Supplementary material folder 1 (BJSM)

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**Figure 1** Primary cam morphology: a concept analysis process (adapted from Walker & Avant)

**Table 1** Definitions: ontology, taxonomy, theory, concept, primitive concept, concrete concept, abstract concept, and concept analysis

<table>
<thead>
<tr>
<th>Term</th>
<th>Definition</th>
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<tbody>
<tr>
<td>Ontology</td>
<td>“An ontology is an explicit specification of a conceptualization. The term is borrowed from philosophy, where an ontology is a systematic account of Existence.” For knowledge-based systems, what “exists” is exactly that which can be represented. [2]</td>
</tr>
</tbody>
</table>
| Taxonomy      | Taxonomy is an orderly classification (of conditions, diseases, living organisms, lists etc) into domains, categories and subcategories. [3,4] The Free Dictionary defines taxonomies as: 1. The classification and naming of organisms in an ordered system that is intended to indicate natural relationships, especially evolutionary relationships. 2. The science, laws, or principles of classification. 3. An ordered arrangement of groups or categories: a taxonomy of literary genres. [3] The Convention of Biological Diversity defines taxonomy as “the science of naming, describing and classifying organisms and includes plants, animals and microorganisms of the world. Using morphological, behavioural, genetic and biochemical observations, taxonomists identify, describe and arrange species into classifications, including those that are new to science. Taxonomy identifies and enumerates the components of biological diversity providing
basic knowledge underpinning management and implementation of the Convention on Biological Diversity. “[5]
“Disease taxonomy plays an important role in defining the diagnosis, treatment and mechanisms of human disease.” [6]

| Theory | “An internally consistent group of relational statements that presents a systematic view about a phenomenon and that is useful for description, explanation, prediction, and prescription or control.” (Page 8 in Walker and Avant) [1] |
| Concept | “A concept is a mental image of a phenomenon, an idea, or a construct in the mind about a thing or an action. It is not the thing or action, only the image of it” (Kaplan, 1964 – The Conduct of Inquiry). Page 63 in Walker and Avant. [1] |
| Primitive concept | “Concepts have different levels of abstractness (Reynolds PA A Primer in Theory Construction. 1971). Primitive concepts are those that have a common shared meaning among all individuals in a culture. For instance, a primitive concept like the colour blue cannot be defined other than by giving examples of blue and not blue.” [1] |
| Concrete concept | “Concrete concepts are those that can be defined by primitive concepts, are limited by time and space, and are observable.” [1] |
| Abstract concept | “Abstract concepts are also capable of being defined by primitive or concrete concepts, but they are independent of time and space (Reynolds, 1971). The concept of temperature, for instance, is abstract, whereas the concept of ‘temperature today in Kansas City’ is concrete because it is dependent on a specific time and place.” (Page 63 in Walker and Avant) [1] |
| Concept analysis | “The only way we will be able to demonstrate the evidence base for our practice is to be able to first describe the phenomena in measurable or at least communicable ways. Concept analysis allows the theorist, researcher, or clinician to come to grips with the various possibilities within the concept of interest—to ‘get inside’ the concept and see how it works. It is a challenging activity but provides an enormous insight into the phenomenon of interest.” (Page 167 in Walker and Avant) [1]
“Concept analysis clarifies the symbols (words or terms) used in communication. The main advantage of concept analysis is that it renders very precise theoretical as well as operational definitions based on the empirical referents for us in theory and research.” (Page 180 in Walker and Avant) [1] |
| Purposes of concept analysis are to: [7] | • distinguish between the defining attributes of a concept and its irrelevant attributes
• develop critical thinking through analysis and synthesis
• identify pertinent areas for research
• refine ambiguous concepts in theory
• help clarify overused, vague or abstract concepts
• develop a rigorous process for operationalising variables e.g. tool development
• develop critical thinking through analysis and synthesis |
| Empirical referent | “Empirical referents are classes or categories of actual phenomena that by their existence or presence demonstrate the occurrence of the concept itself. Kissing might be used as an empirical referent for the concept of ‘affection’. “
“Empirical referents are not tools to measure the concept. They are the means by which you can recognize or measure the defining characteristics or attributes. Thus, the empirical referents relate directly to the defining
attributes, not the entire concept itself.” (Page 179-180 in Walker and Avant)

[1]

Examples of concept analyses in the health care literature (table 2)

<table>
<thead>
<tr>
<th>Concept(s)</th>
<th>Author(s)</th>
<th>Journal</th>
<th>Year</th>
</tr>
</thead>
<tbody>
<tr>
<td>How is exercise different from physical activity? [9]</td>
<td>Dasso</td>
<td>Nurs Forum</td>
<td>2019</td>
</tr>
</tbody>
</table>
Primary cam morphology taxonomy as an important concept for femoroacetabular impingement syndrome (including figure 2A and 2B)

**Taxonomy for primary cam morphology**

![Taxonomy Diagram]

*Figure 2A* Proposed primary cam morphology taxonomy as an important concept for femoroacetabular impingement syndrome

![ICD-11 for Mortality and Morbidity Statistics](image)

*Figure 2B* Femoroacetabular impingement syndrome ICD-11 taxonomy: “Impingement syndrome of the hip”; FA34.5 [16]
Concept analysis step 5 to 7 (including table 3)

Step 5: Model case

We describe a model case of primary cam morphology in a 15-year old male football player (table 3) to inform the concept of primary cam morphology.

Table 3 Model and additional clinical cases to illustrate the concept of primary cam morphology

<table>
<thead>
<tr>
<th>Clinical case</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Model case:</strong></td>
<td>A 15-year-old male football player reports occasional left groin (anterior hip) stiffness associated with high-intensity practice or games for 1 month. He also reports a feeling of restricted ‘hip’ movement but cannot explain exactly what he feels. He is a striker who kicks with his right foot, and has played competitive football at his club’s a football academy since age 9. He denies any previous hip or groin injuries. Hip and groin examination findings are unremarkable apart from lower left hip internal rotation of 20° on the left compared to 35° on the right side. He agrees to further special investigations: anterior posterior pelvis (AP-pelvis) and lateral radiographs of both hips. The radiologist reports a left hip cam morphology visible on both AP-pelvis and lateral radiographs, measuring the alpha angles in both views (65 degrees and 58 degrees respectively). The player, his parents and the club’s sports physician are all keen to know more about the current joint status and agree to a 3 Tesla MR imaging of the player’s left hip. (The radiologist asked for two morphological sequences: three-dimensional (3D) water selective fluid (WATSf) to image joint cartilage and bone, and 3D proton density fat saturation (PDFS) to image the physeal scar. The radiologist also performed 3D multiplanar reconstructions and acquired radial images around the femoral neck at 30° intervals. [17]) The alpha angle (63 degrees), epiphyseal hypertrophy and extension are maximal at the 1 o’clock position</td>
</tr>
<tr>
<td><strong>Additional case – border line:</strong> Primary mixed morphology</td>
<td>A 19-year-old female rugby player reports occasional left anterior groin stiffness associated with high-intensity practice or games. She also reports a feeling of restricted ‘hip’ movement but cannot explain exactly what he feels. She is a fly-half and kicks with her right foot, and has played competitive rugby at her club’s rugby academy since age 9. She denies any previous significant hip or groin injuries. Hip and groin examination findings are unremarkable apart from slightly lower hip internal rotation of 25° on the left compared to 35° on the right side. She agrees to further special investigations: anterior posterior pelvis (AP-pelvis) and lateral radiographs of both hips. The radiologist reports left hip mixed cam and pincer morphologies visible on both AP-pelvis and lateral radiographs and measured the alpha angles in both views (67 degrees and 63 degrees respectively). The player, her parents and the club’s sports physician are all keen to know more about the current</td>
</tr>
</tbody>
</table>
joint status and agree to a 3 Tesla MR imaging of her left hip. (The radiologist asked for a morphological sequence: three-dimensional (3D) water selective fluid (WATSf) to image joint cartilage and bone. The radiologist also performed 3D multiplanar reconstructions and acquired radial images around the femoral neck at 30° intervals. [17])

Additional case – contrary: Hip dysplasia

A 15-year-old male ballet dancer reports occasional left groin stiffness associated with high-intensity practice or performance. He also reports an occasional feeling of restricted ‘hip’ movement but cannot explain exactly what he feels. He joined the ballet school, practicing and performing more than 10 hours per week since age 9. He denies any previous significant hip or groin injuries. Hip and groin examination findings are unremarkable, including normal bilateral hip range of motion tests (internal rotation of 40°). The flexion adduction internal rotation (FADIR) and flexion abduction external rotation (FABER) special tests are both normal. He agrees to further special investigations: AP-pelvis and lateral radiographs of both hips. The radiologist reports normal head-neck femoral morphology on both AP-pelvis and lateral radiographs with signs of left hip dysplasia; she measured normal alpha angles in both views (48 degrees and 49 degrees respectively); Lateral center-edge angles (LCEAs) were 19 degrees and 23 degrees for the left and right hips respectively. The dancer, his parents and the ballet school’s sports physician are all keen to know more about the current joint status and agree to a 3 Tesla MR imaging of his left hip. (The radiologist asked for two morphological sequences: three-dimensional (3D) water selective fluid (WATSf) to image joint cartilage and bone, and 3D proton density fat saturation (PDFS) to image the physeal scar. The radiologist also performed 3D multiplanar reconstructions and acquired radial images around the femoral neck at 30° intervals. [17]) MR imaging confirmed a small anterior labral tear.

Additional case – contrary: Secondary cam morphology

A 13-year-old male presents with a two-month history of intermittent left groin and knee pain and an associated limp made worse by playing sports. He also reports an occasional feeling of restricted ‘hip’ movement but cannot tell exactly what he feels. He kicks with his right foot and plays occasional football with his friends, no more than 1 hour per week since the age of 11. He denies any previous significant hip and groin injuries. He is overweight; his knee examination is normal. The left hip has reduced hip flexion and rotation range of motion (internal and external rotation) compared to the right hip. The FADIR test is painful on the left but the FABER special test on the left is normal. His left leg is 2.5 cm shorter compared to the right leg. He and his parents agree to further special investigations: anterior posterior pelvis (AP-pelvis) and lateral radiographs of both hips. The radiologist reports a posterior slipped capital femoral epiphysis.
Step 6: Additional cases

We wrote corresponding narratives for additional cases, describing borderline cases, related cases, contrary cases to further illustrate the concept of primary cam morphology. This is an important step as it may be difficult to determine the defining attributes that most closely represent primary cam morphology. We therefore describe additional cases to help refine the best fit defining attributes. [1] We describe three additional cases to inform the concept: (1) Primary mixed morphology, (2) Hip dysplasia, and (3) Secondary cam morphology due to slipped capital femoral epiphysis (table 3).

Step 7: Antecedents and consequences

The science concerning primary cam morphology, including its aetiology and prognosis, is not settled. No high-quality prospective studies with an adequate follow up time exist on primary cam morphology aetiology or prognosis. This concept analysis will inform higher quality future research, including expert opinion and consensus agreement (or expert dissent for discussion) on taxonomy, terminology, definitions and imaging outcome measures. A collaborative approach to multi-cohort prospective aetiology and prognosis studies provides the opportunity to share higher quality, uniform research data.

Antecedents

Three important antecedents were identified in our concept analysis: (1) young adolescents with no other disorders of the hip (absence of conditions associated with secondary cam morphology), (2) an open femoral capital physis with epiphyseal hypertrophy and/or extension as a result of (3) high-load physical activity (shear-type load) as the likely causative risk factor (volume and type of load are not well understood; probably external rotation with flexion—axial and rotational forces combined), and other unconfirmed risk factors.

No other disorders of the hip or any co-occurring hip impingement/pathology (absence of conditions associated with secondary cam morphology)

Primary cam morphology is cam morphology that develops in the absence of other hip pathology; we excluded all articles on secondary primary cam morphology from the primary cam morphology risk factor systematic review and this concept analysis. Secondary cam morphology is caused by pre-existing hip disease or trauma including, Perthes disease, slipped capital femoral epiphysis (SCFE), healed proximal femoral fractures, avascular necrosis or osteophytes.

An open femoral capital growth plate (when does primary cam morphology develop?)

It is still unclear exactly when and how primary cam morphology develops. We conclude from cross sectional studies and a small number of prospective cohort studies that primary cam
morbidity likely develops during skeletal maturation when the femoral capital physis is still open. The included articles refer to (early) adolescence / childhood / maturation / young adulthood and additional long-term and multi-centre studies are needed to investigate this further.

‘Future research recommendations: We recommend large-scale, interdisciplinary research on aetiology and prognosis for each of the listed hip-related pain conditions. (For example, the relationship between bony morphology and other factors related to these conditions or movement-related factors relative to each hip-related pain condition.)’ [18]

High-load physical activity (shear-type) as the likely causative risk factor (volume and type of load are not well understood; probably external rotation with flexion—axial and rotational forces combined), and other unconfirmed risk factors.

Primary cam morphology develops gradually during skeletal maturation as a result of physiological skeletal response to physical load (athletic activity) on the femoral capital physis, hence the term primary (including idiopathic) cam morphology. The exact mechanism of primary cam morphology development is unknown. Our systematic review of risk factors for primary cam morphology has identified several factors associated with primary cam morphology. These include demographic risk factors, environmental/lifestyle risk factors (sport, physical activity and dance) and a variety of other risk factors. The science of its causal inference, understanding why primary cam morphology occurs, is unclear. A detailed analysis of these factors will be presented in the report of the primary cam morphology risk factors systematic review.

Consequences

A consequence of primary cam morphology could be motion dependant abutment against the acetabular rim, described as femoroacetabular impingement (FAI). However, in large population-based prospective studies, end-stage osteoarthritis was the sequelae in < 11% of hips with cam morphology. [19,20] Furthermore, in two smaller prospective studies, > 84% of hips defined as having cam morphology did not develop hip pain. [21,22] A combination of risk factors, including primary cam morphology, may cause hip disease in some individuals, including: (1) femoroacetabular impingement syndrome (combination of symptoms, including pain, stiffness, reduced range of motion, signs and hip morphology changes on imaging); (2) tissue damage, including labral, and cartilage, and (3) early hip joint osteoarthritis. [18] (figure 3).

Femoroacetabular impingement (FAI) with cam or mixed morphology

FAI refers to the unwanted compression of soft tissue (labrum; cartilage; joint capsule) between the femur (head; head-neck junction; neck) and the acetabulum (usually the acetabular rim). FAI
with cam morphology refers to asymptomatic motion-related abutment of cam morphology against the acetabular rim.

**Soft tissue damage: labral; chondral**

Cam morphology has been associated with hip joint soft tissue damage, including labral and chondral tissue using normal MRI [23], MRI T1ρ relaxation time [24,24–27], and T2-mapping. [28]

**FAI syndrome (symptoms: pain, stiffness, other)**

FAI syndrome is a triad of symptoms (most often motion-related or position-related pain in the hip or groin), clinical signs (most commonly a positive flexion adduction internal rotation – FADIR test) and imaging findings (cam and/or pincer morphology). [29]

**Osteoarthritis**

Patients with cam deformity and decreased internal rotation were at significantly higher risk of developing end stage osteoarthritis (odds ratio 25.21) in a large cohort of individuals with early onset hip pain with osteoarthritis. [30]

**Other consequences**

Other possible consequences of primary cam morphology include limited hip range of motion, changes in hip mechanics and biomechanics and muscle recruitment patterns. [31] The data are equivocal and mostly cross sectional, [32] and a detailed analysis of primary cam morphology consequences is beyond the scope of this paper.

**Figure 3 Pathogenesis (antecedents and consequences) of primary cam morphology**

![Pathogenesis diagram](image-url)
References


