Evolution of the term and definition of dysplasia of the hip – a review of the literature

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Abstract

There is no consensus on the definition of dysplasia of the hip (DH). Past and present concepts used to describe DH do not form a complete view of the pathology. Moreover, some authors still present the disease as congenital, not developmental. This prompted authors to analyze the evolution of the definition of DH. Based on the biomedical databases 500 articles and books in the field of hip dysplasia were found and analyzed. Fifteen definitions of hip dysplasia met inclusion criteria, subsequently were analyzed and presented in chronological order. The analysis revealed that currently there is no single, universal definition of hip dysplasia in the aspect of morphological, clinical, and radiological studies. Despite the widely-used term of DH, it is described imprecisely and in different ways. Therefore, it is necessary to develop a multidisciplinary definition of this pathology covering all aspects of hip disorders considered valid in modern orthopaedics.

Key words: hip dysplasia, developmental dysplasia of the hip, congenital hip dislocation, acetabular dysplasia.

Introduction

The definition of dysplasia of the hip (DH) is not universally agreed upon [1–30]. Usually, the term dysplasia is used with reference to infants born with dislocation or instability of the hip, which in the residual form are defined as hip dysplasia. A broader definition of DH mentions the abnormal growth of the hip. The abnormal development of the joint refers not only to the osseous structures, but also other tissues (including soft tissues) forming the structure of the hip. In the literature there can be found more precise terms of DH, which are intended for detailed description of this pathology, such as subluxation (incomplete dislocation, incomplete contact between articular surfaces of the acetabulum and the femoral head), dislocation (loss of contact between the articular surfaces of the acetabulum and the femoral head) or hip instability (a possibility to cause subluxation or dislocation of the hip by using passive balancing force).

These terms frequently used in clinical practice and literature reports do not, however, form a complete picture of the problem which hip dysplasia creates. Moreover, some authors still treat the pathology as a congenital, rather than a developmental disorder [2–6, 19, 20]. The fact that hip dysplasia is a heterogeneous phenomenon prompted the authors of

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Bartosz Musielak 58a/6 Poznańska St Phone: +48 600 302 158 60-853 Poznan, Poland E-mail: ortopediadziecieca@ gmail.com this paper to analyze the evolution of its definition over the centuries.

The aim of the study was to analyze changes in definitions of hip dysplasia and their evolution based on a review of international and local literature.

Material and methods

Based on the databases of Google Scholar, PubMed, Cochrane and the authors' own sources, 485 articles and books in the field of hip dysplasia were found and analyzed. Qualified for the study were only those research papers that met the basic inclusion criterium, which was the presence of a definition of hip dysplasia. Rejected were those publications that described the characteristic features of dysplastic joints but did not contain a definition. Based on these criterium, 15 definitions were accepted and then arranged in chronological order.

Results

The first descriptions of congenital hip dislocation appeared as early as in the time of Hippocrates (400-300 BC). Hippocrates claimed that dislocation of the hip in children developing in the womb [1, 2] may have occured as a result of a fall, stroke, or compression [1, 2]. In later years, authors did not report any interest in the subject of defining DH. In modern times the first who presented the case of hip dysplasia and gave it a name was Palletta [30] and Dupuytren [3, 4]. In 1832 Dupuytren described a dislocation of the hip at birth and called it "primary or congenital". He wrote: "There is a species of displacement of the upper extremity of the femur, of which I have not found any mention in authors [...]. This displacement consists in a transposition of the head of the femur, from the cotyloid cavity on to the external iliac fossa (dorsum) of the ilium, a transpostion which exists at birth, and which appears due to a defect in the depth or completeness of the acetabulum, rather than to an accident or disease [...], which I shall name [the disease - ed.] original or congenital dislocation". In the further part of the description of the case Dupuytren mentions characteristic symptoms (mostly the same which are nowadays observed in patients with dysplasia of the hip) [3, 4] (Table I).

One of the first to describe hip dysplasia was also Phelps [2, 5]. In 1891 in "The Journal of Bone and Joint Surgery" [5] Phelps described a case of hip dislocation occurring in a patient and tried to identify its causes. He wrote:

• "The acetabulum is seen to be angular in shape, small, and undeveloped, with the remains of the ligamentum teres".

- "Congenital dislocation of the hip is produced, I believe, by injury at birth; injury *in utero*, or disease *in utero*; rhachitis; hereditary influences, and, in exceptional cases, if such there are, by arrest of development of the acetabulum".
- "I am inclined to believe [...] that the deformity is more frequently produced by violence of some description, or by pathological changes being a result of inflammation, than from any other cause".

Another definition of hip dysplasia can be found in the collections of "California State Journal of Medicine" [6]. Pahl, describing cases of congenital dislocation of the hip, proposed one of the first, yet simple, definitions of dysplasia, describing it as a condition in which the femoral head is not located in the hip socket after birth. He based this definition on the features which he observed during his study.

The debate on the term "hip dysplasia" can also be found in the orthopaedic world. Lorenz, Austrian orthopaedician, who had tremendous influence on the development of diagnosis and treatment of hip dysplasia in infants in the end of the XIXth century, implemented the term "socalled congenital hip dislocation" in 1920 [30-32]. Ortolani [7, 8] in his article from 1937 suggested using the term congenital hip dysplasia instead of congenital dislocation because, according to him, not all of the improperly formed hip joints are a result of congenital dislocation. On the other hand, Howorth [9] in the sixties of the last century thought that the word displacement is accurate and convenient as a group term for both subluxations and dislocations, whereas the word congenital is appropriate only when it is known that displacement was present at birth. For displacements, especially subluxations, recognized some time after birth, more suitable is the term infantile displacement [9].

In Poland one of the first and most important researchers of hip dysplasia was Dega. His first research on the subject was published already in the twenties of the XXth century. Nevertheless, it is difficult to find in his output a clear definition of this pathology [10]. However, on the basis of his own observations the "father of Polish orthopaedics" (how Dega was called) described some features of hip dysplasia, which he published in 1968 in a textbook "Orthopaedics and Rehabilitation" [10]. Dega wrote: "As soon as a child begins to walk, the shallow and steep acetabulum cannot keep the femoral head in its place. Depending on the degree of dysplasia the head ejects slowly and gradually from the acetabulum [...]. The shape of the acetabulum becomes oval and takes on the appearance of a Gothic pointed arch. Its upper rim disappears. The bottom of the acetabulum

Characteristics of hip dysplasia in particular definitions					
Study	Year of publication	DH as a congenital or developmental disease	Congruency of the hip joint	Acetabular dysplasia or changes	Other features
Dupuytren	1832	Congenital	Dislocation	Present	_
Phelps	1891	Congenital	Dislocation	Present	Cause of dysplasia: injury at birth, injury in utero, disease in utero, rhachitis, others
Pahl	1905	Congenital	Not included	Absent	-
Epps, Bowen	1995	Developmental	Subluxation, dislocation	Absent	Intracapsular displacement
Sollazzo <i>et al</i> .	2000	Congenital	Instability, subluxation, dislocation	Absent	Lax hip capsule
American Academy of Pediatrics	2000	Developmental	Not included	Absent	-
Czubak	2000	Developmental	Congruent, no dislocation	Present	Shallow acetabulum
Timmler <i>et al</i> .	2003	Developmental	Not included	Present	Malfunctioning biomechanical forces, disorder of the muscles surrounding the hip joint
Bowen, Kotzias-Neto	2006	Developmental	Not included	Present	_
Canale, Beaty	2007	Congenital	Congruent, subluxation, dislocation	Present	-
Hefti	2007	Developmental	Not included	Present	Impaired ossification of the lateral acetabular epiphysis
Herring	2008	Developmental	Instability, subluxation, dislocation	Present	Lax hip capsule
Sionek <i>et al</i> .	2008	Developmental	Predispo- sition to dislocation	Absent	-
Dormans	2009	Developmental	Not included	Present	Radiological definition
Shi et al.	2012	Developmental	Not included	Present	Lax hip capsule, secondary deformity of the femur

 Table I. Hip dysplasia definition overview

thickens. The acetabulum becomes shallow. The crushed articular labrum curles either towards the acetabulum, or outside. Adipose tissue accumulates in the bottom of the acetabulum" [10]. What is important, Dega introduced a radiological classification of dysplasia, in which he divided the defect into four basic categories: dysplasia (without dislocation), subluxation, supraacetabular dislocation, and total hip dislocation.

The definition of dysplasia on the basis of the above mentioned observations of Dega as well as on his own was formulated by Czubak [11, 12] who described it as a condition in which there are symptoms of abnormal development of the hip joint, but without femoral head dislocation in relation to the acetabulum, which is shallower than normal.

Undoubtedly, a breakthrough in the diagnosis of hip dysplasia occurred in the eighties of the twentieth century. In 1984 Graf [13], an Austrian orthopaedic surgeon, introduced an ultrasound classification of dysplastic changes, and began to promote this method in the early diagnosis of DH. The method gained popular support shortly after its dissemination. This event had considerable impact on the further development of views on the definition and naming of hip dysplasia.

By the end of the nineties, a Yugoslav orthopaedician Klisic took part in the ongoing discussion about hip dysplasia [14]. He proposed a new definition of DH, which was accepted by all the major orthopaedic societies. He also introduced the term developmental dysplasia of the hip [14], writing: "The modern term "Developmental Displacement of the Hip" (DDH) is much better [than the previous one – ed.]. It realistically indicates a dynamic disorder as the baby develops, of getting better or getting worse". Further in his report in "The Journal of Bone and Joint Surgery" Klisic described the concept of developmental dislocation combining the earlier thesis of Michele (from 1962) and Howorth (from 1963) as universal, including all variations of the disorder (dislocation, subluxation, dysplasia), regardless of whether it developed in the antenatal or postnatal period [14]. The aim of the change of the name was to include in the definition all the possible forms of deformity of the hip joint, not only developed in the prenatal period, but also after birth [15].

Based on progress in the diagnostic methods and treatment as well as on progress in the understanding of the pathophysiology of hip dysplasia, after 1990 numerous modern definitions of the disease, with various origins (etiological, biomechanical) were formed. Nowadays, development of the molecular methods of evaluation of processes taking place in the bony structures [33] gives an opportunity to define hip dysplasia at the cellular level. Unfortunately, they lack a common strategy of defining the dysplastic deformities of the hip joint.

In the textbook "Tachdjian's Pediatric Orthopaedics" [16], which is a kind of "bible" of the contemporary generation of pediatric orthopaedic surgeons, Herring in DH definition emphasized that dysplasia comprises the whole spectrum of developmental disorders of the hip that present in different forms at different ages with common etiology which is excessive laxity of the hip capsule, with failure to maintain the femoral head within the acetabulum. Moreover Herring wrote in his definiton that "the syndrome in the newborn consists of instability of the hip, such that the femoral head can be displaced partially (subluxated) or fully (dislocated) from the acetabulum by an examiner. The hip may also rest in a dislocated position and be reducible on examination. Over time, the femoral head becomes fully dislocated and cannot be reduced by changing the position of the hip" [16].

In 1995 Bowen [17] identified DH as a spectrum of intracapsular displacement of the femoral head from its normal relation in the acetabulum, leading to interruption in the development of the hip before, during or just after birth. In later years, Bowen, along with Kotzias-Neto, wrote in the book "Developmental Dysplasia of the Hip" [18] (2006) that the term hip dysplasia refers to inadequate development of the hip joint, including the acetabulum, femoral head, or both at the same time.

In 2000, Sollazzo *et al.* created their own definition of DH [19]. It said that congenital hip dysplasia is a condition in which the physiological seating of the femoral head in the acetabulum is impaired. It manifests itself in several ways, ranging from mild instability of the femoral head with slight capsular laxity, permitting minimal lateral displacement, through moderate lateral displacement of the femoral head, without loss of contact of the head with the acetabulum, up to complete dislocation of the femoral head from the acetabulum [19].

In turn, in the textbook "Campbell's Operative Orthopaedics" from 2007, edited by Canale and Beaty [20], there is a definition according to which congenital hip dysplasia involves subluxation (partial dislocation) of the femoral head, acetabular dysplasia, and complete dislocation (displacement) of the femoral head from the true, original acetabulum.

In the same year, Hefti [21] described hip dysplasia as inadequate development of the hip joint with impaired ossification of the lateral acetabular epiphysis.

Many orthopaedic surgeons dealing with patients with DH tried to describe (usually on the basis of autopsy or radiological examinations) the features of dysplastic acetabulum [7, 10-12, 15, 16, 18, 20, 22]. Dormans [22] is an example of this. Not only did he make the definition of the hip joint, which he described in 2009 as a whole set of disorders of the hip during its development occurring in different forms and at different ages, but he also described the features of the dysplastic hip joint: thickening of the adipose tissue (pulvinar) inside the acetabulum; teres ligament hypertrophy, sometimes occupying the entire space of the acetabulum; transverse ligament hypertrophy, which may reduce repositioning; hourglass shape of the lower part of the joint capsule; a more oblique acetabular roof; a shallow shape of the acetabulum, with thickened medial wall [22].

In the American Pediatric Society there is the current definition of DH [23] according to which it is a condition in which the femoral head has an abnormal relationship to the acetabulum.

In Poland, apart from the definition of Dega and Czubak [11], another can be found created by Timmler *et al.* in 2003 [24]. The Poznan researchers described hip dysplasia as a dynamically changing disorder of the joint, covering all its components: the acetabulum, the proximal end of the femur and the joint capsule, as well as the muscles surrounding the joint. The malfunctioning biomechanical forces cause the changes in the osteochondral components what leads to progressive deformity of the previously well-formed structures in the fetal period.

The problem of dysplasia of the hip joint is recognized in a slightly different way by successive Polish authors – Sionek *et al.* [25]. Their definition of the hip joint from 2008 says that DH is malformation of the hip in the period of intrauterine life, in the perinatal period, as well as in the first weeks after birth, which may lead to its dislocation.

Discussion

The widespread use of the term hip dysplasia and acetabular dysplasia led the authors of this paper to review the literature in order to get a detailed answer on the proper meaning of the term. As the above shows, the definition has undergone gradual evolution since the first description of hip dysplasia. First definitions [1-4] described it only as an impaired configuration of the joint caused by injury, or as a congenital dislocation of the hip. The first who drew attention to an extremely important developmental factor in the abnormal construction of the hip was probably Phelps [2, 5], who pointed out that one of the causes of dysplasia is inhibition of the development of the acetabulum [5]. The developmental aspect was also noted by Howorth [9] who differentiated developmental dislocation or subluxation of the hip (calling it "infant dislocation") from congenital dislocation. Further definitions of hip dysplasia also put clear emphasis on the aspect of development, largely determining the final architecture of the hip [14-18]. Modern terminology of hip dysplasia, starting from Klisic [14], is based on the assumptions of development, including the development of phylogenetic bipedal gait. Contemporary definitions also describe in detail the morphology of the dysplastic hip joint showing the particular components of the joint whose construction is defective. It should be noted that none of the listed definitions mention any genetic factors affecting the development of hip dysplasia.

Many orthopaedicians carried out research on hip dysplasia, which had an influence on the dynamic development of what is known about the pathomorphology, diagnosis and treatment of this condition. They did not, however, take any attempts to create their own definition of DH, according to our knowledge. It is worth mentioning

Despite the widely-used term of Hip Dysplasia, it is described imprecisely. Therefore, it is necessary to develop a multidisciplinary definition of this pathology covering all aspects of hip disorders and considered valid in modern orthopedics.

such researchers as: Barlow [26] - who devised a basic test used during examination of newborn babies for hip dysplasia; von Rosen [27] – inventor of the splint for the treatment of children with dysplasia; Browne [28, 29] – father of pediatric surgery in the UK, inventor of the splint named after him and used in the treatment of clubfeet; Zahradníček – a Czech professor who lived and worked in the first half of the twentieth century, mentor of the Czech pediatric orthopaedics belonging to the group of world-class surgeons, and known as the creator of one of the corrective osteotomies of the proximal end of the femur. Despite the enormous contribution to the development of knowledge on dysplasia (e.g. in his publications Browne described the influence of limb positioning in the uterus on the development of hip dislocation, and the co-occurrence of other, particular defects, such as talipes valgus), none of the above-mentioned researchers attempted to define this pathology. In their publications [26–29] they usually called the disorder congenital dislocation of the hip.

In this context it is puzzling why the researchers dealing with hip dysplasia on a daily basis proposed no definition of this pathology.

All the analyzed definitions of hip dysplasia refer to the hip joint in the stage of development, but there are no definitions of the deformity of the hip after completion of its growth. It seems that the morphological and clinical picture of hip dysplasia should be then supplemented by the secondary changes such as loss of articular cartilage, subchondral sclerosis of the acetabulum or thickening of its bony roof [34]. Another aspect relating to the hip joint after completion of its growth is frequently observed, so-called, secondary' hip dysplasia, resulting in the thickening of the acetabular roof in the course of development of osteoarthritis, which leads to the dislocation of the femoral head to the side, and ultimately upwards. Such a dislocation is one of the features of hip dysplasia.

In conclusion, in the light of the above discussion it is clear that there is currently no single, universal definition of the DH in the aspect of morphological, clinical and radiological studies. Despite the widely-used term of hip dysplasia, it is described imprecisely and not unanimously (Figure 1). It seems that it is necessary to develop a multidisciplinary definition of hip dysplasia covering all aspects of the hip joint disorders and considered as valid in modern orthopedics. This will enable establishing a confident diagnosis of the disease, choosing the best course of treatment and determining its outcome.

Conflict of interest

The authors declare no conflict of interest.

Figure 1. Hypothesis of the study

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