since it is a painless pathology, being the baby asymptomatic and also because there is no definitive test or clinical finding [20].

During the first 6 weeks of life, radiographs are not useful, because the femoral head is composed of radiolucent cartilage and ultrasonography, although it can help, has a high false-positive rate during this period. Thus, a careful physical examination combined with high clinical suspicion is the best initial approach for the early diagnosis of DDH, and should be done in all newborns [7,21].

The evaluation begins with a careful clinical history, focused on potential risk factors, continuing with a general physical examination, which may present congenital deformities associated, such as congenital torticollis, and a specific examination of the hips, with the observation of asymmetries-the asymmetric inguinal skinfolds, sometimes caused by hip dislocation and limb shortening, it is a classic reference, although it does not have a true diagnostic value, because has low sensitivity and specificity; the shortening of the limb can be observed by Galeazzi's test, in which the child is lying in a supine, and with flexion of the hips and knees and the heels supported on the table, and any difference in knees height are determined. [2] The limitation in passive hip abduction, less than 70º, is a positive sign for instability and may be asymmetric in cases of unilateral DDH. The click felt with hip movement, with no other findings on physical examination, is considered benign [21].

The Barlow and Ortolani tests are the reference methods for the early diagnosis of DDH in the newborn. The Barlow test is a provocative test capable of identifying posterior sublimations or dislocation. With one hand stabilizing the pelvis whilst the other grasps the knee and flexes the hip to 90 °, the examiner makes hip adduction and slight pressure in the posterior and lateral direction. The Barlow test is considered positive if the hip can be popped out of the socket with this maneuver. The dislocation will be palpable. In turn, the Ortolani tests already dislocated hips, in which the hip flexion maneuver is performed with abduction and anterior translation, reducing the dislocated hip. Both tests should always be tested independently [22,23].

At 3 months of life, limited hip abduction becomes the most reliable sign associated with AD, since with the baby's muscle development the Barlow and Ortolani tests lose diagnostic sensitivity. Hip abduction may be decreased bilaterally, and thus without asymmetry, but should not be understood as normal, but as probable bilateral AD [22,23].

Image

After physical examination, if there is a suspicion of DDH, must be to performed a complementary imaging exam. An imaging study has also been recommended in the first 6 months of life for all babies with important risk factors, however, it is controversial which ones to consider [24]. The European Society of Pediatric Radiology mentions only positive first-degree family history and pelvic presentation at birth as the only ones with indication for diagnostic hip ultrasound in newborns with normal physical examination. Ultrasound universally to all newborns is not recommended, as it leads to an excess of diagnosis and treatment, without significant decrease in the prevalence of late hip dysplasia and surgical need [25].

Hip ultrasonography is the examination of choice in the first months of life, since the ossification center of the femoral head appears only between 4 and 6 months of life, making the pelvic radiography imprecise and of little use. However, in the first 6 weeks, the baby's physiological and benign hyperlaxity may lead to false positives on ultrasound. Thus, it is recommended to perform hip ultrasound after 6 weeks of life, to all those with family history, pelvic presentation or documented instability on examination to the newborn [26].

Several sonographic techniques have been described, being the one described by Graf the most commonly used. It evaluates the morphology of the hip through two obtained angles, the alpha angle and the beta angle. It is a static and quantitative evaluation method [27]. In turn, Terjesen [28] proposed a dynamic evaluation method, in which it evaluates functional instability and percentage of the covered femoral head, quantitatively and qualitatively. Both methods present reliable results, with no data supporting one method to the detriment of the other [29] (Figure 1, table 1).

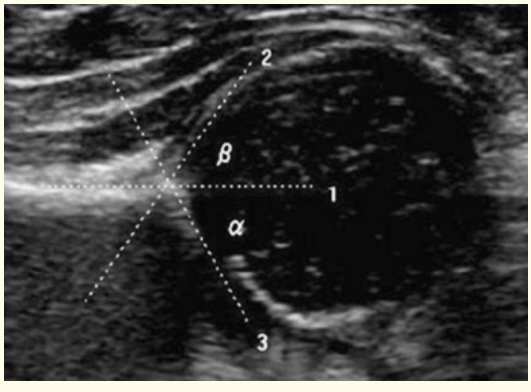

Figure 1: Ultrasound Alpha and Beta angles.

Grade	Alpha angle	Beta angle	Alteration	Treatment
I	>60º	<55º	Normal	No treatment
II	43 to 59º	55º to 77º	Ossification delay	IIa-alfa 50º to 59º, <3m-No treatment. IIb-alfa 50º to 59º, >3m-Pavlik. IIc-alfa 43º to 49º - Pavlik.
III	<43º	>77º	Subluxation	Pavlik
IV	No measurement	No measurement	Dislocation	Pavlik

Table 1: Graf classification system, based on ultrasonographic angles of the hip.

Pelvic radiography is the main method of evaluation of the hip after the 4th to 6th month of life, being possible at this age to draw lines and angles that give estimates of important anatomical parameters. The anterior-posterior incidence is generally sufficient [30]. These parameters may suggest acetabular dysplasia, evaluated by acetabular index or by positional deviations of the femur head drawing Hilgenreiner and Perkins lines, incongruence in the normal relationship between acetabulum and femur head, observed by the Shenton line and insufficient acetabular coverings of the femur head, by Wiberg's center-lateral angle [31] (Figure 2).

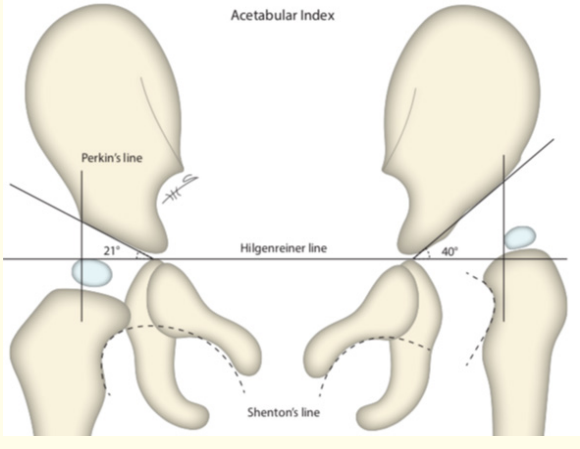

Figure 2: Basead on Pediatric Radiology (2019) 49:1652-1668.

Treatment

The success of the treatment of DDH depends on its early detection. When instituted in the early stages of the disease, it may present good results, however, in later stages and older ages, there may be permanent dysplastic changes and worse outcomes [33]. The potential for acetabular correction decreases exponentially after 3-4 years of age [33]. Thus, the therapeutic goal varies according to the age of diagnosis and consequent remodeling potential. In younger ages it is intended a stable articulation, with the femoral head concentrically reduced, for a normal development of the joint, without sequelae, while in older children, the changes are permanent, and so the goal to prevent or delay the OA of the hip and the need of THA [31].

The start of treatment in the baby is controversial, since about 80% of newborns with hip instability documented in the first life exam resolve spontaneously in the first 2 months of life [8]. A common approach is to immediately treat the dislocated hips at rest-positive Ortolani, and to maintain expectant attitude in the hips that are reduced but can dislocate with provocative maneuver-Barlow positive. If it remains unstable at 3 weeks, then treatment must be started [34]. A recent study concluded that in unstable hips, but without dislocation at rest in newborns, the beginning of treatment after 2 to 8 weeks of life decreased overtreatment, without significantly increasing the number of late dysplasia or surgical need [25].

Up to 6 months, the first line of treatment is immobilization in abduction, and there are several options for this, such as Pavlik or Von Rosen orthosis. There is no evidence in the literature to support any, with Pavlik's orthosis being the most popular, both in the luxated and sub-luxated hips [35]. It is a safe and effective method with success rates exceeding 90% [36]. This immobilization device keeps hips in flexion and abduction, and the joint remains reduced and stabilized. Hip abduction should not, however, be higher than 60º, as excessive abduction increases the risk of avascular hip necrosis (NAV) [37]. NAV is a rare complication with the correct use of orthosis, and it is always iatrogenic, not being associated with the DDH [37]. Another complication, also rare, is femoral nerve palsy, caused by excessive hip flexion.

This treatment should be closely monitored, and if after 2 to 4 weeks there is no improvement, on physical examination or ultrasound, should be discontinued, to avoid the risk that the lateralized femoral head causes a supracetabular erosion, and a more invasive treatment should be done [36]. If it presents a favorable evolution, it should be maintained until a stable and normalized joint is reached, and there is no duration defined in the literature [38]. Treatment is less effective at advanced ages, and it is not recommended after 6 months of age, in cases of high dysplasia of acetabular coverage, or in irreducible hips by inverted labrum. Bilaterality as a risk factor for failure is controversial [39].

When Pavlik's orthosis fails to achieve reduction or the child is older than 6 months, the hip should be reduced, closed if possible, in the operating room, followed by pelvipodalic cast immobilization for 2 to 3 months. The use of cutaneous traction previously to reduction, although in the past it has been very popular, it has been abandoned, because there is no evidence to reduce the risk of NAV and is difficult to be accepted by the child and family members [38].

Intraoperative arthrography is recommended to decide between closed or open reduction [33,40]. Arthrography has the advantage of allowing evaluation of the soft tissue and cartilaginous contours of the femoral head and acetabulum, and is especially use-

ful when the epiphysis of the proximal femur is not yet ossified. In fact, it allows to understand if a concentric and stable closed reduction of the hip is possible, or if an open reduction is necessary, in order to remove obstacles to the reduction or to perform a capsulorraphy to increase stability [33]. It is well known that the most important factor in determining the outcome of treatment with closed reduction of DDH is the quality of the initial reduction. Ahmed and colleagues demonstrated that hips reduced in a closed manner and confirmed with intraoperative fluoroscopy, and then perform arthrography, it showed that in about 20% of them the reduction was not concentric. In these cases, it was necessary to perform an open reduction by medial approach, in order to release the obstacles that avoid the concentric reduction of the joint [40]. There are several adaptive changes secondary to DDH that can block concentric reduction of the hip. These soft tissue obstacles can be identified through arthrography and include an inverted labrum, hypertrophied teres ligament, constriction of the capsule by the iliopsoas tendon, hypertrophy and shortening of the transverse acetabular ligament, and hypertrophy of the intra-articular fibroadipose tissue called pulvinar [40].

Once again, immobilization in extreme abduction can lead to NAV and should be avoided. Thus, immobilization should take into account a limited abduction with hip flexion at 90-100º. In cases where extreme abduction or internal rotation of more than 10 to 15º is required to maintain the reduction, the reduction is considered unstable, and therefore adductor tenotomy should be performed to increase the safety zone [40].

Magnetic resonance imaging is the gold standard to confirm the quality of reduction after closed reduction, as it does not present radiation, gives detailed information of the surrounding soft tissues and also allows the evaluation of the perfusion of the femoral head [41].

When closed reduction is not enough to achieve a concentric reduction, there is indication for open reduction. This is more common in older children, typically older than 18 months. Although rarely performed in children under 6 months of age, it should perform in all children in whom closed joint reduction is not possible [5].

Several approaches have been described. The impossibility to perform osteotomy or capsulorrhaphy during the medial approach, limits this approach to children under 18 months of age. Although minimally invasive, has sometimes been associated with NAV, due to injury of the vessels responsible for femoral head irrigation [42]. New arthroscopic techniques for release the medial structures have been described [43]. The anterior Smith-Petersen approach allows the reduction of the joint and concomitant capsulorrhaphy or pelvic osteotomy, and this approach is generally used in children older than 18 months. After open reduction, a period of 6 weeks should be followed with pelvopodalic immobilization in flexion, internal rotation and abduction at 30º [44].

The early treatment of DDA at young ages, even if well-succeeded, does not exclude the possibility of residual dysplasia in the future. Some series show that at 19% of the patients successfully treated by the Pavlik method and up to 33% by open reduction presented residual dysplasia [45]. These series support the need for patient follow-up until it reaches skeletal maturity.

In persistent acetabular dysplasia, the treatment is more invasive, undergoing a surgical correction with the aim of creating a joint that approaches normal anatomy, with a mechanically stable environment, in order to avoid early arthrosis in adulthood. Acetabular, femoral, osteotomy of both may be required [46].

Femoral osteotomy can be used to correct excessive femoral anteversion or femoral valgism, which, although controversial, has often been associated with DDH. The procedure shouldn't be performed before 3 years of age and it is recommended a prior pelvic x-ray with the hip in abduction and internal rotation to evaluate the potential correction of the joint congruence [46]. Excessive femoral anteversion has been pointed out as the main cause for recurrent subluxations [47]. Femoral derotation and varization osteotomy can then improve joint stability, increase the percentage of femoral head with acetabular coverage, and stimulate the remodeling of the dysplastic acetabulum to an anatomy closer to normal, especially in children under 4 years of age. [47] Trendelenburg gait is a common complication in the first 2 years after surgery, due to changes in the relationship of the abductor arm with the joint, however, with femoral growth it is spontaneously treated [46].

There is no consensus in the literature regarding the remodeling capacity of the acetabulum, the ideal age for a pelvic osteotomy, the most indicated type of osteotomy, the safety and long-term clinical and radiological outcomes of each osteotomy. In general, in children between 12 and 18 months of age who need an open reduction of the hip, pelvic osteotomies are rarely indicated, considering

the remodeling potential. Between 18 months and 4 years of age, the need for a pelvic osteotomy at the time of open reduction is controversial. Some authors advocate delaying the osteotomy and deciding the need for it based on serial radiographs to assess the quality of remodeling. However, other authors believe that performing pelvic osteotomy at the time of open reduction maximizes the likelihood of developing a normal acetabulum. In children older than 4 years, pelvic osteotomy is routinely performed, given the unpredictable remodeling potential. In children older than 8 years, the need for intervention is based on symptoms, severity of dysplasia, and laterality [48].

Within the acetabular procedures, several surgical techniques were described, divided into two groups: reconstruction osteotomies, that preserve acetabular cartilage, and salvation osteotomies, that do not preserve de articular cartilage [33].

In reconstruction osteotomies it is a pre-surgical requirement that hip is reduced concentrically, otherwise procedures are generally ineffective [33,46]. In this group are included reorientation osteotomies (Salter, triple osteotomy, Bernese periacetabular osteotomy (PAO)), that increase lateral and anterior coverage of the femoral head, by changing acetabular direction without change the size or shape of the acetabulum, and acetabuloplasty (Dega, San Diego, Pemberton), that change the morphology and offer a higher rate of correction of acetabular dysplasia in comparison with to the first ones [33,48].

Salter osteotomy is indicated in concentrically reduced hips in which there is a deficiency of anterolateral coverage, and can be performed between the ages of 18 months and 8 years, although the results are better when performed in children under 4 years of age, since it maintains greater flexibility of the pubic symphysis. The existence of a shallow acetabulum is a relative contraindication for this osteotomy [48]. In this procedure, a transverse osteotomy of the iliac bone is performed from the sciatic notch toward the region immediately above the anterior inferior iliac spine. Then the acetabulum is redirected to cover the anterolateral defect, with the symphysis pubis serving as a rotating hinge. This osteotomy is not stable, so it needs to be stabilized by bone grafting, usually a wedge taken from the iliac crest, and internal fixation with 2 or 3 K-wires [48,49].

Triple pelvic osteotomy is indicated in older children and adolescents with open triradial cartilage. This osteotomy overcomes the disadvantages of Salter osteotomy, as the acetabular fragment has more freedom of movement and is not dependent on the flexibility of the pubic symphysis [48,49]. This procedure is suitable for the treatment of complex hip dysplasia associated with congenital, neuromuscular and teratological conditions [49]. Triple pelvic osteotomy is unstable, since it violates the posterior column and creates pelvic discontinuity, and therefore requires rigid fixation and cast immobilization in the postoperative period [48,49]. There are several modifications of the original technique described, which had the main objective of compromising less the pelvic stability and improving the mobility of the acetabular fragment [48,49].

PAO is indicated in skeletally mature adolescents and adults with symptomatic acetabular dysplasia. This procedure combines a complete osteotomy of the pubis, an incomplete osteotomy of the ischium and a biplanar osteotomy of the ilium, allowing considerable correction of the acetabular version and of the anterior and lateral coverage [48,49]. The continuity of the posterior column is maintained with this osteotomy, which contributes to its stability, avoiding the need for post-operative immobilization. Another advantage of this procedure is that it preserves the shape of the pelvis, allowing for a normal vaginal delivery in the future [48].

Reshaping osteotomies of the acetabulum are incomplete osteotomies of the innominate bone, from which the acetabular roof is bent to increase coverage anteriorly and laterally. They are intrinsically stable, so they do not require fixation [48].

Pemberton osteotomy is indicated for patients with congenital dislocation or subluxation of the hip, aged 1 to 14 years, since in this case the rotation movement occurs at the level of the triradiate cartilage (in older children there is also some movement in the sacroiliac joint and pubic symphysis) [48,49]. A bicortical and curvilinear osteotomy is performed, starting between the anterior iliac spines and ending at the posterior column above the posterior branch of the triradiate cartilage, parallel and approximately 1 cm proximal to the joint capsule. The osteotomy opening is maintained with a bicortical graft [48,49].

The Dega osteotomy, like the Pemberton osteotomy, is indicated in children before skeletal maturity, with open triradial cartilage, where most of the rotation movement will occur. However, in ages below 4 years it is not recommended, as other osteotomies may be more effective [49]. In this procedure the osteotomy begins anteriorly, above the anterior inferior iliac spine, toward the

sciatic notch, ending about 1-1.5 cm anterior to it. The osteotomy is bicortical in its anterior portion, however a variable amount of posteromedial cortical of the inner table is left intact [48,49]. After opening the osteotomy, the gap is filled with bicortical bone graft, whose size and positioning, more anterior or posterior, will depend on the type of coverage needed in each case, which makes this osteotomy quite versatile [48,49].

The San Diego osteotomy is a modification of the Dega osteotomy, originally described for the treatment of acetabular dysplasia with essentially posterolateral coverage defects, although nowadays its indication has expanded, and it is also recommended in the treatment of idiopathic DDH with anterior and anterolateral coverage defects [48,49]. The main difference between these two acetabuloplasties is that in the San Diego procedure, the inner cortex of the ilium is kept intact, with the exception of its most anterior portion (above the anterior inferior iliac spine) and posterior portion (at the sciatic notch), where the osteotomy is bicortical [48,49]. By changing the positioning and size of the bicortical wedges of the bone graft, that fill the osteotomy opening, the surgeon can alter the shape of the acetabulum to specifically treat the type of dysplasia in each case [48].

Salvage osteotomies are indicated in cases of severe, symptomatic, and irreducible acetabular dysplasia, i.e., in which it is not possible to obtain a congruent reduction between femoral head and acetabulum [46]. These procedures increase the load surface by decreasing de overpressure and do not preserve the articular cartilage as the contact surface between the femoral head and the acetabulum. However, the articular capsule interposed between the femoral head and the ilium (in the case of Chiari osteotomy), or bone graft (in shelf osteotomy) suffers a cartilaginous metaplasia resembling articular cartilage [33].

Either the Chiari osteotomy (medial displacement of the cancellous bone of the ilium) or the shelf osteotomy (iliac graft placed in the superior margin of the acetabulum) can be used to improve the coverage of the femoral head and increase hip stability in cases where degenerative changes of the hip are already present and reorientation osteotomies are no longer recommended [49].

Pelvic osteotomies, however, do not completely prevent the development of early OA, with studies pointing to the need for THA in 23% of patients who have been treated with Salter osteotomy, and 38% of those treated with PAO [50].

Follow-up

It is consensual that children with a history of AD should be followed until they reach skeletal maturity, because, even achieved a successful treatment, it is still common to have residual acetabular dysplasia [45]. Clinical evaluation with radiography at 6 and 12 months and clinical at 2, 5 and 12 years is therefore recommended [34]. However, radiographic monitorization, due to inherent radiation, is more controversial. A recent study proposed that a normal pelvis face x-ray at 2 years of age does not require radiographic follow-up until skeletal maturity is reached [32].

Conclusion

DDH is the main cause of THA in young adults. Its early diagnosis and treatment are essential for a good final result. All newborns should be evaluated, with a careful and specific physical examination, for possible hip instability, and suspected cases closely monitored. Pelvic presentation at birth and positive family history should be risk factors to be taken into account, increasing clinical suspicion. Ultrasonography is the technique of choice to evaluate DDH up to 6 months of age, and pelvic radiography is the preferred complementary examination from 6 months to skeletal maturity. Pavlik's orthosis is a safe and successful treatment in most cases, and the treatment of choice in early cases, reserving more invasive and surgical procedures for refractory or later ages cases.

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