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THE RELATIONSHIP BETWEEN KINAESTHESIA, MOTOR PERFORMANCE, PHYSICAL FITNESS AND JOINT MOBILITY IN CHILDREN WITH AND WITHOUT JOINT HYPERMOBILITY IN NIGERIA

by

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Declaration

I, Ebuka Miracle Anieto, hereby declare that the work on which this dissertation/thesis is based is my original work (except where acknowledgements indicate otherwise) and that neither the whole work nor any part of it has been, is being, or is to be submitted for another degree in this or any other university.

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Abbreviations

AMEDA	Active Movement Discrimination Assessment
ANCOVA	Analysis of Covariance
BJHS	Benign Joint Hypermobility Syndrome
BMI	Body Mass Index
BOT-2	Bruininks-Oseretsky Test of Motor Proficiency 2
CENTRAL	Central Register of Controlled Trials
CINAHL	Cumulative Index to Nursing and Allied Health Literature
DCD	Developmental Coordination Disorder
EDS	Ehlers Danlos Syndrome
EMBASE	Excerpta Medica Database
GJH	Generalized Joint Hypermobility
HSD	Hypermobility Spectrum Disorder
H/Q	Hamstring/Quadriceps
ICF	International Classification of Functioning, Disability and Health
JPS	Joint Position Sense
KOOS	Osteoarthritis Outcome Score
MABC-2	Movement Assessment Battery for Children-2
MAND	McCarron Assessment of Neuromuscular Development
MEDLINE	Medical Literature Analysis and Retrieval System Online
NM	Normal Mobility
NSMDA	Neurological Sensory Motor Developmental Assessment
PAR-Q	Physical Activity Readiness Questionnaire
PDMS-2	Peabody Developmental Motor Scales 2
PERF-FIT	Performance and Fitness battery
PRFD	Peak Rate of Force Development
PVJD	Peak Vertical Jump Displacement
SIS	Skill Item Series
TGMD-2	Test of Gross Motor Development 2
WHO	World Health Organization

ABSTRACT

Background: Some children with generalized joint hypermobility (GJH) develop symptoms at some point, which could negatively affect their overall health status. When GJH presents with symptoms, it is described as benign joint hypermobility syndrome (BJHS). Some of the symptoms may include pain, soft tissue injuries, and early onset osteoarthritis. The factors that may predict the development of symptoms in people with GJH have not been established. It is important to explore and identify the factors that modulate the clinical outcomes of children with joint hypermobility, and the factors that predispose some children to developing BJHS). Exploring these factors will help in establishing indicators to observe in longitudinal studies to identify causality, and in developing interventions that will be specifically targeted at influencing those modulators.

Aims: to determine the relationship between kinaesthesia, motor performance, fitness and joint mobility in children, and to also determine if kinaesthesia, motor performance, fitness are different in children diagnosed with GJH compared to those who have normal mobility.

Methods: a cross-sectional, analytical study was conducted involving children from two primary schools in South-Eastern part of Nigeria. The Beighton criteria were used for the classification of GJH, while using a cut-off of ≥ 6 out of the 9-maximum score. The children were allocated into two groups: children with GJH, and children with normal mobility (NM). Motor performance, fitness and kinaesthesia were measured in all the children. Motor performance and fitness were measured using the performance and fitness battery (PERF-FIT), while kinaesthesia was measured using wedges. Partial correlation was used to evaluate the relationship between the outcomes, while controlling for age and BMI. The non-parametric ANCOVA test (Quade's test) was used to evaluate the differences in the outcomes (motor performance, fitness, and kinaesthesia) between children with GJH and children with NM, while also controlling for age and BMI.

Results: A total of 91 children (51.6% girls, and 48.4% boys) participated in the study. The mean age of the children was 8.20 ± 1.98. GJH was identified in a total of 35 (38.46%) children, while 56 (61.54%) children had normal mobility. GJH was more frequent in females (60.0%) than in males (40.0%). There was no statistically significant correlation between joint mobility and kinaesthesia. There was also no statistically significant correlation between joint mobility and motor performance items, as well as the fitness items. There was a statistically significant positive correlation between kinaesthesia and some motor performance items including ball bounce, ball throw and dynamic balance, as well as a significant negative correlation between kinaesthesia and one fitness item- ladder run. Furthermore, there was a statistically significant positive correlation between age and kinaesthesia (correct wedges discrimination). The study also showed that motor performance items, kinaesthesia, and most fitness items, did not differ significantly between children with GJH and children with NM.

Conclusion: Joint mobility may not have a significant influence on motor performance and fitness in children that are still at their early stage of growth. Kinaesthesia may be an important factor to consider in children, as it had significant correlations with some motor performance and a fitness item. Furthermore, kinaesthesia is better when the children are older.

Keywords: Generalized Joint Hypermobility, Benign Joint Hypermobility Syndrome Kinaesthesia, Motor Performance, Physical fitness

CHAPTER 1: Introduction

Joint hypermobility is a condition characterized by excessive passive and/or active range of motion of the joints beyond normal limits along physiological axes (Engelbert et al., 2005; Rombaut et al., 2010). This could be peripheral (limited to the hands and feet), localized (involving a single joint) or generalized (involving multiple joints), referred to as Generalized Joint Hypermobility (GJH) (Castori et al., 2017; Smits-Engelsman et al., 2011). GJH is commonly associated with some genetic disorders such as Ehlers-Danlos syndrome, Marfan syndrome and Osteogenesis imperfecta (Tofts et al., 2008), however, it could be present in the absence of the listed disorders (Castori et al., 2017). GJH has been reported in the literature using different terms. Notably, GJH that is accompanied with musculoskeletal symptoms such as pain was clinically referred to as benign joint hypermobility syndrome (BJHS), provided that the individuals have no signs of any known diseases that could be of rheumatological, neurologic, skeletal or metabolic origin (Remvig et al., 2007). Furthermore, Castori et al. (2017) classified joint hypermobility into three categories which are a) individuals with asymptomatic GJH, b) individuals with a well-known syndrome which is accompanied by GJH (e.g., the hypermobility type of Ehlers Danlos Syndrome), and c) individuals with symptomatic GJH who did not meet the criteria for a syndrome, which they named hypermobility spectrum disorder (HSD).

GJH is mostly classified using the Beighton criteria (Beighton et al., 1973). The Beighton criteria, originally developed as a screening tool classifies GJH based on the individual's score out of a maximum of 9-points from the following tests; passive thumb apposition to the flexor part of the forearm, passive elbow hyperextension for beyond 10 degrees, passive knee hyperextension for beyond 10 degrees, passive little fingers dorsiflexion for beyond 90 degrees, and forward trunk flexion, with the knees straightened while placing the palms to easily rest on the floor (Beighton et al., 1973).

GJH, which is usually idiopathic, is reportedly present in about 14% of the children within the age bracket of 4 to 7 years in the Netherlands (Larsson et al., 1987; Rikken- Bultman et al., 1997; Wordsworth et al., 1987). The prevalence of GJH, measured using the Beighton criteria was reported to be very high (43% of individuals studied) in a West African population (western

Nigeria) of individuals aged 6 to 66 years (Birrell et al., 1994). GJH has been reported to be more prevalent in females than in males (Birrell et al., 1994; Cheng et al., 1991; Jansson et al., 2004; Larsson et al., 1993; Wordsworth et al., 1987).

Over time, some children with GJH develop symptoms (a sign of BJHS/HSD), such as pain from ligaments or soft tissues, joint subluxation, generalized fatigue, skin striations and papyraceous scar tissue formations on the skin (Scheper et al., 2013b; Grahame, 1990). A decrease in bone mineral density and blood pressure as well as an increased skin extensibility and fatigue are some other clinical features seen in children with BJHS/HSD (Engelbert et al., 2003). As a result of the pain and joint instability associated with BJHS/HSD, the individuals have a higher risk of joint damage for example dislocations, premature osteoarthritis (Beighton et al., 2011) and abnormal postures given to the abnormal weight-bearing on the joint articular surfaces (Booshanam et al., 2011). It is important to evaluate the factors that modulate the progression of the usually asymptomatic GJH to BJHS/HSD, which could present with serious symptoms.

1.1 Generalized joint hypermobility and Kinaesthesia

GJH accompanied with symptoms may have negative neurological implications (Rombaut et al., 2010). One of the major neurological implications is impairment of the conscious sense of active or passive movements and the direction of the movement, known as kinaesthesia (Rombaut et al., 2010). Kinaesthesia is a sense of position, movement, weight, force, effort, pressure, segment (size/shape) of the body, and balance (Stillman, 2002). Some authors have attempted to define kinaesthesia as movement sense, and proprioception as either position sense or both, which do not take the other senses (e.g., effort, pressure etc.) into account. Therefore, it is safer to consider kinaesthesia and proprioception as the same (Stillman, 2002). The kinaesthetic system is important in maintaining joint stability (Riemann & Lephart, 2002), hence, kinaesthetic deficits may predispose individuals with symptomatic GJH to more severe musculoskeletal injuries (Rombaut et al., 2010). Reduced proprioception leads to a poor detection of the body position during movement, which could lead to falls and abnormal postures during the performance of functional activities (Ribeiro & Oliveira, 2007). A study involving a population of adults reported that the proprioceptive feedback in individuals with symptomatic GJH is significantly decreased, signaling a neurophysiological impairment (Sahin et al., 2008), which is attributed to diminished muscle spindle activation by the γ-motoneurone (Hurley et al., 1997). Similarly, reduced knee proprioception was reported among children between 9-13 years with symptomatic GJH (Fatoye et al., 2009). A systematic review conducted by Smith et al. (2013) also revealed that individuals with symptomatic GJH present with a statistically significant poorer lower limb joint position sense (JPS) ($p<0.001$) and threshold detection to movement ($p<0.001$) than those with normal mobility. Perceived effort, a function of kinaesthesia, is also impaired in individuals with symptomatic GJH (Smith et al., 2013). There is a paucity of studies that investigated kinaesthesia in individuals with asymptomatic GJH.

1.2 Generalized joint hypermobility and motor performance

GJH may lead to an impairment in motor performance (Engelbert et al, 2005). Some studies reported that children with GJH present with motor delay before they reach two years, while some children have persistently a significant motor delay (both fine and gross motor skills) when tested at the age of 5 years (Jaffe et al., 1988; Tirosh et al., 1991). A number of explanations to the motor deficits have been reported. They include the association of GJH with congenital benign hypotonia (Murray & Woo, 2001), reduced proprioception (Hall et al., 1995), and muscle weakness (Hanewinkel-van Kleef et al., 2009). A study involving 59 infants that were 18 months of age who were followed up for 3.5 years reported that fine and gross motor skills were delayed in those with GJH (Tirosh et al., 1991). The common motor skills impaired in children with GJH include balance, hopping, running, performing jumps, easy change of direction during movement, limiting their ability to participate in sports (Adib et al., 2005; Murray, 2006).

1.3 Generalized joint hypermobility and physical fitness

Children with symptomatic GJH have been reported to present with reduced physical fitness (Engelbert et al., 2006). Absolute peak oxygen consumption and relative peak oxygen, which are outcomes for measuring exercise capacity were found to be decreased in children with symptomatic GJH (Engelbert et al., 2006). Symptomatic GJH was also independently associated with fatigue, physical deconditioning in a group of professional dancers who benefit from being hypermobile, despite their regular training (Scheper et al., 2013a). The reduced physical fitness reported in children with symptomatic GJH may be as a result of the deficits in motor skills which leads to activity limitations (leading to deconditioning) or due to pain (Engelbert et al., 2006). Studies reporting the relationship between GJH (both symptomatic and asymptomatic), and physical fitness are scarce.

1.4 Study aims and objectives

1.4.1 Aim

The overall aim of the study was to determine the magnitude and direction of the relationship between kinaesthesia, motor performance, physical fitness and joint mobility in children. This is to ascertain whether these factors modulate the progression of GJH to BJHS/HSD in children in future longitudinal studies.

1.4.2 Objectives

The specific objectives of the study were:

1. To determine the relationship between joint mobility, kinaesthesia, motor performance and physical fitness in children.
2. To determine the relationship between kinaesthesia and age in children.
3. To determine the differences in motor performance, physical fitness and kinaesthesia between children with and without GJH.

1.5 Study hypothesis

1. There will be a significant correlation between joint mobility, kinaesthesia, motor performance and physical fitness in children.
2. There will be a significant correlation between kinaesthesia and age in children.
3. There will be significant differences in the motor performance, physical fitness, and kinaesthesia between the two groups of children with and without GJH.

1.6 Significance of the study

Since GJH usually presents with no symptoms or discomfort, the hypermobility and co-occurring microtrauma are often missed during clinical assessment (Fatoye et al., 2008). This has not been helpful, as some children with GJH develop limiting symptoms over time, and some developing BJHS (Scheper et al., 2013b). Some factors may modulate the progression of joint hypermobility, and an early intervention targeted at those factors would be beneficial in preventing the progression of the condition. Several studies have flagged reduced kinaesthesia as a factor that impacts the clinical outcome of individuals with joint hypermobility (Rombaut et al., 2010; Scheper et al., 2013b). Reduced kinaesthesia has also been associated with decreased motor coordination and physical fitness (Scheper et al., 2013b). The overall clinical outcomes associated with joint hypermobility may result in activity limitations and participation restrictions. It is important to explore and identify the factors that modulate the clinical outcomes (motor performance, and fitness) of children with GJH. This will facilitate identifying childhood factors that predispose individuals to chronic conditions such as osteoarthritis, poor coordination, joint dislocation, reduced fitness, etc. Given the high prevalence of GJH in Nigeria (Birrell et al., 1994), it was important to conduct this study within the Nigerian context to raise awareness to promote early identification and management of GJH symptoms. This study therefore added to the body of evidence on the relationship between kinaesthesia, motor performance, physical fitness and joint hypermobility, which may provide directions for future studies.

CHAPTER 2: Literature Review

2.1 Introduction

The literature review targeted at identifying studies that conceptualized Generalized Joint Hypermobility, Benign Joint Hypermobility Syndrome, Kinaesthesia/proprioception, motor performance and physical fitness. The review also explored the existing knowledge on the relationship between these concepts to identify the gaps in literature, which informed the direction of this research project. However, a systematic approach was not used.

A literature search was carried out in the following databases; MEDLINE (PubMed), Cochrane CENTRAL, CINAHL, EMBASE and Google Scholar. References of identified studies were also searched. The keywords that were used for the search included; kinaesthesia, proprioception, movement sensation, position sense, motor performance, motor skills, motor activity, physical fitness, physical functional performance, hypermobility, joint laxity, benign hypermobility syndrome, Ehlers-Danlos syndrome type 3, BJHS, GJH.

2.2 Conceptual framework

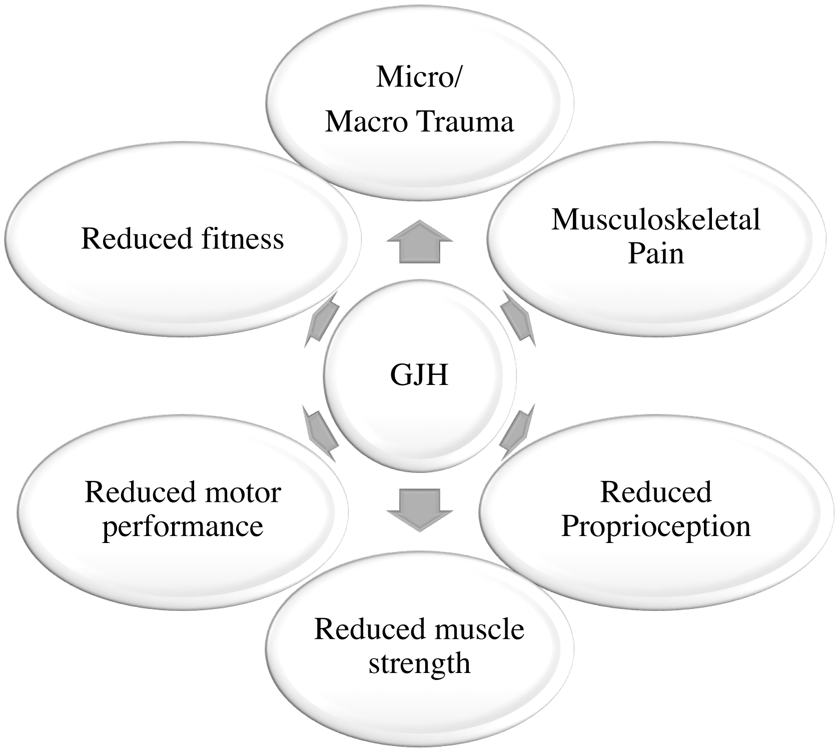

Figure 1: Conceptual framework showing the link between the variables of interest (Castori et al., 2017).

2.2.1 Generalized Joint Hypermobility

Generalized Joint Hypermobility (GJH) is a hereditary disorder characterized by the ability to move the joints of the body beyond the normal range of motion (Juul-Kristensen et al., 2017). It is tagged as 'generalized' depending on the number of joints involved. Clinical terms such as joint-laxity, double-jointedness, joint hyperlaxity and joint instability are used synonymously with GJH (Castori et al., 2017). In addition to the inherited types of GJH, there are acquired types that involve broad inflammatory or degenerative joint illnesses, musculoskeletal tissues, and peripheral nerves, in addition to hypothyroidism and some other endocrine abnormalities. Also of note is that malnutrition in children may be a contributing factor to secondary GJH occurrence (Hasija et al., 2008). The British Society of Rheumatology denoted the Beighton classification criteria of positive tests in four or more out of nine tests as an acceptable diagnosis of GJH (Remvig, Jensen, & Ward, 2007). GJH is sometimes linked with musculoskeletal features such as pain, which Simmonds, & Keer (2007) highlighted as hypermobility syndrome (HS). Some authors refer to the associated musculoskeletal complaints as ‘Benign’ joint hypermobility syndrome (BJHS) to contrast the condition with more severe and sometimes life-threatening conditions with similar hypermobility presentations such as the Ehlers-Danlos syndrome (EDS), Marfan syndrome, osteogenesis imperfecta, (Grahame, 2000). According to the Villefranche nosology, the Ehlers-Danlos syndrome (hypermobility type) has similar presentations as GJH, however, it is also accompanied with features such as smooth, soft and fragile skin, and musculoskeletal symptoms (Beighton et al., 1998). The other types of EDS such as the classical and vascular EDS, also present with GJH, however, have more cutaneous involvement than the hypermobility type (Tinkle et al., 2017).

2.2.1.1 Associated symptoms of Generalized Joint Hypermobility

Children with GJH are usually without symptoms. However, a substantial body of evidence supports the development of a variety of musculoskeletal features and problems that may be regarded to as secondary presentations of GJH (Castori et al., 2017). Some of the symptoms when GJH becomes symptomatic include the following:

a. <u>Soft tissue injuries</u>

Due to the excessive movements in hypermobile joints, they may be prone to macro- and microtrauma. The macrotrauma may include but not limited to subluxations, dislocations, and other injuries of the soft tissues (e.g., muscle tear, and damages to either the tendons, ligaments, synovium, and cartilages) (Castori et al., 2017). The macro damages are usually as a result of single or repetitive trauma to the joints given to the beyond normal range of motions along non-physiological axes, which may be exacerbated by joint instability (Castori et al., 2017). Macrotrauma is often characterised by intense pain, function loss, and the need for immediate medical attention. Small, subtle, and insinuating injuries are known as microtrauma, and they are often not seen by the subject or the healthcare providers when they occur. The risk of recurring or persistent pain, as well as the possibility of early joint deterioration, may increase with time (i.e., early osteoarthritis) due to chronic microtrauma (Castori et al., 2017). Though it is well known that GJH is a risk factor for musculoskeletal-related pain, neither chronic pain nor early onset osteoarthritis are universally considered to be mandatory complications of GJH. Recurrent microtrauma and occasional/recurrent macrotrauma can also result to localised joint problems such as temporomandibular joint issues (De Coster et al., 2005) and hip labral tear (Groh and Herrera, 2009).

b. Long-term pain

As a result of having a predisposition to trauma, occasional and recurring musculoskeletal pain is sometimes associated with GJH, which is then referred to as BJHS (Remvig, Jensen, & Ward, 2007). It is possible for GJH to lead to chronic pain that lasts for years or even decades (Castori et al., 2016). Hyperalgesia may be a form of pain sensitization in people who have Ehlers-Danlos Syndrome (EDS) (hypermobility type) and long-term pain, according to some early research (Di Stefano et al., 2016). Cazzato et al (2016) found that individuals with common EDS types (hypermobile type, classical type, and vascular type) had a high rate of small fibre neuropathy, which warranted the thought that a dysfunctional connective tissue and abnormal pain processing could be linked.

c. Reduced proprioception

GJH is usually associated with decreased proprioception in certain joints (Smith et al., 2013) and reduced muscle strength, especially in symptomatic persons (Rombaut et al., 2012; Scheper et al., 2016). Decreased proprioception and reduced muscle strength have a strong relationship and may create a vicious cycle of growing limitations in daily activities in people with GJH. Although the mechanisms underlying the link between impaired proprioception, reduced muscle strength, and GJH are unknown, their co-existence should be put into consideration in developing rehabilitation approaches (Scheper et al., 2016). Furthermore, the combination of GJH and its neuromuscular characteristics may also affect children's motor performance. Some studies reported a link between GJH and developmental co-ordination disorder (DCD) (Ghibellini et al., 2015). The pathogenesis of DCD is also unknown, although one explanation could be the interaction of impaired proprioception and reduced muscle strength during the formation of representations of movements (Wilson et al., 2013).

d. Psychological symptoms

A systematic review reported that people with GJH have a higher risk of developing anxiety, panic disorders and depression compared with individuals with normal mobility (Smith et al., 2014).

Furthermore, a population-based nationwide cohort study reported an increased incidence of psychiatric disorders which includes depression, anxiety, and attention deficit disorder in the population of individuals with joint hypermobility (Cederlof et al., 2016). Cederlof et al (2016) explained that the burden of the disease (hypermobility syndrome) may lead to a low quality of life, which could lead to the risk of depression. They also explained that the other psychiatric disorders could have a genetic influence.

e. Other musculoskeletal presentations

Persons with GJH frequently exhibit a number of minor musculoskeletal characteristics, which could be as a result of interplays between soft tissues and other mechanical forces (e.g., body weight, impact of gravity, recumbent preferred position, and sporting activities) during the early stages of growth (Castori et al., 2017). Some of the musculoskeletal characteristics include flexible pes planus, valgus deformities (at the elbows, hind-feet, and halluces), mild to moderate degree scoliosis, exaggerated kyphosis and lumbar lordosis, as well as plagiocephaly (Morlino et al., 2016; Tinkle, 2010). Some GJH-related genetic disorders are linked to a significant decrease in bone mass, a higher risk of fractures and lengthy bone abnormalities. The pleiotropic effect of the causal gene obscures any pathogenic link between GJH and low bone mass in these circumstances. In phenotypes where the decline in bone mass is very minimal, it is difficult to classify it as a pleiotropic effect. In this case, the loss of bone mass is usually milder, not clearly linked with high fracture risk, and could the results of so many factors (Dolan et al., 2003; Gulbahar et al., 2006). It could be partly related to the impaired proprioception/kinaesthesia, reduced muscle strength, and reduced activity that frequently characterise GJH regardless of the underlying cause (Castori et al., 2017).

2.2.1.2 Pathogenesis of GJH

A specific pathophysiologic mechanism by which individuals develop GJH and BJHS have not been determined due to the varied clinical features and lack of reliable biomarkers. Given that only a small percentage of patients with GJH develop musculoskeletal symptoms, reduced proprioception and reduced muscle strength, a variety of genetic and environmental factors may play a role in its development (Kumar & Lenert, 2017).

a. Genetic factors

The involvement of genetic factors in the development of GJH and BJHS is debatable, and it is changing as genetic testing becomes more widely used in research and clinical settings. Although some pedigree investigations have revealed a weak, autosomal dominant form of inheritance with varied clinical expression, the great majority of cases do not seem to be connected to any detectable mutation (Castori et al., 2017). A mutation in the TNXB gene, which codes for the extracellular matrix glycoprotein (Tenascin X), has been found in less than 10% of cases. Skin hyperextensibility, velvety skin, and being easily bruised are more common in these patients (Bristow et al., 2005). Several uncommon mutations have been detected in certain patients with BJHS, but the clinical value of genetic tests for them has not been determined, in contrast to Ehlers-Danlos Syndrome etc. (Chiarelli et al., 2016).

b. Environmental factors

A study highlighted the importance of localised biomechanical overloading and chronic injuries of the soft tissues related to joint laxity and instability in addition to the genetic factors, due to assumed differences in collagen composition (Kumar, & Lenert, 2017). Collagens are vital components of the ligaments, tendons, bones, and skin which are usually reduced or altered in individuals with the hypermobile type of EDS (Hermanns-Lê, & Piérard, 2007). Hence, repetitive microtrauma can produce kinematic changes, which can lead to joint overload and soft tissue injury, resulting in arthralgias and musculoskeletal pain (Kumar, & Lenert, 2017). Studies have also shown that altered gait and posture have a broader impact. Patients with BJHS have reduced joint momenta in the lower extremities, needing more force to maintain stability, according to two investigations comparing patients with asymptomatic GJH to those with BJHS (Chiarelli et al., 2016; Nikolajsen et al., 2013).

2.2.1.3 Epidemiology/Prevalence of GJH in children

There is no agreement on the Beighton cut-off score for the diagnosis of GJH, whilst some studies placed the cutoff as ≥ 4 joints involvements others placed it as ≥ 5 joints involvements (Smits-Engelsman et al., 2011). This has led to a wide disparity in the prevalence of GJH reported by several studies. A study including 6,022 children in the United Kingdom reported a prevalence of

27.5% and 10.6% in females and males respectively, when the Beighton cut-off was placed as ≥ 4 affected joints (Clinch et al., 2011). The study also reported a statistically significant relationship between hypermobility in females and outcomes such as body mass index, physical activity, and maternal education, whilst there was no significant association in males (Clinch et al., 2011). Another study including 778 school-aged children from east-central Europe reported a prevalence of 19.2%, 9.5% and 5.7% for Beighton cut-offs of ≥ 4, ≥ 5, and ≥ 6 respectively (Gocentas et al., 2016). Smits-Engelsman et al (2011) reported a prevalence of 9.1% and 35.6% for Beighton cut-offs of ≥ 7 and ≥ 5 respectively in a study that involved 551 children in Dutch elementary schools. Notably, a study conducted in a West African population (western Nigeria) including 204 individuals within the age range of 6 to 66 years recorded a very high prevalence of 43%, using a Beighton cut-off of ≥ 4 (Birrell et al., 1994). A study from South Africa included 1081 individuals of all ages reported a prevalence of 20% in females and 6% in males. The study did not set any criterion but considered Beighton scores of 0, 1, or 2 as normal scores (Beighton et al., 1973). Literature on the prevalence of GJH in African populations is limited. It is recommended that more studies should be conducted using a higher cut-off Beighton score.

2.2.1.4 Diagnosis of Generalized Joint Hypermobility

The following three methods are common for the assessment and diagnosis of GJH: Beighton criteria (Beighton et al., 1973), Carter and Wilkinson criteria (Carter & Wilkinson, 1964), Rotes-Querol method (Rotes-Querol, 1957). Other assessment methods include that of Kirk et al (1967), Beighton and Horan (1970), Beighton et al (1997) and Grahame et al (2000).

a. Beighton criteria (Beighton et al., 1973)

They described GJH as having a high score out of a maximum of 9 points of the following items;

- Passive thumb apposition to the flexor part of the forearm. A score of 2 is given when it's positive bilaterally, and 1 if it is on one side.
- Passive elbow hyperextension for beyond 10 degrees. A score of 2 is given when it's positive bilaterally, and 1 if it is on one side.
- Passive knee hyperextension for beyond 10 degrees. A score of 2 is given when it's positive bilaterally, and 1 if it is on one side.

- Passive little fingers dorsiflexion for beyond 90 degrees. A score of 2 is given when it's positive bilaterally, and 1 if it is on one side.
- Forward trunk flexion, with the knees straightened while placing the palms to easily rest on the floor. A score of 1 is given if it's positive, and 1 if it is on one side.

There was no specified cut-off score. However, the investigators recognized lower scores such as 0, 1, 2 as normal. Figure 2 shows the various items of the Beighton criteria.

b. <u>Beighton et al. (1998)</u>

The authors recommended a score of ≥ 5 positives tests out of the 9 score criteria reported by Beighton, Solomon & Soskolne (1973).

b. <u>Carter and Wilkinson criteria (1964)</u>

The authors described GJH as scoring > 3 out of 5 tests. The tests include:

- Passive thumb(s) apposition to the flexor part of the forearm.
- Passive fingers hyperextension to the point that they could lie parallel to the extensor part of the forearm.
- Passive hyperextension of the elbow(s) for > 10 degrees.
- Passive hyperextension of the knee(s) for > 10 degrees.
- Passive dorsiflexion of the ankle, and foot eversion in excess of the normal range (estimated at 18 degrees).

c. <u>Rotes-Querol (1957) method</u>

Rotes-Querol (1957) described 10 tests and categorized GJH in grades. Scores of 0-2 = Grade I; 3-5 = Grade II; 6-7 = Grade III; 8-10= Grade IV. The 10 tests include:

- Passive thumb(s) apposition to the flexor part of the forearm > 185 degrees.
- Passive hyperextension of the elbow(s) for > 10 degrees.
- Passive hyperextension of the knee(s) for > 5 degrees.
- Passive dorsiflexion of the second finger to the extent that the angle between the distal phalanx and the support-table is > 100 degrees.

- Forward trunk flexion, with the knees straight while placing the palms to easily rest on the floor.
- Shoulder external rotation > 90 degrees.
- Rotation of the cervical vertebrae > 90 degrees and side flexion of the cervical vertebrae > 50 degrees.
- Bilateral hip abduction > 90 degrees.
- Dorsiflexion of the metatarsophalangeal joint > 90 degrees.
- Lumbar vertebra lateral flexion with the head and column below the horizontal plane.

e. <u>Grahame et al (2000)</u>

They recommended a score of ≥ 4 positives tests out of the 9 score criteria reported by Beighton, Solomon & Soskolne (1973).

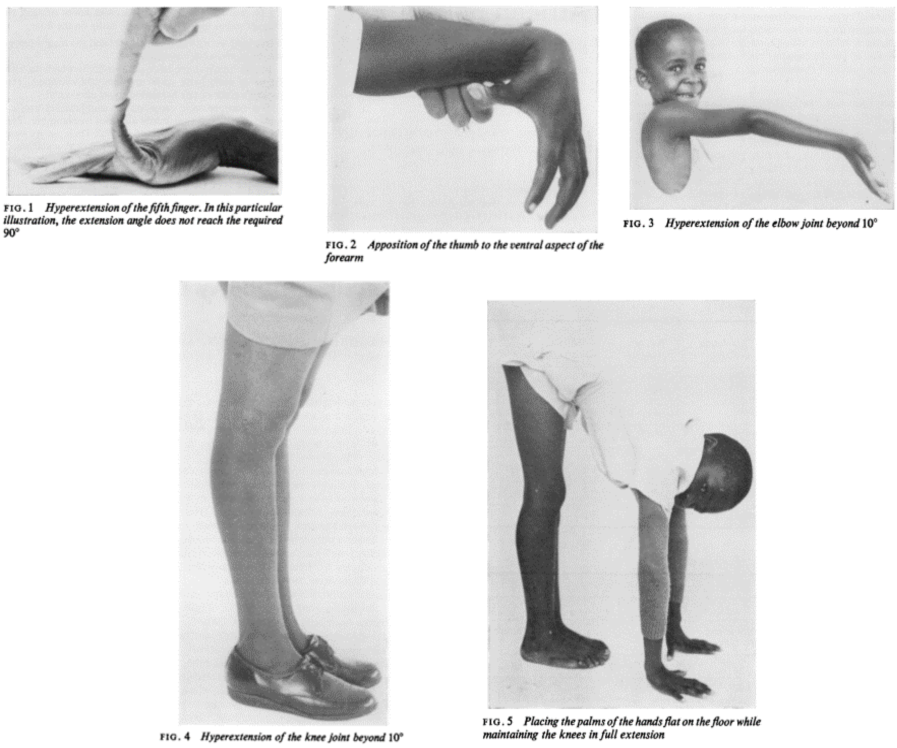

Figure 2: The Beighton criteria (Beighton, Solomon & Soskolne, 1973)

2.2.1.5 Treatment of symptomatic GJH

Given to the symptoms that could be associated with GJH, the International Classification of Functioning, Disability and Health (ICF) has been adopted as a guideline (Atkinson and Nixon-Cave, 2011). The World Health Organisation (WHO) described disability as an encompassing term housing physical function, activity and participation, and environmental and other personal factors (WHO, 2015). Impairments in the ICF domain of body and function may lead to limitations in activity performance and restrictions in participation (e.g., decreased physical activity performance and reduced social participations) in individuals with symptomatic GJH. Therefore, physiotherapy treatment is warranted for this population group.

A randomised comparative trial was conducted to compare a six-weeks generalized physical therapy programme targeted at improving muscle strength and physical fitness via a physiotherapy programme with the aim of correcting the motion-control of joints with symptoms in children with GJH (Kemp et al., 2010). The study showed a significant improvement in both the children's and parents-assessed pain scores in the intervention group. Furthermore, a randomized controlled trial (RCT) including adults with BJHS reported a significant improvement in proprioception and knee pain in the intervention group that received proprioceptive, balance and plyometric exercises, compared with the control group that did not receive exercises (Sahin et al., 2008). Reduction in pain may result to an improved proprioception because pain could hinder the performance of proprioceptive tasks (Ferrell et al., 2004).

A guideline from the American College of Foot and Ankle Surgeons also recommended the use of orthotics and/or sensible footwear for children with flexible pes planus associated with pain in the background of GJH (Evans & Rome, 2011). It was reported that the use of orthotics could enhance the gait efficiency in children with GJH (Evans & Rome, 2011; Morrison et al., 2013). The use of foot orthoses facilitated a reduction in cadence and increase in double support duration, and consequent enhancement of stability during gait performance in children with DCD presenting with joint hypermobility (Morrison et al., 2013).

A few cohort studies including children and adults with GJH have reported that strength exercises, core stability exercises, endurance training, and patients' education could improve perceived performance of activities of daily living, muscular strength, endurance, proprioception, pain and a significant decrease of kinesiophobia in individuals with GJH and BJHS (Bathen et al., 2013; Ferrell et al., 2004; Moller et al., 2014; Rahman et al., 2014).

The evidence on the benefits of exercise for the management of GJH-related symptoms is still growing, and more rigorous trials have been recommended by researchers. An in-depth understanding of the features associated with GJH and how they are related is necessary to allow for a more targeted, outcome-specific management.

2.2.2 Kinaesthesia

Kinaesthesia is a functional sensory aggregation comprised of three distinct sub-senses. The feeling of orientation and position of the limbs and body in space is one of the most mentioned in literature. The second kinaesthetic sense allows us to perceive limb movement, while the third sense allows us to feel the force generated by our own muscles as well as the effort exerted when the muscle force is being generated (Proske, 2006; Proske & Gandevia, 2009). Humans can actively engage with the surroundings, attaining planned body movements, using the information provided by these three basic body senses. Kinaesthetic assessments are critical in evaluating rehabilitation outcomes, especially in clinical practice. Impaired kinaesthesia has been demonstrated to be an essential injury risk factor in sports (Riemann & Lephart, 2002). Measures of kinaesthesia in functional motions, on the other hand, help us understand the impact of practice and other intrinsic elements like weariness (Rosker & Sarabon, 2010).
Protocols for measuring kinaesthesia have been utilised extensively in research and therapeutic practice. Motor control research has aided in the understanding of kinaesthesia in sports. Rather than measuring more complex sensory motor function, no techniques have been devised to isolate the kinaesthetic sense (Rosker & Sarabon, 2010). Proprioception is used interchangeably with kinaesthesia in the literature.

2.2.2.1 Measurement of Kinaesthesia

Measurement of kinaesthesia/propioception is sometimes based on an assessment in the context of purposeful and consciously perceived motion. This could include methods for evaluating limb position in space, movement, perceived effort, and tracking tests (Chung et al., 2006). Some of the assessment methods include:

a. Joint position sense assessment

Position sense tests have been the most commonly utilised kinaesthetic sense assessment instrument (Rosker & Sarabon, 2010). In practise and research, three basic kinaesthetic testing approaches are commonly used. Two of them evaluate the capacity to recognise and replicate a given joint angle, with one being an active joint position reproduction test and the other a passive joint position reproduction test (Alvemalm et al., 1996). The ability to perceive the initiation of limb movement is the subject of the third method (Streepey et al., 2010). The functional aspect of kinaesthetic perception that these three fundamental approaches measure differs. Position awareness may be measured using active and passive joint repositioning methods, while movement sense can be assessed using the third method at varied velocities, directions, and ranges of motion. Using the appropriate approach and protocol design, distinct sensory domains can also be stressed or excluded from the measurement (Rosker & Sarabon, 2010).

As a standard procedure in active joint repositioning methods, it is usual practice to move the limb into a predetermined reference position. Most measurements begin with a precise positioning of the measured limb. Memory of the position of the limb/joint is required of the subject in this test. Thereafter, the limb is returned passively to its initial position. Participants are first shown a reference position and then asked to perform movements into a position to match the reference position (Alvemalm et al., 1996). Active joint position sensation can be measured by comparing these two values. Some authors have proposed functional enhancements. For example, the cooking posture in arm throwing has been employed as a position of reference (Tripp et al., 2009). The participant is then asked to position to whole upper limbs to match that reference position. Using this method, it is possible to determine how much influence different joints have on gross limb kinaesthesia (Tripp et al., 2006). Muscle spindle and Golgi tendon organs aid in the detection of the reference location when the limb is actively moving.

The second technique utilizes passive movement, whereby the participant is expected to point out when he reaches the reference position (Niessen et al., 2009). The clinician or the device begins to move the limb through its range of motion and the participant is expected to stop the movement when the limb position matches the reference position. The inaccuracy or acuity of passive joint position perception is then represented as the discrepancy between the marked position and reference position. The muscles of the evaluated body part stay completely relaxed throughout this testing technique. Despite the fact that proprioceptors in muscles are assumed to be dormant, they might produce inconsistencies in joint position sensing because they will feel stretches or loosening. The sensory error could be caused by an additional sensory information caused by passive stretching and thixotropy (Proske, 2006; Proske et al., 1993). Joint receptors may also have a role in passive joint position sense.

A third concept (Brindle et al., 2010) is the perception of passive movement. The limb is usually placed in the position of interest, such as at the end range or in a functionally relevant position. The movement is then initiated by the evaluation apparatus. The participant is then expected to detect movement as quickly as possible and report it. Stop buttons or position markers are typically utilised to provide exact recording of the position the movement was felt. A measure of passive movement sense is the range of motion the limb travelled before being detected. A variety of passive movement speeds are used in these tests (Brindle et al., 2010). The acuity is improved by using faster movements (Ashton-Miller et al., 2001).

b. Measurement of perceived force/effort

Methods for assessing force perception/perceived effort have been created specifically for the purposes of research and rehabilitation. There are four major ways for assessing force sense that are currently recognised. The first method relies on the participant's ability to apply a wide range of force. Specific force levels are assigned numbers. The participant is asked to describe the level of force he believes he is producing during the force production tasks (Eisler, 1965). The second way involves using the contra-lateral limb to do force matching tasks. The limb being tested (representing the force reference) is under load, and the opposite hand must match the force level as closely as feasible (Cafarelli, 1982). Similar studies utilising only one limb have been conducted, in which the individual was required to create a predetermined degree of force with feedback or without feedback input (Bock et al., 2005; Smits-Engelsman & Duysens, 2008; Smits-Engelsman et al., 2003; Smits-Engelsman et al., 2005; Smits-Engelsman et al., 2008). The third type of test measures the ability to recognise change in force. The force produced is usually influenced by the rise or reduction in force. The sensation of force is represented by the smallest change noticed (Pang et al., 1991). The fourth method employs active tracking techniques to provide functional and active force production measurements (Chung et al., 2006). Sensory drive originating mostly from Golgi tendon organs and cerebral representations of movement is assumed to underpin force perception (Jones, 1995). More functional assessment of kinaesthetic sense can be performed by integrating joint position and force sense measures. The perception of force can be effectively lowered by simply unloading the measured limb, allowing for a simple yet effective technique of measuring the contribution of individual senses to a particular movement. These techniques necessitate the use of specialised equipment.

c. Active Movement Extent Discrimination Assessment

The Active Movement Extent Discrimination Assessment (AMEDA) was created by Waddington and Adams (1999) using the absolute judgement approach to assess participants' ability to discern between ankle inversion angles using proprioceptive information. Active movements are used in the AMEDA testing. Before beginning to collect data, each participant is given a session to become familiar with the AMEDA apparatus. During this session, they are informed that they will be subjected to, for instance, five motion displacement distances, in order, starting with the smallest (moving to position 1) and working their way up to the largest (moving to position 5), three times: a total of fifteen different movements. Following that, participants are required to complete 50 testing trials in which each of the five positions is shown ten times in a random order. Participants are tasked with making a judgement on the ranked number (1, 2, 3, 4, or 5) of every test movement after experiencing it and reverting to the start position, with no feedback on whether or not their judgement is correct. That is, following the familiarisation trials, participants must use their knowledge of the five movement extents to identify each stimulus and make a numeric judgement (1, 2, 3, 4, or 5) distinguishing each perceived stimulus. This is a single stimulus or absolute judgement test, in which a single stimulus is presented on each trial and a single response is given. An average of ten minutes is needed to perform a single joint proprioception assessment (Waddington & Adams, 1999).

The AMEDA technique influenced the method of assessing kinaesthesia in this study, which is the use of wedges to determine the degree of angulation at the ankle joint. The advantage of using the AMEDA technique is that it assesses kinaesthesia in multi-joint motions while involving the muscles (Wang et al., 2021). A notable rationale for using the wedges is that it does not require the use of specialized equipment as compared to other techniques.

2.2.3 Motor performance

A motor skill is a characteristic that requires the muscles of the body to perform specific movements in order to accomplish a specified task (Schmidt, 1976). Walking, running, or cycling are examples of some of these tasks. The nervous system, and skeletal muscles must all work together in order to accomplish these tasks (Schmidt, 1976). The goal of training a motor skill is to enhance the ability to do the skill with a high rate of success, precision, and reduced energy use. Motor performance refers to the act of successfully executing a motor skill or task. Motor learning occurs when a given motor skill is repeatedly practiced, resulting in a considerable improvement in performance. Motor learning is the process of a person's ability to improve a task over time as a result of repeated practice or exposure (Karni et al., 1998). There are two major types of motor skills: the type that demands using of large muscle groups (e.g, running, jumping, kicking etc.) is called gross motor skills, while the type that engages smaller muscle groups to perform finer movements (e.g., tying of shoelace, brushing of teeth, playing a guitar etc.) is called fine motor skills.

2.2.3.1 Measurement of motor performance

There are several existing assessment batteries for estimating motor performance in children. They include Movement Assessment Battery for Children-2 (MABC-2), McCarron Assessment of Neuromuscular Development (MAND), Bruininks-Oseretsky Test of Motor Proficiency 2 (BOT-2), and Test of Gross Motor Development 2 (TGMD-2).

a. <u>Movement Assessment Battery for Children-2 (MABC-2)</u>

The Movement Assessment Battery for Children-2 (MABC-2) (Henderson et al., 2007) is a discriminative and evaluative tool used for measuring gross motor skills, fine motor skills, and balance. The components tested include aiming and catching, static and dynamic balance. It uses the traffic light system to rate children's motor performance as; green= normal, amber= at-risk, and red=definite motor impairment (which is a score <15%). It can be used for children between three to 16 years old. The normative sample included 1172 children from the UK.

b. <u>McCarron Assessment of Neuromuscular Development</u>

The McCarron Assessment of Neuromuscular Development (MAND) (McCarron, 1997) is an evaluative assessment tool used for estimating gross and fine motor skills. The items tested include: coordination, jumping, static and dynamic balance. The diagnostic criteria is thus: total score of 70-85 rated as mild, 55-69 rated as moderate, <55 rated as severe disability. It can be used for individuals between 3 to 25 years old. The normative sample included 2000 participants from the USA.

c. <u>Bruininks-Oseretsky Test of Motor Proficiency 2</u>

The Bruininks-Oseretsky Test of Motor Proficiency 2 (BOT-2) (Bruininks & Bruininks, 2005) is a discriminative and evaluative tool used for assessing gross and fine motor skills. The items tested include; coordination, balance, running, speed, agility, and strength. It can be used for individuals between 4 to 21 years-old. The normative sample included 1520 children from the USA.

d. <u>Test of Gross Motor Development 2</u>

The Test of Gross Motor Development 2 (TGMD-2) is a discriminative and evaluative tool used for assessing gross motor skills. The items tested include; running, galloping, hopping, leaping, jumping, sliding, object control (e.g., batting, dribbling, catching, kicking, throwing, and rolling) (Ulrich & Sanford, 1985). It can be used for children between three to 10 years-old. The normative sample included 1208 children from the USA (Ulrich & Sanford, 1985). The TGMD-2 was revised following recommendations by experts and clinicians in the field of motor development to create the TGMD-3. Two skills from the TGMD-2, the leap and underhand roll, were removed and replaced with the skip, underhand throw, and striking with one hand in the TGMD-3. Minor scoring explanations and wording were also changed to in order to simplify it for academics, instructors, and professionals in the field when it comes to scoring (Ulrich, 2013).

2.2.3.2 Problems with the assessment batteries for motor performance

Generally, the described assessment batteries were aimed at a specific target group and thus have specific contents. Most of them are product-oriented tests that refer to a norm and often used or referred in Western countries, which makes them valid within those contexts. There were no motor performance tests with the normative data referred in an African context. The performance and fitness battery (PERF-FIT) (Smits-Engelsman, 2018) was developed to bridge that gap, and we utilized it for this study (see methods section 3.4.2 for the description of PERF-FIT).

2.3 Review of empirical literature

2.3.1 GJH and Kinaesthesia/proprioception

Seven studies (Barrack et al., 1983; Blasier et al., 1994; Clayton et al., 2021; Fatoye et al., 2009; Hall et al., 1995; Mallik et al., 1994; Rombaut et al., 2010) reported on the differences in kinaesthesia/proprioception in individuals with GJH compared to those with normal mobility. All the studies reported that kinaesthesia/proprioception was significantly reduced in individuals with GJH when compared with those with normal mobility. However, only one (Fatoye et al., 2009) of the studies involved children as participants. The studies were not adequately powered as their sample sizes ranged from 24 participants (Barrack et al., 1983) to 66 participants (Fatoye et al., 2009). In terms of study settings, all the studies were conducted in the Western context (developed countries), and none was conducted in low-resource settings (e.g., Africa). Consequently, all the studies used expensive equipment (e.g., two-joint robot manipulandum, isokinetic dynamometer, purpose-built motorized proprioception measuring device, a stepper motor controlled by a computer etc.) to evaluate kinaesthesia/proprioception, which may not be feasible for use in low-resource contexts. The studies were heterogenous in terms of the outcome measure use for representing kinaesthesia/proprioception. Joint position sense was the outcome measure used to represent kinaesthesia/proprioception in most of the studies (Barrack et al., 1983; Fatoye et al., 2009; Mallik et al., 1994; Rombaut et al., 2010). Furthermore, most of the studies (Blasier et al., 1994; Clayton et al., 2021; Fatoye et al., 2009; Hall et al., 1995; Mallik et al., 1994) used the Beighton criteria to determine GJH.

2.3.2 Motor performance in children with or without GJH

Six studies (Davidovitch et al., 1994; De Boer et al., 2015; Engelbert et al., 2005; Jaffe et al., 1988; Juul-Kristensen et al., 2009; Tirosh et al., 1991) reported on the differences in motor performance in individuals with GJH compared to those with normal mobility. Two of the studies (Jaffe et al., 1988; Tirosh et al., 1991) reported that children/infants with GJH had significant delay in motor development as compared to children/infants with normal mobility, however, one of the studies (Jaffe et al., 1988) reported that they had normal motor function after 6 months. The two studies involved infants and toddlers (eight months to 3.5 years) and did not follow them up to school age to ascertain if the motor delay persisted, therefore, it is difficult to conclude that the motor delay noted in the infants had a link with the joint hypermobility. Four studies (Davidovitch et al., 1994; De Boer et al., 2015; Engelbert et al., 2005; Juul-Kristensen et al., 2009) reported that there was no significant difference between the motor performance of children with GJH and children with normal mobility. All the studies involved children with sample sizes ranging from 56 children (Engelbert et al., 2005) to 524 children (Juul-Kristensen et al., 2009). Again, all the studies were conducted in developed countries, and none was conducted in low-resource settings (e.g., Africa). The studies were also heterogenous in terms of the outcome measurement battery used for evaluating motor performance. The measurement batteries used in the studies included; the Bayley Scales of Infant Development II (De Boer et al., 2015; Engelbert et al., 2005), the Movement Assessment Battery for Children-2 (De Boer et al., 2015; Engelbert et al., 2005), Paediatrician-performed physical and neurological examination (Jaffe et al., 1988), Eye-hand board (30 seconds) test and a test of visual-motor integration (Davidovitch et al., 1994), the Nelson hand reaction test (Juul-Kristensen et al., 2009), the Hoskins-Squires test for measuring gross motor and reflex development, the Bruininks-Oseretsky Pegboard test, the block tower assessment, and the Beery-Buktenica visual-motor integration test for fine motor performance (Tirosh et al., 1991). The studies also used varying methods to describe GJH, which include; the Beighton criteria (Davidovitch et al., 1994; De Boer et al., 2015; Juul-Kristensen et al., 2009), the Bulbena criteria (Engelbert et al., 2005), and the Carter and Wilkinson criteria (Davidovitch et al., 1994; Jaffe et al., 1988; Tirosh et al., 1991).

2.3.3 Physical fitness in children with or without GJH

Two studies (Engelbert et al., 2006; Juul-Kristensen et al., 2012) reported on the differences in motor performance in individuals with GJH compared to those with normal mobility. One of the studies (Engelbert et al., 2006) reported that maximal exercise capacity is markedly reduced in children with GJH and hypomobility, as compared to age-matched and gender-matched control with normal joint range of motion. The study stated that deconditioning is the most likely cause of their participant's decreased exercise tolerance. The second study (Juul-Kristensen et al., 2012) reported that there was no significant difference between the physical fitness of children with GJH and children with normal mobility. However, the study used self-reported method for assessing physical fitness, which is a subjective measure. The two studies included both children and adults. The studies were also conducted in developed countries, and none was conducted in low-resource settings (e.g., Africa). The outcome measures that were used in the studies included; self-reported physical activity (Juul-Kristensen et al., 2012), maximal/peak exercise capacity (Engelbert et al., 2006). The method of diagnosing GJH used in the studies were the Beighton criteria (Juul-Kristensen et al., 2012), and goniometry (Engelbert et al., 2006).

2.4 Summary of the identified gaps in literature

The prevalence of GJH appears to be higher in Africa. However, GJH has not been given much attention in clinical practice (especially in low-income settings) because of its usual asymptomatic nature and because of the limited knowledge on its mechanism, diagnosis and progression. There is also an existing discrepancy in literature on the diagnostic criteria for GJH, which resulted to a varying report of its prevalence.

Some clinical outcomes like proprioception, motor performance, and fitness have been highlighted in literature as contributors to the progression of GJH. However, the correlation between these outcomes and how they are associated with GJH have not been substantially established, as varying evidence exists. There is also a paucity of studies that evaluated physical fitness in children with GJH. Furthermore, the assessment batteries that were used to generate the norm reference for these outcomes are not culturally and environmentally sensitive as they were developed within the Western (high-income) context. This questions their validity for use among people in low-income and lower-middle income settings (e.g., Nigeria). The performance and fitness assessment battery (PERF-FIT) was developed to address that, but it has not been used to assess those clinical outcomes (motor performance, and physical fitness) in Nigeria. Our study therefore also provided additional data to be used for reference values for motor performance and physical fitness for children in a low resource setting.

CHAPTER 3: Methodology

3.1 Research design

A cross-sectional analytical design was used for the study. It is a design where data are collected from an entire study population at a single time point (Busk, 2005). This design was considered most appropriate as it allowed for an easy evaluation of the specific outcomes at a certain moment. It also allowed for the exploration of relationships between the outcomes at a specific timepoint.

3.2 Study participants

The primary respondents for the study were school-age children within the age bracket of six to eleven years. They were recruited from one public and one private primary schools in Onitsha city, Anambra State, South-Eastern Nigeria.

3.2.1 Inclusion and exclusion criteria

3.2.1.1 Inclusion criteria- Hypermobility group

- Children between the age range of 6-11 years.
- Children with GJH (Beighton score ≥6).

Children were considered because GJH occurs more frequently in children than adults (Bird, 2005). Furthermore, the age range is appropriate because joint laxity in children increases and peaks at the adolescent age, afterwards it starts declining (Bird, 2005). There are also risks (for example, pain) that children may develop as they grow older, therefore, this study serves as a pre-measurement for a larger longitudinal study. The screening for inclusion was done by the principal researcher and the trained research assistants after the recruitment.

3.2.1.2 Inclusion criteria: Typically developing group

- Age range of 6-11 years.
- Children without GJH (Beighton score ≤5).

3.2.1.3 Exclusion criteria: Both groups

- Children who have high risk level and poor safety as it pertains to physical activity. This was assessed using The Physical Activity Readiness Questionnaire (PAR-Q) (Appendix 9).
- Children with disabilities that are limited in their ability to understand the testing instructions or the performance of the activities (e.g., cognitive impairment, gross motor impairment etc.). This was assessed through physical examination by the research team who were physiotherapists, and corroborated reports from their parents and/or teachers. However, efforts were made to ensure that the excluded children did not feel othered. For example, they were allowed to perform any of the activities they could attempt without scoring them.

3.3 Sample size and sampling

A convenience sampling technique was utilized for the study, as the participants were recruited based on their willingness to participate. The sample size was calculated through a power analysis that showed that a total sample size of 90 is needed for a medium effect size (d= 0.6), at a power of 80% while alpha is set at 0.05. The G-power analysis software version 3.1 was used for the sample size calculation (Faul et al., 2007). The G-power analysis software calculated the required sample size a priori when given alpha, power and effect size, for a t-test. See Figure 3.

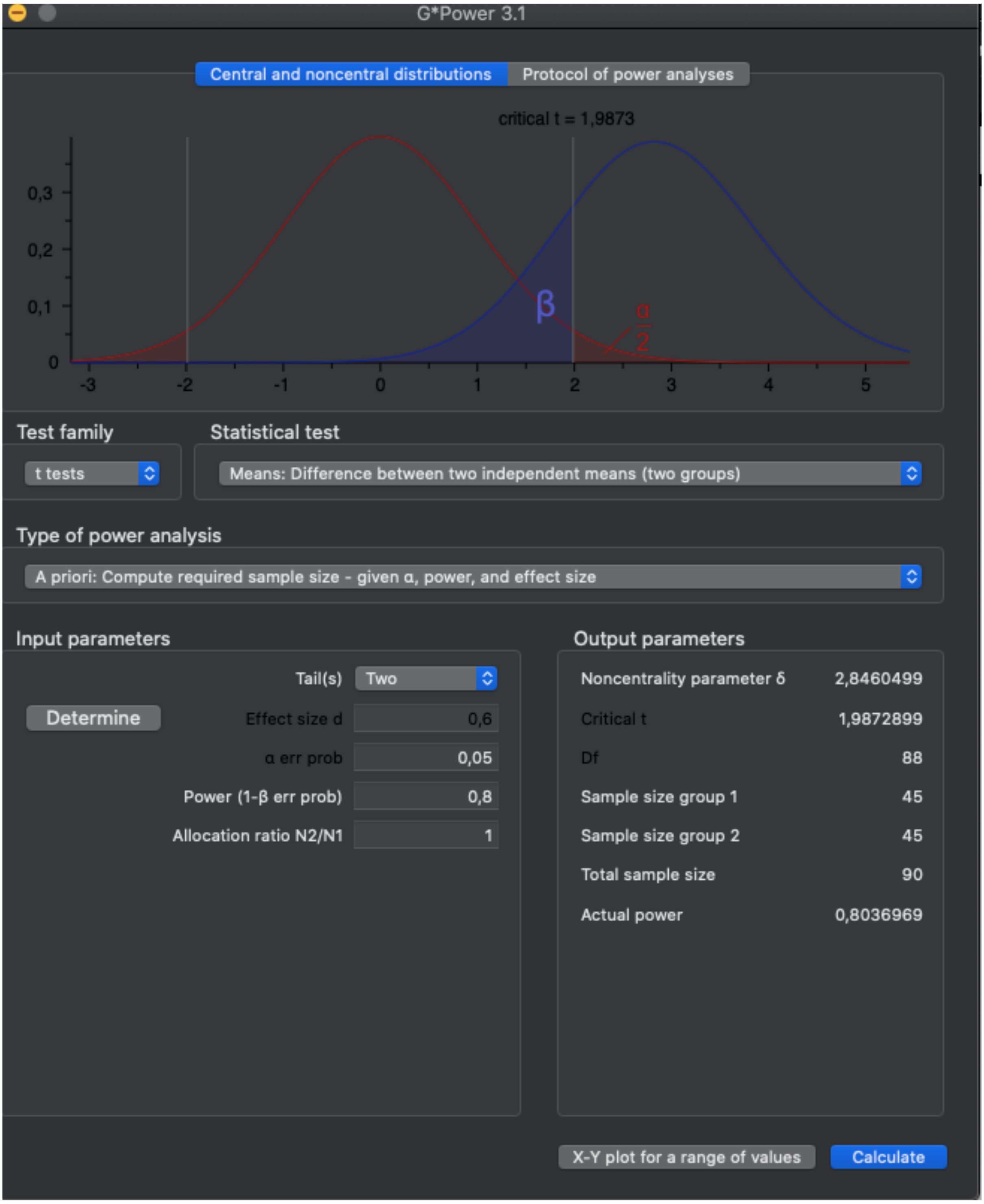

Figure 3: G-power analysis for the estimation of the sample size

3.4 Instrumentation & questionnaires

The following assessment batteries were used to assess the various outcomes:

3.4.1 The Physical Activity Readiness Questionnaire

The Physical Activity Readiness Questionnaire (PAR-Q) was used to screen the children to ascertain their eligibility of participating in physical activity considering that some of the tests were physically demanding. The questionnaire was completed by the parents of the children. The criterion for exclusion was if the parents answered 'yes' to any of the 7-item questions that are contraindications to physical activity performance (Shephard, 1988). The PAR-Q has been reported as a minimum criterion for determining enrollment into moderate intensity physical activity programmes (Terbizan et al., 2002). A study evaluated the validity of the PAR-Q by comparing the outcome of the screening with PAR-Q with a physician's examination. The study evaluated the sensitivity and specificity of PAR-Q in relation to the physician's examination, and recorded a 100% sensitivity and 81.4% specificity (Shephard, 1988).

3.4.2 Beighton criteria

GJH was assessed using the Beighton criteria (Beighton, 1973). The Beighton criteria were chosen because it has the best studied clinimetric properties for classifying GJH. Furthermore, the Beighton criteria have been reported as a valid instrument for measuring joint hypermobility (Smits-Engelsman et al., 2011). The Beighton score is comprised of 9 score points. One point is given for specific excess joint manoeuvres the child can do. These activities include: 1) the ability to put both hands on the floor with the both knees held straight. 2) the ability to hyperextend the elbows more than 10 degrees. 3) the ability to hyperextend the knees more than 10 degrees. 4) the ability to bend the thumbs back onto the front of the forearm. 5) the ability to bend the little finger to a right angle (90º) to the back of the hand.

In a study by Smits-Engelsman et al (2011) evaluating the validity of the Beighton score for children with joint hypermobility, the cut-off value for the diagnosis of joint hypermobility in children was ≥ 5 points. However, a higher cut-off score was recommended especially for children (Grahame, 1990, Smits-Engelsman et al., 2011). Therefore, the cut-off for the diagnosis of joint hypermobility was set at ≥6 points for this study.

On measure the range of motion in the knee and elbow, a goniometer of 15cm length, and 360^o was used to measure the range of motion of the elbow, knee. A 12 cm and 180^o was used for the 5th metacarpophalangeal (MCP) joints. For the elbow, the measurement was done with the child sitting on a chair, the shoulder in a 90^o anteflexion and forearm supinated. For the knee, the child was lying supine with the legs placed in a horizontal position. For the 5th MCP joints, the child was on a chair with the forearm resting on a table, arm kept at an 80^o abduction, elbow at 90^o flexion and forearm pronated. The measurement was done by physiotherapists who were trained on goniometric measurements, and it was taken twice for each child to ensure reliability. The Beighton criteria are shown in Appendix 11.

3.4.3 The performance and fitness battery (PERF-FIT)

The PERF-FIT is a valid assessment battery used to measure motor skill and physical fitness in elementary school aged children within the age bracket of 5-12 years (Smits- Engelsman et al., 2020a). The assessment battery was developed to be both culturally and economically valid for use in low-resourced countries (Smits-Engelsman et al., 2020a; Smits-Engelsman et al., 2020c). The assessment battery comprises of two subscales: a) The performance part: Motor skill subscale: made up of 5 skill item series, and b) Fitness part: Agility and power subscale made up of 5 items. The PERF-FIT scoring pattern and form is shown in Appendices 12 and 13, which were drawn from the PERF-FIT manual (Smits-Engelsman, 2018).

3.4.3.1 Performance part: Motor skill subscale

The motor skill subscale or the skill item series (SIS) comprises of five items which include a) jumping, b) hopping, c) throwing and catching, d) bouncing and catching, and e) balance. These activities were administered to the children with the difficulty level increased progressively, which is referred to as task loading. The activities were started with the simplest task and then progressed to the most difficult task within a skill series. The tasks in a particular skill series were discontinued if the child fails to attain the minimum scores for that attempt. The trial was discontinued when a child attained the maximum points at the first attempt. The performance of the children in the trials were then scored for each skill series.

3.4.3.2 Fitness part: Agility and power subscale

This component is comprised of five items. The items include running, stepping, side-jump, overhead throw, and long jump. The activities were demonstrated to the children prior to the test. Each child was given two test trials at a 15 second interval. The best performance of the children during the two trials were then scored and used for the final analysis.

3.4.4 Wedges

Kinaesthesia was assessed using wedges (Ituen et al., 2020). Wedges of varying degrees: 1.5º, 3º, 4º, 4.5º, 5º, 6º, 9º, 12º, but equal lengths were positioned on the floor. The use of wedges for assessing kinaesthesia is based on the principle of discrimination of the position of the limbs. A study reported that use of wedges is a valid outcome instrument for measuring kinaesthesia (Ituen et al., 2020). The wedges were presented in pairs while the children were in standing position with their eyes covered with a blindfold. This was to prevent the use of visual senses in the discrimination of the limb position. Pairs of wedges with different angles, and two pairs of the same angle were presented for the test. The wedges were presented in a random order. The children were asked to identify the heel that was higher without using visual cues. They were asked to identify the more elevated heel by raising their ipsilateral hand. The children were given one point for correct answers and zero point for incorrect answers. A penalty score was given for incorrect answers based on the angle variance the child could not spot. An incorrect answer for the largest wedge difference amounted to the highest penalty score. The scores were then summed up and used for the final analysis (Appendix 14).

3.4.5 Height Scale

A height scale calibrated in centimetres was used to measure the height of the children.

3.4.6 Weight scale

A weight scale calibrated in kilograms was used to obtain the weight of the children.

3.4.7 Body Mass Index

Following the recommendation by the WHO, the BMI was calculated whilst taking age, gender, weight and height into consideration (World Health Organisation, 1995). The age-gender specific BMI centiles (Must et al., 1991) were recommended for global use by the WHO. From the recommendation, the BMI of a child is represented as centiles to demonstrate how the BMI is comparable to existing norms gotten from population-based studies. For example, a child whose BMI is on the 60th centile is heavier than 60 out of 100 other children of the same age. The BMI centile classification are: Underweight= ≤ 2nd centile, Normal weight= 3rd to 90th centiles, Overweight= 91st to 97th centiles, Obese= ≥ 98 centile. We used an age-gender specific BMI calculator developed by the National Health Service, United Kingdom for the calculation of the children's BMI centiles (National Health Service, 2022).

3.5 Research procedures

3.5.1 Ethical approval and institutional permission

Ethical approvals were sought and obtained from the Faculty of Health Sciences Human Research Ethics Committees (HREC), University of Cape Town (HREC Ref: 490/2021) (Appendix 1), and the Ethical Committee University of Nigeria Teaching Hospital, Enugu (Appendix 2). Permission was also sought and obtained from the Anambra State Universal Basic Education Board (Appendix 4) and the head of the target primary schools (Appendix 8).

3.5.2 Research personnel training

The research assistants were physiotherapy students in their clinical classes (4th and 5th year) with experience in working with children, from the Department of Medical Rehabilitation, College of Health Sciences, Nnamdi Azikiwe University, Awka, Nigeria. They were trained on the testing procedures prior to the data collection. The training was done physically through demonstrations of each of the tests. The students were asked to perform the various tests (Beighton, PERF-FIT, wedges) to ensure they fully understood the procedures. Permission was obtained from the Head, Department of Medical Rehabilitation to involve the students.

3.5.3 Recruitment, informed consent and assent

The information for recruitment was given by word of mouth by the principal researcher, as parents of the children were addressed during a Parents Teachers Association (PTA) meeting. A written advert for the recruitment was also given to the parents during the meeting (Appendix 5). An informed consent document detailing the study procedure, risks and benefits, and declaration sheet were also given to the parents of the children by the principal researcher and the trained research assistants (Appendix 6). The parents were given 2 weeks to decide and return the consent form through their children. The parents were well informed of their rights to refuse the participation of their children or withdraw their consent at any time without any consequences on them or their children. After receiving the parents' consent, each child whose parents gave consent was given an informed assent declaration form to fill (Appendix 7). A research assistant was present to explain to the children if they needed assistance with the assent form.

Children whose parents gave informed consent and who gave assent were recruited. A test for GJH was conducted for each child using the Beighton's criteria (Beighton, Solomon & Soskolne, 1973). Children that met the criteria for GJH (Beighton score ≥6) were recruited. Afterwards, an age and sex matched children from the same class who did not meet the criteria for GJH (Beighton score ≤5) were recruited as a reference group. The reference group were recruited to facilitate a comparison of the specific outcomes. We ensured that no participant was in a dependent relationship with the research team involved in the recruitment. We also ensured that the participants were not involved in previous research that may result to the present study becoming an additional burden.

3.6 Data collection

The data that were collected included anthropometrics and demographics (sex, age, class/grade level, height, weight, BMI (calculated as the weight divided by the square of height), motor performance and physical fitness using PERF-FIT, kinaesthesia using the wedges, and Beighton score. The tests (Beighton, PERF-FIT, kinaesthesia tests) were administered to all the children with the help of research assistants within the school premises. Stations for each test were set up and the children rotated through them in no particular order. The data was recorded in hard copy recording sheets and then transferred to a Microsoft Excel spreadsheet in a passworded personal computer. See the data recording sheet in Appendix 15.

3.7 Data management

The data with codes were stored in a passworded storage system. The data was accessed only by the principal investigator and co-investigators and were not shared with any third party. Upon study conclusion, the data will be stored for a minimum of five years.

3.8 Data analysis

The descriptive statistics of median, interquartile range, frequency, and percentage were used to describe the anthropometric characteristics of the children, their motor performance, physical fitness, kinaesthesia, and Beighton score. The normality of the data was evaluated using the Shapiro-Wilks test. Shapiro-wilks test showed that the data were not normally distributed. Therefore, the non-parametric statistical tests were used for the analysis. Partial Spearman's rank correlation was used to determine the relationship between the variables of interest (motor performance, physical fitness, kinaesthesia and joint range of motion). The Fisher's Exact test was used to determine the gender-specific difference in the prevalence of GJH. The Mann-Whitney U test was used to determine the between-group (hyper and normal mobility) differences in the anthropometrics of the children. The normal mobile and hypermobile children differed significantly in age and BMI. Therefore, age and BMI were considered as covariates. The non-parametric ANCOVA test (Quade's test) was used to compare the fitness, and kinaesthesia of the normal mobile and hypermobile children, whilst adjusting for age and BMI. Because the Quade's test does not give the value of the adjusted medians (corrected for age and BMI) we used the parametric ANCOVA only to determine the direction of the differences (i.e. data presented as adjusted mean and standard error). Spearman correlation was used to explore the relationship between the children's kinaesthesia and age. Alpha was set at 0.05 (for a 95% confidence level).

3.9 Ethical considerations

The ethics guidelines contained in the Declaration of Helsinki (World Medical Association, 2013) were strictly adhered to while conducting this study.

3.9.1 Benefits

The population from which study participants were drawn will likely benefit from the research. The study provided evidence on the relationship between joint hypermobility, kinaesthesia, motor performance, and physical fitness in children, which will inform clinical practice and provide foundation for future studies. Referrals to the appropriate healthcare practitioners (such as physiotherapists, paediatricians) working in public hospital facilities were given to parents for two children with conditions that needed attention (one with wrist drop and the other with Erb's palsy) in the course of the study. Recommendations for physical activities were given to the schools and to children and their parents.

3.9.2 Risks

Given to the COVID-19 pandemic, there was a possible risk of transmission since the study required human to human contact. Worthy of note, the risk of transmission of COVID-19 to children has been reported to be low (Lee et al., 2020). The outcome assessments did not have a risk level that was above minimal. This implies that the level of discomfort from the assessment was not greater than those involved during the performance of the activities of daily living. The physical risks during the tests included pain, fatigue, and falls. The psychological risks included embarrassment due to failure of the tests, emotional distress. The social risk included stigmatization when diagnosed with GJH.

3.9.3 Managing risks

The WHO's guideline for minimizing the risk of transmitting COVID-19 were followed strictly (World Health Organization, 2020). The risk management strategies (both for the researchers and participants) included use of alcohol-based hand sanitizers (60-95% alcohol), adequate physical distancing (2 metres apart), use of face masks by the research assistants. Any instrument (e.g., goniometer) that was used for more than one child was sanitized with alcohol before it was used on another child.

The floor of the testing area was non-slip to minimize chances of injury. To minimize the risk of embarrassment or humiliation, the weight and height measurements for each child were done in a private space to maintain the dignity and confidentiality of measurements. The children rotated through the various test stations (Beighton, PERF-FIT motor, PERF-FIT fitness, and kinaesthesia stations) for the measurements and testing. The researcher and assistants cheered up the children to prevent emotional distress due to perceived failures in any of the tests. The diagnosis of GJH and description of group allocation were concealed from the participants to prevent stigmatization.

Before testing, the fitness evaluation using PAR-Q was used to ensure that each child was fit to participate in the physical activities.

3.9.4 Non-maleficence

Considering that the children may be distracted from their school work if the data collection was done during school time, the time allocated for their physical education and holiday periods were utilized for the data collection. Where it was not feasible to leverage the physical education time, the data was collected after school. Plans were made with the teachers to use the best possible time for the assessment purposes.

3.9.5 Justice

The respondents involved in the study were given fair treatment. There was no form of discrimination based on race, tribe, gender, social status, religious belief or study outcome.

3.9.6 Privacy and Confidentiality

All information provided by the parents, teachers or the children was strictly kept confidential. Names of the participants and their schools were stored in a separate audit and encrypted file. Codes were used for the identity of the participants.

3.10 Dissemination of information

The study results may be published in a peer-reviewed journal with wide readership. The results may also be presented in any local, regional or international conference. The study also forms part of a larger longitudinal study.

3.11 Reimbursement

The candidate is the recipient of the Mandela Rhodes Scholar award and sponsored by the Mandela Rhodes Foundation. The foundation provided funding for the purposes of the research as per submission of the budget (Appendix 16). No monetary incentives were given to the participants.

3.12 Conflict of Interest

There was no conflict of interest.

CHAPTER 4: RESULTS

4.1 Study sample

A total of 250 children were eligible to be included in the study from the two schools. However, a total of 130 informed consents and assents were received. Therefore, a total of 130 children from the two schools were included in the study. Thirty-nine (30%) children did not complete the tests, and therefore were excluded from the final analysis. Data from 91 children were analyzed and the results presented. The flow chart of the study process could be seen in Figure 1.

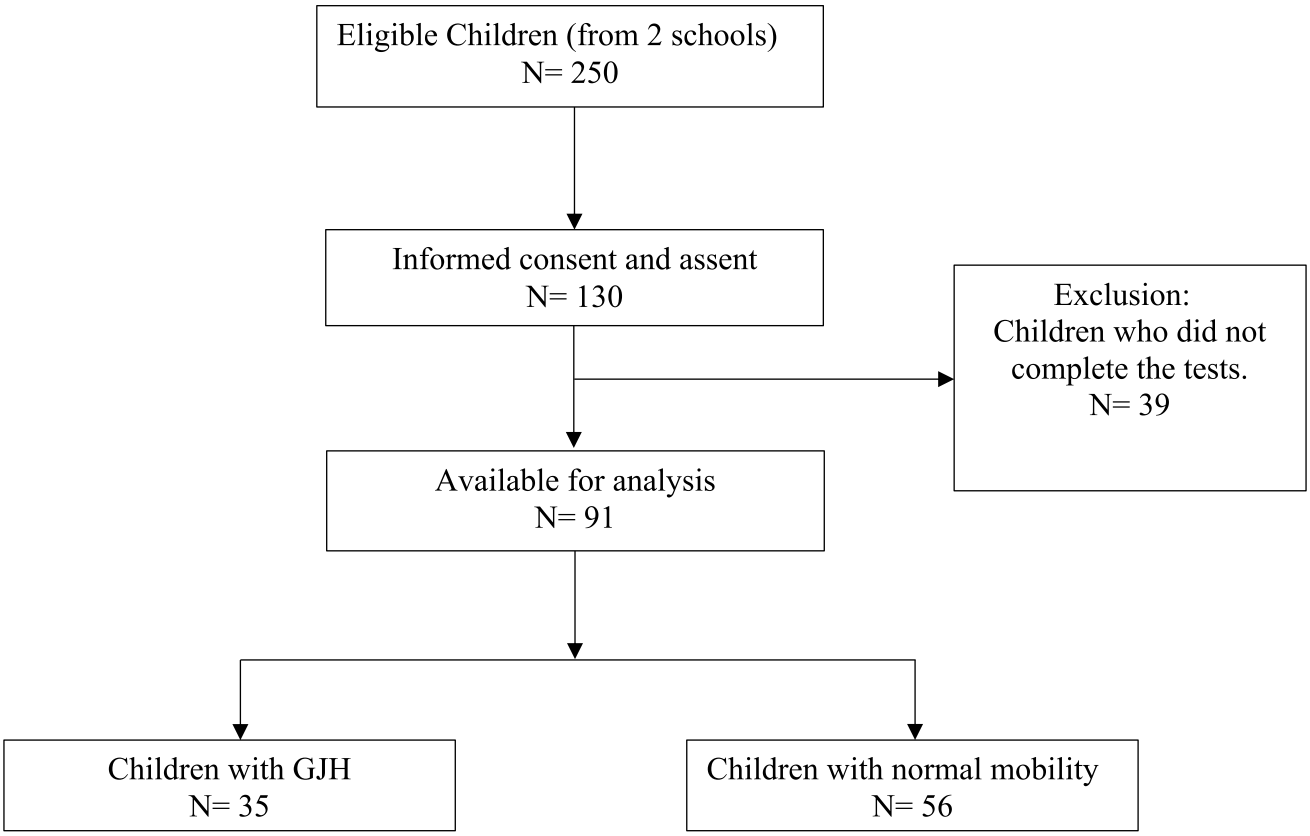

Figure 4: Flow chart of study process

4.2 Sociodemographic and anthropometric characteristics of the participants

Descriptive statistics of mean and standard deviation as well as frequency and percentage were used to describe the demographics and anthropometric characteristics of the participants.

Data from a total of 91 children (51.6% girls, and 48.4% boys) were analyzed. The mean age of the children was 8.2 years. Their mean waist circumference was 59.5. Following the World Health Organisation (WHO) age-gender specific BMI classification system, 53 (58.2%) children had normal weight. Further details on the descriptive statistics of the demographics and anthropometrics of the participants could be found in Table 1.

Table 1: The demographics and anthropometrics of the participants

Variable	Median (IQR)	Minimum	Maximum
Age (years)	8.20 (4)	6	11
Weight (Kg)	28.00 (17.00)	15	53.50
Height (m)	1.31 (0.21)	1.04	1.58
BMI (Kg/m^2)	16.12 (5.96)	11.57	31.13
Waist (cm)	60.00 (9.00)	50	75
	Frequency (%)		
BMI Classification			
Underweight	22 (24.2)		
Normal weight	53 (58.2)		
Overweight	10 (11.0)		
Obese	6 (6.6)		

Key: NA- Not applicable; SD- Standard deviation

4.3 Prevalence of GJH

GJH was identified in a total of 35 (38.46%) children, while 56 (61.54%) children had normal mobility, following the Beighton cut-off criteria of the presence of hypermobility in ≥ 6 joints. The prevalence rate is presented in a pie chart in Figure 5.

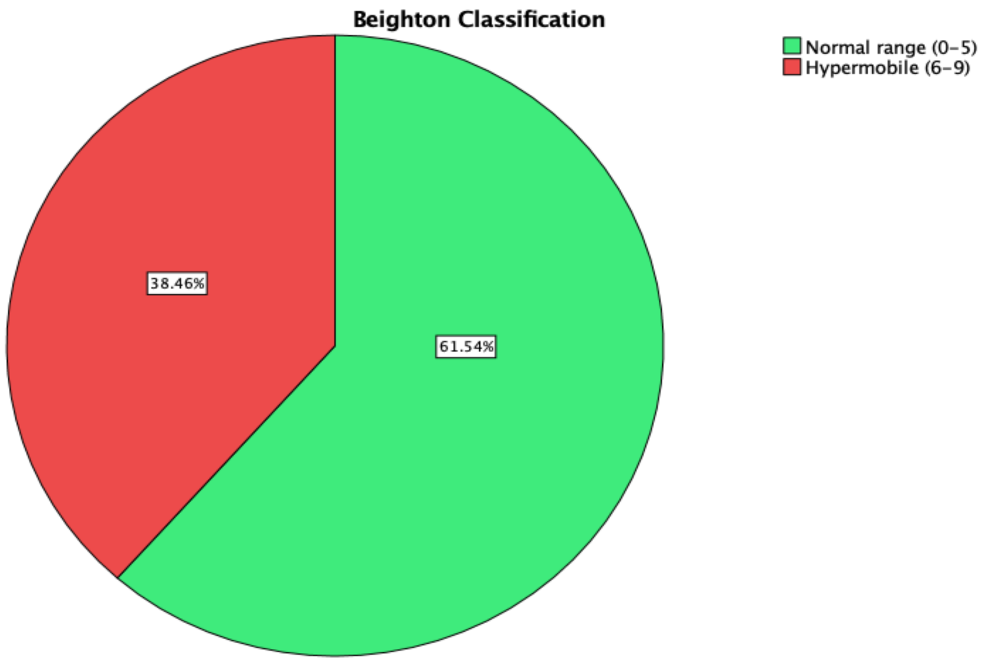

Figure 5: Prevalence of GJH

4.3.1 Gender-specific prevalence of GJH

Even though hypermobility was found in 60% of the females and 40% of the males, the difference did not reach a statistical significance (X^2= 1.589, p=0.281), when analyzed with a Fisher's Exact test. The details of the results are presented in Table 2.

Table 2: The association between gender and GJH

Beighton Classification	**Male**	**Female**	**Total**	X^2	*p*-value
Normal mobility	30	26	56	1.589	0.281
%	53.6%	46.4%			
GJH	14	21	35		
%	40.0%	60.0%			
Total	44	47	91		

4.3.2 Comparing anthropometrics of the children with and without GJH

Shapiro-Wilks test showed that the data for the anthropometrics were not normally distributed. Therefore, the non-parametric Mann-Whitney U test was used to determine the differences between the median scores of the anthropometrics of children with and without GJH. There were significant differences between the median age, weight, height, BMI, and waist circumference of the children with GJH and children with normal mobility (NM). Given to these significant differences, age and BMI were considered as covariates. Weight, height and waist circumference were not considered as covariates since they are related with BMI. Details of the comparison are presented in Table 3.

Table 3: Differences in the anthropometrics of children with GJH and NM

Variables	GJH (n=35) Median (IQR)	NM (n=56) Median (IQR)	U	p-value
Age	6.00 (3)	9.50 (4)	499.00	<0.001
Weight	22.00 (10.00)	32.25 (17.75)	442.00	<0.001
Height	1.22 (0.20)	1.37 (0.20)	483.00	<0.001
BMI (Wt/H^2)	13.73 (5.23)	17.60 (5.66)	620.00	0.003
WC	55.00 (7)	62.50 (9)	435.50	<0.001

IQR= Interquartile range, WC= Waist circumference, U= Mann-Whitney U, Wt= Weight, H= Height

4.4 Relationship between kinaesthesia, joint mobility, motor performance and physical fitness in children

There was no statistically significant correlation between the joint mobility (Beighton score) and kinaesthesia (correct wedges, and penalty scores). There was also no statistically significant correlation between the joint mobility (Beighton score) and motor performance items (ball bounce, ball throw, static balance, dynamic balance, and total jump & hop), as well as joint mobility (Beighton score) and fitness items (ladder run, ladder steps, side jump, long jump, and overhand throw). There was a statistically significant positive correlation between kinaesthesia (correct wedges scores) and some motor performance items including ball bounce, ball throw, and dynamic balance. This suggests that motor performance may be better when kinaesthesia is better and vice versa. However, there was no statistically significant correlation between kinaesthesia (correct wedges scores) and jump & hop, and static balance. There was a statistically significant correlation between kinaesthesia (correct wedges scores) and one fitness items- ladder run. However, there was no statistically significant correlation for the other items; ladder steps, side jump, long jump, and overhand throw. The details of the results are presented in Table 4.

Table 4: Correlations between kinaesthesia, Beighton score, motor performance and fitness.

Correlations

Control Variables			Beighto score N=91	Correct Wedges N=91	Penalty scores N=91	Ladder run N=91	Ladder steps N=81	Side jump N=91	Long jump N=91	Overh and throw N=91	Ball bounce N=91	Ball throw N=91	Total jump& hop N=90	Static balance N=91	Dynamic balance N=90
BMI & Age	Beighton score	Corr.	1.000	-.043	.068	-.028	-.127	.111	.145	-.193	-.040	-.075	.096	.027	.119
		Sig.	.	.690	.527	.798	.264	.301	.175	.070	.707	.486	.373	.798	.270
		df	0	87	87	87	77	87	87	87	87	87	86	87	86
	Correct Wedges	Corr.	-.043	1.000	-.979	-.226	-.128	.203	.197	.116	.310	.265	.105	.144	.223
		Sig.	.690	.	<.001	***.034**	.262	.056	.065	.279	***.003**	***.012**	.332	.177	***.036**
		df	87	0	87	87	77	87	87	87	87	87	86	87	86
	Penalty scores	Corr.	.068	-.979	1.000	.203	.160	-.209	-.209	-.149	-.329	-.281	-.083	-.130	-.248
		Sig.	.527	<.001	.	.056	.158	.049	.049	.164	.002	.008	.441	.224	.020
		df	87	87	0	87	77	87	87	87	87	87	86	87	86
	Ladder run	Corr.	-.028	-.226	.203	1.000	.298	-.375	-.232	-.067	-.237	-.296	-.186	-.205	-.219
		Sig.	.798	.034	.056	.	.008	<.001	.029	.530	.026	.005	.083	.054	.040
		df	87	87	87	0	77	87	87	87	87	87	86	87	86
	Ladder steps	Corr.	-.127	-.128	.160	.298	1.000	-.491	-.274	-.068	-.295	-.244	-.104	-.109	-.311
		Sig.	.264	.262	.158	.008	.	<.001	.015	.553	.008	.030	.361	.338	.005
		df	77	77	77	77	0	77	77	77	77	77	77	77	77
	Side jump	Corr.	.111	.203	-.209	-.375	-.491	1.000	.443	.022	.338	.244	.255	.306	.486
		Sig.	.301	.056	.049	<.001	<.001	.	<.001	.840	.001	.021	.017	.004	<.001
		df	87	87	87	87	77	0	87	87	87	87	86	87	86
	Long jump	Corr.	.145	.197	-.209	-.232	-.274	.443	1.000	.270	.442	.325	.241	.295	.474
		Sig.	.175	.065	.049	.029	.015	<.001	.	.010	<.001	.002	.024	.005	<.001
		df	87	87	87	87	77	87	0	87	87	87	86	87	86
	Overhand throw	Corr.	-.193	.116	-.149	-.067	-.068	.022	.270	1.000	.174	.245	.024	-.082	.088
		Sig.	.070	.279	.164	.530	.553	.840	.010	.	.103	.021	.822	.448	.413
		df	87	87	87	87	77	87	87	0	87	87	86	87	86
	Ball bounce	Corr.	-.040	.310	-.329	-.237	-.295	.338	.442	.174	1.000	.814	.278	.109	.320
		Sig.	.707	.003	.002	.026	.008	.001	<.001	.103	.	<.001	.009	.309	.002
		df	87	87	87	87	77	87	87	87	0	87	86	87	86
	Ball throw	Corr.	-.075	.265	-.281	-.296	-.244	.244	.325	.245	.814	1.000	.281	.134	.287
		Sig.	.486	.012	.008	.005	.030	.021	.002	.021	<.001	.	.008	.210	.007
		df	87	87	87	87	77	87	87	87	87	0	86	87	86

	Total jump&hop	Corr.	.096	.105	-.083	-.186	-.104	.255	.241	.024	.278	.281	1.000	.446	.479
		Sig.	.373	.332	.441	.083	.361	.017	.024	.822	.009	.008	.	<.001	<.001
		df	86	86	86	86	77	86	86	86	86	86	0	86	85
	Static balance	Corr.	.027	.144	-.130	-.205	-.109	.306	.295	-.082	.109	.134	.446	1.000	.545
		Sig.	.798	.177	.224	.054	.338	.004	.005	.448	.309	.210	<.001	.	<.001
		df	87	87	87	87	77	87	87	87	87	87	86	0	86
	Dynamic balance	Corr.	.119	.223	-.248	-.219	-.311	.486	.474	.088	.320	.287	.479	.545	1.000
		Sig.	.270	.036	.020	.040	.005	<.001	<.001	.413	.002	.007	<.001	<.001	.
		df	86	86	86	86	77	86	86	86	86	86	85	86	0

*Significance (2-tailed), alpha set at 0.05

4.5 Relationship between kinaesthesia and age in children

There was a statistically significant positive correlation between age and kinaesthesia (correct wedges discrimination). There was a negative correlation between age and the penalty scores, however, it did not attain statistical significance. The result suggests that kinaesthesia is better when children are older. The details of the results are presented in Table 5.

Table 5: Correlation between kinaesthesia and age in children

Correlations					
			Age	**Correct Wedges**	**Penalty scores**
Spearman's rho	**Age**	Corr. Coefficient	1.000	.211*	-.199
		Sig. (2-tailed)	.	**.045**	.059
		N	91	91	91

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

4.6 Differences between the outcomes of children with GJH and NM

4.6.1 PERF-FIT Scores; Agility and power test items (fitness)

There was a significant difference between the error-corrected ladder steps scores of children with GJH and children with NM. The children with GJH performed better in the ladder steps, as it took them a shorter time (seconds) to perform. There were no significant differences between the error-corrected ladder run scores, side jump scores, long jump scores, and overhand throw scores of children with GJH and children with NM. More details on the agility and power items scores for both groups could be seen in Table 6.

Table 6: Adjusted mean and standard error of the Agility and power test items (fitness) scores of the total group, children with NM, and children with GJH

Items	All Mean^a (SE)	NM Mean^a (SE)	GJH Mean^a (SE)	F	p
Ladder run (secs.)	7.95 (0.13)	8.28 (0.17)	7.62 (0.21)#	1.150	0.286
Ladder steps (secs.)	15.51 (0.37)	16.65 (0.46)	14.37 (0.62)#	3.997	***0.049**
Side jump (counts)	21.24 (0.57)	20.04 (0.74)	22.45 (0.96)	2.357	0.128
Long jump (cm)	117.69 (2.04)	114.66 (2.64)	120.73 (3.43)	2.075	0.153
Overhand throw (cm)	208.45 (4.91)	210.37 (6.34)	206.53 (8.24)	0.071	0.790

NB: The best scores of the ladder run, and ladder steps scores were corrected for error for each participant, where 1 mistake is given 0.5 points and the total points were added to the best score. SE- Standard error. a- mean adjusted for Age and BMI. #- Higher score indicates poorer performance. *-significance, alpha set at 0.05.

4.6.2 PERF-FIT Scores; Motor skill performance

There were no statistically significant differences between the ball bounce scores, ball throw scores, static balance scores, dynamic balance scores, and jump (one and two legged) scores of children with GJH and children with NM. More details on the motor skill performance scores for both groups could be seen in Table 7.

Table 7: Adjusted mean and standard error of the motor skill performance scores of the total group, children with NM and children with GJH

Items	All (n=81) Mean[a] (SE)	NM (n=51) Mean[a] (SE)	GJH (n=30) Mean[a] (SE)	F	*p*
Ball bounce (counts)	36.29 (1.27)	36.84 (1.64)	35.75 (2.13)	0.786	0.378
Ball throw (counts)	34.96 (1.24)	36.09 (1.61)	33.82 (2.09)	0.385	0.537
Static balance (secs)	56.56 (1.24)	55.51 (1.61)	57.61 (2.09)	0.001	0.971
Dynamic bal. (counts)	29.20 (0.53)	29.05 (0.68)	29.35 (0.89)	0.144	0.706
Jump (one and two legged)	58.00 (0.48)	57.68 (0.61)	58.32 (0.81)	0.197	0.658

NB: The best possible scores for the items are; Ball bounce= 50 counts, Ball throw= 50 counts, Static balance= 60 seconds, Dynamic balance= 32 counts, Jump (one and two legged) = 60 counts. a- mean adjusted for Age and BMI. Alpha was set at 0.05.

4.6.3 Wedges Scores (Kinaesthesia)

There were no statistically significant differences between the correct wedges scores, and the penalty scores of children with GJH and children with NM. More details on the wedges scores for both groups could be seen in Table 8.

Table 8: Adjusted mean and standard error of the wedges scores of the total group, children with NM and children with GJH

Items	All (n=81) Mean[a] (SE)	NM (n=51) Mean[a] (SE)	GJH (n=30) Mean[a] (SE)	F	*p*
Correct wedges (counts)	19.09 (0.20)	19.12 (0.25)	19.06 (0.33)	0.005	0.945
Penalty score (counts)	3.11 (0.48)	2.79 (0.61)	3.43 (0.80)	0.085	0.771

NB: The best possible scores for the items are; Correct wedges= 21 counts, penalty score= 0 counts. a- mean adjusted for Age and BMI. Alpha was set at 0.05.

CHAPTER 5: DISCUSSION

The study aimed to determine the relationship between kinaesthesia, motor performance, fitness and joint mobility in children, and to determine if kinaesthesia, motor performance and fitness differ between children with GJH and those with normal mobility. The study also explored the relationship between kinaesthesia and age.

5.1 Summary of findings

The study results showed that about one third of the children had GJH, and more than half of them were females. The children with GJH were significantly younger than those with normal mobility. There was no statistically significant correlation between joint mobility and kinaesthesia if corrected for age and BMI. There was also no statistically significant correlation between joint mobility and motor performance items, as well as the fitness items if corrected for age and BMI. There was a statistically significant correlation between kinaesthesia and some motor performance items including; ball bounce, ball throw , and dynamic balance , as well as one fitness item- ladder run. All indication better performance on the wedges is associated with better motor skills Furthermore, there was a statistically significant correlation between age and kinaesthesia (older children more correct wedges discrimination).

The children with GJH performed better than the children with NM in one fitness item (ladder steps), while there was no statistically determined differences in the other fitness items and all the motor performance items. There was also no significant difference in kinaesthesia between children with GJH and children with NM.

5.2 Demographics and anthropometrics of the children

The sample size of the study, the age and gender distribution of the study were reported in the results section 4.2. Majority of the children (58.2%) had normal weight and only a few (6.6%) were obese, while 24.2% were underweight. This is in consonance with the findings of a cross-sectional study of 1,599 children from the Southern part of Nigeria that reported 2.8% children were obese and 13.0% underweight, while the majority (72.9%) had normal weight (Ene-Obong et al., 2012). The age and BMI of the children with GJH differed significantly from that of those with NM, as children with GJH were younger and had lower BMI than those with NM. Therefore, age and BMI were considered as covariates. The age difference is in agreement with the findings of a review that reported that hypermobility decreases as age increases (Remvig et al., 2007).

5.3 Prevalence of GJH

The widely used criteria for the diagnosis of GJH are the Beighton criteria. However, there is an ongoing debate on the cut off score that should be used for the diagnosis. A lower cut-off score results to a higher prevalence. Controlling for the influence of gender, age and ethnicity may be hard, however, a stricter Beighton cut-off score could improve both the sensitivity and specificity of the Beighton criteria in the diagnosis of GJH (Smits-Engelsman et al., 2011). Therefore, we used a Beighton cut-off score of ≥ 6 for the diagnosis of GJH.

The prevalence of GJH was 38.5%, which is relatively high despite the strict cut-off score used for this study. A review reported that there are racial differences to GJH, as some races (e.g. Africans) have a higher prevalence compared to others (Remvig et al., 2007). The review cited a study conducted in South-Western Nigeria that reported a prevalence of 43% in a population comprising of children and adults (Birrell et al., 1994). However, the study used a cut-off score of ≥ 4, which should have contributed to the very high prevalence rate. A more recent study involving children (7-10 years old) from the South-Southern part of Nigeria similarly reported a prevalence of 32% while using a cut-off score of ≥ 6 for the diagnosis of GJH (Ituen, 2016). No study has compared the regional/ethnic differences in the prevalence of GJH in Nigeria. A large population-based study may be needed to establish the prevalence of GJH in Nigeria.

The findings of this study also showed a higher prevalence of GJH in females (60%) than in males (40%), however these frequencies were not significantly different. This result is in consonance with the results of other studies that reported that GJH was more prevalent in females than in males (Birrell et al., 1994; Ituen, 2016; Remvig et al., 2007). However, the studies used lower cut-off scores of ≥ 4 (Birrell et al., 1994; Remvig et al., 2007), and ≥ 5 (Ituen, 2016) for the diagnosis of GJH, they had larger sample sizes. More studies with stricter cut-off scores are needed to enable homogenous comparisons.

5.4 Relationship between kinaesthesia, joint mobility, motor performance and physical fitness in children

There was no significant correlation between the joint mobility and kinaesthesia in children. This is in contrast with some studies that reported that kinaesthesia significantly decreases as joint mobility increases (Barrack et al., 1983; Blasier et al., 1994). Again, the varying results may be a result of the differences in age of the sample, and the outcome measurement i§ nstrument used. The children involved in our study are young and are still developing, therefore, kinaesthetic deficits developing at that age are unlikely. There was also no significant correlation between joint mobility, and the motor performance of the children. This is in consonance with other studies that reported that there is no significant relationship between joint mobility and motor performance (De Boer et al., 2015; Davidovitch et al., 1994; Engelbert et al., 2005). GJH does not seem to have impact on motor performance of children that do not have CNS-related problems (Juul-Kristensen et al., 2009).

There was also no significant correlation between joint mobility and the fitness of the children. This is consistent with the findings of Juul-Kristensen et al. (2012) who reported that physical fitness was not compromised in children (mean age=10.2 years) with GJH, however, it was compromised in adults (mean age 40.3 years). This suggests that the negative impact of GJH on physical fitness may be dependent on age.

There was a significant positive correlation between kinaesthesia (correct wedges scores) and some of the motor performance items including ball bounce, ball throw, and dynamic balance. This suggests that an increase in kinaesthesia may lead to an improvement in motor function or vice versa. The result is consistent with existing evidence that kinaesthesia is essential for the neural control of human movement, and that any kinaesthetic impairment may result to reduced motor control (Allum et al., 1998; Dietz, 2002; Rossignol et al., 2006). Similarly, there was a significant correlation between kinaesthesia (correct wedges scores) and one fitness item, ladder run. However, there was no statistically significant correlation for the other items; ladder steps, side jump, long jump, and overhand throw. Studies that explored the correlation between kinaesthesia and fitness in children are scarce, however, a study involving adult women (mean age=28.7 years), reported that there was a significant positive correlation between fitness (aerobic capacity) and kinaesthesia (Allison et al., 2016). These results suggest that kinaesthesia may be a moderating factor in the motor performance and fitness of individuals with GJH. More rigorous studies (e.g., longitudinal studies and randomized controlled trials) are needed to establish the influence of kinaesthesia on the development of symptoms in some individuals with GJH.

5.5 Relationship between kinaesthesia and age in children

There was a statistically significant positive correlation between age and kinaesthesia (correct wedges discrimination), while there was a negative correlation between age and the penalty scores, however, it did not attain statistical significance. The result suggests that kinaesthesia may improve as age increases in children. In general, little is known about how the kinaesthetic sense develops with age (Smits-Engelsman & Duysens, 2008). The development of kinaesthetic sense in typically developing children has not been adequately documented when compared to other senses such as vision and audition. Previous research has found that kinaesthetic sensitivity and acuity develop in middle childhood and continue to improve at a rate comparable to adult levels during late adolescence (Goble et al., 2005; Pickett & Konczak, 2009).

Despite that no statistically significant relationships between GJH and the clinical outcomes (kinaesthesia, motor performance, and physical fitness) were established in this study, it is imperative to study the changes that may develop as the children get older. Therefore, this study serves as the baseline of a longitudinal study, which may reveal how these clinical outcomes modulate the development of complaints in children with GJH.

5.6 Differences in fitness between children with GJH and NM

Most fitness items (ladder run scores, side jump scores, long jump scores and overhand throw) were not different between mobility groups. Only the ladder steps scores were significantly different between the two groups (GJH and NM). The result is not conclusive that GJH may have a negative impact on activities that require power and agility.

This result is not in consonance with the findings of Engelbert et al (2006), who reported that children with GJH had lower physical fitness as compared to children with NM. They inferred that deconditioning may explain the decreased physical fitness marked in children with GJH because of a possibly low engagement in exercise (Engelbert et al., 2006). The discrepancy may be because of the age of the children, as they are still developing and do not have symptoms that could have limited their physical activity to the point of deconditioning. Moreover, the difference living circumstance between children in Western-Europe and Nigeria, will also have impacted the level of physical activity. The longitudinal study will explore the changes in the children's physical fitness as they grow.

5.7 Differences in Motor performance between children with GJH and NM

There were no significant differences in the scores of the motor performance items between children with GJH and children with NM. The result suggested that GJH may not have a significant impact on motor performance in a random young, asymptomatic population of children. This result is consistent with other studies that reported that there were no significant differences in the motor performance scores of children with GJH and children with NM (Engelbert et al., 2005; Juul-Kristensen et al., 2009; Tirosh et al., 1991). A study that involved infants aged 8 to 14 months reported that there was a significant motor delay in majority of the infants with GJH, however, most of them had normal motor function after 6 months (Jaffe et al., 1988). It may be safe to state that motor performance may not be impaired as a result of GJH in younger children that are still developing (Juul-Kristensen et al., 2009), however, it will be worthwhile to follow-up the children to investigate what happens over time. Another point to note is that most children with GJH are asymptomatic by definition, therefore, it could be assumed that the articular differences as a result of GJH may not be significant enough to impact the motor performances of children who have normal CNS development. A longitudinal study is necessary to determine if the motor performances of children deteriorate over time due to GJH.

5.8 Differences in kinaesthesia between children with GJH and NM

There were no significant differences between the correct wedges scores, and the penalty scores of children with GJH and children with NM. This suggests that GJH may not have negative impact on kinaesthesia. This result is in contrast with the results of some studies that reported that kinaesthesia was significantly reduced in individuals with GJH (Fatoye et al., 2009; Rombaut et al., 2010). The varying results may be as a result of the different methods of measuring kinaesthesia in these studies. While our study assessed kinaesthetic senses using wedges (weight bearing), the two studies (Fatoye et al., 2009; Rombaut et al., 2010) assessed kinaesthetic sense at the knee using a purpose-built motorized device, and an isokinetic dynamometer system respectively. There may be need for a longitudinal study and/or a randomized controlled trial to establish to what extent and at what age the microtrauma caused by GJH may lead to kinaesthetic decline in children with GJH.

5.9 Limitations of the study

The study used a cross-sectional design, which cannot be used to establish cause and effect. Therefore, the results of this study are suggestive and not conclusive.

Sufficient time was provided for rest between the various tests, and we also watched out for signs of fatigue. However, given that some of the tests are physically demanding and the number of tests carried out each day, there was a chance of underperformance as a result of fatigue.

Furthermore, given that the outcome data were not normally distributed and the need to adjust for age and BMI, a relatively new test (Quade’s test) was used to compare the outcomes between the mobility groups. However, the test has a limitation that it does not give the value of the adjusted medians. Therefore, ANCOVA was used to determine the values of the adjusted means of the outcomes so as to determine the direction of the differences.

CHAPTER 6: CONCLUSION

6.1 Conclusion

Some children with GJH develop limiting symptoms at some point in their lifetime, which is then referred to as BJHS/HSD. Some factors modulate the progression of joint hypermobility, and an early intervention targeted at those factors would be beneficial in preventing the progression of the condition. Therefore, this study evaluated the relationship between kinaesthesia, motor performance, fitness and joint mobility in children, and also evaluated if these outcomes differed between children with GJH and children with normal mobility.

The study showed that there was no significant relationship between joint mobility and motor performance, as well as physical fitness in the children. There was also no significant relationship between joint mobility and kinaesthesia in the children. However, there was a significant positive relationship between kinaesthesia and some of the motor performance and some of the physical fitness items. This suggests that whilst joint mobility may not have a significant influence on motor performance and fitness, an increase in kinaesthesia may lead to an increase in motor performance and physical fitness or vice versa. Notably, the study showed that kinaesthesia is better when the children are older.

The study also showed that there was no significant difference between the motor performance of children with GJH and children with NM. This suggests that the biomechanical features of the joints may not have influenced motor performance of developing children that do not have problems with their CNS. The study also showed that most fitness items, and kinaesthesia did not differ significantly between children with GJH and children with NM.

6.2 Recommendations

A large population-based longitudinal study will be necessary to evaluate how kinesthesia, motor performance and physical fitness change over time in children with GJH and in children with NM. This may help to establish the specific outcomes that modulate the development of symptoms in children with GJH, which will help in making clinical decision on the management plan of children with BJHS.

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Appendices

Appendix 1: UCT HREC Ethics Approval

UNIVERSITY OF CAPE TOWN
Faculty of Health Sciences
Human Research Ethics Committee

Room G50- Old Main Building
Groote Schuur Hospital
Observatory 7925
Telephone [021] 406 6492
Email: hrec-enquiries@uct.ac.za
Website: www.health.uct.ac.za/fhs/research/humanethics/forms

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27 September 2021

HREC REF:490/2021

Prof B Smits-Engelsman
Department of Health & Rehab Sciences
Division of Physiotherapy-F45 OMB
Email: bouwienengelsman@icloud.com
Student: antebu001@myuct.ac.za

Dear Prof Smits-Engelsman

PROJECT TITLE: THE RELATIONSHIP BETWEEN KINAESTHESIA, MOTOR PERFORMANCE, PHYSICAL FITNESS AND JOINT MOBILITY IN CHILDREN WITH AND WITHOUT JOINT HYPERMOBILITY IN NIGERIA-MASTERS CANDIDATE-MR EBUKA ANIETO

Thank you for your response letter, addressing the issues raised by the Faculty of Health Sciences Human Research Ethics Committee (HREC).

It is a pleasure to inform you that the HREC has **formally approved** the above-mentioned study.

This approval is subject to strict adherence to the HREC recommendations regarding research involving human participants during COVID -19, dated 17 March 2020: 06 July 2020 & 01 July 2021.

Approval is granted for one year until the 30 September 2022.

Please submit a progress form, using the standardised Annual Report Form if the study continues beyond the approval period. Please submit a Standard Closure form if the study is completed within the approval period.
(Forms can be found on our website: www.health.uct.ac.za/fhs/research/humanethics/forms)

The HREC acknowledge that the student: Mr Ebuka Anieto will also be involved in this study.

Please quote the HREC REF 490/2021 in all your correspondence.

Please note that the ongoing ethical conduct of the study remains the responsibility of the principal investigator.

Please note that for all studies approved by the HREC, the principal investigator **must** obtain appropriate institutional approval, where necessary, before the research may occur.

HREC/REF 490/2021sa

Appendix 2: Ethics approval from the University of Nigeria Teaching Hospital

UNIVERSITY OF NIGERIA TEACHING HOSPITAL

ITUKU - OZALLA, P.M.B. 01129, ENUGU

TEL: 024-252022, 252073, 252172, 252134, FAX 042-252605

cmdunth2019@gmail.com

SULEIMAN UMAR MNI
Chairman UNTH Management Board

UGWU BERNARD C. AHAN, FCAI
Director of Administration/Secretary
UNTH Management Board

Dr. OBINNA D. ONODUGO MBBS, FWACP, FICP, MBA
Chief Medical Director

Dr. E.O.V. UGWU MBBS, MPH, MPA, FWACS, FMCOG, FICS
Chairman MAC & Director Clinical Services & Training

Our Ref: UNTH/CSA/329/VOL.5

Date: 10th November, 2020

NHREC/05/01/2008B-FWA00002458-1RB00002323

ETHICAL CLEARANCE CERTIFICATE

TOPIC: THE RELATIONSHIP BETWEEN KINAESTHESIA, MOTOR PERFORMANCE AND PHYSICAL FITNESS IN CHILDREN WITH AND WITHOUT JOINT HYPERMOBILITY.

BY: **EBUKA MIRACLE ANIETO**

FOR: A DISSERTATION FOR MASTER'S DEGREE IN PHYSIOTHERAPY OF THE DEPARTMENT OF HEALTH AND REHABILITATION SCIENCES, FACULTY OF HEALTH SCIENCES, UNIVERSITY OF CAPE TOWN, SOUTH AFRICA.

This research project on the above topic was reviewed and approved by the University of Nigeria Teaching Hospital Health Research Ethics Committee. This certificate is valid for one year from date of issue. Please note that the Committee Reserves the Right to monitor the Conduct of the study at any time for strict Compliance to the Protocol.

Prof. G.N. Adimora
Chairman, Health Research Ethics Committee

Date: 16/11/2020

Scanned with CamScanner

Appendix 3: UCT confirmation of insurance

Marsh

Fagma Jordaan
Marsh Proprietary Limited
50 Dorp Street
Stellenbosch,7600
P.O Box 414,
Stellenbosch, 7599
South Africa
Tel +27 21 834 1300
Fax +27 21 887 0518
www.marsh-africa.com

2 July 2021

TO WHOM IT MAY CONCERN

UNIVERSITY OF CAPE TOWN: CONFIRMATION OF INSURANCE ON CLINICAL TRIALS

STUDY TITLE: The relationship between kinaesthesia, motor performance, physical fitness and joint mobility in children with and without joint hypermobility in Nigeria

Marsh (Pty) Ltd acts as insurance brokers to University of Cape Town and confirms cover as follows:

INSURED	UNIVERSITY OF CAPE TOWN
CLASS	No fault compensation for clinical trials
PERIOD OF INSURANCE	01 March 2021 to 28 February 2022 (both days inclusive)
NUMBER OF PARTICIPANTS	128
PRINCIPAL INVESTIGATOR	Prof. Bouwien Smits-Engelsman
INSURER	Lloyds
POLICY NUMBER	BOWLT2100124
LIMIT OF INDEMNITY	USD 5 000 000 any one claim and in the aggregate per annum
TERITORIAL LIMITS	World-wide, Excluding USA/Canada

We trust that you will find this to be in order, should you require any additional information, please do not hesitate to contact the writer.

Yours sincerely,

Fagma Jordaan, Divisional Manager
Marsh Africa, Public Enterprises
Tel: +27 21 833 4891| Mobile: +27 76 169 4778| Fax: 0866231027Email: fagma.jordaan@marsh.com
www.africa.marsh.com

Appendix 4: Letter to the Anambra State Universal Basic Education Board

DHRS
Department of Health and Rehabilitation Sciences
FACULTY OF HEALTH SCIENCES

University of Cape Town · iYunivesithi yaseKapa · Universiteit van Kaapstad

Divisions of • Communication Sciences & Disorders • Disability Studies • Nursing & Midwifery • Occupational Therapy • Physiotherapy

F45 Old Main Building, Groote Schuur Hospital
Observatory, Cape Town, South Africa, 7925
Telephone: +27 (0) 21 406 6401.
Website: www.dhrs.uct.ac.za

10/06/2021

Dear Sir/Madam,

I am Ebuka Miracle Anieto, a post-graduate student at the Department of Physiotherapy, University of Cape Town, South Africa. As part of the requirements to obtain my Master's of Physiotherapy degree, I will be undertaking a study evaluating **the relationship between perception of movement, motor performance and physical fitness in children with and without joint hypermobility.** I will be assisted by some research assistants which will either registered physiotherapists or physiotherapy students at the clinical stage of their training.

Joint hypermobility is a condition characterized by excessive movement in the joints, which may involve a single or multiple joints in the body. The condition has been associated with difficulties with perceiving the position and movement of the limbs, poor motor performance, and poor physical fitness. Healthcare professionals feel that children with this condition need to be treated, however, there are no tangible research that has clearly highlighted what the treatment goal should be. That is why we are conducting this study.

Ethical Approval for the study will be obtained from the Faculty of Health Sciences Human Research Ethics Committee, University of Cape Town, and Ethics committee, University of Nigeria Teaching Hospital, Enugu, Nigeria. Two schools under the Anambra State Universal Basic Education Board will be selected and all children in grades 1-5 will be eligible to participate in the study.

Informed consent

The participation of a child will be determined by his/her parents/guardian. The child will also give an assent on the day the assessment will be carried out. The child is free to decline participation and he /she can withdraw from the study at any time without any consequence on the child. The parents and teachers are also free to decline without any consequence.

Assessments

The assessments will be carried out within the school premises, in a safe, comfortable, and well-ventilated place outdoors provided by the school. Arrangements will be made with the schools so that the assessment can be carried out during physical education sessions, break or after school hours. This is to ensure that the study does not affect their class work. The visit in each school will be 4 days in a week for 2 months (July 2021 to September 2021). We will be spending about 4 hours in the schools per visit.

We will ask the children to perform some activities to assess their motor skills, movement perception, and fitness. For example, we will ask each child to run, hop, throw and catch a ball, to stand and hop on one leg. The tests will take about 20-30 minutes. We will also check the weight, and height of the children. The children's elbow joints, knee joints and 5th finger joints will also be measured. Each child will also perform some simple tasks with wedges, and these may take another 20 minutes. These activities are fun, easy, and safe for the children.

Possible risks

Some of the assessments are physical test like running and hopping. This can make the children tired during or after the test. Other physical risks during the tests may include; pain, and falls. The psychological risk may include; embarrassment due to failure of the tests, emotional distress. The social risk may include stigmatization when diagnosed with GJH. Given to the COVID-19 pandemic, there is a possible risk of transmission, since the study will require human to human contact.

To reduce the risks, the children will be allowed to rest when they are tired and will also be provided with water when they demand for it. We will also have a first aid kit with us when they perform the tests and we will always be present when the children carry out the tests. The floor of the testing area will be non-slip, devoid of sharp objects or any other materials that can cause physical injury. The children will be cheered up to prevent emotional distress due to failures in some of the tests. The diagnosis of GJH and description of group allocation will be concealed from the participants to prevent stigmatization. Also, there will be temperature assessment with a non-contact infrared thermometer, use of alcohol-based hand sanitizers (60-95% alcohol), adequate physical distancing (2 metres apart), strict use of face masks by the research assistants and child-specific face shields will be provided for each of the children.

Benefits

The parents of each child will be given a report on his/her motor performance, movement perception, and physical fitness. If any of those outcomes are found to be poor, the children and their parents will be advised by the researcher (Ebuka) on how best to handle it. We will also give their parents referrals to an appropriate healthcare professional if need be.

UCT No-fault insurance policy

If the child experiences a deterioration in his/her health or well-being due to unforeseen sensitivity related to participation in the study, medical care will be provided immediately by the university.

According to the Association of the British Pharmaceutical Industry 1991, UCT will compensate without having to prove it is the university's fault. A trial-related injury is defined as one caused by activities related to our study. If an injury or abnormal side-effects does occur, one of the researchers must be notified immediately.

UCT reserves the right to not provide compensation for participants who become injured as a result of not following the instructions given to them whilst taking part in the study however you still have the right to lawfully claim compensation for any injuries where you prove negligence was not the cause of the injury.

If you would like more information, please contact me; Ebuka Miracle Anieto, tel.: +2348069535182, Email: antebu001@myuct.ac.za

If you will like to speak to someone from UCT physiotherapy department, you may contact my supervisors: Dr. Niri Naidoo, Email: niri.naidoo@uct.ac.za, and Prof. Bouwien Smits-Engelsman, Email: bouwienengelsman@icloud.com .

If you will like to speak to UCT Human Research Ethics Committee, please contact Prof. Marc Blockman (Chairperson of the University of Cape Town Human Ethics Research Committee tel: 021406338. Email marc.blockman@uct.ac.za)

Yours Faithfully
Ebuka Miracle Anieto

Appendix 5: Advert for recruitment

DHRS
Department of Health and Rehabilitation Sciences
FACULTY OF HEALTH SCIENCES

University of Cape Town • iYunivesithi yaseKapa • Universiteit van Kaapstad

SPES BONA

Divisions of • Communication Sciences & Disorders • Disability Studies • Nursing & Midwifery • Occupational Therapy • Physiotherapy

F45 Old Main Building, Groote Schuur Hospital
Observatory, Cape Town, South Africa, 7925
Telephone: +27 (0) 21 406 6401.
Website: www.dhrs.uct.ac.za

I am Ebuka Miracle Anieto, a post-graduate student at the Department of Physiotherapy, University of Cape Town, South Africa. As part of the requirements to obtain my Master's of Physiotherapy degree, I will be undertaking a study evaluating the relationship between perception of movement, motor performance and physical fitness in children with and without joint hypermobility.

Joint hypermobility is a condition characterized by excessive movement in the joints, which may involve a single or multiple joints in the body. The condition has been associated with difficulties with perceiving the position and movement of the limbs, poor motor performance, and poor physical fitness. Healthcare professionals feel that children with this condition need to be treated, however, there are no tangible research that has clearly highlighted what the treatment goal should be. That is why we are conducting this study.

I am seeking for primary and secondary school children within the age range of 6-10 years to participate in the study. All the participants will perform some tests (mostly physical activity related) which has been described in the information sheet attached to this advert.

If you are willing to give permissions for your child to participate, please fill the informed consent declaration form given to you and return it to the teachers through your child.

Participation in the study is completely voluntary, and you have the right to withdraw your child at any stage of the study. Feel free to ask any question at any time, and you will definitely receive answers.

If you would like more information, please contact me; Ebuka Miracle Anieto, tel.: +2348069535182, Email: antebu001@myuct.ac.za

Appendix 6: Parents Infromation Sheet and consent form

DHRS
Department of Health and Rehabilitation Sciences
FACULTY OF HEALTH SCIENCES

University of Cape Town · iYunivesithi yaseKapa · Universiteit van Kaapstad

Divisions of • Communication Sciences & Disorders • Disability Studies • Nursing & Midwifery • Occupational Therapy • Physiotherapy

F45 Old Main Building, Groote Schuur Hospital
Observatory, Cape Town, South Africa, 7925
Telephone: +27 (0) 21 406 6401.
Website: www.dhrs.uct.ac.za

10/06/2021

Dear Parent/Guardian,

I am Ebuka Miracle Anieto, a post-graduate student at the University of Cape Town, South Africa. As part of the requirements needed to obtain a Master's degree in Physiotherapy, I am carrying out a study to evaluate the relationship between the awareness of the position and movement of the limbs, motor skills and physical fitness in children with and without excessive joint movement. I am working with a team comprised of my supervisors- Prof Niri Naidoo and Prof Bouwien Smits-Engelsman, and five research assistants who are registered physiotherapists or physiotherapy students at the clinical stage of their training.

Some children have larger mobility in their joints than others. A small part of the children with very mobile joints develop complaints and we are trying to find out why some children do develop complaints while others do not. It could be that they don't feel the movements in their joints well or maybe they don't have muscles that are strong enough to stabilize the joints. Therefore, we want to understand the things that result to these complaints.

What am I asking you as parent(s)/guardian(s) to do?

I am asking for your consent to involve your child in the study to assess his/her physical fitness, motor performance and awareness of the limb position/movement.

If you agree, would you kindly sign the declaration below and please fill the questionnaire attached? The first part of the questionnaire is about your child and the second part is about you. I am asking these questions to know more about you and your child.

What does your child have to do?

We will ask your child to perform some activities like running, hopping, throwing and catching a ball, standing and hopping on one leg. The tests will take about 20-40 minutes. Your child's elbow joints, knee joints and 5^{th} finger joints will also be measured and your child will also perform some simple tasks to test if he/she feels the difference in height while standing on two wedges, and these tests may take another 20 minutes. We will also measure your child's height and weight.

We have no intention of interrupting your child's education in any way. Therefore, we will plan with the teachers so that the activities will be performed during physical education sessions, break times or after school. The data collection will be for a period 2 months (July 2021 to September 2021). However, the time commitment for each child will be about 40 minutes for just 2 days.

If your child agrees to participate he/she will also be asked to sign a form to show that he/she has agreed. Your child has the right to discontinue the test at any time.

What are the risks involved and what will ensure their safety?
Given to the COVID-19 pandemic, there is a possible risk of transmission, since the study will require human to human contact. Worthy of note, the risk of transmission of COVID-19 to children has been reported to be low.
The tests do not have a risk level that is above minimal. This implies that the level of discomfort expected from the assessment is not greater than those involved during the performance of the activities of daily living. The physical risks during the tests may include; pain, fatigue, and falls. The psychological risk may include; embarrassment due to failure of the tests, emotional distress. The social risk may include stigmatization when diagnosed with GJH.
The WHO's guideline for minimizing the risk of transmitting COVID-19 will be followed strictly (World Health Organization, 2020). The risk management strategies (both for the researchers and participants) will include; temperature assessment with a non-contact infrared thermometer, use of alcohol-based hand sanitizers (60-95% alcohol), adequate physical distancing (2 metres apart), strict use of face masks by the research assistants and child-specific face shields will be provided for each of the children. Any instrument (e.g. goniometer) that will be used for more than one child must be sanitized with alcohol before it will be used on another child.
The floor of the testing area will be non-slip, devoid of sharp objects or any other materials that can cause physical injury. The children will be cheered up to prevent emotional distress due to failures in some of the tests. The diagnosis of GJH and description of group allocation will be concealed from the participants to prevent stigmatization.
The research personnel to administer the tests will be physiotherapists with experience in handling paediatrics. The fitness testing using PAR-Q will be used to ensure that each child is fit to participate in the physical activities.
What are the benefits of participating?
You will be given a report on your child's motor skills and physical fitness. If any of those outcomes are found to be poor, you will be advised on how best to handle it. We will also give you referrals to an appropriate healthcare professional if need be.
What happens if I do not want my child to take part in the study?
Your consent will determine whether your child will participate or not. There will be no form of compulsion or coercion. There will be no consequence if you do not give your consent. Refusal to take part in this study will not affect the quality of education your child will receive. If you agree and then you change your mind later, your child will be excluded from the study immediately with no consequence.
What about confidentiality and privacy?
None of the information obtained during the study will be disclosed to a third party. Both the information you provide through the questionnaire and child's results from all the tests will be stored and managed under a password-controlled system. We will use codes for identification (names will be stored separately). Only the principal researcher and his supervisors will have access to the data. The tests will be conducted with the supervision of the researchers. Your child's peers will not be allowed to observe.
As a researcher and healthcare professional, I may not be able to hold as confidential, information about known or reasonably suspected incidents of deliberate neglect or physical, sexual or emotional abuse of a child. If I am given such information, I may report it to relevant authorities such as child welfare.
Will I be paid for taking part in the study?
I am not providing any money to the parents or the children as a reward for participating in this study. However, all the chil dren who participate in the study will be given snacks (fruits) at the end of the tests.

If you would like further information, please contact me.

You can visit me at your child's school. I will be there from Monday to Thursday 8am-12pm. Telephone: +2348069535182. Email: antebu001@myuct.ac.za. Feel free to ask any question, which will be definitely answered.

If you will like to speak to someone from UCT physiotherapy department, you may contact my supervisors: Dr. Niri Naidoo, Email: niri.naidoo@uct.ac.za, and Prof. Bouwien Smits-Engelsman, Email: bouwienengelsman@icloud.com .

If you will like to speak to UCT Human Research Ethics Committee, please contact Prof. Marc Blockman (Chairperson of the University of Cape Town Human Ethics Research Committee tel: 021406338. Email marc.blockman@uct.ac.za)

Consent form

I ……………………………………………(name and surname), am the parent/guardian of …………………………………………………………………………………………name and surname of child in grade ……………

I have read through the attached letter and I understand what is required of me and my child. I do not feel that I am forced to have my child participate, and I am doing so out of my own free will. I know that I can withdraw my child at any time that I so wish. I understand that withdrawal will have no consequences on the child.

PLEASE INDICATE YOUR CHOICE BY TICKING ONE BOX BELOW AND SIGNING NEXT TO THE BOX.

YES, I AGREE to let my child to take part in the study ☐

Signature……………………………………

NO, I DO NOT AGREE to let my child to take part in the ☐ study

Signature……………………………………..

Thank you

Please return this form to the researcher

Appendix 7: Child Information Sheet and assent form

DHRS
Department of Health and Rehabilitation Sciences
FACULTY OF HEALTH SCIENCES

Divisions of • Communication Sciences & Disorders • Disability Studies • Nursing & Midwifery • Occupational Therapy • Physiotherapy

F45 Old Main Building, Groote Schuur Hospital
Observatory, Cape Town, South Africa, 7925
Telephone: +27 (0) 21 406 6401.
Website: www.dhrs.uct.ac.za

10/06/2021

Dear Learner,

My name is Ebuka Miracle Anieto and I am a student from the University of Cape Town. I and my colleagues want to come to your school to find out how well you perform your motor skills and physical activities.

Motor skills are activities you perform with your arms, hands and legs . For example, you use your hands to catch a ball, throw a ball, while you use your legs to kick a ball, jump and run around the playground. These are things you do everyday, some children do them well while others may find them difficult. We will also measure how flexible you are and if you feel well how your limbs move. In our study, we want to find out if children who are very flexible are better or not in certain activities (running, jumping, catching hopping, standing on a wegde). We like to ask if you will help us with this study so we can learn more about why some very bendy children have difficulties with motor skills and fitness.

What do I have to do?

We will show you pictures or demonstrate the activiites. We will ask you to Run, jump, hop, catch, throw a ball, stand on one leg, bend your arms and legs, grasp your toes. We will also have wedges (see picture) put under one foot and you will be asked to tell us which leg is higher.




.

What are the risks?

The activities you will be asked to perform are like the ones you do everyday. They will not harm or hurt you. We will be around when you perform the activities to make sure that you are safe.

You can stop and rest when you are tired. We will give you water for you to drink if you need it. If you fall and get hurt we have a first aid kit to help you. We will tell your teacher and parents if you get injured so that we can make plan to help you.

Can I disagree?

You are allowed to say that you don't want to be part of the study. You can stop at any time if you wish to and you don't have to do all the things you are asked to do. All you have to do is say so and you would not get into trouble.

Before you agree to participate, you can ask us questions or talk to your parents if there is anything you do not understand.

We will not tell anyone the information you give to us without your permision unless there is something that could cause harm to you or someone else. We may have to tell people that are responsible for taking care of children.

If you want more information, you or your parents can contact my supervisors:

Dr. Niri Naidoo, Email: niri.naidoo@uct.ac.za, and Prof. Bouwien Smits-Engelsman, Email: bouwienengelsman@icloud.com ., Department of physiotherapy, University of Cape Town. or Prof. M. Blockman at the University of Cape Town, Human Research Ethics Committee Tel. 021406338. Email: marc.blockman@uct.ac.za

Child Assent Form

Declaration

I agree to show the researchers how I do these activities and allow them to measure me with their test instruments. I may be tired after the tests. I understand that I do not have to do all the things, I can stop at any time and no one will be angry with me. I understand that the researcher will tell my parents how well I performed.

If you understand and agree to show us how you do some of these activities please write your name in the box below.

If you understand and DO NOT want to show us how you do these activities, please write your name in the box below.

Appendix 8: Headmasters/School principals Information Sheet

DHRS
Department of Health and Rehabilitation Sciences
FACULTY OF HEALTH SCIENCES

Divisions of • Communication Sciences & Disorders • Disability Studies • Nursing & Midwifery • Occupational Therapy • Physiotherapy

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F45 Old Main Building, Groote Schuur Hospital
Observatory, Cape Town, South Africa, 7925
Telephone: +27 (0) 21 406 6401.
Website: www.dhrs.uct.ac.za

10/06/2021

Dear Headmaster,

I am Ebuka Miracle Anieto, a post-graduate student at the University of Cape Town, South Africa. As part of the requirements needed to obtain a Master's degree in Physiotherapy, I am carrying out a study to evaluate the relationship between joint mobility, motor performance and physical fitness in children.

Some children have larger mobility in their joints than others. A small part of the children with very mobile joints develop complaints and we are trying to find out why some children do develop complaints while others do not. It could be that they don't feel the movements in their joints well or maybe they don't have muscles that are strong enough to stabilize the joints. Therefore, we want to understand the things that may result to these complaints in some children.

What am I asking you as the headmaster to do?

I am asking for your permission to recruit children from your school for the research project. Next, I would also want you to assist in providing information on the time allotted for the children's physical education, and break, so we will not disturb the education of the children (or as little as possible). The testing will take about 40 minutes for each child.

If you would like further information, please contact me.

Telephone: +2348069535182. Email: antebu001@myuct.ac.za.

If you will like to speak to someone from UCT physiotherapy department, you may contact my supervisors: Dr. Niri Naidoo, Email: niri.naidoo@uct.ac.za, and Prof. Bouwien Smits-Engelsman, Email: bouwienengelsman@icloud.com .

If you will like to speak to UCT Human Research Ethics Committee, please contact Prof. Marc Blockman (Chairperson of the University of Cape Town Human Ethics Research Committee tel: 021406338. Email marc.blockman@uct.ac.za)

Yours Faithfully

Ebuka Miracle Anieto

Appendix 9: PAR-Q Form

PAR-Q Form

Name:______________________________ Date:______________

DOB:______________ Height:______________ Weight:______________

Health Care Provider:______________________________ Phone: ______________

Questions

Has your health care provider ever said that you have a heart condition and that you should only perform physical activity recommended by a doctor?	Yes	No
Do you feel pain in your chest when performing physical activity?	Yes	No
Have you experienced chest pain when NOT performing physical activity in the last month?	Yes	No
Do you lose your balance because of dizziness or have you lost consciousness recently?	Yes	No
Do you have any bone or joint problems (back, knee, hip, etc.) such as arthritis, which could be aggravated through physical activity?	Yes	No
Is your doctor currently prescribing you medications for high blood pressure or a heart condition?	Yes	No
Is there any reason why you should not participate in physical activity?	Yes	No

Reason: ______________________________

If Yes to Any Questions: ______________________________

If No to All Questions: ______________________________

______________________ ______________________

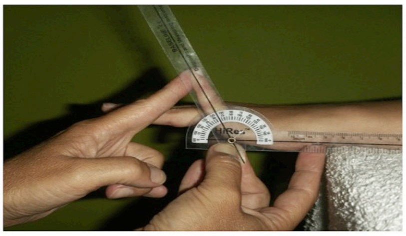
Name Guardian Name

Appendix 10: The 9-point Beighton score for joint hypermobility

Description	Bilateral Testing	Scoring [max. point]
Passive dorsiflexion of the 5th metacarpophalangeal joint to ≥ 90 degrees	Yes	2
Passive hyperextension of the elbow ≥ 10 degrees	Yes	2
Passive hyperextension of the knee ≥ 10 degrees	Yes	2
Passive apposition of the thumb to the flexor side of the forearm, while the shoulder is flexed at 90 degrees, elbow is extended and hand is pronated	Yes	2
Forward flexion of the trunk, with the knees straight, so that the hand palms rest easily on the floor.	No	1
Total		9

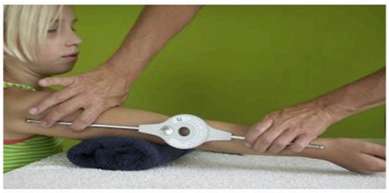
Appendix 11: Protocol for Beighton score (Smits-Engelsman et al., 2011)

1. Passive dorsiflexion of the fifth metacarpophalangeal joint. Score is positive if ≥ 90° (Bilateral testing)



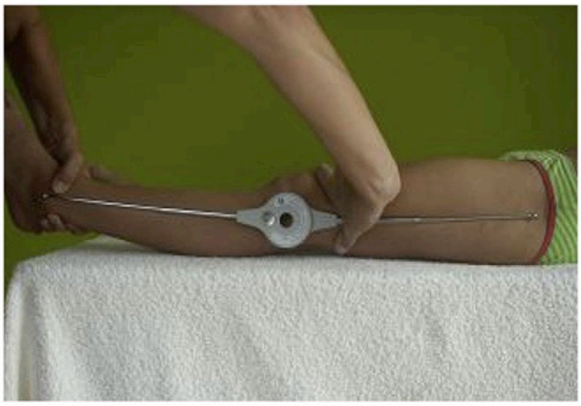
Test position	Motion tested	Positioning Goniometer	Anatomical landmarks	Method
Sit on chair at the short side of the table with arm in 80° abduction, elbow flexed 90°, forearm resting on table, forearm pronated.	Passive Dorsiflexion Digiti 5.	MCP 5.	Dorsal side Metacarpalia 5; in the length of Digiti 5.	Lateral method.

2. Passive hyperextension of the elbow. Score is positive if ≥ 10° (Bilateral testing)



Test position	Motion tested	Positioning Goniometer	Anatomical landmarks	Method
Sit on chair with shoulder 90° anteflexion, forearm supinated	Passive hyperextension of elbow.	Lateral epicondyl Humerus.	Humurus pointed at tub major humeri; Radius pointed at proc styloideus.	Lateral method.

3. Passive hyperextension of the knee. Score is positive if ≥ 10° (Bilateral testing)



Test position	**Motion tested**	**Positioning Goniometer**	**Anatomical landmarks**	**Method**
Lying backwards with legs in horizontal position.	Passive hyperextension knee.	Lateral femur epicondyl.	Femur pointed at trochanter major; Fibula pointed at lateral malleolus.	Lateral method.

4. Passive apposition of the thumb to the flexor side of the forearm, while shoulder is 90° flexed, elbow extended and hand pronated. Score is positive if the whole thumb touches the flexor side of the forearm. (Bilateral testing)

Score: Positive

Score: Negative

5. Forward flexion of the trunk, with the knees straight. Score is positive if the hand palms rest easily on the floor.

Score: Positive

Score: Negative

Scoring

One point may be gained for each side for item 1-4 (max 2 per item if left and right are positive) and only one point in total for item 5.

The maximum hypermobility score is nine points (if all items are positive).

Appendix 12: PERF-FIT Scoring (Smits-Engelsman et al., 2018)

Subtest Agility and Power

1. Running in agility ladder 2 trials (Time in 0.01 s and number of mistakes)
2. Stepping in agility ladder 2 trials (Time in 0.01 s and number of mistakes)
3. Side jumps 2 trials (Number of correct jumps in 15 seconds)
4. Long Jump 2 trials (Distance in cm)
5. Throwing for distance: sandbag 2 trials (Distance in cm)

Subtest Motor Skill Performance

Bounce and catch 10 trials: 5 items 10 point=50 points max score.
1. Bounce the ball to the floor and catch it with two hands
2. Bounce the ball on the floor and catch with the preferred hand
3. Bounce the ball on the floor and catch with the non-preferred hand
4. Bounce on the floor with the preferred hand, clap and catch with the preferred hand.
5. Bounce on the floor with the non-preferred hand, clap and catch with the non-preferred hand.

Throw and catch 10 trials: 5 items 10 point=50 points max score.
1. Throw the ball in the air and catch it with two hands
2. Throw the ball in the air and catch with the preferred hand
3. Throw the ball in the air and catch with the non-preferred hand
4. Throw the ball in the air, clap and catch with the preferred hand
5. Throw the ball in the air with the non-preferred hand, clap and catch with the non-preferred hand.

Static balance: 2 items for each leg 15s; 2 legs x 2 items x15s= 60 seconds max time score;
1. Balance Hug Knee Static R and L (timed max 15s per leg),
2. Balance Grasp Foot Static R and L (timed max 15s per leg).

Dynamic balance: 4 items x 8 points =32 max balance score.
1. Balance Hug Knee Dynamic (count 8)
2. Balance Grasp Foot Dynamic (count 8)
3. Balance Pick up Cans Dynamic R and L (count 8: per leg).

Jump: 4 items 8, 4, 4, 4 points=20 points max score.
1. Jumping in each square (8 jumps)
2. Jumping every other square (4 jumps)
3. Jumping over four 5 cm foams (4 jumps)
4. Jumping over four 10 cm foams (4 jumps).

Hop: 4 items for each leg 8, 4, 4, 4 points=20 points per leg 40 points max score.
1. Hopping each square (8 hops)
2. Hopping every other square (4 hops)
3. Hopping over four 5 cm foams (4 hops)

4. Hopping over four 10 cm foams (4 hops).

FATIGUE AND REST

In order to alternate between more and less fatiguing items the following order of the items and rest periods for the full test procedure are recommended:

- Two slow practice trials of the agility ladder are given as general warming-up for the test
- Running (two slow practice trials of the agility ladder are given as general warming-up for the test, give a minimum 15s rest between the 2 test trials)
- Stepping (minimum 15s rest between the 2 test trials)
- Side jump (minimum 15s between the 2 test trials)
- Long jump (minimum 15s between the 2 test trials)
- Throw sandbag (minimum 15s between the 2 test trials)
- Series Bounce and Catch (no rest needed)
- Series Throw and Catch (no rest needed)
- Series Hug knee (Preferred leg first, no rest needed) Static and Dynamic
- Series Grasp toe (Preferred leg first, no rest needed) Static and Dynamic
- Series Pick up cans (Preferred leg first, no rest needed)
- Series Jumping (4 items) (minimum 15s between the 4 items)
- Series Hopping (4 items) (alternate right and left leg and minimum 15s between the items performed on the same leg).

Appendix 13: PERF-FIT SCORE FORM

<table>
<tr><th colspan="6">Score form Agility and Power PERF-FIT©</th></tr>
<tr><td colspan="2">Name:</td><td colspan="2">ID:</td><td colspan="2" rowspan="2">Remarks</td></tr>
<tr><td colspan="2">Tester:</td><td colspan="2">Test date:</td></tr>
<tr><td rowspan="2">Agility ladder Running (0.01 s)
Always 2 trials</td><td>Trial 1
Time (0.01 s)</td><td>#Mistakes</td><td>Rest 15s
Trial 2
Time (0.01 s)</td><td>#Mistakes</td><td>Extra trial Running
Needed? 3 mistakes or more</td></tr>
<tr><td></td><td></td><td></td><td></td><td>Yes/No</td></tr>
<tr><td rowspan="2">Agility ladder Stepping (0.01 s)
Always 2 trials</td><td>Trial 1
Time (0.01 s)</td><td>#Mistakes</td><td>Rest 15s
Trial 2
Time (0.01 s)</td><td>#Mistakes</td><td>Extra Trial Needed? 3 mistakes or more</td></tr>
<tr><td></td><td></td><td></td><td></td><td>Yes/No</td></tr>
<tr><td rowspan="2">Extra trial Running Only if needed</td><td>Time (0.01 s)</td><td>#Mistakes</td><td rowspan="2">Extra trial Stepping Only if needed</td><td>Time (0.01 s)</td><td>#Mistakes</td></tr>
<tr><td></td><td></td><td></td><td></td></tr>
<tr><td rowspan="2">Side Jump
Always 2 trials</td><td>Trial 1
(# in 15 s)</td><td>Rest 15s Trial 2</td><td rowspan="2">Long jump Distance cm
Always 2 trials</td><td>Trial 1
distance cm</td><td>Rest 15s Trial 2 cm</td></tr>
<tr><td></td><td></td><td></td><td></td></tr>
<tr><td rowspan="2">Throw sandbag 2Kg Kneeling
Always 2 trials</td><td>Trial 1
Kneeling
distance cm</td><td>Trial 2
Kneeling
distance cm</td><td rowspan="2">Throw tennis ball If space available</td><td>Trial 1
distance cm</td><td>Trial 2
distance cm</td></tr>
<tr><td></td><td></td><td></td><td></td></tr>
</table>

Score form SIS Bouncing and Throwing PERF-FIT©

Name:			ID:		
Tester:			Test date:		
Ball Skills Bounce and catch *Max score 10 catches per item* Preferred R/L	**Item 1** # Bounce and catches with 2 hands *6 out of 10 continue*	**Item2** # Bounce and catches with Preferred hand *6 out of 10 continue*	**Item 3** # Bounce and catches with Non - preferred hand *6 out of 10 continue*	**Item4** # Bounce and catches with Preferred hand with clap *6 out of 10 continue*	**Item 5** # Bounce and catches with Non-preferred hand with clap *End of series*
SIS Bounce and Catch					
Ball Skills Throw and Catch *Max score 10 catches per item* Preferred R/L	**Item 1** # Throw and Catches 2 hands *6 out of 10 continue*	**Item2** # Throw and Catches Preferred *6 out of 10 continue*	**Item 3** # Throw and catches Non preferred *6 out of 10 continue*	**Item4** # Throw and catches with Preferred hand with clap *6 out of 10 continue*	**Item 5** # Throw and catches with Non-preferred hand with clap End of series
SIS Throw and Catch					

Score form SIS Balance PERF-FIT©

Name **ID**	**Tester** **Date**	**Remarks** **Shoes Y/N** **Protocol 1 2**

Knee Hug Static R *If 15s no second trial*	Item R Leg Trial 1 *Max 15 s*	Item R Trial 2 *Max 15 s*	**Knee Hug Static** L *If 15s no second trial*	Item L Trial 1 *Max 15 s*	Item L Trial 2 *Max 15 s*
Knee Hug Dynamic *If 8 steps no second trial*	Item **Knee Hug** Walk Trial 1 *Max 8 steps*		**Knee Hug Dynamic** *If 8 steps no second trial*	Item **Knee Hug** Walk Trial 2 *Max 8 steps*	
Grasp Foot Static Right *If 15s no second trial*	Item R Trial 1 *Max 15 s*	Item R Trial 2 *Max 15 s*	**Grasp Foot Static** Left *If 15s no second trial*	Item L leg Trial 1 *Max 15 s*	Item L leg Trial 2 *Max 15 s*
Grasp Foot Dynamic *If 8 steps no second trial*	Item **Grasp Foot** Walk Trial 1 *Max 8*		**Grasp Foot Dynamic** *If 8 steps no second trial*	Item **Grasp Foot** Walk Trial 2 *Max 8*	
Pick up 4 cans Right Stance # of right performed cans	Item R Stance Leg Close to far *Max 4*	Item R Stance Leg Far to close *Max 4*	**Pick up 4 cans** Left Stance # of right performed cans	Item L Stance Leg Close to far *Max 4*	Item L Stance Leg Far to close *Max 4*

Score form SIS Jumps Hops PERF-FIT©			
Name:		ID:	
Jump in **each square** **Item 1** Trial 1 *Max score 8 (no 2nd trial)*	Rest 15s **Item 1** Trial 2 *Stop if not 5 points*	Jump in **every other** square (red cross) **Item2** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 2** Trial 2 *Stop if not 3 points*
Jump in every other square over **5cm foam** **Item 3** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 3** Trial 2 *Stop if not 3 points*	Jump in every other square over **10cm foam** **Item 4** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 4** Trial 2 End of series
Hop in **each square** **Item 1 Right** Trial 1 *Max score 8 (no 2nd trial)*	Rest 15s **Item 1 Right** Trial 2 *Stop if not 5 points*	Hop in **every other** square (red cross) **Item2 Right** Trial 1 *Max score (no 2nd trial)*	Rest 15s **Item 2 Right** Trial 2 Stop if not 3 points
Hop Every other square over **5cm foam** **Item 3 Right** Trial 1 *Max score 4 (no 2nd trial*	Rest 15s **Item 3 Right** Trial 2. *Stop if not 3 points*	Hop every other square over **10cm foam** **Item 4 Righ**t Trial 1 *Max score 4 (no 2nd trial*	Rest 15s **Item 4 Right** Trial 3 End of series
Hop in **each square** **Item 1 Left** Trial 1 *Max score 8 (no 2nd trial)*	Rest 15s **Item 1 Left** Trial 2 *Stop if not 5 points*	Hop in **every other** square (red cross) **Item2 Left** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 2 Left** Trial 2 *Stop if not 3 points*
Hop every other square over **5cm foam** **Item 3 Left** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 3 Left** Trial 2 *Stop if not 3 points*	Hop every other square over **10cm foam** **Item 4 Left** Trial 1 *Max score 4 (no 2nd trial)*	

Appendix 14: Wedges (Ituen et al., 2020)

Fig. 1. Wedges with different angles: 3º, 6º, 9º and 12º.

The children in standing position will be given combinations of six wedges of varying degrees (1.5º, 3º, 4º, 5º, 6º, 9º). The wedges will be presented in pairs while the children will be in standing position, in a way they will not be able to see their feet. The children will be asked to make a distinction between constant stimuli and other stimuli at varying levels. Pairs of wedges with different angles, and a pair of the same angle will be presented for the test. The wedges will be presented in a random order. The children will then be asked to spot the heel that is higher without using visual cues. They will be asked to identify the more elevated heel by raising the ipsilateral hand. The children will be given one point for correct answers and zero point for incorrect answers. A penalty score will be given for incorrect answers based on the angle variance the child could not spot. An incorrect answer for the largest wedge difference will amount to the highest penalty score. The scores will then be summed up and used for the final analysis.

		Set 1		
Set 1	**Left(°)**	**Right(°)**	**Difference (°)**	**Correct(1) /mistake (0)**
1	9	12	3	
2	12	3	9	
3	3	1.5	1.5	
4	1.5	6	4.5	
5	6	9	3	
6	9	9	0	

7	9	4.5	4.5	
	Set2			
Set 2	**Left(°)**	**Right(°)**	**Difference (°)**	**Correct (1)/mistake (0)**
1	3	3	0	
2	3	12	9	
3	12	6	6	
4	6	4.5	1.5	
5	4.5	9	4.5	
6	9	6	3	
7	6	1.5	4.5	
	Set3			
Set 3	**Left(°)**	**Right(°)**	**Difference (°)**	**Correct(1) /mistake (0)**
1	3	4.5	1.5	
2	4.5	6	1.5	
3	6	3	3	
4	3	9	6	
5	9	9	0	
6	9	3	6	
7	3	9	6	

Appendix 15: Data recording sheet

Scoring sheets

ID Number:

Sociodemographic and Anthropometrics

Child's name:
Age:
Sex:
Class:
Weight:
Height:
Parents phone number:
Tester:

1. Score sheet for Beighton score

Table 1: Score Sheet for Goniometric assessment

Description	Bilateral Testing (Yes/No)	Score (Right & Left)
Passive dorsiflexion of the 5th metacarpophalangeal joint to ≥ 90 degrees		
Passive hyperextension of the elbow ≥ 10 degrees		
Passive hyperextension of the knee ≥ 10 degrees		
Passive apposition of the thumb to the flexor side of the forearm, while the shoulder is flexed at 90 degrees, elbow is extended and hand is pronated		
Forward flexion of the trunk, with the knees straight, so that the hand palms rest easily on the floor.		
Total		

2. Wedges

		Set 1		
Set 1	**Left(°)**	**Right(°)**	**Difference (°)**	**Correct(1) /mistake (0)**
1	9	12	3	
2	12	3	9	
3	3	1.5	1.5	
4	1.5	6	4.5	
5	6	9	3	
6	9	9	0	

7	9	4.5	4.5	
	Set2			
Set 2	**Left(°)**	**Right(°)**	**Difference (°)**	**Correct (1)/mistake (0)**
1	3	3	0	
2	3	12	9	
3	12	6	6	
4	6	4.5	1.5	
5	4.5	9	4.5	
6	9	6	3	
7	6	1.5	4.5	
	Set3			
Set 3	**Left(°)**	**Right(°)**	**Difference (°)**	**Correct(1) /mistake (0)**
1	3	4.5	1.5	
2	4.5	6	1.5	
3	6	3	3	
4	3	9	6	
5	9	9	0	
6	9	3	6	
7	3	9	6	

3. Score sheet for PERF-FIT

<table>
<tr><td colspan="6">Score form Agility and Power PERF-FIT©</td></tr>
<tr><td colspan="2">Name:</td><td colspan="2">ID:</td><td colspan="2" rowspan="2">Remarks</td></tr>
<tr><td colspan="2">Tester:</td><td colspan="2">Test date:</td></tr>
<tr><td rowspan="2">Agility ladder Running (0.02 s)
Always 2 trials</td><td>Trial 1
Time (0.01 s)</td><td>#Mistakes</td><td>Rest 15s
Trial 2
Time (0.01 s)</td><td>#Mistakes</td><td>Extra trial Running
Needed? 3 mistakes or more</td></tr>
<tr><td></td><td></td><td></td><td></td><td>Yes/No</td></tr>
<tr><td rowspan="2">Agility ladder Stepping (0.02 s)
Always 2 trials</td><td>Trial 1
Time (0.01 s)</td><td>#Mistakes</td><td>Rest 15s
Trial 2
Time (0.01 s)</td><td>#Mistakes</td><td>Extra Trial Needed? 3 mistakes or more</td></tr>
<tr><td></td><td></td><td></td><td></td><td>Yes/No</td></tr>
<tr><td rowspan="2">Extra trial Running
Only if needed</td><td>Time (0.01 s)</td><td>#Mistakes</td><td rowspan="2">Extra trial Stepping
Only if needed</td><td>Time (0.01 s)</td><td>#Mistakes</td></tr>
<tr><td></td><td></td><td></td><td></td></tr>
<tr><td rowspan="2">Side Jump
Always 2 trials</td><td>Trial 1
(# in 15 s)</td><td>Rest 15s Trial 2</td><td rowspan="2">Long jump Distance cm
Always 2 trials</td><td>Trial 1
distance cm</td><td>Rest 15s Trial 2 cm</td></tr>
<tr><td></td><td></td><td></td><td></td></tr>
<tr><td rowspan="2">Throw sandbag 2Kg Kneeling
Always 2 trials</td><td>Trial 1
Kneeling
distance cm</td><td>Trial 2
Kneeling
distance cm</td><td rowspan="2">Throw tennis ball
If space available</td><td>Trial 1
distance cm</td><td>Trial 2
distance cm</td></tr>
<tr><td></td><td></td><td></td><td></td></tr>
</table>

Score form SIS Bouncing and Throwing PERF-FIT©					
Name:			ID:		
Tester:			Test date:		
Ball Skills Bounce and catch *Max score 10 catches per item* Preferred R/L	**Item 1** # Bounce and catches with 2 hands *6 out of 10 continue*	**Item2** # Bounce and catches with Preferred hand *6 out of 10 continue*	**Item 3** # Bounce and catches with Non - preferred hand *6 out of 10 continue*	**Item4** # Bounce and catches with Preferred hand with clap *6 out of 10 continue*	**Item 5** # Bounce and catches with Non-preferred hand with clap *End of series*
SIS Bounce and Catch					
Ball Skills Throw and Catch *Max score 10 catches per item* Preferred R/L	**Item 1** # Throw and Catches 2 hands *6 out of 10 continue*	**Item2** # Throw and Catches Preferred *6 out of 10 continue*	**Item 3** # Throw and catches Non preferred *6 out of 10 continue*	**Item4** # Throw and catches with Preferred hand with clap *6 out of 10 continue*	**Item 5** # Throw and catches with Non-preferred hand with clap End of series
SIS Throw and Catch					

Score form SIS Balance PERF-FIT©

Name **ID**	**Tester** **Date**	**Remarks** **Shoes Y/N** **Protocol 1 2**

Knee Hug Static R *If 15s no second trial*	Item R Leg Trial 1 *Max 15 s*	Item R Trial 2 *Max 15 s*	**Knee Hug Static** L *If 15s no second trial*	Item L Trial 1 *Max 15 s*	Item L Trial 2 *Max 15 s*
Knee Hug Dynamic *If 8 steps no second trial*	Item **Knee Hug** Walk Trial 1 *Max 8 steps*		**Knee Hug Dynamic** *If 8 steps no second trial*	Item **Knee Hug** Walk Trial 2 *Max 8 steps*	
Grasp Foot **Static** Right *If 15s no second trial*	Item R Trial 1 *Max 15 s*	Item R Trial 2 *Max 15 s*	**Grasp Foot** **Static** Left *If 15s no second trial*	Item L leg Trial 1 *Max 15 s*	Item L leg Trial 2 *Max 15 s*
Grasp Foot Dynamic *If 8 steps no second trial*	Item **Grasp Foot** Walk Trial 1 *Max 8*		**Grasp Foot Dynamic** *If 8 steps no second trial*	Item **Grasp Foot** Walk Trial 2 *Max 8*	
Pick up 4 cans Right Stance # of right performed cans	Item R Stance Leg Close to far *Max 4*	Item R Stance Leg Far to close *Max 4*	**Pick up 4 cans** Left Stance # of right performed cans	Item L Stance Leg Close to far *Max 4*	Item L Stance Leg Far to close *Max 4*

Score form SIS Jumps Hops PERF-FIT©			
Name:		ID:	
Jump in **each square** **Item 1** Trial 1 *Max score 8 (no 2nd trial)*	Rest 15s **Item 1** Trial 2 *Stop if not 5 points*	Jump in **every other** square (red cross) **Item2** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 2** Trial 2 *Stop if not 3 points*
Jump in every other square over **5cm foam** **Item 3** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 3** Trial 2 *Stop if not 3 points*	Jump in every other square over **10cm foam** **Item 4** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 4** Trial 2 End of series
Hop in **each square** **Item 1 Right** Trial 1 *Max score 8 (no 2nd trial)*	Rest 15s **Item 1 Right** Trial 2 *Stop if not 5 points*	Hop in **every other** square (red cross) **Item2 Right** Trial 1 *Max score (no 2nd trial)*	Rest 15s **Item 2 Right** Trial 2 Stop if not 3 points
Hop Every other square over **5cm foam** **Item 3 Right** Trial 1 *Max score 4 (no 2nd trial*	Rest 15s **Item 3 Right** Trial 2. *Stop if not 3 points*	Hop every other square over **10cm foam** **Item 4 Righ**t Trial 1 *Max score 4 (no 2nd trial*	Rest 15s **Item 4 Right** Trial 3 End of series
Hop in **each square** **Item 1 Left** Trial 1 *Max score 8 (no 2nd trial)*	Rest 15s **Item 1 Left** Trial 2 *Stop if not 5 points*	Hop in **every other** square (red cross) **Item2 Left** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 2 Left** Trial 2 *Stop if not 3 points*
Hop every other square over **5cm foam** **Item 3 Left** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 3 Left** Trial 2 *Stop if not 3 points*	Hop every other square over **10cm foam** **Item 4 Left** Trial 1 *Max score 4 (no 2nd trial)*	Rest 15s **Item 4 Left** Trial 2 End of series

Appendix 16: Budget

Item	Estimated costs	Comments
Infrared Thermometer	R1000	Due to the risk of COVID-19 pandemic, we will be using an infrared thermometer to assess the temperature of the children to determine and minimize the risk of infection.
Digital Weighing Scale	R600	A weighing scale will be used to measure the weights of the children. This is one of the outcomes that will be used to determine their body mass index (BMI).
Height Scale (Stadiometer)	R500	A height scale will be used to measure the heights of the children. This is also an outcome that will be used to calculate their BMI.
Goniometers	R500	Three Goniometers of specific sizes (12, 8 and 6 inches) suitable for the target joints will be needed for measuring the subject's joint range of motion.
PERF-FIT Instrument		Provided by Bouwien Smits-Engelsman
Wedges		Provided by Bouwien Smits-Engelsman
Printing of documents	R818.4	PERF-FIT (4), Beighton (1), consents (3), assent (3)= 11 pages i.e. R0.60/page = R6.6/participant; R6.6* 124 participants= R818.4
Face shields and masks	R7812	Face shields priced at R63 each will be provided for the 124 subjects to reduce the risk of COVID-19 transmission.
Hand sanitizer	R70	A 500ml, 80% alcohol hand sanitizer will be provided for both the researchers and the subjects to reduce the risk of COVID-19 transmission.
Transport to and from study setting (schools)	R1800	Commercial transport system will be used to travel to the study setting. i.e. R5/km x 12 km x 5 days x 6 weeks: R1800
Incentives	R1500	Because of the exertive nature of some of the tests, fruits, snacks and water will be provided for each child. i.e. R30/Child; R30*124 subjects= R3720

Miscellenous	R1000	
Total	R15,600.4	