

Definition of Developmental Dysplasia of the Hip: Literature Review

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Abstract

Existing definitions of developmental dysplasia of the hip (DDH) lack uniformity. This study reviewed the main articles discussing DDH definition to make a uniform, feasible, and clinically relevant definition. Electronic databases with some orthopedic books were used to search for the relevant articles. The articles included were 26 (13 from journals and 13 from books). They were published in the last three decades, in the English language, and related to the human species. The retrieved descriptive data used to define DDH were summarized and sorted according to the concepts they contained (they were 12 concepts). Most of them gave more than one set of data to define DDH. From the summary of these data, it can be concluded that DDH can be defined as a developmental spectrum of abnormal formation and alignment of hip joint components that present in healthy infants.

Keywords: Definition, developmental dysplasia of the hip, systematic review

INTRODUCTION

Definition, in general, is an explanation of the meaning of a word or phrase.^[1] In the medical field, the definition of a term is a statement expressing the essential nature of something.^[2]

Developmental dysplasia of the hip (DDH) is considered an uncommon medical problem; however, in the orthopedics field, it is a common one and forms a large portion of pediatric orthopedic practice. This condition, i.e. DDH, refers to a wide spectrum of pathologic conditions, ranging from a subtle acetabular dysplasia to an irreducible hip dislocation.^[3,4]

The term is defined simply as an abnormality of the hip joint where the hip joint has not formed normally in babies and young children, the socket portion (acetabulum) is shallow and does not fully cover the ball portion, and the ball is loose in the socket and results in an increased risk for joint dislocation. It may occur at birth or develops during a child's 1st year of life.^[5] This definition is present in public health websites.

The precise definition of hip dysplasia is controversial, and it encompasses a spectrum of abnormalities, rather than a single condition.^[6] Existing definitions of DDH lack uniformity. The literature has no clear, widely accepted definition of the condition. Therefore, it is necessary to develop a

multidisciplinary definition of this pathology covering all aspects of hip disorders and is considered valid in modern orthopedics.^[7]

Objectives

This study aimed to perform a systematic review of the articles discussing DDH, to map evidence on the main concepts of the condition's definition, and to come up with a suggestion for a feasible and clinically relevant definition.

METHODS

Study design, protocol, and registration

The study was done according to the Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) checklist.^[8,9] A review protocol was prepared before conducting the study and starting the search process [Supplementary File 1]. This protocol was not registered in the PROSPERO site for protocol registration because this

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site does not currently accept registrations for scoping reviews, literature reviews, or mapping reviews. It was registered prospectively before starting the review process with the Open Science Framework on June 19, 2020 (<https://osf.io/fe96b>).

Information sources

Electronic databases such as PubMed, Europe PMC, Cochrane Library, IMEMR, and Google Scholar were used to search for relevant articles. The keywords or phrases in the search process were selected according to the Medical Subject Headings (MeSH), and they were: developmental dysplasia of the hip, developmental dislocation of the hip, congenital dysplasia of the hip, congenital dislocation of the hip, and hip dysplasia with the term definition.

In addition to the electronic database search, 13 books were also involved in the search process. These books were all of the orthopedics, and some of them were a specialty in pediatric orthopedics.^[10-22] Only the definition and the descriptive data of DDH were taken from these books.

Eligibility criteria

Inclusion criteria were journaled articles discussing general aspects of DDH (e.g., review, update, or current concepts) or book chapters discussing the definition or description of the condition. The articles included were those published in the last three decades (i.e., between 1990 and 2020) and related to the human species. The search process was restricted to those articles published in the English language only.

Exclusion criteria were grey literature, studies that discuss a specific subject other than definition (e.g., risk factors, screening, diagnostic or therapeutic method, genetics), and case reports because these articles described DDH in their introduction and usually had been taken from other references.

Search strategy

To identify potentially relevant documents, the search process was done from June 20, 2020, to June 25, 2020. The search was done by the author. The Peer Review of Electronic Search Strategies checklist was taken into consideration during the search process.^[23] An example of the search strategy in PubMed is found in Supplementary File 2.

Data extraction and charting

The variables extracted from the articles were collected, sorted, and charted using Microsoft Office Word software sheet (Microsoft Corporation, Microsoft Office Word, 2019. Redmond, Washington, USA). A primary chart [Supplementary File 3] for journal articles was designed and contained the following fields of data:

- First author name
- Title of the article
- Publication date
- Type of the article
- Source of the article
- Descriptive data for DDH.

The extracted variables from the books were arranged in another primary chart [Supplementary File 4]. It contained the following fields:

- First editor name
- Book name (with edition)
- Publication details (date, publisher, and place)
- Chapter (with pages numbers) include descriptive data
- Descriptive data for DDH.

Data items further charting and result synthesis

The descriptive data (the aim of this review) were those phrases and statements that had a relation with the definition, description, and clarification of the condition. They have been reviewed, summarized, and sorted into a single secondary chart for both journals and books [Supplementary File 5]. The text of each article was sorted and bulleted according to the concepts it contained (author conclusion). Each concept was considered a sort or type of data. For each type, a short title was made and listed in a special column in this secondary chart.

The short titles of data concepts (types of data) were arranged in a tertiary chart [Supplementary File 6]. The summarized data of the articles were included in this chart to know how each type of data (concept) was used by different articles.

According to the scoping review methodology, an assessment of the quality (risk of bias) of the included studies was not performed.^[8,9]

RESULTS

Selection of sources of evidence

To start with, the search process for the electronic database by using the developmental dysplasia of the hip as a MeSH with the term definition yielded a single article only. A modification was made by excluding the term definition from the search process. Then, a total of 8910 articles were identified from the preliminary (first look) search; 8897 were from electronic databases and 13 from books. The electronic databases articles were as follows: 3986 from PubMed, 2886 from Europe PMC, 1890 from Google Scholar, 85 from Cochrane Library, and 50 from IMEMR.

Based on the title, 164 articles were preliminary eligible. The rest 8746 articles were excluded according to the specific filters and advanced search builders used during the search procedure. Cochrane Library and IMEMR websites had no articles eligible for the present study.

Sixty of the eligible articles were considered duplicates, and they were also excluded. Thirty-two of them had more than one version, and the other 28 articles were found in more than one database source. The author gave priority to those articles found in PubMed and books.

The remaining 104 articles underwent further filtration by checking their abstracts (second look). Sixty-four of them

were further excluded because they were not relevant to the query of this article. The remaining 40 articles were found to be relevant.

The full text of these articles was retrieved and underwent a further precise look (third look). The descriptive data related to DDH of 14 of them were found to be a complete replication of data from other references. They were also excluded. The remaining 26 articles were found sufficient for final eligibility, and their data were considered as sources of evidence for this review [Figure 1]. Thirteen of them were from journals (9 from PubMed,^[24-32] 3 from Europe PMC,^[3,33,34] and 1 from Google Scholar^[35]) and the remaining 13 were from books.

Characteristics of sources of evidence

The sources of evidence were arranged chronologically for both journals' and books' articles [Supplementary File 6]. Four of them were published between 1990 and 1999; three journal articles and one book article. Between 2000 and 2009, further, four articles (three from journals and one from a book) were found. Eighteen articles (7 from journals and 11 from books) were found to be published between 2010 and 2020.

Half of the articles (13 out of 26) were from journals: ten were in the form of review articles, one was a multicenter study, one was guidelines (from the American Academy of Pediatrics), and the other one was Editorial discussing a comparative study (congenital dysplasia of the hip [CDH] or DDH). The other half were books' articles, eight were in the specialty of pediatric orthopedics, and the rest four were of general orthopedics.

Characteristics of the data

Eleven (42.3%) sources of evidence gave a special heading for the definition. They were seven journals (6 were review articles and one multicenter study) and four books (three books were in the pediatric orthopedic specialty and one of general orthopedics). The other sources (15 out of 26 = 57.7%) gave the descriptive data for the DDH in their introduction part.

The descriptive data for the DDH given by the articles were of different types (concepts). In summary, they were of 12 types [Table 1].

Most of the articles (23 out of 26) gave more than one set of data to describe the DDH. Only three articles gave a single

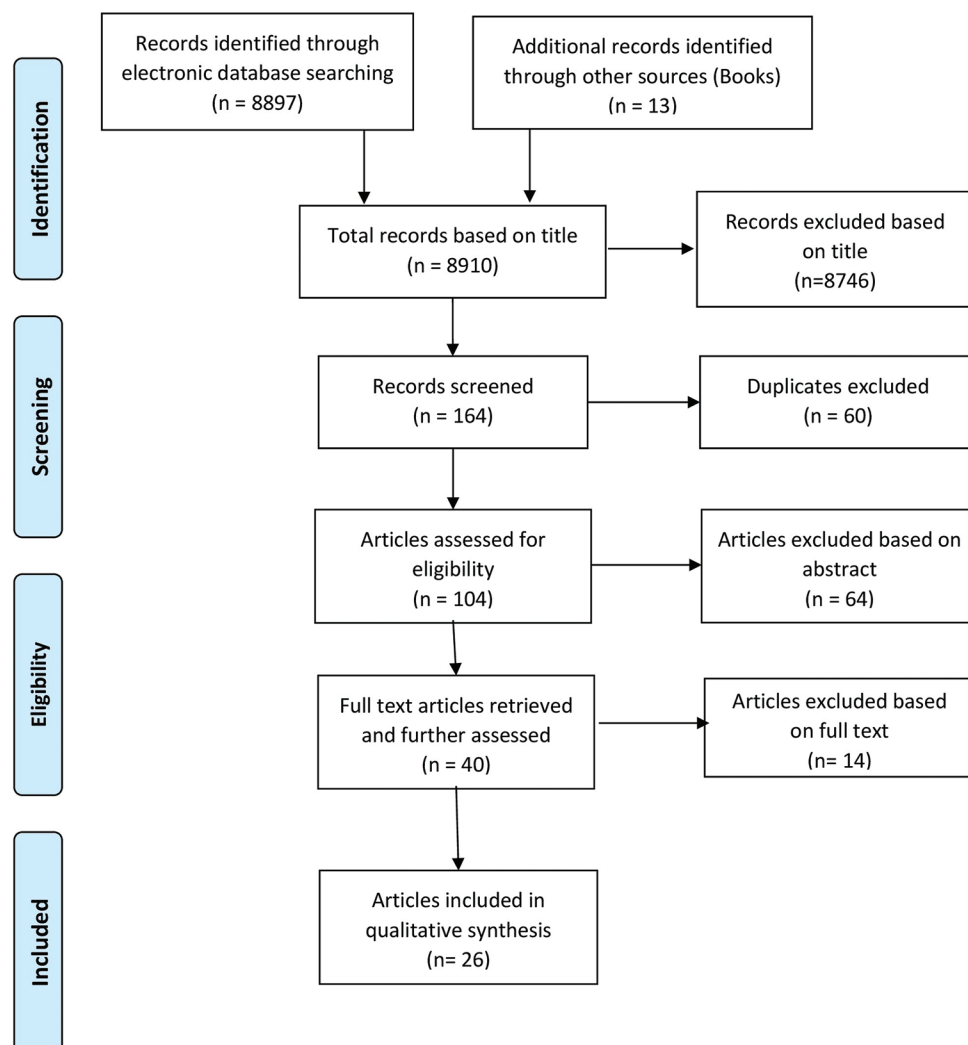


Figure 1: Flow diagram of the search strategy

Table 1: Frequency of types of data

Type of data	Frequency (%)
DDH term replaces CDH	16 (61.5)
Developmental not congenital	4 (15.4)
Dysplasia means	7 (26.9)
Spectrum	16 (61.5)
Relationship of femoral head to acetabulum	6 (23.1)
Forms of DDH	19 (73.1)
Typical DDH dislocation not teratogenic	12 (46.2)
Age of presentation	8 (30.8)
Untreated cases predispose to	5 (19.2)
Others	7
Period when abnormalities occur	3 (11.5)
Primary and secondary pathology	3 (11.5)
Variable at presentation	1 (3.8)

DDH: Developmental dysplasia of the hip, CDH: Congenital dysplasia of the hip

form of data (one journal article,^[28] one general orthopedics book,^[14] and one pediatric orthopedics specialty^[11]). One pediatric orthopedics book gave eight forms of descriptive data.^[20]

DISCUSSION

The term DDH was suggested by Klisic in 1989 to change the previous misleading term (CDH) because the condition proved not to be true congenital and it is of variable pathology, not always reaching a dislocation stage.^[36] Since that, most orthopedicians are satisfied with this new term. However, the words used by Klisic in his brief report for the new term DDH were Developmental *Displacement* of the Hip. He used the word *displacement* instate of *dislocation*. Now, most orthopedicians use the word dysplasia instate of displacement to indicate the DDH term. Musielak *et al.*^[7] in their article was not clear how the words used by Klisic (developmental displacement of the hip = DDH) were changed to “developmental dysplasia of the hip” (DDH).^[7] However, the term DDH with the words “developmental dysplasia of the hip” was adopted by the American Academy of Pediatrics and has been used in their guidelines published in 2000.^[27]

All the articles used as a source of evidence for this review used the words *Developmental Dysplasia of the Hip* apart from a single journal review article that used the word *Developmental Dislocation of the Hip*.^[25]

There were 12 types of data used in the articles to describe DDH [Table 1]. Most of the articles used more than one type. One of the objectives of this article was to map the main concepts and data types used to describe DDH to make a complete picture of the DDH description subject.

Developmental dysplasia of the hip replaces congenital dysplasia of the hip

Sixteen out of the 26 (61.5%) articles mentioned that DDH replaced CDH terms (11 journal articles and 5 books). Fourteen

of them gave explanations on why this change occurred. The explanations were mainly of two things (as mentioned by Klisic):

First, not all cases are detectable at birth, but some develop over the first few months of life.^[13] Hence, there may be a late diagnosis in patients previously normal clinically and/or radiologically.^[17,21,25,30] Second, the condition is a spectrum of abnormalities rather than an overt dislocation.^[10,25,29-31,33,35]

Yamamuro in his article gave another explanation. He said that DDH is not caused by a single gene but by multiple genetic factors.^[28] The multigenetic disease (such as osteoporosis and rheumatoid arthritis) is not called congenital. Its manifestations and progression are subjected to perinatal environmental factors.

Developmental, not congenital

Embryologically, the limb bud first appears at 4 weeks of gestation. The component of the hip joint – the femoral and acetabular – develops from the same primitive mesenchymal cells. Around the 7th–8th weeks of gestation, a cleft develops, defining the future femoral head and the acetabulum. The hip joint is fully formed by the 11th intrauterine week.^[22,24,27] As a result, the structures that make the hip joint are normal during embryogenesis and the hip is always located early in the embryological stage. Further growth and development come from the concentric location of the femoral head within the acetabulum. Hip dysplasia or dislocation may then occur *in utero*, prenatally, or subsequently with further development postnatally due to a variety of reasons, the chief being the fetal position and presentation at birth (e.g., malposition of the femoral head, abnormal forces acting on the developing hip) and the laxity of the ligamentous structures around the hip joint resulting in instability.^[18,21,24,26] Hence, in conclusion, the condition (i.e. DDH) is a developmental disorder.

Hip dysplasia

Seven (26.9%) articles gave special definitions for hip dysplasia as a whole. Four of them were general in their description (abnormality of development or formation of the hip joint),^[16,20,24,26] and the other three were more specific (abnormality in size, shape, and anatomical orientation of the acetabulum and/or the femoral head).^[29,33,35] For purpose of summarization and to make the sentence that will be used for definition as short as possible, it is wise to take the summary of these descriptions (i.e., abnormal formation of the hip joint).

Spectrum

Sixteen (61.5%) articles included in this review mentioned that DDH encompasses a wide range of hip problems^[24] or pathology,^[17] especially anatomical abnormalities and alterations affect growing hip,^[3,10,11,29,30,34,35] resulting in a spectrum of deformities and disorders^[13] in different forms and presentations,^[25,20] at different ages.^[21,32] Novacheck mentioned that it is a spectrum of both time and severity, and Gosselin *et al.* generalized it more that it is a spectrum of physical

and imaging findings.^[19,26] In summary, it is a spectrum of abnormalities whether anatomical, clinical, or radiological. Weinstein *et al.* in their article gave three forms of definitions for DDH (clinical definition, anatomical definition, and radiological definition).^[22]

Relationship of the femoral head to the acetabulum

Six (23.1%) articles in their description for DDH mentioned that the femoral head has an abnormal structural relationship with the acetabulum (i.e., abnormality of the hip joint congruency).^[20,27,30,31] This abnormal relationship may be in the form of partial or complete displacement.^[15,16] In summary, there is an abnormal relationship between the femoral head and the acetabulum.

Forms of developmental dysplasia of the hip

Most of the articles included in this review (19 out of 26 = 73.1%) gave the spectrum (i.e., the forms or types) of DDH. Thirteen articles gave four forms and five articles gave three forms. Only one article gave five forms of DDH.

Those who gave four forms had different terms:

1. Dysplasia (malformed), instability, subluxation, dislocation^[3,18,19,24,27,29,31,34,35]
2. Dysplastic, dislocatable, dislocated reducible, dislocated unreducible^[13,10]
3. Dysplasia, instability, dislocated reducible, dislocated unreducible^[21]
4. Unstable, subluxable, or dislocatable, subluxated, dislocated.^[20]

Those who gave three forms had the following terms:

1. Dislocated, located but unstable or dislocatable, dysplastic^[25]
2. Unstable, subluxation, dislocation^[33]
3. Dysplasia, subluxation, dislocation.^[14,22,15]

Nemeth *et al.* in their article gave five terms:^[30]

1. Dislocated, dislocatable, subluxed, subluxable, dysplastic.

Hence, in general, the terms used as forms of DDH in all articles appear to be confusing.^[18,22] This problem can be solved by re-checking and summarizing the definitions of the terms used for that.

Dysplasia (malformed)

Any abnormality (inadequate and delayed) in the development of the acetabulum (shallow shaped), femoral head, or the surrounding soft tissues (as ligament laxity) without displacement of the femoral head, present at birth that can usually be visualized radiographically but may or may not cause instability on clinical examination.^[11,19,20,22,24,25,29-31,35]

Instability (unstable joint)

The hip is in a located anatomical position at rest, but the tight fit between the femoral head and the acetabulum is lost, and the femoral head can move within (subluxable) or outside (dislocatable) the acetabular socket by an externally

applied force of a provocative maneuver (like Barlow's test) during clinical examination.^[24,25,27,29,31,33,35]

Subluxation

Partial displacement of the femoral head from its central anatomical location within the acetabulum to the margin of the hip joint. The articular surfaces are not in perfect contact with each other's. It can be detected during Ortolani and Barlow maneuvers as a soft clunk, and radiographically, there will be a disturbed Shenton line.^[18-22,24,29-31,33,35]

Subluxable

The femoral head is located in normal relation to the acetabulum at rest and certain position (flexion and abduction) but has partial (incomplete) loss of contact in other positions or if induced with provocative maneuvers.^[20,30]

Dislocatable

The hip rests in a located position at rest and becomes dislocated on clinical examination and provocative maneuvers.^[25,13]

Dislocation (luxation)

Complete loss of contact between the articular surfaces of the femoral head and the acetabulum. This dislocation may be reducible (dislocated reducible) or irreducible (dislocated irreducible) by provocative tests. The irreducibility may be due to teratogenic cause (not considered a type of DDH) or occurs in typical DDH dislocation later on in older children due to secondary changes in the joint.^[13,20,21,24,25,27,29,30,33,35]

Jackson *et al.*^[31] in their article mentioned another three terms used in DDH in addition to the forms (dislocation, dysplasia, instability, subluxation). They were (equivocal examination, hip click, hip clunk, limited abduction). However, these terms are clinical and used during the examination of the infant. In this review, they were not considered as forms of DDH.

From the proceeded discussion for the forms of DDH, the data can be summarized in the following manner:

Dysplasia is a malformation of the acetabulum, femoral head, or surrounding soft tissue that may not or may cause joint instability (subluxable or dislocatable).

Subluxation is a partial loss of contact between the articular surfaces of the femoral head and the acetabulum.

Dislocation is the complete loss of contact between the articular surfaces which can be reducible or irreducible.

From this summary, it can be concluded that the forms of DDH are three (dysplasia, subluxation, dislocation) [Figure 2].

Typical developmental dysplasia of the hip dislocation (not teratologic)

Authors of 12 of the articles (46.2%) included in this review stressed the differentiation between two types of dislocations that occur at birth and infancy: typical and teratologic. The typical dislocation of DDH is usually reducible in newborn (but become irreducible in older children due to secondary changes); hip joint has a good range of motion; presents in healthy

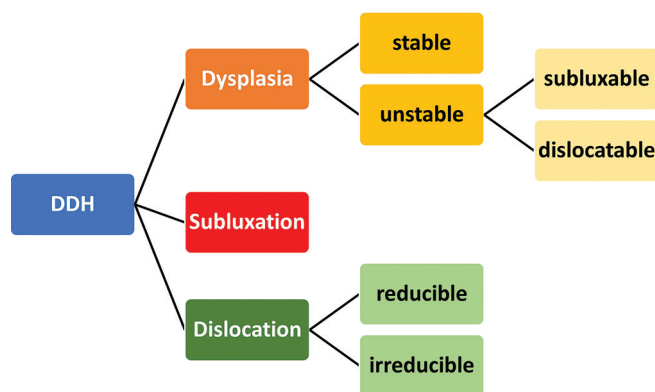


Figure 2: Forms of developmental dysplasia of the hip

infants; and may occur *in utero*, at birth, or after birth. While the teratologic dislocation of the hip is usually characterized by that it is usually severe (high grade), fixed (unreducible), have limited range of motion (stiff), occurs early in utero (prenatally) and is present at birth, and is associated with other malformations such as lumbosacral agenesis, neuromuscular disorders (like spina bifida) and joint contracture syndromes (like arthrogryposis).^[3,14,17,21,24,25,27,29,30,33,35]

Age of presentation

Eight articles (30.8%) determined the age of presentation of the patients with DDH. They all agree that DDH is a disease of infancy and early childhood period. It presents at birth and neonatal period whether diagnosed or not.^[13,15,20,24,30-32,35]

Untreated cases

The authors of five articles (19.2%) gave the natural history of the disorder. The sequel of untreated DDH may be in their lower back (hyperlordosis or degenerative scoliosis), ipsilateral or contralateral hips premature early-onset degenerative changes, and painful arthritis with limitation of hip movements, especially abduction), knees deformities and osteoarthritis, limb length inequality, and gait disturbance. This depends on the bilaterality, severity, and formation of pseudojoint (false acetabulum) between the femoral head and the iliac bone.^[12,19,22,29,34]

Others

Three articles (11.5%) mentioned that the pathological changes that occur in DDH are mainly in the acetabulum and are primary while those in the femoral head are secondary.^[20,22,33] However, this is a pathological note and not important in the subject of definition. Others mentioned that these pathological changes usually occur during infancy and early development.^[30,32,35] Novacheck mentioned in his description that DDH is variable at presentation. This can be included in the previous discussion (spectrum).^[26]

CONCLUSION

From the summary of these data, it can be concluded that DDH can be defined as developmental abnormal formation of the hip joint resulting in a spectrum of pathologies with an abnormal

relationship of femoral head and acetabulum (in the form of dysplasia, subluxation, or dislocation) that presents in healthy infants and presented at birth, infancy, or early childhood but leads to different sequel in adulthood if not been treated. In other words, and in a shorter form, it is a developmental spectrum of abnormal formation and alignment of hip joint components that presents in healthy infants.

Limitations

The study was done according to the PRISMA-ScR checklist.

However, because all requirements were not assumed, the author found it suitable to apply the term literature review in the state of systematic scoping review in the title of this study.

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