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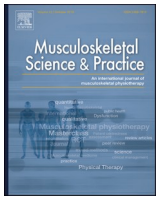

Original article

Development of an aid to detect adults acetabular hip dysplasia (the ALPHA alert): A mixed methods study

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ABSTRACT

Objectives: To identify the signs and symptoms that people living with acetabular hip dysplasia (AHD) describe and to provide an aid for translating the findings into practice.

Methods: A three-phased mixed methods study. Phase 1 employed an open-question online survey that enabled people with AHD (aged ≥16 years) to describe features associated with their condition. Responses were thematically analysed. A Phase 2 survey used these themes to establish how common those features were. Phase 3 created a mnemonic that prompts clinicians to suspect AHD.

Results: Ninety-eight respondents completed Phase 1 and sixty-two completed Phase 2. From the responses, five themes were identified: Demographic and Diagnostic Profile; Characteristics of Posture and Gait; Pain; Childhood Hip and Family History; and Hip Joint Characteristics. Within these themes, 19 common signs and symptoms were reported, represented by the ALPHA mnemonic. ALPHA describes a young age at onset of problems (**A**ge), a limp (**L**imp), progressing pain (**P**ain), a history of childhood and family hip anomalies (**H**istory) as well as hip joint hypermobility and instability (**A**rticulation).

Conclusion: The findings extend current understanding of AHD indicators. ALPHA alerts clinicians to suspect a diagnosis of AHD. ALPHA may facilitate timelier referral of patients for diagnostic X-Ray and appropriate treatment. Future studies should evaluate its clinical utility.

1Introduction

Hip dysplasia is a multifactorial condition that describes a treatable hip joint instability. Commonly referred to as Developmental Dysplasia of the Hip (DDH), the condition is recognised as having two patterns of onset; an instability at infancy and an instability that occurs initially during adolescence or young adulthood (Pun 2016). The two onset patterns indicate that the condition affects the hip at different stages of skeletal development. This has led to well understood features of infant onset DDH, such as causes and risk factors, to be applied to hip dysplasia of later onset, yet the connection between the two onset patterns, if indeed there is one, has not yet been determined. To ensure clarity therefore, this paper makes an age-related distinction in using the term DDH to refer to the condition in infancy and Acetabular Hip Dysplasia (AHD) to refer to an adolescent or young adult onset. Where reference is made more generally to these conditions presenting at various ages, the broader term Hip Dysplasia (HD) is used.

Acetabular Hip Dysplasia (AHD) is recognised by a shallow, often steeply orientated acetabulum resulting in activity-limiting hip joint instability (Troelsen 2012). It is a common cause of hip pain in young adults and has been shown to occur in over 40% of patients less than 50 years of age undergoing hip joint replacement surgery (Muddalum et al., 2023). AHD is a leading precursor of premature, secondary osteoarthritis (OA) (Wyles et al., 2017). It is accepted that early recognition and treatment are key to successful outcomes of corrective surgery (Steppacher et al; Lerch et al., 2017) and whilst diagnostic X-Ray measures are well recognised, patient presentation at the pre-X-Ray stage is poorly understood (Gambling and Long 2019). Consequently, there may be uncertainty or difficulty for some clinicians with limited knowledge of AHD, to recognise when referral for diagnosis by X-Ray is required. Currently, what is known about pre-X-Ray presentation is based on Nunley et al., 's 2011 study of fifty-seven patients with symptomatic AHD. Patient reported symptoms included daily moderate-to-severe pain that was commonly localised to the groin (72%) or the lateral aspect of the hip (66%) and hip pain that was related to activity (88%). On examination, 48% had an associated limp, 38% showed a positive Trendelenburg sign and 97% had a positive impingement test. The average time between patients' onset of symptoms and AHD diagnosis

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was longer than 5 years. These findings are useful, but because of an overlap of symptom presentation, they do not provide guidelines that sufficiently distinguish AHD or hip joint instability from other hip conditions such as femoroacetabular impingement (FAI) (Olsen 2023). Another larger study would not only establish whether the findings of Nunley et al. (2011) would be replicated in a second group of people living with AHD, but it could also expand the findings to identify nuances of AHD that would provide much needed guidelines for differential diagnosis (Olsen 2023). The primary aims of this study were therefore to a) identify the signs and symptoms that people living with AHD describe and b) provide an aid for translating the findings into practice.

2. Methods

An exploratory, 3-phase, mixed methods approach was used. In Phases 1 and 2, surveys were used to identify the signs and symptoms that people living with AHD associated with their condition. In Phase 3 the results of both surveys were used to create an approach for translating the findings into practice. The study received ethical approval from the institutional review board and the relevant Health Board's Research and Development Research Review Committee (Reference: RD/1527/17). This study is reported according to the Consolidation criteria for Reporting Qualitative research (COREQ) checklist (Tong et al., 2007).

2.1. Study sample

Recruitment occurred over a three month period. Invitations to participate in Phase 1 and Phase 2 surveys were posted on the web page of four online hip dysplasia patient support groups (STEPS Charity Worldwide; The Periacetabular Osteotomy United Kingdom Based Group (PAO UK); Developmental Dysplasia of the Hip UK(DDH UK) and Adult Hip Dysplasia Support Group). Each survey included an information sheet followed by the survey questions and was live for two weeks. Consent was confirmed by completing and submitting the survey and AHD diagnosis was self-declared. The surveys then asked questions aimed at exploring further details about participants' diagnosis. This confirmed their self-declaration. Whilst a large proportion of respondents (54% in Phase 1 and 71% in Phase 2), had already received corrective or replacement surgery for their AHD, all respondents were asked to provide information based on the signs and symptoms they associated with their AHD *before* surgery. Investigators acknowledged that relying on participants' memory risks recall bias, but excluding those who had received surgery would have severely limited the participant pool. Troublesome symptoms for which participants had clear memories were considered key to building a picture of AHD features.

2.2. Phase 1 and phase 2 surveys: design and administration

The Phase 1 survey (Table 2) was developed following a systematic review of literature that aimed to identify AHD indicators. The findings of the review showed that as well as symptoms that included an insidious onset of moderate to severe activity-related pain (Nunley et al., 2011), there was tentative evidence of an association of AHD with Gluteus Medius changes (Liu et al., 2012), heritability and recurrent risk (Li et al., 2013; Carroll et al., 2016), and hyperlaxity (Bilsel et al., 2016; Samper et al., 2015). The survey was piloted and refined in collaboration between the lead author (EE) and two people living with AHD (TG and MJS). Open-ended questions were employed as they are effective where there is insufficient knowledge of how respondents perceive and describe a disorder (Bengtsson 2016). Themes and their items generated by Phase 1 data were used to develop a second survey (Phase 2) aimed at quantifying each items' occurrence. Phase 2 therefore used mainly closed questions that required respondents to provide their agreement or disagreement with a given list of focussed statements. These symptom-related statements required a yes/no response or ticking a choice answer from a given list of possibilities. Five-point Likert-type scales that used "Strongly Agree"; "Agree"; "Neither Agree nor Disagree"; "Disagree" or "Strongly Disagree" were also applied to record whether respondents had experienced specific signs and symptoms. Additionally, open questions were presented so that respondents could add details if they chose to (Table 3). The Bristol Online Survey (BoS) platform (now JISC Online Surveys) administered the questionnaire surveys and automatically compiled response data into spreadsheet format.

2.3. Phase 1 and phase 2 surveys: data analyses

An inductive, six-stage thematic approach (Braun and Clarke, 2006) was used for Phase 1 and Phase 2 qualitative data analysis. This involved data familiarisation, coding (item identification), identifying, reviewing and defining themes, and organising the results. For the Phase 2 ordinal data, analysis was conducted using frequency counts and percentages. The resulting values identified the occurrence of agreement between respondents around a particular issue. For this purpose, Likert scale data indicating 'strongly agree' and 'agree' were merged. Analysis was completed by the lead author (EE) and the procedure was supported throughout by TG, who analysed approximately 10% of both Phase 1

Table 1
Phase 1 and phase 2 respondent demographics.

Demographic		Phase 1 n = 98	Phase 2 n = 62
General Characteristics	Mean Age	36.7 years (range 16–61 years)	35.7 years (range 16–60 years)
	Males	2 (plus 1 female to male transgender)	2
	Deciding on or awaiting surgery	45 (46%)	18 (29%)
	Post corrective or hip joint replacement surgery	53 (54%) (of those declaring years since surgery (n = 78), the mean was 6.6 years)	44 (71%)
	Hips affected bilaterally	69 (70%)	41 (66%)
Age at initial diagnosis	Birth - <2 years	n = 17 (17%)	n = 14 (23%)
	2–12 years	n = 14 (14%)	n = 7 (11%)
	13–19 years	n = 8 (8%)	n = 2 (3%)
	20–29 years	n = 25 (26%)	n = 10 (16%)
	30–60 years	n = 34 (35%)	n = 29 (47%)
Country of residence	United Kingdom	n = 71 (72%)	n = 56 (90%)
	United States	n = 20 (20%)	n = 5 (8%)
	Republic of Ireland	n = 0 (0%)	n = 1 (1%)
	Canada	n = 1 (1%)	n = 0 (0%)
	Australia	n = 1 (1%)	n = 0 (0%)
	Slovenia	n = 1 (1%)	n = 0 (0%)

Table 2
Phase 1 Survey. Summary and Theme Development (total n = 98).

Section 1: Demographic Profile *Section Questions*	*Sample Responses*
• Male/female? • In which hip/hips do you have Hip Dysplasia? • At what age were you first diagnosed with DDH or hip dysplasia?	Demographic Details for Age, Location, Treatment Stage presented in Table 1. • Preponderance of female participants (n = 96/98, 98%) • Bilateral (n = 69/98, 70%) • Greater number of participants receiving diagnosis of AHD after the age of 2 years (n = 81/98, 83%) ❶
Section 2: Earliest Memories *Section Questions*	*Sample Responses*
• What are your earliest memories of having a hip problem? • Looking back on your childhood, were there any movements/activities you struggled to do or any activities that you found easier than other people seemed to?	*"Earliest memory is experiencing strange twinges and shooting nerve pain and sensations when lying on back with both legs straight, or lying on affected hip side to sleep ….. It was not everyday, but intermittent❸ … ….. symptoms became more often and collapses started ❺ …" (R1:96).* *"I could never sit cross legged at school, I used to dread story time at primary school as I found sitting on the floor unbearable ❹. Physical exercise wise I was able to run/swim/play sports …. It wasn't until I looked back did I realise that my hips had not been normal" (R1:40).*
Section 3: **Pre-Surgery/Current Signs and symptoms** *Section Questions*	*Sample Responses*
• Please can you give a brief description of your symptoms, how they have developed since they first started and how they have changed over time? • How has DDH/hip dysplasia affected your life from the time you were diagnosed up to now?	*"My hip problems (pain, loss of range of movement) were looked at, but my gait (leaning forwards, limping)❷ has never been addressed ❶. It feels like I have a whole package of bad posture/gait"❷ (R1:28)* *"My symptoms worsened over the years and I was walking with a pronounced limp"❷ (R1:33)* *"My parents and family had concerns because of leg different lengths❷ and pain❸ was told I had flat feet". (R2:31)*
Section 4: Experiences of Diagnosis *Section Questions*	*Sample Responses*
• Please tell us about your experience of getting your diagnosis.	*"I couldn't walk ❷ so my parents took me to see various GP's, doctors, health visitors etc. …. I went so often. I have had hundreds of xrays and mri scans"❶. (R1:11)* *"I was sent from surgeon to surgeon (each not knowing what was going on). I got diagnoses from cancer to a shrug of the shoulder. A team of surgeons then looked at my case and sent me to a surgeon … that specialized in PAO. … It was a draining and humiliating process that made me feel like no one knew what was going on. … The diagnosis was 10 years in the making." ❶ (R1:21)* *"Over the years I went to see my GP multiple times❶, however as I moved around a lot it was always a different GP. I was always sent for physio and it never helped." (R1:33)* *"My experience is a huge lack of awareness from all doctors and other AHPs ❶ about dysplasia in ADULTS. They tried to control my worsening symptoms over many years with simply analgesia and PT [physiotherapy] which did not help".(R1:54).* *"Overall I was so disappointed that the NHS [National Health Service] service missed my condition on several occasions ❶ and the lack of experience of the condition is worrying" (R1:66).*
Section 5: Any Other Information *Section Questions*	*Sample Responses*
• Is there any other information about your experience of living with DDH/Hip Dysplasia which you feel is important to include?	*"I am hypermobile ❺ and was a competitive gymnast for years. I was able to do all three splits easily, among other things … … My three daughters ❹ … are 20, 17, and 15. I'm sure all three are also hypermobile, and occasionally they have complained of mild, non-debilitating hip pain" (R1:88).* *"I went many times to the Doctors ❶ with pain in my leg/hip/knee/groin ❸ but despite my eldest son having bilateral congenital CDH ❹, it was never suspected with me. Eventually, after my* 4th *son also was born with one dislocated hip ❹, I asked for my hips to be x-rayed" (R1:101)* *"I made sure our children were extensively tested for DDH as babies and young children ….. there is always that niggling anxiety in the back of your mind about whether you've passed anything on to your children"❹ (R1:93).*

Key: DDH: Developmental Dysplasia of the Hip.
Sample Responses Key: Anonymisation coding: Phase 1 participants identified by the prefix 'R1' which is followed by an individualised number (e.g., R1:46). *Numbering within sample quotes* illustrate how items of interest (signs and symptoms) were identified and grouped to form themes.
❶ Theme 1: Demographic and Diagnostic Profile ❹ Theme 4: Childhood & Family History.
❷ Theme 2: Characteristics of Posture & Gait ❺ Theme 5: Hip Joint Characteristics.
❸ Theme 3: Pain.

and Phase 2 data sets. Agreement was evident between the analyses of EE and TG, confirming analytical consistency and appropriateness of the emerging themes and their items. Subsequent discussions with DA assisted in refining theme titles.

2.4. Phase 3 ALPHA alert development

Following discussions between EE, DA, TG and MJS, the themes and their items were used to develop a mnemonic labelled 'ALPHA' that facilitates the translation of findings into practice. Evidence suggests that mnemonics are valuable in providing an alert that supports memory recall (Dresler et al., 2017). For instance, a single word such as F.A.S.T. (face, arms, speech, time) is an effective alert of stroke symptoms (Chen et al., 2022). EE therefore re-examined the themes and their items (Table 4) and considered the assessment of hip conditions generally, alongside the recognised indicators of differential diagnosis (Dick et al., 2018). Then to translate the study's findings into practice, EE developed a single term mnemonic to inform clinicians of patient-reported signs and symptoms (theme items) associated with AHD presentation.

Table 3
Responses to the Phase 2 Survey. Question wording is provided, along with numerical summary of features' occurrence and sample quotes (total n = 62).

Theme 1: Demographic and Diagnostic Profile *Questions*	*Occurrence*	*Quotes (Additional Comments)*
• How old were you when you received your first diagnosis of hip dysplasia? • How long did it take you to get your initial diagnosis of hip dysplasia? • If there are any features or indications of your hip problem which you feel were ignored by the clinicians	Mean age = 22 years Range = 0–54 years <5 years: n = 47 >5 years: n = 15 Severity/chronicity of pain: n = 29	*"Even if symptoms were hard to diagnose, the length of time and persistence in symptoms should have been a red flag". (R2:11)* *"I don't understand how so many specialists can have seen X-rays over the years and no one picked up the dysplasia until a hip specialist looked at it". (R2:2)* *"I was 'diagnosed' for a number of years by GP as having 'growing pains' and 'just being a bit of a hypochondriac' and having a 'low pain threshold' (R2:14)* *"Pain, Symptoms, Mobility, Quality of life, Impact of deformity on other areas of the body … all ignored (R2:14)*
Theme 2: Characteristics of Posture and Gait *Questions* Please rate your level of agreement with the following:	*Occurrence*	*Quotes (Additional Comments)*
• My poor posture causes me to have back pain	n = 44 agree (71%)	*"I stand with most of my weight on my left leg". (R2:16)* *"I was a dancer as a child and when I think back now I wasn't symmetrical in my movements. My high kicks were different on each leg". (R2:51)*
• My hip has caused me to walk with a limp	n = 54 agree (87%)	*"My symptoms worsened over the years and I was walking with a pronounced limp". (R2:11)*
• My hip has caused me to waddle, hobble, shuffle or roll when I walk	n = 48 agree (77%)	*"Preferred to sit in a 'w' position. Waddled when walking or running". (R2:58)*
• My pelvis drops down on one side when I walk	n = 38 agree (61%)	*"… my gait (leaning forwards, limping) has never been addressed. It feels like I have a whole package of bad posture/gait …" (R2:47)*
Theme 3: Pain *Questions* Please indicate your level of agreement with each of the statements	*Occurrence*	*Quotes (Additional Comments)*
• My pain developed over months or even years from a mild niggle to severe pain	n = 54 agree (87%)	*"I feel like the pain in my hip accelerated very quickly, from an annoying [s] hearing pain initially to a debilitating constant pain that was impacting on everyday life. In the space of a few months my pain really intensified" (R2:2).*
• My pain is worsened by any physical activity, over-use or exercise	n = 56 agree (90%)	*"Have to ensure hip angle not closed when sitting for long periods". (R2:12)*
• My pain increases with turning/twisting/pivoting/ cutting	n = 52 agree (84%)	*"Sitting straight in a normal chair was difficult, I always wanted my legs curled under me.." (R2:48)*
• My pain increases with prolonged sitting	n = 49 agree (79%) (avoids deep hip flexion)	
Theme 4: Childhood Hip and Family History *Questions*	*Occurrence*	*Quotes (Additional Comments)*
As a child or adolescent, were there movements/activities related to your hip that your found difficult or were unable to do?	Problems sitting cross-legged n = 36 agree (60%) Considered to be hyperflexible n = 32 agree (52%)	*"I could never sit cross-legged and was given special permission to sit how I wanted (with legs in a W)" (R2:47)* *"Couldn't sit cross-legged, even as a child. Always sat in W position. I found sitting on the carpet at primary school very uncomfortable." (R2:53)* *"I now know that I'm hypermobile and this was a red herring in my diagnosis as when I saw physios they said there was nothing wrong with my hip as I had a very good ROM in the hip joint." (R2:55)*
Please list any of your relatives who have or have had hip problems and the treatment they received	Relatives with known hip problem n = 57 (total) Reported by 34 (55%) respondents Between 1 and 6 relatives listed by each	*"grandmother, her sister & her grandaughter, all had hip issues, with cousin having SUFE also - when initially diagnosed …" (R2:44)* *"Mother said she "had funny feet" as a child but didn't elaborate. My sister has hip pain and mild dysplasia … My Niece (25) has occasional hip pain and a slightly "dysplasia" gait." (R2:47)*
Theme 5: Hip Joint Characteristics *Questions*	*Respondent Agreement Count*	*Quotes (Additional Comments)*
Please list any aspects of your hip problem which you feel are important for clinicians to know about during their diagnostic assessment	Continued Hypermobility n = 28 agree (45%) Hip Sounds (lock; pop; crack; clunk) n = 56 agree (90%) Hip instability n = 44 agree (71%) Hip joint pain/niggles as a child but no major problems until some years later n = 23 agree (37%) Hip joint problems began in teens/as an adult n = 27 agree (44%) Problems in both hips (Bilateral symptoms) n = 7 (11%)	*"felt like "Barbie hips" they would dislocate easily and pop out of socket." (R2:63)* *"Aching and clunking made worse by exercising". (R2:38)* *"Hip clunks when moving, groin pain, leg gives way when put weight through left leg.". (R2:66)* *"When I walk, I feel as if my femur is out of socket at times." (R2:43)* *"At 12/13 I saw my first consultant who I described in both sides, hip pain, cracking, popping, locking of hip feeling that the hip wasn't in the right place frequently …..instability in my hips as though my legs would 'just go from under me' …" (R2:14)* *"I pressed for something else and he agreed to x ray, which immediately showed bilateral hip dysplasia." (R2:11)* *"The person I saw first was the hip specialist and he later diagnosed me with bilateral hip dysplasia …." (R2:14)*

Key: Slipped Upper Femoral Epiphysis (SUFE); Developmental Dysplasia of the Hip (DDH).

3. Results

Ninety-eight respondents completed Phase 1 and sixty-two completed Phase 2. Demographic details are presented in Table 1.

Phase 1 generated detailed data that uncovered new knowledge of AHD presentation. Table 2 illustrates how these signs and symptoms were identified and how they were grouped into themes. Following the details of respondents' demographic and diagnostic profiles, sample

responses are shown for questions within each survey section. Topics are numerically labelled to show how themes emerged from the data. Respondents described a range of features considered to be the start of their hip problems. In childhood, hip pain, participation in high level or intensive sport, abnormal movements related to their hip joint instability or hypermobility, and the inability to maintain certain postures (such as cross-legged sitting and positions of deep hip flexion), were all reported to be associated with the eventual onset of longer-term hip problems. There were repeated reports of progressive hip pain and of respondents' concerns being triggered when they experienced their hip 'popping out of its socket' or 'locking' and 'clicking'. Many described themselves as being hypermobile or hyperflexible, having 'weak' hips or a leg length discrepancy. Additionally, there were recurrent comments suggesting a familial pattern of hip problems.

Five principal themes were generated from Phase 1 data. Theme items identified the signs and symptoms respondents associated with AHD. These were used to construct the Phase 2 questionnaire which was used to identify the occurrence of signs and symptoms. The results are shown in Table 3 which also provides sample quotes from the responses to Phase 2 open questions. These illustrate respondents' support for the occurrence of theme items.

3.1. Signs and symptoms associated with AHD

3.1.1. Theme 1: demographic and diagnostic profile

This theme comprised four patient-reported characteristics. Firstly, there was a predominance of female respondents, with only 2 respondents being male in both Phase 1 and Phase 2. Secondly, 69 Phase 1 respondents (70%), and 41 Phase 2 respondents (66%) indicated bilateral symptoms. Mostly, these respondents described how the problems affected only one hip initially and that the contralateral hip became problematic some years later. The third notable characteristic was the young age at which respondents experienced hip problems. Whilst the largest proportion of respondents in both Phase 1 and Phase 2 received their AHD diagnosis after the age of 30 years (n = 34 or 35%; n = 29 or 47% respectively), respondents reported having experienced hip problems for years or even decades before receiving their AHD diagnosis. This highlights the fourth feature of this theme; delayed diagnosis where respondents emphasised their frustration at being told that their problems were due to, for instance, 'growing pains' or 'muscle weakness'. Some stated that they simply felt they were being 'fobbed off'. Of the 87 Phase 1 respondents who described their experiences of diagnosis, 78 (90%) listed a range of clinicians from various professions who they repeatedly consulted before their diagnosis was determined. These included general practitioners (GPs), physiotherapists, radiologists, orthopaedic surgeons and other secondary care specialists.

3.1.2. Theme 2: Characteristics of Posture & Gait

Phase 1 survey responses included various descriptions of persistent postural abnormalities and associated back pain that participants related to their hip problems. Features of gait were also described. These issues were explored in greater detail in the Phase 2 survey. Approximately 72% of respondents agreed that their hip-related poor posture caused their back pain and 69% confirmed having postural asymmetry (Table 3). Posture was also affected by hip pain that was said to be triggered by prolonged sitting and standing, causing respondents to fidget, put weight on one side or lean against a wall, enabling pressure to be taken off their painful hip. Others described slouch-sitting or favouring a foot-raised, open hip-angle position to avoid painful deep hip flexion. Most Phase 2 respondents agreed that they had noticeable gait changes, with 88% reporting a limp. A waddle, hobble, shuffle or rolling gait was described by 77% and 59% of respondents also agreed that their pelvis dropped on one side when walking.

3.1.3. Theme 3: pain

Pain impacted on almost all areas of life including education, career progression, participation in social and sporting activities and relationships. Phase 1 respondents reported various aspects of their pain onset and development using a wide range of terms. Phase 2 established whether common descriptors could be identified for the nature and development of pain between people living with AHD. The results showed 87% indicated that hip pain which had started as a mild 'niggle', developed into intense pain that changed from an occasional discomfort to a constant, debilitating problem (Table 3). Additionally, 79% agreed that sitting and deep hip flexion beyond 90°, required for upright sitting, squatting and lifting legs up to the chest, was difficult or painful (Table 3). Sitting in a reclined position that limited hip flexion was, however, described as a position of ease that helped in the management of hip pain.

3.1.4. Theme 4: Childhood Hip and Family History

In Phase 1, respondents often expressed memories of hip-related experiences that they felt were different from other children. This included the inability to sit cross-legged during childhood; an issue for many as it was a requirement of story-time and assemblies during primary school. Such issues did not appear to prevent respondents from involvement in other activities expected of their age; indeed, most described themselves as being highly active, keen athletes, gymnasts or dancers. For many, these sporting skills appeared to be enhanced by their hypermobility. Nevertheless, it appears that with the passage of time, respondents associated their early hypermobility with later hip pain. Phase 2 was used to gain an accurate insight into the commonality of these features of childhood history. Sitting cross-legged was reported to increase hip pain by 60% of respondents and 52% described themselves as hyperflexible or 'double-jointed'.

A familial association of hip problems was indicated in Phase 1 data. For instance, respondents who also had children with AHD, described how they referred to their children's hip dysplasia diagnoses to justify their own need for diagnostic hip X-Ray. Phase 2 respondents were therefore asked to provide information about any relatives who were known to have had hip problems. In response, some 55% named between 1 and 6 relatives with a known hip problem (Table 3). The data showed that during their initial clinical assessment, respondents either did not think to mention or were not given the opportunity to explain

Table 4
Summary of themes and their related items.

Theme 1: Demographic and Diagnostic Profile	Theme 2: Posture and Gait	Theme 3: Pain	Theme 4: Childhood and Family History	Theme 5: Hip Joint Characteristics
Young age	Limp – leg length discrepancy/ Trendelenburg gait	Mild, insidious onset becoming persistent/intense	Any childhood hip concerns	Hypermobility
Females (predominantly but not exclusively)	Waddle, hobble	Groin/hip region	Hyper-flexibility/hypermobility	Hip joint Instability
Commonly bilateral hip problems	Forward lean posture (tight hip flexors)	Increases with deep hip flexion/ activity	Preference for W-sitting	Audible hip sounds
Repeated clinical appointments	Postural asymmetry		DDH/AHD or THR/OA at young age in relatives	Inability to sit cross-legged

Key: Developmental Dysplasia of the Hip (DDH); Acetabular Hip Dysplasia (AHD); Total Hip Replacement (THR); Osteoarthritis (OA).

these childhood or familial features.

3.1.5. Theme 5: hip joint characteristics

Hypermobility persisting into adolescence and adulthood was common and considered by some to be initially beneficial in achieving gymnastic abilities or notable 'trick' movements. This was particularly evident in those phase 1 participants who reported a pattern of hip problems featuring two onset periods: firstly, an infant-onset of hip problems, that was followed by years of problem-free hips. Subsequently, a second onset of hip problems in adolescence or young adulthood was reported. Fifty percent of these respondents also described favouring unusual or W-sitting positions, that involved sitting on the floor with the hips internally rotated, knees flexed forward, and feet splayed laterally. Hypermobility was often identified in Phase 1 as being linked with respondents' hip joint instability including dislocation, subluxation or the sensation of the hip 'popping out of the socket'. This feeling of instability was commonly associated with hip sounds, which included 'clunking', 'popping', 'cracking' and 'clicking'. Phase 2 showed that some of the respondents who indicated that they were hypermobile, stated that as adolescents, hypermobility and hip joint instability were such that they could make their hips dislocate at will. Most agreed that their hips felt unstable (77%), and overall, hip sounds were reported by 91% of respondents (Table 3).

The five themes provide an extended clinical picture of AHD presentation prior to radiographic diagnosis and are summarised in Table 4.

3.2. Phase 3: translation of findings for clinical application

To aid the translation of findings into practice, the data were synthesised to create the ALPHA alert mnemonic (Fig. 1). Each letter of the word ALPHA acts as a reminder of a clinical feature that people living with AHD commonly associated with the condition. During patient examination, the occurrence of a combination of ALPHA features should prompt suspicion of AHD presence, alerting the clinician that referral for X-Ray evaluation should be considered.

4. Discussion

Familiarity with the AHD clinical picture is vital for facilitating early suspicion of the condition's presence. Translation of knowledge from this study into clinical use requires an approach that is not onerous for clinicians to apply to their diagnostic evaluation, and it has importance for individual and societal benefits (van der Laan and Boenink 2015). Willmen et al. (2021) demonstrated how support systems that accelerate referral of patients to the appropriate specialist, avoided unnecessary patient assessments and repeated evaluation, providing health economy benefits in the case of rare conditions. Additionally, the Department of Health 2006 expressed how improving our understanding of disease, its recognition and prevention will provide benefits to society's health and wellbeing. From respondents' experiences of living with AHD, we have extended current understanding of AHD presentation by describing and summarising patient-reported signs and symptoms (Table 4). We have translated these findings into a mnemonic for use in clinical practice (Fig. 1). The demographic profile of our respondents corresponds with other studies (Okano et al., 2015; Loder and Skopelja 2011; Lee et al., 2013; Nunley et al., 2011). Firstly, most participants (more than 65% in both Phase 1 and Phase 2) received their initial AHD diagnosis during or after adolescence (Table 1) having had no previous history of infant DDH. This concurs with the findings of a study of 245 patients with hip dysplasia in which 36% had a history of infant DDH and the remaining 64% had an initial onset as adolescents or young adults (Okano et al., 2015). Secondly, the high response rate of females (97%) in our study is consistent with current understanding of infant DDH occurring more frequently, but not exclusively in females (Loder and Skopelja 2011). Lee et al. (2013) specifically investigated demographic differences between AHD and DDH and identified that whilst overall, both conditions were more prevalent in females, AHD showed a higher rate of male occurrence than DDH. Thirdly, our data also showed a common incidence of bilateral presentation (more than 65%) which is consistent with other AHD studies. A Japanese study of 187 women and 19 men with pre-arthritic AHD showed that 84% presented bilaterally (Okano et al., 2008). The importance of acknowledging bilateral presentation is that during physical assessment, clinicians typically compare limbs to evaluate the degree of difference caused by the pathology (Gaskell 2013). Such differences may be unrecognisable if dysplasia exists in both hips. Fourthly, in terms of delayed diagnosis, respondents described having repeated clinical appointments before the correct AHD diagnosis was recognised, resulting in costly and inappropriate use of health resources. Similarly, Nunley et al. (2011) showed that patients in their study were assessed by up to eleven clinicians, often from different professions, before receiving the correct AHD diagnosis, which can delay appropriate treatment.

Our findings indicate that people living with AHD experienced other abnormalities of posture and gait that included postural asymmetry, leg

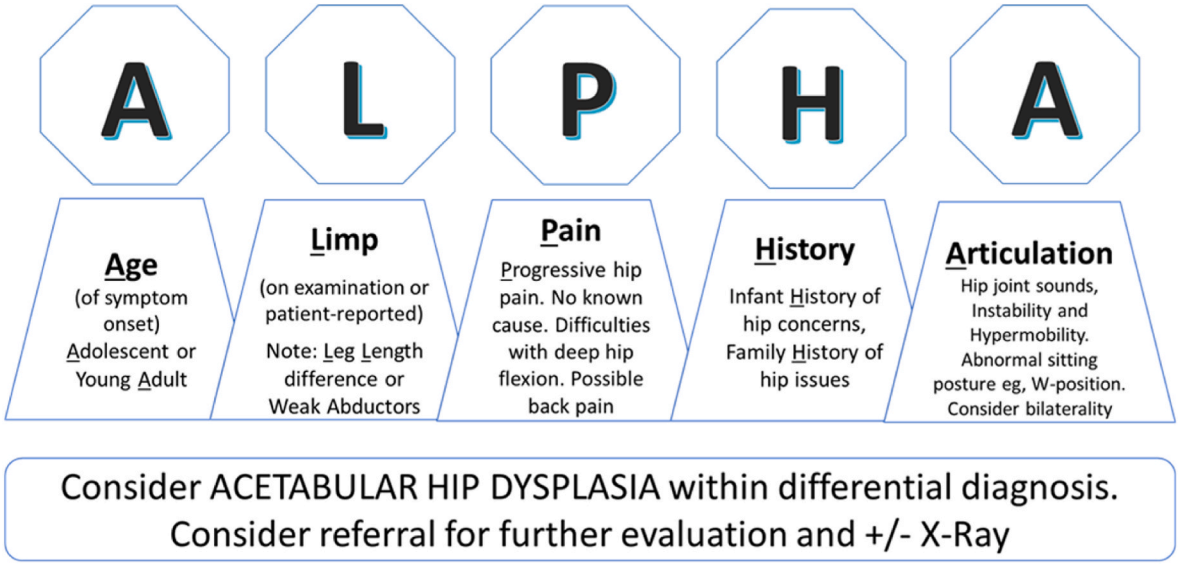

Fig. 1. The ALPHA Hip Dysplasia Alert: A prompt for differential diagnosis.

length discrepancy or pelvic dropping on one side resulting in limp. This latter point adds support to the findings of Liu et al. (2012), who identified an association between AHD and a reduction in cross-sectional area and radiological density of Gluteus Medius. This muscle is essential for gait and hip stability (Palastanga et al., 1998). Whilst cross-sectional and density measures are beyond the typical, pre-X-Ray assessment of patients with hip pain, it is possible that weakness in the Gluteus Medius is identifiable during physical assessment of gait and hip strength.

Our findings support those of Nunley et al. (2011) in that pain onset was most described as gradual, progressing over time and often becoming severe and unrelenting. Activity-related pain was common with aggravating features identified as prolonged walking, standing and sitting. Hip pain characteristics alone are, however, unlikely to be a helpful indicator as they are not specific to AHD. People with a variety of hip conditions, such as OA, FAI and various soft tissue injuries often characterise their hip pain in a similar way (Bisciotti et al). Our study revealed respondents associated their hip pain with the difficulty of cross-legged sitting and any activity involving deep hip flexion, and with features of their hip joint that include hypermobility, hip sounds and joint instability (Theme 5).

A history of childhood hip issues or a family history of hip problems were reported in our study. Siblings of individuals with hip dysplasia have previously been shown to be at least 10 times more likely to have the condition than siblings of non-hip dysplasia families (Li et al., 2013). Moreover, Carroll et al. (2016) identified that more than a quarter (27%) of close relatives of patients with infant DDH have unsuspected radiographic or 'occult' hip dysplasia. These individuals were commonly under 30 years and after this age the majority developed symptoms. Although no definitive evidence of a genetic link has been found, heritable traits have been indicated through patterns of occurrence evident within various ethnic groups. For instance, Corrigan and Segal (1950) estimated a 6% incidence of AHD in a small village population of 1253 from Island Lake, Manitoba, USA for which an hereditary association was identified. By contrast, in their thorough systematic review, Loder and Skopelja (2011) highlighted a near zero (0.06%) incidence of AHD in an African population.

Our study identified three hip joint characteristics that were features of AHD. Firstly, hypermobility, also referred to as hyper-flexibility, hyper-laxity or being 'double-jointed' was reported as being associated with AHD by some 45% of Phase 2 respondents. This concurs with previous studies that have shown AHD to occur more frequently in patients with hyper-flexibility, generalised laxity and shoulder instability (Bilsel et al., 2016). Additionally, patients with a history of hip dysplasia were five times more likely to present with a flexible flat foot, compared to those without hip dysplasia (Samper et al., 2015). Our study showed an association between hypermobility and uncomfortable hip joint instability, which included recurrent hip joint dislocation, subluxation or the feeling of the "hip popping out of its socket" which was reported by almost three-quarters of respondents (71%). Many opinion pieces and descriptive reports have suggested the value of conducting physical tests for hip instability in the evaluation of hip pain in young adult patients (Schmitz et al., 2020; Nepple et al., 2017), yet patient-reported symptoms of joint instability in AHD have not commonly been reported in the literature. In a review of 194 hip arthroscopies however, Hoppe et al. (2017) evaluated 3 physical tests of hip instability, the abduction–hyperextension–external rotation (AB-HEER) test, the prone instability test and the hyperextension–external rotation (HEER) test. The tests demonstrated high sensitivity and specificity, accurately reflecting the reference standard of intraoperative instability, particularly when the findings of all 3 tests were combined. However, negative test results could not confirm the absence of hip instability and the study suggested that diagnosis would require the application of other assessment measures. Our findings provide insight into the presentation that people living with AHD report. The respondents' descriptions of hip instability concur with X-Ray evidence relating to the anatomical abnormalities in AHD, where a deficiency of the bony acetabulum results in poor femoral head coverage that compromises hip stability (Troelsen 2012). The third notable feature of hip joint characteristics was that respondents describing hip joint instability frequently related the symptom to a clunk, click or pop. Joint locking, snapping or popping have previously been reported (Nunley et al., 2011) but, as a symptom of AHD, there is no mention that these features might accompany hip joint instability.

Whilst the signs and symptoms listed within each theme improve the understanding of AHD presentation, similar symptoms are described in other hip disorders. It is therefore vital for AHD to be considered within the differential diagnosis in patients presenting with hip problems. The ALPHA alert mnemonic (Fig. 1) should help to increase clinical suspicion of AHD and referral for X-Ray should be considered. Future studies will test both the wider agreement amongst patients of how ALPHA reflects their AHD presentation and the effectiveness of the ALPHA as a workable alert for AHD and differential diagnosis.

4.1. Limitations

The first possible limitation is that participants who had received joint surgery for their hip dysplasia were asked to report their pre-surgery signs and symptoms. The mean time since surgery for those disclosing the relevant date was 6.6 years (Table 1). The accuracy of these participants' recall may therefore have affected what was reported, but as alluded to previously, troublesome symptoms for which participants had clear memories were considered key to building a picture of AHD features. Other potential limitations include recruiting participants from online hip dysplasia patient support groups as it assumes access to technology. Poor digital literacy and digital deprivation may have excluded some members of the AHD population. The biggest age group of active new media users (70% worldwide) are, however, young adults in the 18–44 year-old age category (Statista 2020, which reflects the age at which people most commonly report their AHD onset (Nunley et al., 2011) and have hip surgery (Clohisy et al., 2007, 2009). The use of these groups and platforms was an effective method of recruiting appropriate participants and enabled an international representation of AHD participants. Phase 2 demographics show however, that 90% of participants were in fact British. The authors acknowledge that this limits the global generalisability of the findings. Also, whilst females have a higher AHD prevalence rate when compared to males, having only two male participants in each phase of our data collection limits evidence of the male perspective. Whether this is because males are less likely to use support groups or that they are less likely to respond to a survey is unknown. Additionally, people accessing patient support groups could be limited to those experiencing on-going problems, evading those with AHD who may have received timely and effective intervention. Nonetheless, the purpose of this study was to address the problems of delayed recognition and misdiagnosis. Details related to the chronicity of the problems from patients living with the consequences of delayed diagnosis of AHD, and the pattern of symptom development over time were therefore important to identify. Hence, people living with long-term problems of AHD were considered key informants.

5. Conclusions

Evidence increasingly points to the importance of early AHD diagnosis for achieving successful surgical outcomes and the need for a better understanding of AHD has previously been highlighted. This study has drawn on the experiences of people living with AHD to develop a mnemonic for increasing diagnostic awareness of the condition, facilitating timelier referral of patients for diagnostic X-Ray to support effective treatment. AHD can have lifelong consequences and if management effectiveness is to improve, clinicians need to have a better understanding of AHD during their examination of young patients with chronic hip pain. ALPHA may facilitate timelier referral of patients for

diagnostic X-Ray and appropriate treatment. Future studies should evaluate its clinical utility.

Declarations of interest

None.

Ethical

The study received ethical approval from the institutional review board and the relevant Health Board's Research and Development Research Review Committee (Reference: RD/1527/17).

CRediT authorship contribution statement

Elizabeth M. Evans: Writing – review & editing, Writing – original draft, Methodology, Investigation, Formal analysis, Conceptualization. **Shea Palmer:** Writing – review & editing. **Tina Gambling:** (deceased), Supervision. **Valerie Sparkes:** Writing – review & editing. **Davina Allen:** Writing – review & editing, Supervision.

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