Proprioception, balance and lower limb strength in Nigerian children (7-10 years) with Generalized Joint Hypermobility and Developmental Coordination Disorder

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Declaration

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Synopsis

Background and justification: African children are reported as having a higher prevalence of generalised joint hypermobility (GJH) than their Caucasian counterparts (1). It is believed that abnormal joint biomechanics as a result of the joint laxity contribute to the damage of joints(2). The ability to perceive movement or position sense at joints (proprioception) is necessary for good postural control and motor performance(3). Sensory receptors carry information from the joints to the central nervous system for interpretation and appropriate motor response. Damage to these receptors or joint pain may have a negative effect on proprioception and motor control. A number of children with GJH also present with poor motor coordination and some may even have Developmental Coordination Disorder (DCD). Children with DCD and GJH also have similar functional difficulties (4). Both groups of children display difficulty in motor activities at school and home and are referred to as clumsy. There is evidence that poor motor coordination seen in children with DCD may be as a result of their inability to adequately control their flexible joints during movement (5). The role proprioception, balance and muscle strength plays in the relationship between GJH and DCD is still not clear.

Aims and objectives: The main aim of this study was to determine whether proprioception, standing balance and strength in the lower limbs was different between children with GJH and children with normal joint mobility (NM). The specific objectives were to firstly identify the prevalence of GJH in a sample of Nigerian children and determine whether age and gender are related with the prevalence of GJH. Secondly, to determine whether having DCD or not was associated with differences in performance on these measures in children with and without GJH.

Method: Children aged 7 to10 years, attending private and public schools in Southern Nigeria participated in this study. Joint hypermobility was measured using the Beighton score with goniometry. A cut-off score of ≥5 hypermobile joints was used to determine group allocation (GJH and NM). This cut-off point was one score higher than the score used by Birrell et al., (1994) to measure hypermobility in a Nigerian sample (≥4 joints) (6). The two groups were then assessed and compared on measures of isomeric lower

limb muscle strength (using a hand-held dynamometer), balance (using the Y-Balance test), and proprioception (measured using a novel experimental test, created for this study, called the Wedge Test). The wedge test comprises of four sets of wooden wedges, each set varying in height: 3mm, 6mm, 9mm and 12mm. The sets were randomly presented. Sets were either the same (e.g. 3mm and 3mm, 6mm and 6mmm, etc.) or different heights (for example, 6mm and 3mm or 9mm and 6 mm, etc.). The children were requested to report the difference in perceived height while standing on two wedges. A penalty score was calculated related to the child's inability to detect the height difference. Thus higher penalty scores were indicative of poorer performance.

The motor performance of the two groups was assessed using the Movement Assessment Battery Children 2nd edition (MABC-2) (two groups were subsequently formed: Typically developing (TD) group, who had MABC-2 scores >16th percentile, and the DCD group who scored ≤ 16th percentile).

Data analysis: Descriptive statistics were used to describe the sample demographic characteristics, motor performance and joint mobility of the children. The Shapiro Wilks test was used to check for normality of the data on proprioception, balance and strength. Mann-Whitney-U test was used to determine whether the ranking of scores in proprioception, lower limb strength and balance were significantly different between GJH and NM groups. The two-way ANOVA was used to examine the influence of two independent variables GJH/NM and DCD/TD on proprioception, lower limb strength and balance.

Results: 162 pupils participated in this study. 84 children (girls =52, boys=32) were classified as having GJH (51.85%) using the selected cut off score (≥5). Using our convention of hypermobility defined as ≥5 joints, we found that children with NM were less able to detect difference in wedge height in comparison children with GJH (U= 2547.5, z= 2.44, p=0.013), suggesting poorer proprioceptive ability. Children with NM and children with GJH were not different in performance on any of the six components of the Y-balance test [right anterior (p=0.87), left anterior (p=0. 96), right posteromedial (p=0.30), left posteromedial (p= 0.16), right posterolateral (p= 0.29), left posterolateral (p=0.63)]. On the six lower limb strength measures the groups were only different in the left knee extensors (p=0.01), right plantarflexors (p=0.28), left plantarflexors (p=0.19),

right dorsiflexors (p=0.05), left dorsiflexors (p=0.10)]. The result of the two-way ANOVA showed that DCD did not have an effect on proprioception (p=0.22), balance (p=0.69), and strength (p=0.59) in the lower limbs of children with GJH.

Conclusion: It seems GJH does not appear to affect balance or motor ability. However the result of better proprioceptive awareness in children with GJH as reported in this study needs to be varied in other future studies. The high prevalence of hypermobility in the sample using a cut off score of ≥5 joints, warrants further investigation as recommended cut off points appear to overestimate GJH in this population. The fact that only two children in the GJH group reported pain contradicts the notion that hypermobility is necessarily associated with more joint pain. However, since this was a relatively young sample, further longitudinal studies are required to give more insight into why some children with joint hypermobility develop symptoms later in life. In practical terms, we suggest that children with GJH be monitored to determine if they eventually develop arthralgia later in life.

Keywords:
Joint hypermobility
Balance
Proprioception
Motor coordination
Developmental Coordination Disorder

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Glossary of abbreviated terms

AMEDA: Active Movement Extent Discrimination Apparatus.
The AMEDA is a measure of proprioception that is conducted using active movements.

BMI: Body Mass Index.
It is measured as weight (kilograms) divided by the height squared (metres).

COG: Centre of Gravity
COG is an imaginary point in space calculated biomechanically from measure forces and moments, where the sum total of all the forces equal zero (7).

COP: Centre of Pressure
COP refers to the location of the global ground reaction force of the centre of mass. An increase in the centre of pressure excursion velocity measures, is an indication of an impaired postural control (8,9).

DCD: Developmental Coordination Disorder
DCD is a neurodevelopmental motor disorder characterised by delayed motor skill acquisition and poor motor performance that occurs in the early developmental period. The deficit in motor skills is sufficient enough to impact the performance of activities of daily living as well as academic performance and it should not be as a result of a medical condition like cerebral palsy or muscular dystrophy (10).

FPS: Faces Pain Scale
The FPS is a questionnaire that can be used to measure pain in children and adults with illiterate adults having a stronger preference for it because of its simplicity (11).

HDCT: Heritable Disorder of Connective Tissue
The HDCT is a group of genetic disorder that affects connecting tissues matrix. They comprise of Ehlers-Danlos syndrome, Marfan syndrome and Osteogenesis Imperfecta (12,13)

HHD: Hand-Held Dynamometer

The HHD is a device that is able to measure the force or power created by contraction of a muscle or muscle group (14,15).

JHS: Joint Hypermobility Syndrome

JHS is a condition in which joint hypermobility is associated with musculoskeletal symptoms in more than four joints, including pain of over a period of more than 12 weeks (16,17).

JPR: Joint Position Reproduction

JPR is a measure of proprioception and it occurs when a previously experienced joint position (target position) is reproduced, either when the joint is passively or actively moved into the same range (18).

JPS: Joint Position Sense

JPS refers to an individual's awareness of (ability to perceive) joint position in space as a result of sensation from the joint receptors (19–21).

MABC-2: The Movement Assessment Battery for Children 2nd Edition

MABC-2 is a norm-referenced assessment measure that can be used to assess movement problems in children, and is the most commonly reported assessment used to determine the presence of DCD in school aged children (22).

NM: Normal Mobility is the acceptable value of movement at a joint and it is measured in degrees

ROM: Range of Motion

ROM is the amount of movement at a joint (23,24).

SEBT: Star Excursion Balance Test

SEBT is a physical test that can be used to measure balance. It involves maintaining a single leg stance while the other leg is used to move the indicator reach (a wooden block) with the foot as far as possible without falling (25,26).

TD: Typically Developing

TD refers to children who follow normal developmental milestones in the appropriate and commonly accepted time frame (27,28).

TTDPM: Threshold To Detection of Passive Motion is a measure of proprioception and it is reported as soon as joint movement and direction is perceived (18). This assessment requires that joint movement be detected under different velocities.

TTLQ: The Teachers Traffic Light questionnaire assists teachers in identifying children that may have functional motor problems. It gives a global impression of the child's motor performance according to the teacher's observation.

1 Introduction

Generalised Joint Hypermobility (GJH) is common among African children and occurs as a result of their inherent ligamentous laxity (6,29,30). It has been reported that the excess movement in the hypermobile joint account for the number of challenges. These includes a higher prevalence of pain and a greater incidence of injury (31,32). In addition, children with hypermobility in the lower limbs joints have reduced proprioception (33,34), weaker lower limb muscles (35), and poorer balance compared to their peers with normal mobility (3,36,37). Whether African children who are hypermobile are affected by these same challenges is not well described in the literature.

In this thesis, we explored the relationship between GJH, proprioception, balance and strength in children aged 7-10 years attending a mainstream school, in southern Nigeria. We further explore the effects of poor motor coordination (Developmental Coordination Disorder) on these aspects in children with and without GJH.

1.1 Defining Generalised Joint Hypermobility

GJH is characterised by increased laxity of the connective tissue (ligaments, joint capsules, tendon) surrounding a joint, which manifests as a more than normal range of motion (ROM) at several joints (13,26). GJH is not considered a pathology, as most individuals with GJH lead a symptom-free life (35,36).

Individuals with GJH are distinct from other groups, who may acquire joint laxity as a result of excessive stretching through their involvement in sports (40,41). Other groups with diagnosed heritable disorders of connective tissues (HDCT), such as Marfan syndrome, Ehlers-Danlos syndrome and Osteogenesis Imperfecta, also experience joint hypermobility (36,38,39).

1.1.1 Prevalence and Epidemiology

The prevalence of GJH is varied and is widely reported to be between 3% and 30%, depending on ethnicity, age and gender (5,16,43). The prevalence of GJH is reported to be higher among Africans and Asians than Caucasians (12,30,44). Though no evidence has been provided for the high prevalence of GJH among Africans and Asians, it is

assumed that it may be as a result of their genetic make-up (45). To date only one documented study on GJH has included a population of children and adults from western Nigeria (6); the other studies have been among adults Nigerian population only (46,47). Although females are more frequently reported to have GJH, not all studies have shown an association between GJH and gender (16,29,48). However, there has been a consistent association between age and GJH with younger children having more hypermobile joints than adults(16,42,49,50).

In this thesis, we explored whether the prevalence of GJH in our sample of Nigerian children is indeed higher compared to other studies, and whether age and gender are related to the prevalence of GJH.

1.1.2 Consequences of GJH

The presence of GJH has been found to have dual consequences. While some individuals with GJH enjoy the positive consequence of GJH and remain asymptomatic throughout life, some individuals experience problems.

A positive consequence of GJH is seen in hypermobile individuals that participate in sports that require lot of flexibility; examples include ballet, and gymnastics. GJH coupled with good motor control enables the performer to achieve complex poses and postures necessary to excel in their sports (5,51). Gymnasts and ballerinas with GJH for example are able to take advantage of the extra degrees of movements at their joints and can use it to exhibit better performance than their colleagues with normal mobility (NM).

There are cases of individuals with GJH, who experience the negative consequence of excessive joint laxity (38,39,52). Several studies have reported that GJH results in abnormal biomechanics at the joint, which may give rise to pain and increase the risk of injuries (31,33,53). It is also believed that some individuals with GJH may develop Joint Hypermobility Syndrome (JHS) (46). JHS has been defined as an association of GJH with musculoskeletal symptoms, such as pain and mild swelling after exercise or activity in more than four joints, including pain over a period of 12 weeks when HDCT and other causes of symptoms have been excluded (16,54).

In some children, laxity in several joints may delay motor development (55) or be associated with motor coordination problems (4,5,56). Motor control requires musculoskeletal, neuromuscular, sensory systems, anticipation, balance and internal representation (57,58), but in a situation where the joint laxity changes the biomechanics of the joint, the motor control process may be affected. The movement patterns of children with GJH have been referred to as clumsy and uncoordinated (36,59). It has been suggested that their clumsy movement patterns seen in children with DCD may be as a result of impaired proprioception (37,60).

We hypothesise that GJH in the lower limbs would affect proprioception and therefore balance control, which is an aspect of motor coordination. However it is not known if GJH on its own can cause a deficit in proprioception thereby leading to uncoordinated movement patterns.

1.2 The Influence of Proprioception on movement

There are various kinds of mechanoreceptors at the joints which provide (the individual) with important proprioceptive information. Proprioception is one's ability to perceive body/joints movements or body/ limb position in space whereas kinaesthesia has been referred to as the awareness of joint motion (18). When a joint is moved or a limb is in a position, the information is passed to the brain through mechanoreceptors embedded in the muscles, ligaments, tendons, capsules and skin surrounding a joint (58,61–63). The proprioceptive information is then organised, interpreted and integrated in a feed forward loop within the central nervous system to produce an appropriate motor response (64). Proprioception thus starts from when the sense organs receives a stimulus to the point where it ends with an appropriate motor response. This function makes proprioception essential in both motor skill learning and motor control (3).

Joint movement and joint position seem to go hand in hand during performance of activities of daily living, indicating a strong association between them and in effect making it difficult to discriminate between proprioception and kinaesthesia (18,34). For the purpose of this thesis we will use the term proprioception since it will give a reflection of both the joint position and joint movement as demonstrated during performance of activities of daily living (18).

It was hypothesised that GJH increases the risk of injury at a joint which could lead to damage of various mechanoreceptors and thus reduce proprioception (5). When GJH is associated with musculo-skeletal symptoms in more than four joints, including pain of over a period of more than 12 weeks It is referred to as Joint Hypermobility Syndrome (JHS) (16). Fatoye et al., (2009) found proprioception to be significantly different between children with JHS and children without JHS (36). The authors suggested that proprioception deficit in participants with JHS maybe linked to their joint laxity.

However, these studies were limited because the participants had pain, which in addition to joint laxity may have altered afferent sensory input (33,60).

1.3 The Role of Balance on Motor Performance

Balance is the ability to maintain one's centre of gravity (COG) within a base of support while performing functional activities (57,65). Control of balance results from integration of multiple systems and overtime with practice, proficiency is achieved (66–68).

Studies have shown that individuals with proprioception deficit present with balance difficulties during functional activities (7,8,69)(67,70–72). Lee et al., (2009) in a study of ten young men with unilateral anterior cruciate ligament injury observed a positive correlation between balance and threshold for detection of passive motion (TTDPDM) (73). TTDPDM is a measure of proprioception and it is reported as soon as joint movemnet and direction is perceived (18,74). Schbert-hjalmarsson and colleagues (2012), studied 20 children with JHS and 24 Typically Developing (TD) children, the children with JHS were limited in balance as compared to the TD children.

It is still not clear if other factors apart from proprioceptive deficit, can negatively impact on balance performance in children with GJH.

1.4 The Effect of Strength on Movement

Having adequate muscle strength is one of the building blocks for motor control. Muscles contain sensory receptors that provide the CNS with information regarding joint position and movement (74,75). It is thus possible that muscle weakness may be a reason why

some individuals with GJH have poor motor control (5,76). This reasoning becomes significant since we have ballerinas and gymnasts who have good muscle strength and can therefore demonstrate good motor control despite their joint laxity (77). Though they are more susceptible to injuries than their non-hypermobile counterparts.

Grahame (1999) suggested that the difference in the motor performance of individuals with GJH may be as a result of variety in the degree of hypermobility (13). For instance, motor control in an individual with a Beighton score of 5 will have a different from an individual with a score of 7 and above (78). Clinch et al., 2011 found a stronger evidence of association between GJH and physical activity on a Beighton cut off of ≥6 compared with ≥4 (29). In addition, it is not clear if the connective tissues of individual who acquire hypermobility through stretching are different from those who are genetically hypermobile (13).

Ageberg et al., (2005), investigated the influence of muscle strength on balance among 36 patients with unilateral, non-operated acute anterior cruciate injury, the study found an association between high muscle strength and good balance performance, implying that muscle strength seems to be significant in balance performance (74). Also, Juul-Kristensen et al., (2009), found that children with GJH and children without GJH were not different in knee function and strength (79).

1.5 Motor Control Disorders and GJH

Clumsy, uncoordinated movements are characteristic of children with Developmental Coordination Disorder (DCD). DCD is a disorder that affects the learning and performance of motor activities of daily living (10). Children with DCD present with a wide range of motor impairments, some may manifest deficiencies in gross motor skills, others may have difficulties in fine motor skills while some may combine difficulties in both areas (80–82).

Perceptual information processing and motor control mechanism are the two path ways that lead to movement and the process is sequential (83). Perceptual processing has to do with the way sensory information is registered, integrated and interpreted while motor control mechanism is the ability to choose an appropriate motor response to the processed

sensory information." A disruption in either/ both of the perceptual processing and motor control mechanism will lead to motor deficit.

Children with DCD have shown deficits in internal modelling, rhythmic coordination, and sensoriperception among many others. Of all the deficits identified in children with DCD, internal modelling and stable coordination pattern have been the most significant (84).

Internal modelling which is also referred to as predictive control of movement is the ability to either copy a motor command already stored in the brain or generate a new one to achieve a desired goal (Wilson et al., 2013).

In performing activities like writing, dressing running that require motor skills, the ability to either generate a copy or new motor command is very important in learning a new motor skill or perfecting an old one (Bo & Lee 2013). However, it is still not clear how biomechanics affects prediction of movement (84).

Kirby and Davies (2007) found an overlap of symptoms between GJH and DCD, with a high percentage of children with DCD also being hypermobile (4). Jelsma and colleagues (2013) also found a high percentage of GJH among children with DCD in their study (64%) and found a negative correlation between their motor performance and joint mobility (5). It is possible that, the similarities in symptoms between JHS and DCD may be as a result of the GJH (5). With high number of children with DCD also found to be hypermobile, it is not known if their poor motor coordination is as a result of joint laxity(5).

As previously highlighted GJH increases the risk of trauma to the joint receptors and may reduce proprioception at the joint. Considering the role of proprioception in motor control, a deficit in proprioception will negatively impact on motor performance (83,84) Studies that have investigated proprioception have reported deficits in children with DCD when compared to typically developing children (85–87). However, all these studies were done in the upper limbs. In addition, none of the studies considered the joint mobility of the participants.

It therefore appears that when the flexibility and motor control of individuals with GJH is optimum, the negative consequence of joint laxity is reduced and the ability to maintain control at the joints is enhanced (5,53).

1.6 Rationale

To date, the only known study of GJH among Nigerian children was reported by Birrell and colleagues (1994). Their study found a high prevalence of GJH among their sample population of children and adults (6). With a documented high prevalence of GJH among a Nigerian population, it will also be necessary to investigate if there will be any association between GJH and motor coordination among children in Nigeria. There are several other issues around GJH that require further investigation. The first relates to the relationship between GJH and pain. There is limited information concerning the association between joint pain and GJH in children. Previous studies linking GJH with pain in children had limitation relating to sample size, methods of assessing hypermobility and pain (29). Evidence suggests that the prevalence of pain among children with GJH varies between 30% to 55% (29) and thus it will be useful to investigate the prevalence of pain in Nigerian children with GJH.

It is believed that controlling extra degrees of movement at the joints requires more postural control (5), but it is known that some individuals with GJH can use their flexibility to gain supremacy in motor performance over their contemporaries who have NM. However, the mechanisms by which they are able to achieve this are not known.

Children with DCD and GJH also have similar functional difficulties (4). Both group of children display difficulty in motor activities both at school and home. Investigating proprioception in the lower limbs of children with DCD will give more insight into how they control degrees of freedom at the joint. In addition, we may gain a better understanding of the relationship between balance impairment and deficits in proprioception.

1.6.1 Research Questions

Therefore, based on the literature, we have identified the following questions:

1. Is there a difference in proprioception (as measured by the wedge test) in children with GJH and children with NM?

2. Is there a difference in balance (as measured by Y-balance) in children with GJH and children with NM?
3. Is there a difference in lower limb strength (as measured by the hand held dynamometer) in children with GJH and NM?
4. In all of the above, does having DCD or not (TD) influence the results?

1.6.2 Hypotheses

It was hypothesised that the children with NM would perform better on the proprioception and balance test than children with GJH. It was also hypothesised that poor motor coordination will impact negatively on the proprioception, balance and strength of children with GJH.

1.7 Aims and Objectives

The aims of this study were to determine whether proprioception, strength in the lower limbs and balance was different between children with GJH and children with NM, as well as, to find out if motor performance had any effect on the abovementioned variables.

The specific objectives were to:

1. Identify the prevalence of GJH in our sample of Nigerian children and determine whether age and gender are related with the prevalence of GJH.
2. Compare four groups of children: with and without GJH and with /without DCD in terms of:
    i. Demographic characteristics (BMI, handedness, age, gender.)
    ii. Lower limb strength using the Handheld Dynamometer
    iii. Proprioception using the wedge tests.
    iv. Balance using the Y-balance test.

1.8 Research Setting

The study was conducted in Uyo Local Government Area, the capital of Akwa Ibom State in Southern Nigeria. Figure one shows the various local government areas in Akwa Ibom state. Uyo covers an area of 187km^2 and has a population of 309,573. Data collection took place in two private owned and one government owned (public) schools. The initial plan was to conduct the study in a public school, where there was access to more children.

However, majority of the parents could not provide basic information on the exact date of birth of the children, which was necessary for the study. This informed the decision to include private schools where the parents could provide information about their children's date of birth. All the schools that participated in the study were English speaking schools. The schools are located in areas of low socioeconomic status with low and medium income earners. The children were active and apparently healthy. Data collection took place over three school terms.

Figure 1.1: The map of Akwa Ibom state, southern Nigeria

1.9 Significance of Study

As physiotherapists are involved in the management of children with GJH, identifying the deficits as it relates to motor performance is important. This will ensure that right form of management is provided for children with GJH. The result of this study will also provide more evidence on the characteristic of GJH among children between the ages of seven and ten years. The information on pain, muscle strength, balance, and proprioception will also enable health care providers to know if children with GJH are different from children without GJH as evidenced in literature.

2 Literature Review

In this section, we reviewed and synthesised information from literature related to the various objectives of this study, such as prevalence of GJH in children, motor coordination and motor control in children with GJH. The review is focused on defining GJH and evaluating the impact of GJH on proprioception, strength and balance in children. We also examined the relationship between motor coordination and GJH.

Data for the review were sourced from several online databases; such Clinical key, PubMed, EBSCOHost, JSTOR, Google Scholar, Science Direct and Wiley online library. Key words that were used to search included 'DCD', 'joint hypermobility', 'muscle strength', 'proprioception' and 'balance.' References of useful articles found from the above databases were further investigated for more leads.

2.1 Joint Hypermobility

Joint hypermobility (JH) is defined as an increase in the joint range of motion (ROM) of an individual when sex, age and ethnicity have been taken into consideration (88). JH can be localised, that is, in one joint, or generalised, across several joints (GJH).

2.1.1 Pathophysiology/underlying mechanisms of GJH

GJH occurs as a result of laxity of the connective tissues of the joint, particularly the ligaments. Joint connective tissues, such as ligaments, joints capsules, and tendons contain collagen that enables them to elongate/stretch during movement. There are two main causes of GJH. The first relates to the hereditary disorder of the collagen, and the second is the purposeful acquisition of increased range by stretching (13,40,89).

2.1.2 Assessment of Generalised Joint Hypermobility

Simple scoring systems are the general means by which researchers have defined GJH. It was first devised by Carter and Wilkinson (1964) than modified by Beighton et al., 1973. The defined GJH as being present when three of the following test is positive, provided both upper and lower limbs are involved,

- Passive apposition of the thumb to the flexor aspect of the forearm.
- Passive hyperextension of the fingers so that they lie parallel with the extensor aspect of the forearm.

- Ability to hyperextend the elbow more than 10^0.
- Ability to hyperextend the knee more than 10^0.
- An excessive range of dorsiflexion of the ankle and eversion of the foot (90).

Other methods of assessing GJH include Comtompasis score and the 10 point hospital del mar (Barcelona criteria) (2,17,91). The scoring systems are most useful in epidemiological studies because they are fast and easy to administer (90). However, when precise quantification of movement at a joint is required goniometry is used.

The Beighton Scale was first devised by Carter and Wilkerson (1964) and later modified by Beighton and Horan (1973) and Grahame and Jenkins (1972) (90). Of all the modifications that were made to the scoring system devised by Carter and Wilkerson (1964), it is the modification by Beighton and Horan (1973) that gives a better reflection of generalised joint laxity because of the inclusion of forward flexion of the trunk and dorsiflexion of the little finger beyond 90^0.

The Beighton score involves allocating a point for each side of the body that is hypermobile. The following joints are tested 1) passive dorsiflexion of the little finger beyond 90 degrees, 2) passive apposition of the thumb to the flexor aspect of the forearm, 3) hyper extension of both elbows beyond ten degrees, 4) hyper extension of the knees beyond ten degrees, 5) forward flexion of the trunk, with knees straight, so that the palms of the hands rest easily on the floor. The score ranges from 0-9, a high score indicating GJH. Beighton scores of ≥ 4 and ≥ 5 have been the most commonly used cut-off scores in studies to define GJH (6,29,33).

Smits-Engelsman et al., (2011) conducted a study of the validity of the Beighton score among 551 children, aged 6-12 years recruited from elementary schools (16). The authors measured passive ROM of major joints in the extremities using goniometry in a standardised manner. 35% of the children scored more than 5/9 on the Beighton score. The children who scored high on the Beighton score also showed increased ROM in other joints measured (16). Juul-kristensen et al., (2007) reported interrater reliability of the Beighton score to be above 0.80 except for little finger and elbow that was ≥ 0.60 (92). In another study of intrarater and interrater reliability of the Beighton score by Remvig et

al., (2007) the Kappa score for intrarater and interrater reliability were 0.75 and 0.78 respectively (93).

2.1.3 Epidemiology of Generalised Joint Hypermobility

The lack of international consensus on the Beighton score cut-off to be used to define GJH has accounted for the variation in the reported prevalence of GJH (3%-33%). For example, Smits-Engelsman and colleagues (2011) considered two different cut off points to identify children with GJH in the sample population (16). Using a Beighton score cut-off of ≥ 5 they found the prevalence was 35.7% but it reduced to 9.5% in the same sample population when a Beighton score cut-off of ≥7 was used.

Seçkin et al.,(2005) reported a prevalence rate of GJH at ≥4=62% and ≥5=15 among 861 Turkish high school students with mean age (S.D.) =15.4 years (1.1) % (43). Birrell and colleagues (1994) used a score of ≥ 4 to define GJH in 204 Nigerian individuals aged range 6-66 years (6). Toit and colleagues (2011) defined GJH among 480 individuals in South Africa, age range of 18-25 years, with a Beighton score cut of ≥ 4 (42). The prevalence of GJH was 26.19% among the sample population. Van Dongen et al., (1999) investigated GJH among 509 South African pregnant women (94). GJH was defined with Beighton score cut-off of ≥5.4.9% of the women were hypermobile. It appears literature on hypermobility among African children is limited. There is no consensus on the Beighton score cut-off for children.

Studies that used a score of ≥4 was reported to be an exaggerated prevalence of GJH and have therefore called for a stricter cut off of the Beighton score (16,29,51). Some studies have shown how prevalence of GJH changes when different Beighton cut off scores is used.

Therefore several authors have called for consensus on the Beighton cut-off point so that hypermobility will not be under or over represented in a population (29,90). However, since joint laxity reduces as age increases it might therefore seem appropriate to have a higher cut-off for children and a lower cut off for adults (6,53). In addition, it has been recommended to take ethnicity as consideration and use a higher cut-off score for Africans and Asians. A Beighton score of ≥7 may be appropriate.

2.1.4 Distinguishing between Generalised Joint Hypermobility and Joint Hypermobility Syndrome

GJH and Joint hypermobility syndrome (JHS) are not the same condition although information about these two terms have been mixed up in literature (90). While GJH presents with only articular features, JHS has shown both articular and extra-articular features.

JHS is characterised by the presence of musculoskeletal symptoms (joint pain, dislocation and subluxation) in more than four joints over a period of 12 weeks in the absence of other rheumatic, neurological, skeletal and metabolic disorders (13,16). Although JHS is a multi-system disorder, the symptoms are mild and not life threatening, for this reason it is also referred to as Benign Joint Hypermobility Syndrome (BJHS) (33,95).

The presence of these extra-articular features made the Beighton scoring system inadequate to diagnose JHS so this led to the formation of the Brighton criteria which comprises of major and minor criteria to identify JHS. The major criteria are, a Beighton score of 4/9 or greater (either currently or historically) and arthralgia for longer than 3 months in 4 or more joints. Minor criteria are, 1) a Beighton score of 1, 2 or 3/9 (0, 1, 2 or 3 if aged 50+). 2) Arthralgia (>3 months) in one to three joints or back pain (>3 months) Spondylosis, Spondylolysis or spondylolisthesis. 3) Dislocation/subluxation in more than one joint, or in one joint on more than one occasion. 4) Soft tissue rheumatism >3 lesions (e.g. epicondylitis, tenosynovitis, bursitis). 5) Marfanoid habitus (tall, slim, span/height ratio >1.03, upper: lower segment ratio <0.89). 7), Arachnodactyly (positive Steinberg/wrist signs). 6) Abnormal skin: striae, hyper extensibility, thin skin, papyraceous scarring. 7) Eye signs: drooping eyelids or myopia. 8) Varicose veins or hernia or uterine/rectal prolapse, (52).

JHS is diagnosed in the presence of two major criteria, or one major and two minor criteria, or four minor criteria. Although there is no biological maker for JHS, it has autosomal dominant inheritance, so two minor criteria will suffice where a family member has similar problem (39,90).

2.2 Generalised Joint Hypermobility and Pain

Laxity of joint makes the joint unstable, predisposing it to joint subluxation and dislocation in the short term and osteoarthritis in the long term. The soft tissues around the joints are not usually strong enough to cope with the laxity and this may also increase the frequency of strain and sprain of the soft tissues during activities (13). The resultant effect of this on the joint is repetitive trauma at the joint and the consequence is pain and dysfunction (31).

Pain is measured using either the self- reported one-dimensional or multidimensional scales (96). Examples of the one dimensional scale include Visual Analogue Scale (VAS), the Numerical Rating Scale (NRS) and the Verbal Rating Scale (VRS), and the multidimensional scales are McGrill pain questionnaire and St. Antonine pain scale. These tools have been found to be reliable and valid measure of pain in both adult and children population (97,98).

The Faces Pain Scale (FPS), NRS, VAS, pieces of hurt, colour scales and adjective scales have been used to evaluate pain in children and FPS is reported to be the most preferred by children, probably because it is simple to administer (99). It has a reliability range of 0.26-0.70. The FPS has six faces (see figure 2.1) with numeric values of 0, 2, 4, 6, 8, and 10 assigned to each (97). The first which has a neutral face measure 0, signifies no pain and the last face measures the most pain I have ever experienced.

Figure 2.1: The Faces pain scale

Tobias et al., (2013), assessed pain with the focus 17 pain questionnaire and reported that adolescents with GJH had higher musculoskeletal pain at the knee joint (8.8%) than the ankle joint (6.8%) (31).

Evidence has shown that children with GJH (age ten and below) experience less pain and dysfunction compared to adolescents (age 13-18 years) with GJH. Smits-Engelsman and

colleagues (2011), argued that pain was not a necessary symptom to assess in the children with GJH in the age range 6-12 years because they were not different in frequency of reported pain after exercise from children with normal mobility (16). Similarly, Juul-Kristensen et al (2009) found the frequency of musculoskeletal pain in eight years old children with and without GJH to be the same (51).

Tobias and colleagues (2013), in their study of 2901 children, first identified children with GJH at a mean age of 13.8 years and found a prevalence of 4.6% among the study population (31). They re-evaluated the children 5 years later at the mean age of 17.8 years for moderate to troublesome musculoskeletal pain which has lasted for over 1 day, 16.1% reported pain at the lower back, 9.5% reported pain at the shoulder, 8.8% reported pain at the knee and 6.8% reported pain at the ankle. They also found that knee pain was higher among the participants with GJH who were also obese. Their study result showed an association between GJH and pain at the shoulder, knee and ankle but not at the lower back, they argued that this association may be as a result of age and Beighton cut-off of ≥6 that was used. This study provides evidence that older children with GJH may experience more pain than younger children with GJH. However, there is an indication that their body mass should also be taken into consideration.

2.3 The Impact of Obesity on Generalised Joint Hypermobility and proprioception

It has been suggested that an increase in the mechanical load on weight-bearing joint can increase the risk of pain and injuries at the joint (76,100). It has also been suggested that the report of musculoskeletal pain will higher in hypermobile individuals who are obese (100). This is because of the impact of excessive body weight on the abnormal joint structure. The effect of this over time is possible deficit in proprioception or balance (76). Clinch et al., 2011 in their study found an association between an association between GJH and obesity (29). Similarly, Tobias and colleagues (2013) found an association between GJH and obesity. Their study found a higher risk of knee pain in adolescents with GJH (31). In contrast, Bout-Tabaku et al., (2014) in their study of 142 adolescents did not find an association GJH and obesity (76).

2.4 Generalised Joint Hypermobility and Motor Control

Effective movement gives children the opportunity to play, interact with their environment, perfect their motor skills and learn new ones (101). For individuals who are involved in competitive sports, success in their field will rely on control and accuracy of movement (18,64). The ability to gain good motor control when performing movement is as a result the brain integrating sensory information on proprioception, balance and strength (3). This cerebral processing shows the complexity of motor performance.

Children with GJH may demonstrate the same level of motor performance and physical activity as their counterpart without GJH (51,102). There has also been a high prevalence of GJH among ballet dancers gymnasts and swimmers, most likely because those sporting activities require joint flexibility for their good performance (5). However, it is important to know if the presence of GJH affects motor performance (coordination and balance) in a negative way. It may be logical to assume that if by reason of trauma or disease condition to a joint there is a breakdown in the passage of information to the CNS and feedback from the CNS, motor coordination will be affected.

2.4.1 The Impact of Proprioception on Generalised Joint Hypermobility

Proprioception has been defined both as joint position sense, and a perception that helps to identify, organise and interpret sensory information from musculoskeletal, vestibular and visual systems (73). Proprioception helps us to know how the body moves as well as the position of body, or body segment in space. That way, proprioception can help an individual internally represent and understand his environment.

Joint receptors, muscle spindles and skin receptors provide sensory information on joint position and joint movement but evidence has shown while the primary ending of the muscle spindle give sensory information on both joint position and joint movement, the secondary ending of the muscle spindle gives sensory information on only joint position (103).

Musculoskeletal mechanoreceptors that are embedded in the muscles, tendon, ligament, and joint capsule, includes Ruffini ending, Pacinian corpuscles and Golgi bodies and free nerve endings (73,104). Pacinian corpuscles are quick adapting mechanoreceptors; they are thought to mediate sensation of joint motion. The Ruffini endings and Golgi tendons

are slow adapting mechanoreceptors which mediate sensation of joint position and most importantly joint movement as they are able to detect change in muscle tension (103).

Michelson & Hutchins, (1995), investigated the distribution and classification of mechanoreceptors in the ankle ligaments (105). Type-1 receptors which provide position sense were less frequently found in the ankle ligament, the authors however found in abundance, Type-11 receptors which is believed to provide a sense of beginning of joint motion and Type-111 receptors which is thought to provide sensation at the extreme of motion. The abundance of Type-111 receptors in the ankle ligaments is very significant, as this should ensure prevention of injuries. In addition the receptors carry sensory information required for a good postural control that ensures one's ability to maintain stable base after a perturbation in order to prevent falls (72).

Based on significance and use, the musculoskeletal and vestibular systems are also referred to as full time proprioceptive receptors, while the visual system is viewed to be a part time proprioceptive receptor (3). However, in standing position and some other dynamic functional activities, the visual and cutaneous receptors at the feet that have otherwise been referred to as part proprioceptive receptors, may find high relevance. They can also help to clarify body segment positions. Full Weight Bearing (FWB) and Partial Weight Bearing (PWB) facilitate different proprioceptive effects. In FWB condition the body weight puts more load on the joint capsule and causes maximum approximation of joint that will enhance the recruitment of sensory receptors necessary for carrying proprioceptive information to the brain, increased stimulation of joint receptors acts as afferent inputs to the muscle spindles thereby increasing the muscle tone (106).

Kinaesthesia is defined as sensory stimuli that result from movement, these stimuli is what the brain uses to tell positions and movement of the limbs and as a result will unconsciously guide motor performance accurately (18). It may therefore be right to say the two terms 'kinaesthesia' and "proprioception" are synonymous (3). When we also consider our everyday activity separating kinaesthesia and proprioception may be difficult, because as movement that ends in a position it will also commence from a position.

2.4.1.1 Measurement of Proprioception

Proprioceptive acuity is demonstrated both by how proprioceptive information is received and how it is integrated and used (18,101). There are three techniques that can be used to assess proprioception, they are: 1) Joint position reproduction (JPR), 2) Threshold to Detection of Passive Motion (TTDPM), and 3) Active movement extent discrimination (AMEDA) (18). These techniques are based on different concepts, assess the different aspects of proprioception and are carried out using different methods.

- Joint Position Reproduction (JPR)

During a JPR test the participant is required to control the level of a stimulus, (which may either be greater or lesser than the reference stimulus) and adjust it to a reference stimulus. It is also referred to as a form of method of adjustments (18). For example, a limb is first moved to a certain position at a certain velocity, which the individual is required to note and reproduce. This first position is the reference position. Moving the limb at the same velocity either from a position greater or less than the reference position, the individual is the required to stop the movement once he perceives the reference position.

In a systematic review of measurement of knee joint position sense, Smith et al., 2013 reported an intrarater reliability ICC range of 0.18-0.88 (101). Active positioning and replication of the knee joint in standing position intrarater reliability ICC range 0.87-0.88. Angle motion chair to measure passive knee position and replication, intrarater reliability ICC range = 0.86, interrater reliability ICC=0.73 (94).

- Threshold to Detection of Passive Motion (TTDPM)

The TTDPM is one form of method of limits and it can be performed either in ascending or descending order (18). In the ascending method, the limb is moved at such a low level that the participant is unable to detect it; the movement is then gradually increased until the participant can just detect it. The reverse is the case for the descending method. In which case the test starts from when movement can be perceived to when it becomes unnoticeable.

Deshpande et al., (2003) used a motorised footplate to investigate movement perception at the ankle in 3 groups of participants, young adults (20-39 years), middle aged adults

(40-59 years), and older adults (≥60 years) (61). The study included a total of 24 participants with 8 participants in each group, they reported a test-retest reliability of TTDPM range of 0.79-0.95 for the three groups (61).

- Active Movement Extent Discrimination Apparatus (AMEDA)

The AMEDA method is called a method of constant stimuli. In this method, movements are presented in pairs (a constant position and variable positions). The individual is expected to make a comparison between the two movements either by reporting a perception of movement (absolute threshold) or a comparison between the constant stimulus and variable stimuli (18). The constant stimuli method is the only method that requires practice trials because it is a measure of discrimination between the constant movement and the variable movement. The standard stimulus is presented randomly; this prevents the participant from anticipating the next stimulus. This way error of expectation and habituation is reduced (18).

In conclusion, of the three technique used in assessing proprioception, it is only the AMEDA technique that gives a picture of the role proprioception plays in motor activities of everyday life (18).

2.4.1.2 GJH and Proprioception

It is suggested that the poor motor control sometimes seen in children with GJH may be linked to dysfunction in proprioception (36,107). The proprioceptive information available to the central nervous system (CNS) on posture, initiation and extremes of joint movements is highly relevant in the coordination of movement. Due to the laxity of the connective tissues in GJH the joint biomechanics is abnormal, this will cause instability, repeated stress on the joints, and the resultant effect will be deficit in proprioception because of the damaged joints receptors (31). In addition, pain can alter afferent sensory input and cause a deficit in proprioceptive awareness. A deficit in proprioception will affect motor coordination and balance (33,60).

More studies have investigated proprioception at the knee joint than other joints in the lower limb, this might be because the knee joint has been reported to be more at risk of injury than the other joints of the lower limb (102). Fatoye et al., (2009) investigated knee joint proprioception in 66 children (8 and 15 years), 29 children had JHS and 37 were healthy with NM. A purpose built motorised proprioception measuring device was to test

JPS and joint movement. Their findings showed that children with JHS had significantly poorer proprioceptive acuity compared to healthy controls (36). The authors were of the opinion that the poor proprioception at the knee might be because of damage joint receptor, although a causal relationship between damaged joint receptors and poor proprioceptive acuity was not established in the study. The result of this study is the only known data investigating proprioception in children with JHS (36). Given that joint laxity can predispose to damage of joint receptors, it will also be important that sensorimotor function of children with GJH be investigated.

Proprioception has been found to reduce as age increases; however what is not known is if this deterioration starts from childhood. A longitudinal study will help answer this question, if this is known appropriate help and advice can be given to individuals who are at risk damaging their joints and ligaments receptors.

Mishra et al., (2013) compared knee JPS and ROM between young adults and elderly subjects, the authors found JPS and joint movement to be poorer in the elderly subjects (108). According to them, the deficit maybe because of aging process.

There is a suggestion that pain can affect afferent input, which will manifest as a deficit in proprioception. Jeremiah and Alexander (2007) found that individuals with GJH who had significantly greater shoulder mobility ($p=0.004$) but were not different in proprioception from individuals without GJH ($p=0.27$). Both groups were pain free (60). They were also of the opinion that pain will be higher among subjects recruited from clinical settings, who may also be experiencing pain.

2.4.2 Relationship between Balance, Proprioception, and Generalised Joint Hypermobility

Static balance is the ability to maintain one's centre of gravity with minimal movement while dynamic balance is the ability to maintain a stable/upright position while moving (108). Balance is significant in the process of motor learning as it ensures that the child continues to participate in activities that will promote motor learning (69,109). Individuals with balance impairments, individuals with GJH may also show reduction in levels of activities and participation, as they do not have the stable platform for motor activities (70).

Keeping balance requires inputs from the visual, proprioceptive and the vestibular systems to the Central Nervous System (CNS) for the required motor response (108). When there is a perturbation there must be a proper integration of sensory inputs by the CNS for good balance performance (8,72). It therefore implies if there is a deficit in proprioception balance will be affected negatively (65). This will ultimately affect the motor control of the individual.

Balance control may also be affected by joint ROM. Falkerslev et al., (2013), found from their study, that the strategy utlilized by children with GJH to achieve balance when the task is difficult is similar to the strategy utilised by typically developing children between ages three and six years who are still developing their balance skills (40). In a group of children 8-16 years, Schubert- Hjalmarsson et al., (2012), found a significant difference between the balance performance of children with JHS and children without JHS (70). The authors were of the opinion that the joint laxity may be responsible for the poor balance performance in the children with JHS. Suggesting that motor control may be poor in individuals with GJH.

However, Juul-Kristensen et al., (2009), did not find a significant difference between the balance performance of eight year old children with GJH and the children without GJH (51). It is believed that GJH may not present with deficit in 8 year old children as prevalence of musculoskeletal pain, injuries and fatigue is low among children below the age of 12 years (16,31).While Juul-Kristensen et al., (2009), in their study did not find a difference in the level of activity between children with and without GJH (51), it was significantly different between children with JHS and children without JHS in the study by Schubert-Hjalmarsson et al., (2012) (70).

2.3.2.1 Measurement of Balance

Balance can be assessed under different sensory conditions (e.g. with eyes opened or closed, soft surface or hard surface) and during static and dynamic conditions. Force platforms and clinical equipment can also be used to assess balance. For the purpose of this study, we chose to evaluate the Y-Balance test, which is a valid instrument that can be used to measure balance.

- Y-balance test

An example of clinical apparatus used to assess balance is the Y-Balance test (Functional Movement.Com Danville, VA). The Y-Balance test is used to measure balance in an individual maintaining single leg stance while the contralateral limb is used to move the indicator reach as far as possible without falling (25).

The Y-balance test consist of a platform for stance and 3 pipes connected to the stance platform, the subject to be tested stands on the stance platform and uses the other limb to move the reach indicator along the calibrated pipe (110). Each reaching direction offers different challenges can be used to identify individuals chronic ankle instability (25). The goal of the task is to have the individual establish a stable base of support on the stance limb in the middle of the testing grid and maintain it through a maximal reach excursion in one of the prescribed directions.

Each reaching direction offers different challenges and requires combinations of sagittal, frontal, and transverse movements. The goal of the task is to have the individual establish a stable base of support on the stance limb in the middle of the testing grid and maintain it through a maximal reach excursion in one of the prescribed directions. It has been reported that the anterior, posteromedial and posterolateral reaches on the Y-balance test can be used to identify individuals chronic ankle instability (25).

Faigenbaum et al., (2014) found the Y-Balance test to be a feasible and reliable test of balance in children between the age 6.9-12.1 years (109). The overall ICC was reported as between good and moderate, anterior (right=0.82, left=0.82), posteromedial (right=0.77, left=0.75), posterolateral (right=0.88, left= 0.77). The ICC within session was 0.995 and between sessions was 0.907-0.974.

2.4.3 The interaction between Strength, Generalised Joint Hypermobility and proprioception

Development of motor control requires that motor skills be reinforced through daily use in purposeful activities (111). However, the strength an individual is able to generate from the muscle determines to a large extent the activity he will be able to participate in (75,112,113). Also, muscles contain receptors that carry sensory information as it pertains

to proprioception to the brain, and this information is used for the maintenance of postural control and balance (73).

Children with JHS have demonstrated neuromuscular deficits such as muscle weakness which has been linked to joint laxity and deficit in proprioception (33,114). Over time, especially in cases where the joint instability leads to frequent injuries, activities will be reduced thereby leading to muscle weakness

2.4.3.1 Measurement of Strength

Parametric measures of muscle strength have been found to be a more objective means of quantifying muscle strength than manual muscle testing (115). It is also a good means of detecting early muscle weakness in pathological conditions.

- The Hand Held Dynamometer (HHD)

The Hand Held Dynamometer (HHD) can be used to evaluate isometric muscle strength and because of the size, ease of use and low cost compared to other isokinetic dynamometers makes it the preferable instrument of use (115).

The HHD has a reported test-retest correlation coefficient reliability range from 0.84-0.99 indicating good to excellent reliability and the median and modal correlations for all the muscle groups of either 0.97 or 0.98 (116).

Measurement of the lower limb strength is necessary as we will also be investigating proprioception at the ankle joint.

2.5 The impact of Motor Coordination Problems in children with Generalised Joint Hypermobility

GJH is not a developmental disorder, but some children with GJH also suffer coordination impairments (117). It is assumed that if proprioception that is required for coordinated movements is reduced in children with GJH, their opportunities to practice and mature in motor skills may be reduced and this may eventually lead to delayed motor development.

Tirosh and colleagues, (1991) refuted this assumption who examined a group of 59 infants at 18 months, who were divided into groups according to their joint mobility and motor development. Group A consisted of 20 infants with GJH and delayed motor development, group B consisted of 19 infants with GJH and normal motor development and group C consisted of 20 infants with normal mobility (NM) and normal motor development. The children were reassessed 3.5 years later on their gross motor skill and integration of their visual and fine motor skill. The result of their study showed that children who had GJH and delayed motor development at 18 months were significantly delayed in their motor performance at five years. The children who had GJH and normal motor development at 18months did not show any significant delay in their motor performance at 5 years. GJH was also reported to have resolved more frequently (84%) in children who had GJH and no motor delay than in infants (40%) whose joint mobility was associated with motor delay at 18 months. The findings from this study contradict the opinion that GJH is associated with motor delay (55). However, this study has also provided evidence that there may be 2 subgroups of GJH. The first consisting of individuals with GJH and normal motor development and the second group consisting of individuals with GJH and motor dysfunction. Tirosh and colleagues (1991) suggested that the motor dysfunction in individuals with GJH and motor deficit may be as a result of an impairment from the CNS (55).

Kirby and colleagues (2005), demonstrated from their study that children with JHS also have difficulties with activities of daily living and academic performance similar to children with Developmental Coordination Disorder (DCD) (56). DCD is a motor control disorder that manifests as a delay or deficit in acquisition and execution of motor skill, in which an individual's motor performance is below the expected chronological age (10). The motor deficit must be sufficient enough to affect activities of daily living and academic performance that must not be explained by the presence of other medical conditions like cerebral palsy and muscular dystrophy (10).

Jelsma et al., (2013) found the prevalence of GJH was twice as high in children referred for DCD as compared with the prevalence of GJH in both the random and matched Typically Developing (TD) groups (5). They reported a negative correlation that was significant between joint mobility and motor coordination. This was not the case in the study by Engelbert and colleagues, their study did not find an association between GJH

and motor development, though they observed that one third of the children with GJH had severe motor delay (89). This evidence gives a possible association between motor coordination and GJH. With the high prevalence of GJH among children with DCD, the evidenced link has however not demonstrated a conclusive association between them. The most prominent reason is the methodological differences in reported studies (118).

In the study by Jaffe et al.,(1988), which consisted of 715 infants within the age range of 8-14months, demonstrated an association between GJH and motor development (119). Apart from the lack of a conclusive association between DCD and GJH, it is also important to know why only some children with GJH have poor motor coordination.

2.5.1 Assessment of Motor Coordination

The Bruininks–Oseretsky test of Motor Proficiency (BOTMP), the Movement Assessment Battery for Children 2nd Edition (MABC-2), are tools that have been used to measure motor coordination in children (120–122).

- Movement Assessment Battery for Children 2nd Edition (MABC-2)

The MABC-2 is a norm referenced tool that has been more frequently by researcher among children of various cultures to assess motor coordination in school aged children (22,123–127). It can also be used to design or plan programme for management of these children (22).

The MABC-2 has 8 items divided into 3 motor skill categories which are: Manual dexterity, Aiming & catching and Balance. The raw score obtained from each item is converted to a standard score, a total test score was then obtained by the summation of the eight task standard scores, the total test score was used to obtain the percentile score, see appendix 10.

A percentile score of ≤ 5 is classified as definite motor problem, a percentile score between the 5^{th} and 16^{th} is classified to be at risk of DCD, while percentile score of >16 is classified as no motor problem. The MABC-2 was also reported as a reliable measure of mild to moderate motor impairments in young children in a research by van Waelvelde et al., 2007 (128). The test retest of the total score yielded an intra-class ICC of 0.88.

2.6 Conclusion

African children are more hypermobile than their Caucasian peers. For individuals with GJH, lax ligaments give them extra degree movement at the joint that can lead to abnormal joint biomechanics, joint dysfunction (pain) or abnormal motor coordination. Taken together, it is therefore important that prompt attention be given to African children with GJH.

It is assumed that controlling extra degrees of movement at the joint should be more difficult in persons with GJH (5), but it is still not known how some individuals with GJH are able to use their flexibility to gain supremacy in motor performance over their contemporaries who have NM (5). Considering that an efficient motor performance is based on good motor control, the available information on motor control in children with GJH is not sufficient. The extent to which the hypermobility affects proprioception, balance and motor coordination in GJH is also not known.

With the evidence of DCD co-existing with GJH the extent of impact of DCD on motor performance in individuals with GJH is also not known.

3 Methodology

3.1 Research Design

A cross-sectional, descriptive study design was used.

3.2 Study Participants

The study participants included pupils between the ages of 7 and 10 years old from two private and one public schools, all located in Uyo Local Government Area, Akwa Ibom State. The following exclusion criteria were applied:

- Children two years older than the standard age of the class.
- Self-reported acute injury or illness on the day of the assessment by the children.
- Diagnosis of a developmental disability such as cerebral palsy, muscular dystrophy, known to affect motor performance as identified by a parent or teacher.

3.2.1 Sample Size Determination

The sample size for this study was calculated using the hypothesis that there would be no difference in the proportion of children with proprioceptive deficits in the two groups, children with JHS and children with NM. Fatoye et al., (2009) demonstrated that children with JHS had significantly poorer proprioception compared to children with NM (36). The calculation was based on two independent group comparisons (assuming normality) with a minimum clinically relevant difference of 3° and an SD of 2.7° in knee joint position sense (JPS). Alpha was set at 0.05 and power level at 90%. A minimum sample size of 27 children in each group was estimated on this basis.

3.3 Instruments[1]

The following instruments were used in the assessment of the participants:

3.3.1 Measuring Body Mass Index-for-age (BMI)

The World Health Organisation (WHO) developed an age-gender specific BMI for school children and adolescents (129). According to the WHO protocol, anthropometric

[1] Additional information concerning each instrument is presented in the Literature review

measurements of the children were taken in standing position. The date of examination and the child's date of birth was used to obtain the exact chronological age at the time of assessment. The WHO Anthroplus software was used to obtain the age-gender specific BMI. After which they were categorised according to WHO classification;

- Overweight: >+1SD.
- Obesity: >+2SD.
- Thinness: <-2SD.
- Severe thinness: <-3SD

3.3.2 Beighton score with Goniometry

The nine-point Beighton score along with goniometry (16) was used to assess joint hypermobility (Appendix 6). Children are given a numerical score of 0 to 9, one point for the ability to perform each of the following tests 1) Passive dorsiflexion of the little finger beyond 90 degrees, 2) Passive apposition of the thumb to the flexor aspect of the forearm, 3) Hyper extensions of both elbows beyond ten degrees., 4) Hyper extension of the knees beyond ten degrees., 5) Forward flexion of the trunk, with knees straight, so that the palms of the hands rest easily on the floor.

Birrell et al., (1994) assessed a sample population of adults and children (6-66 years) for JH in south west Nigeria using a Beighton score cut-off of ≥4. We decided to use a Beighton score cut-off of ≥5 since our study population consisted only of children who have been reported to be more hypermobile than adults.

A 15cm, 360 degrees goniometer was used in this study to assess the range of motion at the elbow, knee and a 180^0 goniometer was used for the 5^{th} metacarpophalangeal joint.

3.3.3 Measurement of pain with Pain Questionnaire

The Faces Pain Scale (FPS) was used to assess pain in this study because it uses pictures which, are easy for the children to give the appropriate response. Participants were first asked if they have pain in any part of their body and they were expected to respond with a yes or no answer. If their response is yes, they are shown a picture of the human body and asked to circle the part of the body they are experiencing pain. After which FPS is used to indicate the intensity of the pain they are experiencing (11).

The faces pain scale (FPS) is a valid and reliable tool that can be used to measure pain in children (98). The FPS has also been compared with the visual analogue scale and verbal scale, the correlation between scores ranged from r=0.69-0.79, p< 0.001 (98).

3.3.4 The Teachers Traffic Light Questionnaire

The Teachers Traffic Light questionnaire (TTLQ) (appendix 10) gives a global impression of the child's motor performance according to the teacher's observation. This assists teachers in identifying children that may have functional motor problems. The teacher uses colour based on the child functional motor performance to indicate if a child has motor coordination problem. RED = has a coordination motor problem, ORANGE= Maybe has a coordination motor problem and GREEN= Does not have a motor coordination problem.

The TTLQ is based on the procedure for identifying DCD, and it was used in this study to screen for children with DCD. Ferguson et al (2013) compared the TTLQ with MABC-2 which is considered as the gold standard for assessing motor coordination. They reported positive predictive value of 54% and negative predictive value of 76%. According to the authors, TTLQ has five times chance of identifying children with motor difficulties.

3.3.5 The Movement Assessment Battery for Children (MABC-2)

The Movement Assessment Battery for Children (MABC-2) is a norm-referenced assessment measure that was used to assess movement problems in children (22). It has been tested to be valid among children with DCD in various cultures (130,131). The test retest of the total score yielded an intra-class correlation (ICC) of 0.88.

The MABC-2 has eight items divided into three motor skill categories which are: Manual dexterity, Aiming & catching and Balance. The raw score obtained from each item is converted to a standard score, a total test score was then obtained by the summation of the eight task standard scores, the total test score was used to obtain the percentile score, see appendix 10.

A percentile score of ≤5 is classified as definite motor problem, a percentile score between the 6^{th} and 16^{th} is classified to be at risk of DCD, while percentile score of >16 is classified as no motor problem. Dutch norm were used because it has been used by

Ferguson and colleagues (2013) to identify South African children with motor impairment (132).

Children with poor motor coordination were identified using the Teacher Traffic Light Questionnaire and the MABC-2. Children who were identified by teachers and had a score of ≤ 16th percentile on the MABC-2 were classified as DCD. Children that achieved percentile score of > 16 who were not identified as having problems on the TTLQ formed the TD group.

3.3.6 Hand-Held Dynamometer

A hand-held dynamometer (HHD) (Lafayette Manual Muscle Testing System, Lafayette Instrument NY) was used to assess maximum isometric muscle contraction. The break method was used, in this case the tester gradually overcomes the muscle force and stops the moment the extremity gives way (115). Three muscle groups were tested in the lower limbs they were knee extensors, plantarflexors and dorsiflexors, the measurement were taken in Newton (133). For each muscle group, the participants were tested thrice on the HHD and the highest score was used for data analysis. As a result of the association between muscle strength and body mass, the muscle strength was normalized with the participant's body weight (134). The subject and dynamometer position is presented in table 3.1.

Table 3.1 Subject position and Dynamometer placement for measurement of strength using HHD

Muscle group	**Subject position**	**Dynamometer position**
Knee extensors	Sitting, knee at 90^0.	Anterior surface of the distant shunt just proximal to the heel.
Ankle dorsiflexors	Supine, foot at 90^0.	Proximal to the metatarsophalangeal joint.
Ankle plantarflexors	Supine, foot at 90^0.	On the ball of the foot.

The HHD has a reported test-retest correlation coefficient reliability range from 0.84-0.99 indicating good to excellent reliability and the median and modal correlations for all the muscle groups of either 0.97 or 0.98 (116).

3.3.7 Measurement of balance using the Y-Balance test

The Y-balance test is unilateral limb stance balance test, which consist of a platform for stance and three pipes connected to the stance platform, the subject to be tested stands on the stance platform and uses the other limb to move the reach indicator along the calibrated pipe (110). The test is disqualified if the child is unable to return the leg to the stance platform. The distance measured in centimetres is recorded. The test is performed three times and the best of the three is normalised with body height or limb length. The reach distance is normalised because of its reported correlation with the body height or limb length (135,136). See appendix eight for the result sheet

In this present study, participants were instructed to stand on the stance platform on one leg then move the reach indicator right anterior then left anterior, right posterior medial and left posterior medial, finally right posterior lateral and left posterior lateral (109). They were expected to return the leg back to the stance platform. The participants had a practice session and three test trials, the best of the test trials was used for the data analysis.

3.4 Experimental Task: Assessment of proprioception

In this present study, AMEDA technique was used to assess proprioception. Proprioception was assessed using wooden wedges. Five wedges of different heights were placed on the floor (3mm, 6mm, 9mm, and 12mm high). The wedges were presented in pair, with the participant in standing position with their eyes closed. The individual was required to make a comparison between constant stimuli and other stimuli at different levels. Along with the pair of wedges with different heights, a pair of wedge of same height that served as the catch was also presented during the course of the test (e.g. 3mm and 3mm). The wedges were presented in variable order (see appendix 11).

Participants were asked to identify the leg that is higher without looking down. The higher leg was identified by lifting up the hand on the side of the leg. The participant's response was scored as correct =1 and incorrect=0, see appendix 11, for score sheet.

A penalty score was awarded to every incorrect response and it was based on the height difference the participant is unable to detect. Inability to detect the highest wedge difference got the highest penalty score. The wedge difference and their penalty score is

as follows: wedge difference of 9mm: penalty score=4, wedge difference of 6mm: penalty score=3, wedge difference of 3mm: penalty score =2 and wedge difference of 0mm: penalty score =1. The individual penalty scores were summed up to get a total penalty and this was used for the data analysis. Higher penalty score indicated poor proprioception and vice versa.

3.5 Procedure

3.5.1 Ethical Approval, Permission and Informed Consent

Ethical approval was obtained from the University of Cape Town, Human Research Ethics Committee (Ref.No: 096/2015, appendix 1), and the Akwa Ibom State Health Management Board (Ref No: MH/PRS/99/V.VI/260, appendix 2). Permission was obtained from the Uyo Education Authority, school proprietors and class teachers (appendix 3). Informed consent was obtained from the parents of all the study participants (appendix 4) and the children all gave assent (appendix 5).

3.5.2 Preparation and Training of Research Assistants

The research assistants consisted of five physiotherapists with clinical experience ranging from 3 to 10 years and four university undergraduate students. The five physiotherapists handled different instruments. The goniometry measurement was the only measurement that was conducted by the more experienced physiotherapists and the principal researcher. The research assistants collected the data on weight and height. All the research assistants were taken through training by the principal researcher.

3.5.3 Pilot Study

The feasibility of the study was assessed in a public school in Uyo Local Government Area. The purpose of the feasibility study was to enable the researcher to gain insight into the possible challenges that might be encountered during the actual data collection and how they could be solved. There were 20 children who took part in the feasibility study (those were children whose parents gave consent), but were not included in the main study. Under the same condition, the 20 children were assessed for JH using the Beighton score with goniometry, muscle strength test for bilateral knee extensor, knee flexors, plantarflexors and dorsiflexors using the HHD (Lafeyette Manual Muscle Testing System, Model 01163, Lafeyette Instrument Company, USA). Proprioception at the ankle was assessed with wedges, dynamic balance was assessed with the Y-Balance test, and

motor coordination was assessed with the MABC-2. The only difficulty encounter was the inability of the parents to provide accurate information about their children. This informed the reason why the main study was carried out in two private schools where the parents were literate. No other change was made in the procedure. Detail of results of the pilot study is in the appendix 12.

3.5.4 Recruitment of Participants

The researcher held an information meeting with teachers before the data collection commenced and information letters and consent forms were given to all the pupils to take home to their parent. The information letter contained the contact details of the principal researcher and supervisor in the event that a parent needs further clarification on the study.

3.5.5 Data Collection

The assessment was divided into 2 parts for two reasons; the first was so that the children will not be kept out of the classes for too long and second was to prevent the children getting tired and injured. Part 1 consisted of testing the participants for GJH using the Beighton score with goniometry, the result of this test led to the formation of two groups; 1) children with normal mobility (NM) 2) children with GJH.

The participants were then assessed for their muscle strength using the HHD and finally the experimental task was done using wedges and the balance test using the Y-balance test. In all the first part of the assessment took between 20-30 minutes.

Part 2 consisted of testing the children's motor coordination using the MABC-2, the test took 20-30 minutes per child.

3.5.6 Data Management

All consent forms, assessment forms and the records of treatment were kept in individual folders for each child. Folders were numbered to protect anonymity. The data were entered into Excel spreadsheets and analysed using STATISTICA™ and IBM Statistical Package for Social Sciences (SPSS).

3.6 Statistical Analysis

Descriptive statistics (frequencies, means and standard deviations) were used to describe the sample demographic characteristics, motor performance and joint mobility of the children. The Shapiro-Wilks test was used to test for normality of data on, balance, proprioception and strength. The Mann Whitney U test was used to determine the difference between joint mobility, proprioception, strength, and balance as data was not normally distributed. Pearson's Chi square test was used to check for association between demographic characteristics and joint mobility. The two-way factorial ANOVA was used to examine the influence of two independent variables (joint mobility and motor coordination) on proprioception strength and balance. Statistical significance was set at $p<0.05$.

3.7 Ethical Considerations

The study adhered to the research ethics guidelines of the Declaration of Helsinki (World Medical Association, 2013).

3.7.1 Autonomy

Before the data collection began, information leaflets and consent were sent to parents of the children recruited to participate in the study. Parents were given the option to decline participation in the study. A negative consent from the parent was without prejudice from the principal researcher and the school. Only children whose parents gave positive consent participated in the study. No child or parent requested for withdrawal from the study after positive consent was given.

3.7.2 Confidentiality

The information provided by the parents and the teachers through the questionnaire were stored under password control access with the principal researcher. All the children that participated in the study were given identification codes, which only the principal researcher and supervisors will be able to link specific information to individuals (for the purpose of referral).

3.7.3 Referral

Parents received a report on their child's performance on the MABC-2. Information sheets on how the children with DCD could be helped at home and in school were given

to teachers and the parents respectively. There was no request for referral to a physiotherapist from any parent.

4 Results

The results of the study are presented in this chapter. The first set of results were on the sample population. This was followed by the joint mobility of the sample population and their performances on proprioception, strength and balance tests. Lastly, the result on the effect of motor coordination on joint mobility was also presented.

4.1 Sample

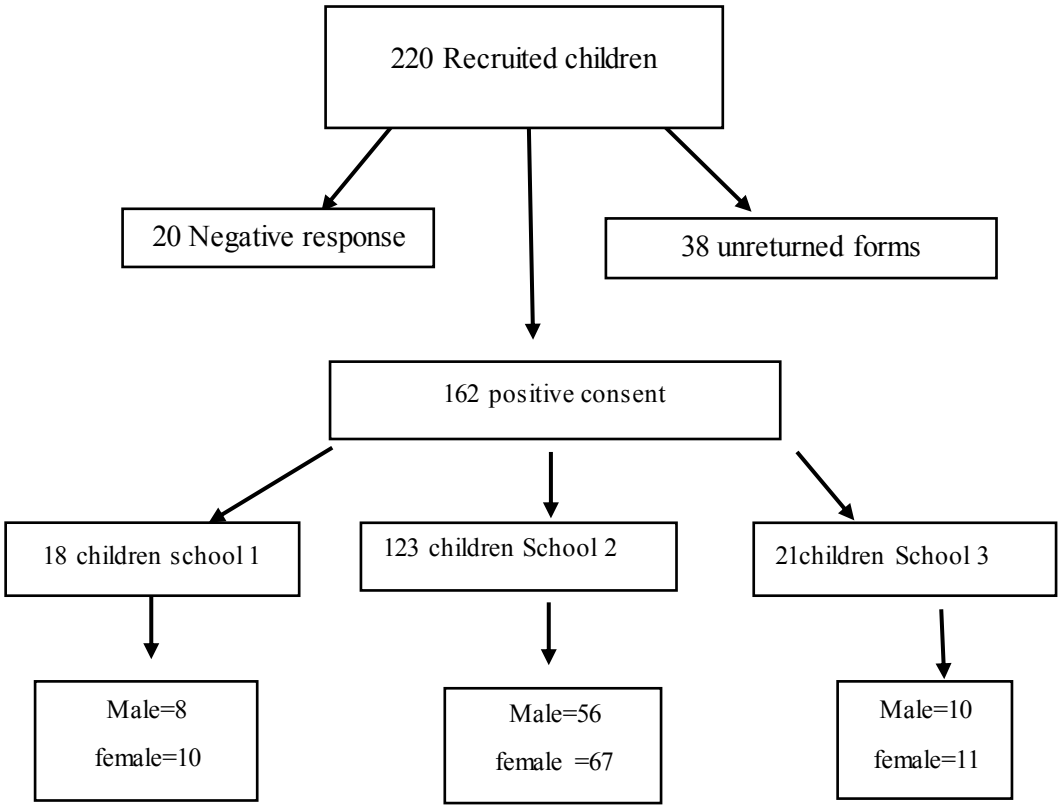

Figure 4.1: Sample distribution among the three schools

According to the WHO age-gender specific BMI classification, 108 children were between the range of +1SD and -2SD, which is categorised as healthy weight. Details of the age-gender specific BMI is presented in table 4.1.

Table 4.1: BMI for the sample population

BMI	Total	Percent
Overweight	30	18.52
Obesity	13	8.02
thinness	2	1.23
Severe thinness	9	5.56
normal	108	66.67
total	162	

160participants were right-handed and two were left-handed.

4.2 Prevalence of Generalised Joint Hypermobility

Using a Beighton score cut- off of ≥5 we identified 84 children (51.8%) who were hypermobile (GHJ group) and 78 (48.2%) who had normal mobility (NM group). With higher cut off scores, the prevalence of GJH dropped, as seen in Table 4.2. The participants on the different Beighton score cut-off were also compared on the various outcomes. The details of the results are presented in appendix 13.

Table 4.2: Prevalence of GJH with different Beighton score cut-off

cut off score	Category	N	%
≥5	NM	78	48.2
	HM	84	51.8
≥6	NM	110	67.9
	HM	52	32.1
≥7	NM	141	87.0
	HM	21	13.0
≥8	NM	153	94.0
	HM	9	6.0

4.3 Differences between Generalise Joint Hypermobility and Normal Mobility groups

We chose to maintain the cut off score of ≥5 joints for further analysis. Groups were compared in terms of demographic characteristics, pain, age-gender specific BMI, performance on the Y-balance test, performance on the lower limb strength tests (using the HHD) and performance on the proprioception test (Wedges).

4.3.1 Demographic characteristics and Body Mass Index of the Generalised Joint Hypermobility and Normal Mobility groups

The overall prevalence of GJH among females (n=52) was higher than males (n=32) (see table 4.3 for details of the result) and the Pearson Chi-square indicated that the association between gender and GJH was significant (chi=4.04, p=0.04).

Table 4.3: Demographic distribution of the GJH and NM groups

Groups	Male	Female	Total
NM	42	36	78
	56.76%	40.91%	
HM	32	52	84
	43.24%	59.09%	
Total	74	88	162

chi=4.04, p=0.04

The GJH group was significantly different from NM group in terms of age (U=2577.5, Z=2.34, p=0.02). As illustrated in figure 4.2, there were more 7 and 8 year olds in the GJH group (64.5%) than in the NM group (38.5%).

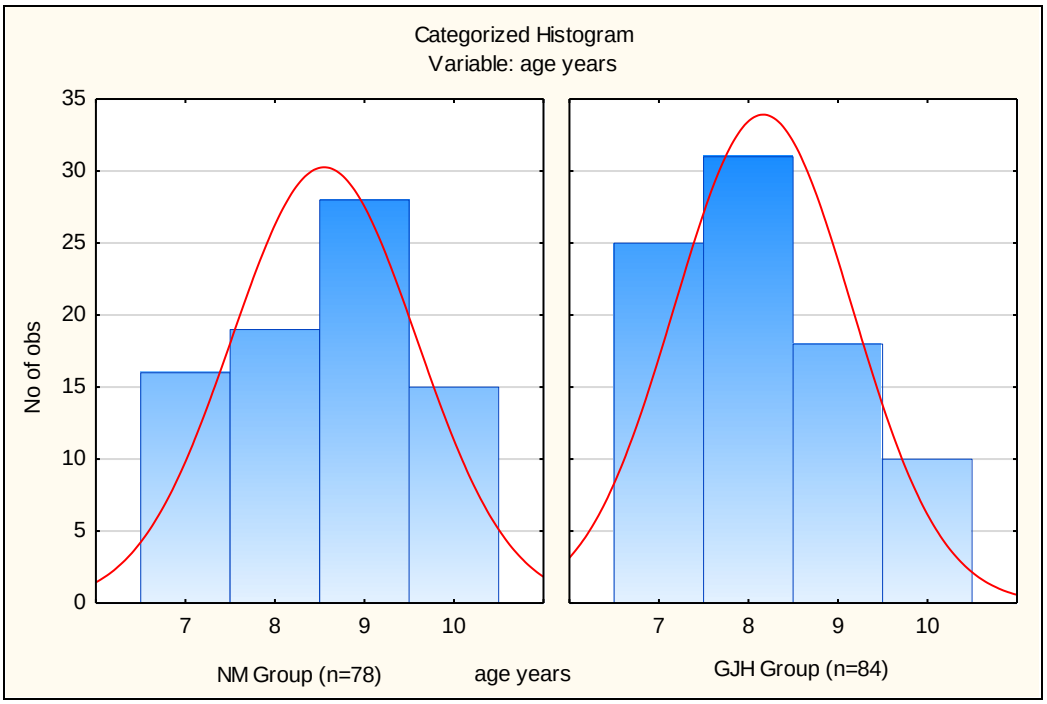

Figure 4.2: Age distribution among the GJH and NM groups

Overweight and obesity were defined using the WHO age-gender BMI classification. 108 children had healthy weight, and nine were severely thin. Details of the result is presented in table 4.4. As presented in table 4.5 the GJH group had more children who were overweight (n=16) and obese (n=10), the NM group had more children with healthy weight. The Pearson chi square did not show any association between joint mobility and BMI (chi-square= 6.73, df=4, p=0.15). The GJH group had higher z score rank sum than the NM group, the Mann-Whitney U test did not find the difference to be significant (u=3037.5, z =-0.96, p=0.34).

Table 4.4: Age-gender specific BMI

Category	Total	Percent
Overweight	30	18.52
Obese	13	8.02
Thin	2	1.24
Severe thinness	9	5.56
Normal	108	66.67

Table 4.5: Comparison of GJH/NM on BMI

category	HM	NM
Overweight	16	14
Obese	10	3
Thin	2	0
Severe thinness	3	6
Healthy weight	53	55

4.3.2 Faces Pain Scale

This study found only two children both from the GJH group that reported joint pain. Both children reported a pain score of 2 (hurts a bit); one at the right elbow and the other at the left wrist joint. The Pearson Chi-square indicated that the association between GJH and pain was not significant (chi=1.88, p=0.17).

4.3.3 Y-Balance Test

The Mann-Whitney U test indicated that the rank sum of distances reached on the Y-Balance were not significantly different between the GJH and NM groups. Details of the result is presented in table 4.6.

Table 4.6: Comparison of the GJH and NM groups on the Y-Balance test

	NM group (n=78)			GJH group (n=84)			Statistics		
Y-balance direction component	**Median**	**Minimum**	**Maximum**	**Median**	**Minimum**	**Maximum**	**U**	**Z**	**p-value**
Right Anterior	0.39	0.07	0.53	0.39	0.26	0.51	3226.0	-0.17	0.87
Left Anterior	0.39	0.08	0.50	0.39	0.23	0.52	3260.5	-0.05	0.96
Right Posteromedial	0.47	0.04	0.65	0.51	0.28	0.66	2964.5	-1.04	0.30
Left Posteromedial	0.50	0.05	0.69	0.53	0.35	0.70	2856.0	-1.41	0.16
Right Posterolateral	0.48	0.04	0.72	0.51	0.36	0.63	2962.0	-1.05	0.29
Left Posterolateral	0.49	0.04	0.67	0.51	0.30	0.67	3130.0	-0.49	0.63

4.3.4 Lower limb strength test

Using the Mann-Whitney U test, the GJH and NM groups were only significantly different in their left knee extensor strength (p=0.01). The median score of the NM group was greater than the GJH score. Details of the result is presented in table 4.7.

Table 4.7: Comparison of the GJH and NM groups on the lower limb strength test

	NM group (n=78)			GJH group (n=84)			Statistics		
Muscle groups	**Median**	**Minimum**	**Maximum**	**Median**	**Minimum**	**Maximum**	**U**	**Z**	**p-value**
Right knee extensors	0.34	0.17	0.60	0.33	0.11	1.76	2737.0	1.81	0.07
Left knee extensors	0.33	0.16	0.58	0.30	0.15	0.62	2482.0	2.66	0.01
Right plantarflexors	0.32	0.14	0.53	0.30	0.13	0.51	2952.5	1.08	0.28
Left plantarflexors	0.31	0.15	0.52	0.29	0.11	0.53	2882.5	1.32	0.19

Right dorsiflexors	0.30	0.16	0.51	0.27	0.14	0.42	2688.0	1.97	0.05
Left dorsiflexors	0.28	0.14	0.45	0.26	0.13	0.40	2789.5	1.63	0.10

4.3.5 The Wedge Test

The wedge set 9mm-9mm, (catch trial), had the highest number of fails (67.9%) overall as participants were not clearly able to determine that these two wedges were the same height. The wedge set with the largest difference, 12mm-3mm and 3mm -12mm had the highest number of correct response (100%). A significant difference was detected between NM and GJH groups on wedge set 9mm-12mm (Chi= 4.09, df=1, p=0.04). Most of participants getting the set incorrect were in the NM group. Details of the result is presented in table 4.8.

Table 4.8 Performance of the GJH and NM groups on the wedge test

Wedge set	Height difference (mm)	All (n=162)		NM (n=78)		GJH (n=84)		stats (NM vs (GJH)
		Correct n (%)	Incorrect n (%)	Correct n (%)	Incorrect n (%)	Correct n (%)	Incorrect n (%)	
3-3	0	74 (45.7)	88 (54.3)	34 (43.6)	44 (56.4)	40 (47.6)	44 (52.38)	Chi=0.26, df=1, p=0.61
3-9	6	154 (95.1)	8 (4.9)	73 (93.6)	5 (6.4)	78 (92.9)	6 (7.1)	Chi= 0.03, df=1, p=0.85
3-12	9	162 (100)	0 (0)	78 (100)	0 (0)	84 (100)	0 (0)	n/a
6-3	3	128 (79.0)	34 (20.9)	57 (73.1)	21 (26.9)	71 (84.5)	13 (15.5)	Chi=3.19, df=1, p=0.07
6-9	3	110 (67.9)	52 (32.1)	50 (64.1)	28 (35.9)	60 (71.4)	24 (28.6)	Chi=0.99, df=1, p=0.32
9-3	6	151 (93.2)	11 (6.8)	70 (89.7)	8 (10.3)	81 (96.4)	3 (3.6)	Chi= 2.85, df=1, p=0.09
9-6	3	92 (56.8)	70 (43.2)	41 (52.6)	37 (47.4)	51 (60.7)	33 (39.3)	Chi= 1.09, df=1, p=0.30
9-9	0	52 (32.1)	110 (67.9)	23 (29.5)	55 (70.5)	29 (34.5)	55 (65.5)	Chi= 0.47, df=1, p=0.49
9-12	3	140 (86.4)	22 (13.6)	63 (80.7)	15 (19.2)	77 (91.6)	7 (8.3)	Chi= 4.09, df=1, p=0.04
12-3	9	162 (100.0)	0 (0)	78 (100)	0 (0)	84 (100)	0 (0)	n/a

12-6	6	157 (96.6)	5 (3.1)	75 (96.2)	3 (3.8)	82 (97.6)	2 (2.4)	Chi= 0.29, df=1, p=0.59

4.3.5.1 Wedge penalty score

A penalty score was awarded to any incorrect response. The higher the wedge difference the higher the penalty that will be awarded. The Mann-Whitney U test showed that the penalty score of the children with NM was significantly different to the children with GJH (U= 2547.5, z= 2.44, p=0.013), indicating that the children with GJH performed better than the NM group on this test. Details of the result is illustrated in figure 4.3.

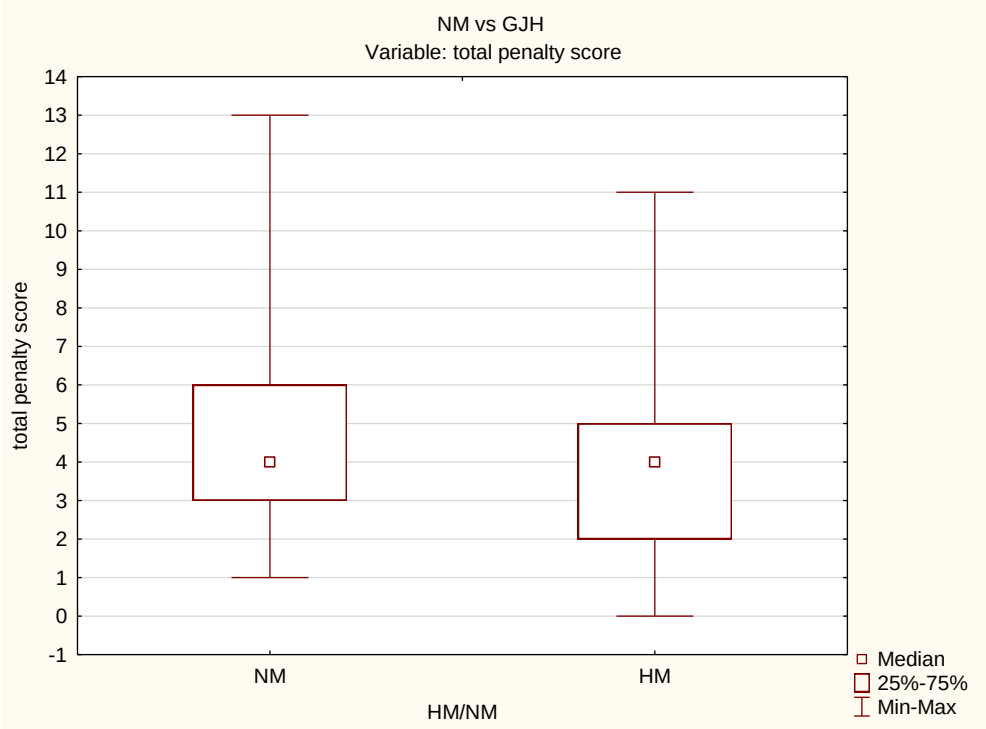

Figure 4.3: Box plot showing penalty score difference between the GJH and NM groups

Differences between groups remained significant when higher cut-off scores were used (≥6, p= 0.04, ≥7: p=0.03,) however differences were not detected at cut off scores of 8 or more p=0.81 possibly to due small sample size of the GJH group.

4.4 Motor performance Developmental Coordination Disorder/Typically Developing

74 participants (45.67%) were classified as having a score ≤16th percentile on the MABC-2 (29 boys, 45 girls). The distribution on MABC-2 scores are presented in table 4.9.

Table 4.9: Motor performance DCD/TD

MABC Percentile Category	Classification	Count	Percent
0.5	RED	2	1.2
1		2	1.2
2		3	1.9
5		14	8.6
SUBTOTAL definite motor problems		21	12.9
6	ORANGE	1	0.6
9		17	10.5
11		1	0.6
16		34	21.0
SUBTOTAL at risk		53	32.7
25	GREEN	18	11.1
37		23	14.2
50		25	15.4
63		7	4.3
75		9	5.6
84		4	2.5
95		1	0.6
98		1	0.6
Subtotal no problems		88	54.3

Using the TTLQ and MABC-2 together, we found 58 children as presented in table 4:10 as who had no problems on either test (TD) and 31 children whom we classified as DCD. Those who did not meet these criteria (n=73) were excluded from the next set of analyses. As expected, groups were significantly different on all aspects of the MABC-2 ($p<0.0001$)

Table 4.10: Performance of the children on TTLQ and MABC-2

TTLQ	MABC-2		
	Green	Orange	Red
Green	58	29	14
Orange	26	22	5
Red	4	2	2

The Pearson Chi-square indicated no significant association between gender and DCD (Chi=0.35, df=1, p=0.55). Although there were older children in the DCD group (median age TD=8 years, IQR: 7-9 vs median age DCD= 9, IQR: 7-10), Mann-Whitney U test indicated that the difference in mean rankings of age was not significantly different between DCD and TD groups (U=1694, z=-0.95, p=0.05).

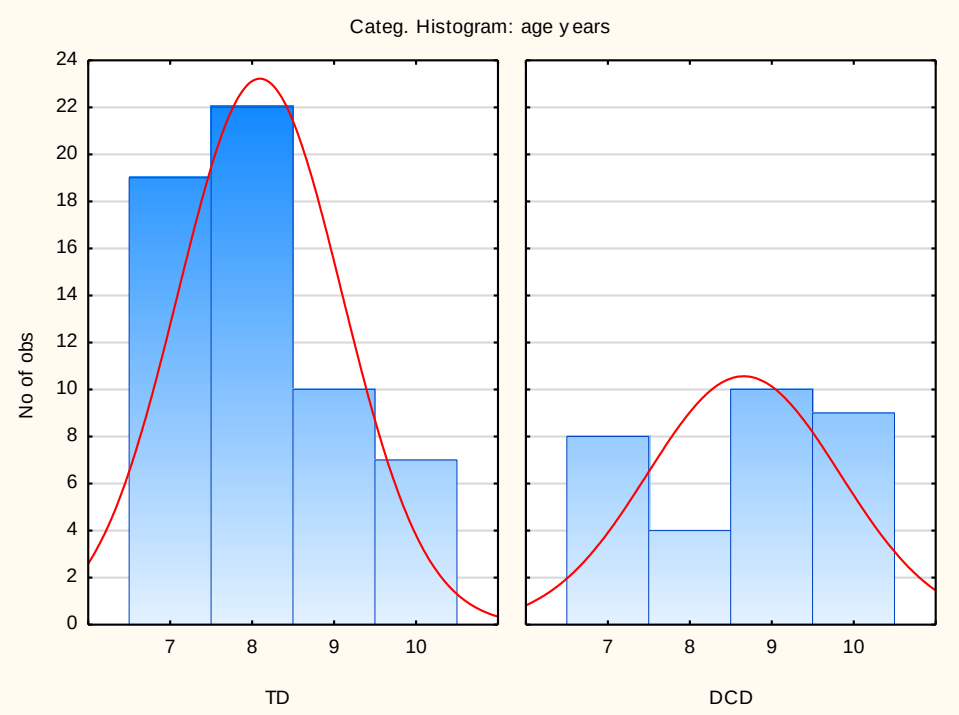

Figure 4.4 Age distribution among the DCD/TD groups

4.5 Effect of Developmental Coordination Disorder/Typically Developing on outcomes

All children with a DCD/TD classification (n=89) were then grouped according to their coordination and joint mobility. This led to the formation of four groups, children with joint hypermobility and DCD [GJHDCD (n=16)], children with GJH and typical motor performance [GJHTD (n=32)] and two normal mobile groups, with DCD [NMDCD (15)], and without [NMTD (n=26)].

Next, we examined the differences between the four groups by testing the effect of DCD/TD and GJH/NM using the two-way ANOVA test.

4.5.1 Y-Balance Test

On examination of the median scores of the Y-balance test (Table 4.11), it is evident that the TD and DCD groups were significantly different [df (6, 80) =3.66, p=0.003)]. However, the interaction effect of TD/DCD and GJH/NM was not significant, suggesting that the four groups were not different in terms of performance on the Y-balance test [df (6, 80) =1.88, p=0.094)] as seen in Table 4.12.

Table 4.11 Median and Interquartile range (IQR) of the GJH/NM and DCD/TD groups on balance

Variable	GJHDCD (n=16)	GJHTD (n=32)	NMDCD (n=15)	NMTD (n=26)
Normalised anterior right(cm) Median (IQR)	0.4 (0.36-0.43)	0.39 (0.37-0.44)	0.36 (0.33-0.39)	0.42 (0.38-0.44)
Normalised anterior left (cm) Median (IQR)	0.41 (0.39-0.42)	0.39 (0.36-0.43)	0.36 (0.32-0.39)	0.42 (0.38-0.44)
Normalised posteromedial right(cm) Median (IQR)	0.5 (0.43-0.51)	0.51 (0.49-0.57)	0.43 (0.38-0.45)	0.51 (0.46-0.56)
Normalised posterolateral right (cm) Median (IQR)	0.49 (0.45-0.53)	0.55 (0.50-0.58)	0.47 (0.45-0.55)	0.53 (0.48-0.59)
Normalised posteromedial left (cm) Median (IQR)	0.48 0.47-0.52)	0.52 (0.49-0.54)	0.46 (0.43-0.48)	0.52 (0.47-0.58)
Normalised posterolateral left (cm) Median (IQR)	0.48 (0.44-0.52)	0.52 (0.49-0.56)	0.47 (0.43-0.51)	0.55 (0.43-0.60)

N=89

Table 4.12 Effect of GJH/NM and DCD/TD on balance

Effect	Wilks Value	F	p
Intercept	0.008	1666.134	<0.001
DCD/TD	0.785	3.656	0.003
GJH/NM	0.940	0.857	0.530
GJH/NM*DCD/TD	0.876	1.883	0.094

F=6, 80, N=89

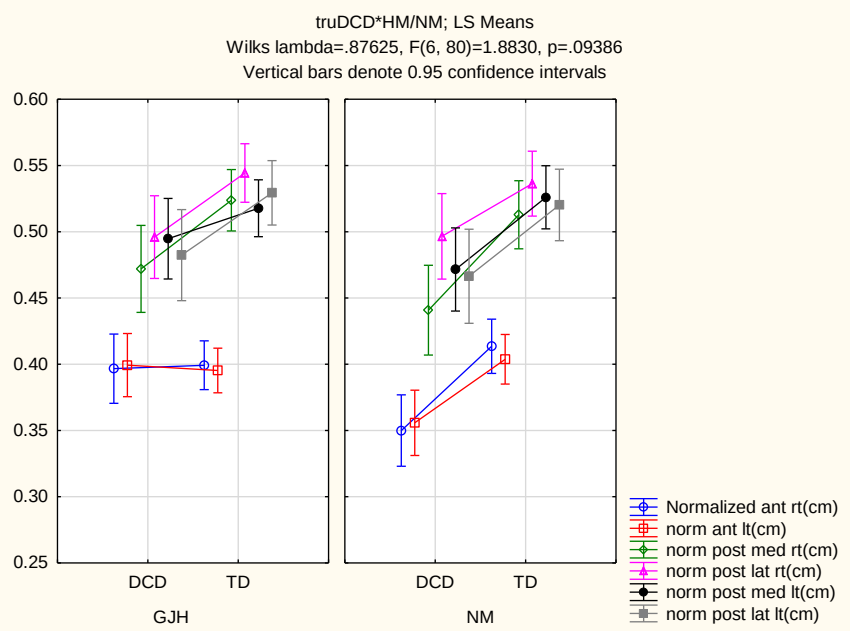

Figure 4.5 Effect of GJH/NM and DCD/TD on balance

4.5.2 Lower limb Strength tests

On examination of the median scores of the various HHD tests (see table 4.13), no differences were found between TD/DCD and NM/GJH groups. Using the two-way factorial ANOVA, joint mobility (NM/GJH) and motor coordination (TD/DCD) was not associated with lower limb strength [F (6, 80) =0.733, p=0.62). Detail of the result is presented in table 4.14.

Table 4.13 Median and Interquartile range (IQR) of GJH/NM and DCD/TD on lower limb strength

Variable	GJHDCD (n=16)	GJHTD (n=32)	NMDCD (n=15)	NMTD (n=26)
Normalised knee extensors right (N/kg)	0.30	0.33	0.33	0.37
Median (IQR)	(0.23-0.37)	(0.25-0.40)	(0.28-0.37)	(0.32-0.44)
Normalised knee extensor left (N/kg)	0.27	0.29	0.35	0.35
median (IQR)	(0.21-0.36)	(0.25-0.36)	(0.27-0.38)	(0.31-0.40)
Normalised plantarflexors right (N/kg)	0.30	0.28	0.29	0.31
median (IQR)	(0.24-0.39)	(0.22-0.37)	(0.22-0.35)	(0.29-0.37)
Normalised plantarflexors left (N/kg)	0.28	0.3	0.29	0.3
median (IQR)	(0.24-0.38)	(0.26-0.35)	(0.25-0.35)	(0.27-0.36)
Normalised dorsiflexors right (N/kg)	0.29	0.27	0.28	0.3
median (IQR)	(0.22-0.32)	(0.23-0.30)	(0.23-0.31)	(0.23-0.35)
Normalised dorsiflexors left (N/kg)	0.25	0.25	0.28	0.3
median (IQR)	(0.21-0.29	(0.23-0.32)	(0.24-0.31)	(0.24-0.35)

Table 4.14 Effect of GJH/NM and DCD/NM on lower limb strength test

Effect	Value	F	p
Intercept	0.039	323.73	<0.001
GJH/NM	0.929	1.009	0.43
DCD/TD	0.913	1.271	0.28
GJH/NM*DCD/TD	0.948	0.733	0.62

F=6,80, p= 0.62

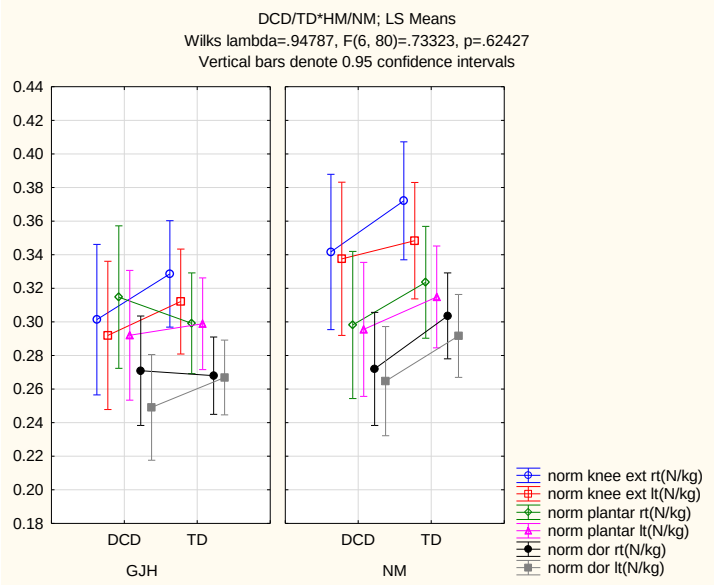

Figure 4.6 Effect of GJH/NM and DCD/TD on lower limb strength

4.5.3 The wedge Test

On examination of the median scores of the various wedge tests, there was a trend for difference between DCD and TD groups on the wedge tests (p=0.05). Using the two-way factorial ANOVA, joint mobility and motor coordination did not have an effect on proprioception at the ankle [F(1,85)= 0.490, p=0.486], as presented in table 4.15.

Table 4.15 Effect of GJH/NM and DCD/TD on proprioception at the ankle

	SS	MS	F	p
Intercept	1334.07	1334.07	237.08	0.00
GJH/NM	0.19	0.19	0.03	0.85
DCD/TD	22.19	22.19	3.94	0.05
GJH/NM*DCD/TD	2.75	2.75	0.49	0.49
Error	478.30	5.62		

F (1,85)= 0.490, p=0.486

Table 4.16 Median, Interquartile range (IQR) of the GJH/NM and DCD/TD on proprioception

Variable	GJHDCD (n=16)	GJHTD (n=32)	NMDCD (n=15)	NMTD (n=26)
total penalty score median (IQR)	2 (1.50-5.50)	4 (2.00-5.00)	4 (4.00-6.00)	4 (2.00-6.00)

5 Discussion

The aims of this study were to determine whether proprioception, strength in the lower limbs and balance was different between children with GJH and children with NM, as well as, to find out if motor coordination had any effect on proprioception, strength and balance.

Our main findings were that children with GJH were significantly better in proprioception than the children with NM. Though the children with GJH had slightly longer reaches on the Y-Balance test than the children with NM in the three directions, the difference however was not significant. Out of the six muscle strength test that was carried out the GJH and NM groups were only different in the left knee extensor strength test. Motor coordination did not have any effect on proprioception, strength, and balance performances in children with NM and GJH.

The nature of the sample are discussed below, the results on the outcome measures that were used. The limitations of the study is presented.

5.1 Sample

The study population consisted of 162 Nigerian children between the ages of 7- 10 years. This study had higher number of female participants (88) than males (74), with a total GJH prevalence of 51.8%. Among the children with GJH the females (59%) were more hypermobile than males (41%). This is similar to the reported prevalence of GJH. Females are more affected than males.

Majority of the children (n=108) had healthy weight and 13 were classified as obese. This result is comparable to the study of Adegoke and colleagues (2009) who studied 720 school children in south west Nigeria and reported the BMI of 77.8% of the children to be less than 18.50 suggesting that overweight was not a problem among Nigerian children. This study did not find an association between BMI and joint mobility. This we found to be comparable to the study result by Clinch et al., (2011), an epidemiological study of GJH among 14 year old children from the UK, they also found no association between BMI and joint laxity (29).

5.2 Prevalence of Generalised Joint Hypermobility

The Beighton score is the generally accepted means of identifying GJH in any study population but the cut-off score used determines what the prevalence rate will be. There has been a lack of consensus about the cut off to be used among different groups of people.

The prevalence of GJH in this study was 51.8%, this is difficult to accept because the widely reported range of GJH is 3%-30%. A very high prevalence of GJH gives the impression that more than half of the population is hypermobile. Birrell and colleagues (1994) used a Beighton score cut off ≥4 in their study among South Western Nigerians (6). They reported a GJH prevalence of 43% among a population of children and adults. For this reason, we considered a Beighton score cut-off of ≥5 to be higher and expected a prevalence lower than the one reported by Birrell et al., (1994). While it may be difficult to control the influence of age, gender and ethnicity on the prevalence of GJH, a stricter Beighton score cut-off may reduce the exaggeration of GJH in the same study population (16,31). For this study if a Beighton score cut-off of ≥6 was used, the prevalence of GJH would have dropped to 32% and it would have dropped further to 13% if a Beighton score cut-off of ≥ 7 was used.

In this study, the prevalence of GJH among the females (32.1%) was significantly higher ($p=0.04$) than the males in accordance with findings from previous studies (16,29,137).

The GJH group was significantly younger than the NM group. The prevalence of GJH among the different age groups in the study population was not significantly different, though there was a decline in prevalence as the age increased. This result is consistent with results from previous study (6,42).

Comparable to the study by Sohrbeck-Nøhr et al., (2014), the result of this study showed that more children with GJH were classified as overweight and obese (138). It is believed that children with GJH have reduced muscle strength that may not allow them to participate in physical activities. As a result they tend to have higher BMI than children with NM (139).

5.3 Generalised Joint Hypermobility and pain

When there is joint laxity, the chances of injuries that can inflict pain is high (31). As a result it was hypothesised that there will be more pain in the GJH group.

The lack of association between pain and GJH may be as a result of the low prevalence of pain in this study. This means the children have asymptomatic joint hypermobility.

Juul-Kristensen, et al., (2012) investigated knee function in 10 year old children and adults with GJH and without GJH. In their study the Visual Analogue Scale (VAS) was used to assess pain, while the children with GJH and without GJH were not significatly different in their report of pain, the adults with GJH and without GJH were significantly different in their report of pain ($p< 0.05$) (79). Similarly, Smits-Engelsman et al.,(2011) did not find a significant difference in frequency of reported pain after exercise and sports in children ages 6-12 years with GJH and without GJH (16). They argued that pain may not be a necessary symptom to assess in children with GJH under the age of 13 years. Tobias et al.,(2013) found an association between GJH and pain, in children ages 13-17 years with GJH, supporting the argument that pain is a significant problem in adolescent with GJH (31). Although the evidence for this is not clear, it may be that the risk of injury in children below age 13 years is low. It is also not known if children will consider pain as abnormal.

5.4 Generalised Joint Hypermobility and obesity

Most of the children in this present study had healthy weight (n=108), therefore it is not likely that their weight will cause pain in any weight bearing joint. It has been reported relationship between overweight and obese in GJH and pain at weight bearing joints (140). This has been attributed to the additional weight on the hypermobile joint causing abnormal joint biomechanics.

5.5 Generalised Joint Hypermobility and Motor Control

5.5.1 Balance

In this study, the balance performance of the GJH and NM groups was tested on the Y-Balance. Although the GJH group had slightly longer reaches in all the three directions than the NM groups, the performances were not significantly different between the two

groups. It was assumed that the laxity at the knees would have given the GJH group longer reaches on the Y-Balance test but it appears from the result of our study that GJH does not seem to make a difference in balance performance.

This result is in line with the study by Juul-Kristensen et al., (2009). The authors investigated balance in eight year old children and found no difference in the balance skills of children with GJH and without GJH (51). They related this to the fact that GJH did not negatively affect the physical activity level of the children with GJH in their study. Involvement in physical activities and ball games give children opportunity to improve their balance skills. Our study participants were physically active children that also participate in ball games. Although the physical activity level of the study population was not assessed, it is likely that the two groups were not different in their level of physical activity. As a result, both groups of children demonstrate same level of balance skills.

It is of interest that in this study, the significant difference in the proprioceptive awareness of the GJH and NM groups did not translate into a difference in their balance performance. It has been argued that the ability to recognise and attend to relevant proprioceptive cues requires training (141). This implies that though the children with GJH in this study had a significantly better proprioceptive ability, they probably lacked the required training that would have enabled them translate it to better balance performance.

The result of the study by Bressel et al, (2007) supports the fact that training is required for proficient balance skills (141). Though their study was among female college athletes, they found that the gymnast and soccer players were not significantly different in static and dynamic balance, but the basketball players demonstrated a poorer dynamic balance compared to the soccer players and gymnasts By virtue of the fact that gymnasts and soccer players have more lower limb activity during their training, they will recognise proprioceptive cues from the lower limbs better (141).

Another consideration in relating the proprioceptive awareness to balance performance might be the demands of the task to be performed. When a task poses a greater challenge on an individual's proprioception, a poor balance performance will be noticed and a less

demanding task will reflect a good balance skill in the same individual (40). The Y-Balance is believed to be a challenging test that can be used to assess balance (66,142). It is likely that both groups found the Y-Balance test equally challenging and therefore did not perform differently.

Individuals classified as obese and overweight have been reported to have poor balance performance because of their sedentary lifestyle (138,143). Though this study had more children with GJH who were obese and overweight, the GJH and NM groups were not significantly different on their BMI. This maybe the likely reason there was no difference in balance performance between the GJH and NM groups.

5.5.2 Proprioception

In this study, the total penalty scores of the NM group were significantly higher than the GJH group. This result suggests that the GJH group children were able to detect movements at the ankle joint better compared to the NM group. This result led to the rejection of our hypothesis and challenges the opinion that proprioception is reduced in children with GJH (36).

It is believed that an individual's ability to detect movement at the joint can be reduced by pain and injuries (64,102,144). It is therefore possible that the low prevalence of pain in this present study may account for the better proprioceptive acuity seen in children with GJH. It is assumed that children with GJH recruited from clinics are likely to report more pain than those recruited from the general population (60). The children in this study were recruited from the general public, so it is not likely that they have suffered trauma that would have led to reduction of their joint receptors. Therefore detecting movement at the ankle will not be difficult. Evidence from studies have shown that the knee and not the ankle joint has higher risk of injury among individuals with GJH (64,102,145). This implies that proprioception at the ankle may be better than the knee in individuals with GJH, and may be the reason why the children with GJH performed better on the wedge test.

It is also possible that the age group of our sample may account for the better performance of the children with GJH on the wedge test. This is because of the

converging evidences that children with GJH below the age of 13 years may not present with pain and sensorimotor deficits (16,29,138,139).

Full Weight Bearing (FWB) and Non-Weight Bearing (NWB) have been found to have different proprioceptive effect (106). In the FWB position, there is maximum contribution of sensory information from the muscle spindles, increased joint approximation and stress on the joint capsules. In relation to this present study, where participants also performed the wedge task in FWB, the GJH and NM groups should have performed alike except if other factors like muscle weakness or fatigue was present (146–148). However, both groups were not different in strength except at the left knee extensors. It is therefore not clear why the GJH performed better than the NM group on the wedge test.

In this study, the wedge test was performed with the participants barefooted, which implies additional sensory input for proprioceptive information for the participants. In as much as studies had tried to limit proprioception to input from muscle and joint receptors only, tactile receptors also contribute to or boost proprioceptive awareness (3). It has been suggested that increased skin extensibility can also contribute to symptoms in GJH (148). It is not known if this serves as a compensatory mechanism by which they get more sensory signals to control their lax joint (3).

5.5.3 Strength and Generalised Joint Hypermobility

It has been suggested that optimal muscle strength is required for good balance skill, this might be because muscle spindles also serve as afferent input for sensory system (141). There has also been reported association between body weight and muscle strength (1,134). This is probably why it is believed that more body weight will generate stronger muscle contraction. Though this study did not find an association between body weight and joint mobility (chi=26.84, df=30, p=0.63), the GJH and NM groups were only different in their left extensor strength test. This result supports evidence that the non-dominate side is usually more hypermobile and weaker (1). Specifically, the knee is more often affected than the ankle (31,102). This may the reason the two groups were not different in their left plantarflexors and dorsiflexors strength.

This result is supported by the finding of Juul-Kristensen et al., (2012).They investigated knee functions in 10 year old children with and without GJH, and found that the

isokinetic strength of the 10 years old children with GJH was not different from the children without GJH (79).

5.6 Association between Motor Coordination and Generalised Joint Hypermobility

The motor performance of the GJH and NM groups was assessed on the MABC-2/TTLQ and this led to the formation of 4 groups, GJHDCD (n=16), GJHTD (n=32), NMDCD (n=15), NMTD (n=26). The findings of this study shows a high number of participants with DCD (random group) and GJH (n=16) supporting the fact that there is increased joint mobility among children with DCD (4,5). However, the extent joint laxity affects the motor performance of children with DCD is not known (5).

The result of this present shows that motor coordination did not have effect on children with GJH.

Poor internal representation of movements and insensitivity to joint movements have been suggested as reasons for poor motor performance in children with DCD (84,149). The studies by Li et al (2015) and Smyth and Mason (1998) both indicated that poor motor performance in children with DCD maybe related to poor proprioceptive awareness (85,149). Wilson and colleagues (2013), had strongly suggested that the poor motor performance in DCD may be a result of poor internal representation (84). Our result however indicates that uncoordinated movement in DCD is not related to abnormal joint biomechanics caused by GJH.

Tirosh et al., (1991) compared the motor performance in a group of children with GJH at the age of 18 months and five years (55). Their findings showed that children with GJH and early motor delay were significantly delayed in both fine and gross motor skills when they were reassessed at five years. The children with GJH and normal motor development at 18 months had no delay in their motor development when they were reassessed at age 5 (55). In their study, Tirosh and colleagues (1991) also found that GJH resolved in some of the children at age 5. But it was found to resolve more among children who had GJH with normal motor development than those with GJH and early motor delay as infants.

It appears not all children with GJH suffer motor difficulties because of joint laxity (51,55,89). It may then seem right to assume that poor motor development is more likely to be as a result of a dysfunction in the CNS than an abnormal biomechanical structure of the joints (118). It can also be assumed that if the motor development is normal, then joint mobility might not necessarily affect motor performance negatively (137).

This was seen in the study of 8 years old Danish school children with GJH by Juul-Kristensen and colleagues (2009), the defined GJH with a Beighton cut-off of ≥6 and they found that children with and without GJH were not different in their motor competence (51). They argued that presence of co morbidities in children with GJH might be a likely reason for poor motor performance in children with GJH. Especially when we have people with GJH perform well in elite sports like ballet, dancing and gymnastics(79).

Studies that reported poor motor performance had children with symptomatic GJH (36,64,150), while the children in this present study were asymptomatic as the prevalence of pain was low. It appears pain is a significant factor when it comes to motor performance in children with GJH (102).

A number of studies have assessed proprioception using techniques that require the limbs to be moved passively (36,71,85,151). It has been recently suggested that proprioceptive test done actively gives the reflection of what happens during functional activities (18). Apparently the results from tests performed actively or passively are different. Pacey et al., (2014) actively assessed proprioception in children with JHS and they found the children to be similar in both the flexion and hyperextension positions (114). Li et al., (2015) compared proprioception in the elbow joints of 30 children with DCD and 30 TD children within the age range of 6-11 years using a passive motion apparatus, their findings showed that the children with DCD were less sensitive to passive motion at their elbow joint (85).

Although the DCDNM group had the lowest reach distance among the four groups, there was no effect of motor coordination on balance in children with GJH. It was expected that the reported balance deficit in children with DCD (8,152–154) will have an impact on balance in children with GJH. However, it appears from the result of this study that GJH may not necessarily be a disadvantage (155). In contrast to the result by Jelsma and

colleagues (2013), they found a significant negative correlation (r_p=0.38, p=0.02) between GJH and DCD (5). Their study included participants from ages 6-16 years, unlike this present study that had children between the ages 7-10 years.

It may seem logical to assume that the age range of this sample population may be the reason children with DCDGJH and TDGJH are not different in their balance performance. Since children from age 12 years and below are still developing their balance skill (79).

Our finding that motor coordination did not have an effect on strength in children with GJH maybe the reason the four groups were not different in their balance performance.

5.7 Study limitations

This is a cross-sectional, descriptive study design, so it is difficult to establish causal relationships between joint mobility and motor coordination.

The protocol of the wedge test is based on the AMEDA technique, which has the advantage of being the only technique that shows the role of proprioception during functional movement. Therefore it is considered to be the most accurate measure of proprioception. Despite the ecological validity of the wedge test it was only the wedge set of 12mm and 9mm out of the 12 wedge sets that demonstrated a significant difference between the GJH and NM groups. It will therefore require that the result of the GJH and NM groups be interpreted with caution.

The children were not blind folded during the wedge test. So, we are not sure if they did not peep when the wedges were placed on the floor for them to step on.

It is also possible that because the test were novel to the children it could have affected the outcome of the study, familiarisation sessions should be given to the children for future study.

The study population was not matched for age and gender. The study population had more females than males who were also more hypermobile than males. The GJH group had more young participants than the NM group.

There is also the possibility of fatigue setting in due to the number of tests that was carried out on each child

6 Conclusion

The aims of this study were to determine whether proprioception, balance and strength in the lower limbs was different between children with GJH and children with normal mobility as well as, to find out if motor performance (DCD) had any effect on the above mentioned variables.

Our study findings showed a difference between the hypermobile children and children with NM in their proprioception ability as measured by the wedge test. Children with GJH were better able to discriminate between wedges of different heights than children with NM. There was no difference in their lower limb strength and balance. No interaction was found between motor coordination and joint mobility on the proprioception, balance and strength test.

6.1 Recommendations

A longitudinal study design may be used to determine the effect of joint mobility on motor coordination over time in children with GJH.

As a result of age and ethnicity, a higher Beighton score cut-off of ≥ 7 may be used to define GJH among Nigerian children in order to avoid an overrepresentation of GJH among the population.

The children should be blind folded during the testing of the wedge test. To ensure that they do not peep while the test is carried out.

Since it was the higher wedges that was able to pick the difference in proprioception between the GJH and NM groups, wedges higher than the ones used in this present study may therefore be considered in future study.

Though this study did not find an effect of DCD on GJH, it may seem appropriate that the Beighton criteria be included in the assessment of children with poor motor coordination as a high number of children with DCD have been found to be hypermobile. So that instructions on joint care can be included in their rehabilitation.

7 References

1. Jindal P, Narayan A, Ganesan S, MacDermid JC. Muscle strength differences in healthy young adults with and without generalized joint hypermobility: a cross-sectional study. BMC Sports Sci Med Rehabil [Internet]. BMC Sports Science, Medicine and Rehabilitation; 2016;8(1):1–9. Available from: http://bmcsportsscimedrehabil.biomedcentral.com/articles/10.1186/s13102-016-0037-x
2. Fatoye F. An Assessment of Neuromuscular Performance, Functional Range of Motion and Quality of Life Characteristrics in Children Diagnosed with Hypermobility Syndrome. 2008.
3. Stillman BC. Making Sense of Proprioception. Physiotherapy [Internet]. 2002 Nov;88(11):667–76. Available from: http://linkinghub.elsevier.com/retrieve/pii/S0031940605601095
4. Kirby a, Davies R. Developmental Coordination Disorder and Joint Hypermobility Syndrome--overlapping disorders? Implications for research and clinical practice. Child Care Health Dev [Internet]. 2007 Sep [cited 2014 Apr 6];33(5):513–9. Available from: http://www.ncbi.nlm.nih.gov/pubmed/17725772
5. Jelsma LD, Geuze RH, Klerks MH, Niemeijer AS, Smits-Engelsman BCM. The relationship between joint mobility and motor performance in children with and without the diagnosis of developmental coordination disorder. BMC Pediatr [Internet]. BMC Pediatrics; 2013 Jan [cited 2014 May 30];13(1):35. Available from: http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=3605363&tool=pmcentrez&rendertype=abstract
6. Birrell FN, Adebajo a O, Hazleman BL, Silman a J. High prevalence of joint laxity in West Africans. [Internet]. Vol. 33, British journal of rheumatology. 1994. p. 56–9. Available from: http://www.ncbi.nlm.nih.gov/pubmed/8162460
7. Geuze RH. Static balance and developmental coordination disorder. Hum Mov Sci [Internet]. 2003;22:527–48. Available from: www.elsevier.com/locate/humov
8. Cherng R-J, Hsu Y-W, Chen Y-J, Chen J-Y. Standing balance of children with developmental coordination disorder under altered sensory conditions. Hum Mov Sci [Internet]. 2007;26:913–26. Available from: www.sciencedirect.com
9. Hertel J, Olmsted-Kramer LC. Deficits in time-to-boundary measures of postural control with chronic ankle instability. Gait Posture. 2007;25(1):33–9.
10. American Psychiatric Association. Dsm V.

http://www.dsm5.org/Documents/Gender%20Dysphoria%20Fact%20Sheet.pdf. 2013. p. 74–7.

11. Wong D, Baker C. Pain in Children: Comparison of Assessment Scales. Pediatr Nurs. 1988;14(1):9–17.
12. Hakim, Alan and Grahame R. Joint hypermobility. Best Pract Res Clin Rheumatol. 2004;17(6):989–1004.
13. Grahame R. Joint hypermobility and genetic collagen disorders: are they related? Arch Dis Child [Internet]. 1999;80(2):188–91. Available from: http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=10325741
14. Scott D a., Bond EQ, Sisto SA, Nadler SF. The intra- and interrater reliability of hip muscle strength assessments using a handheld versus a portable dynamometer anchoring station. Arch Phys Med Rehabil. 2004;85(4):598–603.
15. Wikholm JB, Bohannon RW. Hand-held Dynamometer Measurements : Tester Strength. 1991;(April).
16. Smits-Engelsman B, Kirby A, Klerks M. Beighton score: a valid measure for generalized hypermobility in children. J Pediatr [Internet]. 2011 Jan [cited 2014 Jan 27];158(1):119–123.e4. Available from: http://www.ncbi.nlm.nih.gov/pubmed/20850761
17. Bulbena A, Gago J, Pailhez G, Sperry L, Fullana M a, Vilarroya O. Joint hypermobility syndrome is a risk factor trait for anxiety disorders: a 15-year follow-up cohort study. Gen Hosp Psychiatry [Internet]. Elsevier Inc.; 2011;33(4):363–70. Available from: http://www.ncbi.nlm.nih.gov/pubmed/21762833
18. Han J, Waddington G, Adams R, Anson J, Liu Y. Assessing proprioception: A critical review of methods. J Sport Heal Sci [Internet]. Elsevier Ltd; 2015;1–11. Available from: http://linkinghub.elsevier.com/retrieve/pii/S2095254615000058
19. Kiran D, Carlson M, Medrano D, Smith DR. Correlation of three different knee joint position sense measures. Phys Ther Sport [Internet]. Elsevier Ltd; 2010;11(3):81–5. Available from: http://www.ncbi.nlm.nih.gov/pubmed/20673855
20. Rombaut L, De Paepe A, Malfait F, Cools A, Calders P. Joint position sense and vibratory perception sense in patients with Ehlers-Danlos syndrome type III (hypermobility type). Clin Rheumatol [Internet]. 2010 Mar [cited 2014 Dec 3];29(3):289–95. Available from: http://www.ncbi.nlm.nih.gov/pubmed/19937459
21. Mir SM, Hadian M-R, Talebian S, Nasseri N. Functional assessment of knee joint

position sense following anterior cruciate ligament reconstruction. Br J Sports Med [Internet]. 2008;42(4):300–3. Available from: http://www.ncbi.nlm.nih.gov/pubmed/18390774

22. Henderson SE, Sugden DA, Barnett AL. Movement Assessment Battery for Children,2nd edn. London Pearson. 2007.
23. Dennis DA, Komistek RD, Stiehl JB, Walker SA, Dennis KN. Range of motion after total knee arthroplasty: The effect of implant design and weight-bearing conditions. J Arthroplasty. 1998;13(7):748–52.
24. Gajdosik RL, Bohannon RW. Clinical measurement of range of motion. Review of goniometry emphasizing reliability and validity. Phys Ther [Internet]. 1987 Dec;67(12):1867–72. Available from: http://www.ncbi.nlm.nih.gov/pubmed/3685114
25. Plisky, Phillip , Gorman Paul, Butler Roberts, Kiesel , Kyle, Underwood, Frank EB. The Reliability of an Instrumented Device for Measuring Components of the Star Excursion Balance Test. North Am J Sport Phys Ther. 2009;4(2):92–9.
26. Filipa A, Byrnes R, Paterno M V, Myer GD, Hewett TE. Neuromuscular training improves performance on the star excursion balance test in young female athletes. J Orthop Sports Phys Ther. 2010;40(9):551–8.
27. Kakebeeke TH, Caflisch J, Chaouch A, Rousson V, Largo RH, Jenni OG. Neuromotor development in children. Part 3: motor performance in 3- to 5-year-olds. Dev Med Child Neurol. 2013;55(3):248–56.
28. Smith, Cindy G. Newborn. online J Clin Innov. 2000;3(7):1–77.
29. Clinch J, Deere K, Sayers A, Palmer S, Riddoch C, Tobias JH, et al. Epidemiology of generalized joint laxity (hypermobility) in fourteen-year-old children from the UK: a population-based evaluation. Arthritis Rheum [Internet]. 2011;63(9):2819–27. Available from: /pmc/articles/PMC3164233/?report=abstract
30. Beighton P, Solomon L, Soskolne CL. Articular mobility in an African population. Ann Rheum Dis. 1973;32(5):413–8.
31. Tobias JH, Deere K, Palmer S, Clark EM, Clinch J. Joint hypermobility is a risk factor for musculoskeletal pain during adolescence: Findings of a prospective cohort study. Arthritis Rheum [Internet]. 2013;65(4):1107–15. Available from: http://www.ncbi.nlm.nih.gov/pubmed/23450628
32. Fatoye FA, Palmer S, van der Linden ML, Rowe PJ, Macmillan F. Gait kinematics and passive knee joint range of motion in children with hypermobility syndrome. Gait Posture [Internet]. Elsevier B.V.; 2011;33(3):447–51. Available from:

http://dx.doi.org/10.1016/j.gaitpost.2010.12.022

33. Sahin N, Baskent A, Cakmak A, Salli A, Ugurlu H, Berker E. Evaluation of knee proprioception and effects of proprioception exercise in patients with benign joint hypermobility syndrome. Rheumatol Int [Internet]. 2008 Aug [cited 2014 Nov 23];28(10):995–1000. Available from: http://www.ncbi.nlm.nih.gov/pubmed/18368409

34. Stillman BC, Tully EA, McMeekan JM. Knee Joint Mobility and Position Sense in Healthy Young Adults. Physiotherapy. 2002;88(9):553–60.

35. Galli M, Rigoldi C, Celletti C, Mainardi L, Tenore N, Albertini G, et al. Postural analysis in time and frequency domains in patients with Ehlers-Danlos syndrome. Res Dev Disabil. 2011;32(1):322–5.

36. Fatoye F, Palmer S, Macmillan F, Rowe P, van der Linden M. Proprioception and muscle torque deficits in children with hypermobility syndrome. Rheumatology [Internet]. 2009 Feb [cited 2014 Jan 27];48(2):152–7. Available from: http://www.ncbi.nlm.nih.gov/pubmed/19088133

37. Ferrell WR, Tennant N, Sturrock RD, Ashton L, Creed G, Brydson G, et al. Amelioration of symptoms by enhancement of proprioception in patients with joint hypermobility syndrome. Arthritis Rheum. 2004;50(10):3323–8.

38. Murray KJ. Hypermobility disorders in children and adolescents. Best Pract Res Clin Rheumatol [Internet]. 2006 Apr [cited 2014 Jun 15];20(2):329–51. Available from: http://www.ncbi.nlm.nih.gov/pubmed/16546060

39. Bravo JF, Wolff C. Clinical study of hereditary disorders of connective tissues in a Chilean population: joint hypermobility syndrome and vascular Ehlers-Danlos syndrome. Arthritis Rheum [Internet]. 2006;54(2):515–23. Available from: http://www.ncbi.nlm.nih.gov/pubmed/16447226

40. Falkerslev S, Baagø C, Alkjær T, Remvig L, Halkjær-Kristensen J, Larsen PK, et al. Dynamic balance during gait in children and adults with Generalized Joint Hypermobility. Clin Biomech (Bristol, Avon) [Internet]. Elsevier Ltd; 2013 Mar [cited 2014 Jan 27];28(3):318–24. Available from: http://www.ncbi.nlm.nih.gov/pubmed/23375787

41. Simonsen EB, Tegner H, Alkjær T, Larsen PK, Kristensen JH, Jensen BR, et al. Gait analysis of adults with generalised joint hypermobility. Clin Biomech [Internet]. Elsevier Ltd; 2012;27(6):573–7. Available from: http://dx.doi.org/10.1016/j.clinbiomech.2012.01.008

42. Toit PJ Du, Krüger PE, Terblanche HC, Jansen DC, Rensburg V, Govender C, et al. Sex differences in the nine-point Beighton hypermobility test scores. African J Phys Heal Educ African J Phys Heal Educ Recreat Danc. 2011;17(1741):603–11.
43. Seçkin U, Tur BS, Yilmaz O, Yağci I, Bodur H, Arasil T. The prevalence of joint hypermobility among high school students. Rheumatol Int [Internet]. 2005;25(4):260–3. Available from: http://www.ncbi.nlm.nih.gov/pubmed/14745505
44. Bird HA. Joint hypermobility Main article Main article. Musculoskeletal Care. 2007;5(1):4–19.
45. Bird HA. Joint hypermobility in children. Rheumatology. 2005;44(6):703–4.
46. SA O, OO A. Frequency of benign hypermobility syndrome in females with knee pain. Glob Adv Res J Med Med Sci. 2014;3(April):76–9.
47. Didia B.C., Dapper D.V.B. BSB. Joint hypermobility syndrome among undergraduate. East Africa Med J. 2002;79(2):80–1.
48. Desiree G.A. Rikken-Bultman, Liduine Wellink PWJ van D. Hypermobility in two Dutch school populations. Eur J Obstet Gynecol. 1997;73:189–92.
49. Jansson A, Saartok T, Werner S, Renström P. General joint laxity in 1845 Swedish school children of different ages: age- and gender-specific distributions. Acta Paediatr. 2004;93(9):1202–6.
50. Qvindesland A, Jo H. Paediatric Rheumatology / Series Editor : P . Woo Articular hypermobility in Icelandic 12-year-olds ´ nsson. J Orthop Sport Phys Ther. 1999;1014–6.
51. Juul-Kristensen B, Kristensen JH, Frausing B, Jensen DV, Røgind H, Remvig L. Motor competence and physical activity in 8-year-old school children with generalized joint hypermobility. Pediatrics [Internet]. 2009 Nov [cited 2014 Jan 27];124(5):1380–7. Available from: http://www.ncbi.nlm.nih.gov/pubmed/19822597
52. Simmonds J V, Keer RJ. Hypermobility and the hypermobility syndrome. Man Ther. 2007;12(4):298–309.
53. Collinge R, Simmonds J V. Hypermobility, injury rate and rehabilitation in a professional football squad--a preliminary study. Phys Ther Sport [Internet]. Elsevier Ltd; 2009;10(3):91–6. Available from: http://www.ncbi.nlm.nih.gov/pubmed/19616177
54. Czaprowski D, Kotwicki T, Pawlowska P, Stolinski L. Joint hypermobility syndrome in children with idiopathic scoliosis. 2012;7(Suppl 1):2011–2.
55. Tirosh E, Jaffe M, Marmur R, Taub Y, Rosenberg Z. Prognosis of motor development

and joint hypermobility. Arch Dis Child. 1991;66(ii):931–3.

56. Kirby A, Davies R, Bryant A. Hypermobility syndrome and developmental coordination disorder: Similarities and features. Int J Ther Rehabil. 2005;12(10):431–7.

57. Allison LK, Fuller K. Chapter 22: Balance and vestibular dysfunction. [Internet]. SIXTH EDIT. Vol. 6th, Umphred's neurological rehabilitation. Elsevier Inc.; 2013. 653-709 p. Available from: http://dx.doi.org/10.1016/B978-0-323-07586-2/00031-5

58. Munn J, Sullivan SJ, Schneiders AG. Evidence of sensorimotor deficits in functional ankle instability: a systematic review with meta-analysis. J Sci Med Sport [Internet]. 2010 Jan [cited 2014 Sep 18];13(1):2–12. Available from: http://www.ncbi.nlm.nih.gov/pubmed/19442581

59. Adib N, Davies K, Grahame R, Woo P, Murray KJ. Joint hypermobility syndrome in childhood. A not so benign multisystem disorder? Rheumatology (Oxford) [Internet]. 2005 Jun [cited 2014 Mar 3];44(6):744–50. Available from: http://www.ncbi.nlm.nih.gov/pubmed/15728418

60. Jeremiah HM, Alexander CM. Do Hypermobile Subjects without Pain have Alteration to the Feedback Mechanisms Controlling the Shoulder Girdle ? Musculoskeletal Care. 2007;8(3):157–63.

61. Nandini Deshpande, Denise Connelly, Elsie Culham PC. Reliability and Validity of Ankle Proprioceptive Measures. Arch Phys Med Rehabil [Internet]. 2003;84(6):828–37. Available from: http://linkinghub.elsevier.com/retrieve/pii/S0003999302049468

62. Caulfield B. Functional Instability of the Ankle Joint. Physiotherapy. 2000;86(8):401–11.

63. Duysens J, Beerepoot VP, Veltink PH, Weerdesteyn V, Smits-Engelsman BCM. Proprioceptive perturbations of stability during gait. Neurophysiol Clin. 2008;38:399–410.

64. Smith TO, Jerman E, Easton V, Bacon H, Armon K, Poland F, et al. Do people with benign joint hypermobility syndrome (BJHS) have reduced joint proprioception? A systematic review and meta-analysis. Rheumatol Int. 2013;33:2709–16.

65. Hatzitaki V, Zlsi V, Kollias I, Kioumourtzoglou E. Perceptual-Motor Contributions to Static and Dynamic Balance Control in Children. J Mot Behav. 2002;34(2):161–70.

66. Coughlan GF, Fullam K, Delahunt E, Gissane C, Caulfield BM, Sci M. A Comparison Between Performance on Selected Directions of the Star Excursion Balance Test and the Y Balance Test. J Athl Train. 2012;47(4):366–71.

67. Kaufman KR, Brey RH, Chou L-S, Rabatin A, Brown AW, Basford JR. Comparison of subjective and objective measurements of balance disorders following traumatic brain injury. Med Eng Phys [Internet]. 2006;28(3):234–9. Available from: http://linkinghub.elsevier.com/retrieve/pii/S1350453305001190

68. Shumway-Cook A, Hutchinson S, Kartin D, Price R, Woollacott M. Effect of balance training on recovery of stability in children with cerebral palsy. Dev Med Child Neurol. 2003;45(9):591–602.

69. Richardson PK, Atwater SW, Crowe TK, Deitz JC. Interrater and Test-Retest Reliability of Two Pediatric Balance Tests. Phys Ther [Internet]. 1990;70:79–87. Available from: http://ptjournal.apta.org/content/70/2/79

70. Schubert-Hjalmarsson E, Öhman A, Kyllerman M, Beckung E. Pain, balance, activity, and participation in children with hypermobility syndrome. Pediatr Phys Ther [Internet]. 2012;24(4):339–44. Available from: http://www.ncbi.nlm.nih.gov/pubmed/22965207

71. Konczak J, Krawczewski K, Tuite P, Maschke M. The perception of passive motion in Parkinson's disease. J Neurol [Internet]. 2007;254(5):655–63. Available from: http://www.ncbi.nlm.nih.gov/pubmed/17420926

72. Horak FB, Henry SM, Shumway-Cook a. Postural perturbations: new insights for treatment of balance disorders. Phys Ther. 1997;77(5):517–33.

73. Lee HM, Cheng CK, Liau JJ. Correlation between proprioception, muscle strength, knee laxity, and dynamic standing balance in patients with chronic anterior cruciate ligament deficiency. Knee [Internet]. Elsevier B.V.; 2009;16(5):387–91. Available from: http://dx.doi.org/10.1016/j.knee.2009.01.006

74. Ageberg E, Roberts D, Holmström E, Fridén T. Balance in single-limb stance in patients with anterior cruciate ligament injury: relation to knee laxity, proprioception, muscle strength, and subjective function. Am J Sports Med [Internet]. 2005;33(10):1527–35. Available from: http://journal.ajsm.org/cgi/doi/10.1177/0363546505274934

75. Ada L, Dorsch S, Canning CG. Strengthening interventions increase strength and improve activity after stroke: a systematic review. Aust J Physiother [Internet]. Elsevier; 2006 Jan [cited 2014 Jun 6];52(4):241–8. Available from: http://linkinghub.elsevier.com/retrieve/pii/S0004951406700034

76. Bout-Tabaku S, Klieger SB, Wrotniak BH, Sherry DD, Zemel BS, Stettler N, et al. Adolescent obesity, joint pain, and hypermobility. Pediatr Rheumatol [Internet].

2014;12(11):1–5. Available from: http://ped-rheum.biomedcentral.com/articles/10.1186/1546-0096-12-11

77. Balter SGT, Stokroos RJ, Akkermans E, Kingma H. Habituation to galvanic vestibular stimulation for analysis of postural control abilities in gymnasts. Neurosci Lett. 2004;366:71–5.

78. Tarara DT, Hegedus EJ, Taylor JB. Real-time test-retest and interrater reliability of select physical performance measures in physically active college-aged students. Int J Sports Phys Ther [Internet]. 2014;9(7):874–87. Available from: http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=4275192&tool=pmcentrez&rendertype=abstract

79. Juul-Kristensen B, Hansen H, Simonsen EB, Alkjær T, Kristensen JH, Jensen BR, et al. Knee function in 10-year-old children and adults with Generalised Joint Hypermobility. Knee [Internet]. Elsevier B.V.; 2012 Dec [cited 2014 Jan 27];19(6):773–8. Available from: http://www.ncbi.nlm.nih.gov/pubmed/22417629

80. Pieters S, Roeyers H, Rosseel Y, Van Waelvelde H, Desoete A. Identifying Subtypes Among Children With Developmental Coordination Disorder and Mathematical Learning Disabilities, Using Model-Based Clustering. J Learn Disabil [Internet]. 2013; Available from: http://www.ncbi.nlm.nih.gov/pubmed/23757349

81. Macnab JJ, Miller LT, Polatajko HJ. The search for subtypes of DCD: Is cluster analysis the answer? Hum Mov Sci [Internet]. 2001;20(1–2):49–72. Available from: www.elsevier.com/locate/humov

82. Smits-Engelsman BCM, Niemeijer AS, Van Galen GP. Fine motor deficiencies in children diagnosed as DCD based on poor grapho-motor ability. Hum Mov Sci. 2001;20(1–2):161–82.

83. Wilson PH, McKenzie BE. Information processing deficits associated with developmental coordination disorder: a meta-analysis of research findings. J Child Psychol Psychiatry [Internet]. 1998 Sep;39(6):829–40. Available from: http://www.ncbi.nlm.nih.gov/pubmed/9758192

84. Wilson PH, Ruddock S, Smits-Engelsman B, Polatajko H, Blank R. Understanding performance deficits in developmental coordination disorder: a meta-analysis of recent research. Dev Med Child Neurol [Internet]. 2013 Mar [cited 2014 Jan 30];55(3):217–28. Available from: http://www.ncbi.nlm.nih.gov/pubmed/23106668

85. Li K-Y, Su W, Fu H-W, Pickett KA. Kinesthetic deficit in children with developmental coordination disorder. Res Dev Disabil [Internet]. Elsevier Ltd.;

2015;38C:125–33. Available from: http://linkinghub.elsevier.com/retrieve/pii/S0891422214005186

86. Coleman R, Piek JP, Livesey DJ. A longitudinal study of motor ability and kinaesthetic acuity in young children at risk of developmental coordination disorder. Hum Mov Sci. 2001;20(1–2):95–110.
87. Visser J, Geuze RH. Kinaesthetic acuity in adolescent boys: a longitudinal study. Dev Med Child Neurol. 2000;42:93–6.
88. Keer R, Simmonds J. Joint protection and physical rehabilitation of the adult with hypermobility syndrome. Curr Opin Rheumatol [Internet]. 2011 Mar [cited 2014 Jun 15];23(2):131–6. Available from: http://www.ncbi.nlm.nih.gov/pubmed/21252682
89. Engelbert RHH, Kooijmans FTC, van Riet a MH, Feitsma TM, Uiterwaal CSPM, Helders PJM. The Relationship Between Generalized Joint Hypermobility and Motor Development. Pediatr Phys Ther [Internet]. 2005 [cited 2014 Feb 17];17(4):258–63. Available from: http://content.wkhealth.com/linkback/openurl?sid=WKPTLP:landingpage&an=00001577-200501740-00005
90. Peter Beighton, Rodney Grahame HB. Hypermobility of joints. Vol. 62, springer - Verlag London Limited. 2012. 1-2 p.
91. Bulbena A, Agulló A, Pailhez G, Martín-Santos R, Porta M, Guitart J, et al. Is joint hypermobility related to anxiety in a nonclinical population also? Psychosomatics. 2004;45(October):432–7.
92. Juul-Kristensen B, Røgind H, Jensen D V, Remvig L. Inter-examiner reproducibility of tests and criteria for generalized joint hypermobility and benign joint hypermobility syndrome. Rheumatology (Oxford) [Internet]. 2007 Dec [cited 2014 Jan 22];46(12):1835–41. Available from: http://www.ncbi.nlm.nih.gov/pubmed/18006569
93. Remvig L, Jensen D V, Ward RC. Are diagnostic criteria for general joint hypermobility and benign joint Are Diagnostic Criteria for General Joint Hypermobility and Benign Joint Hypermobility Syndrome Based on Reproducible and Valid Tests ? A Review of the Literature. J Rheumatol. 2007;34(4):798–802.
94. Van Dongen PWJ, De Boer M, Lemmens WAJG, Theron GB. Hypermobility and peripartum pelvic pain syndrome in pregnant South African women. Eur J Obstet Gynecol Reprod Biol. 1999;84(1):77–82.
95. Russek LN. Examination and treatment of a patient with hypermobility syndrome. Phys Ther. 2000;80(4):386–98.

96. Berthier F, Potel G, Leconte P, Touze M-D, Baron D. Comparative study of methods of measuring acute pain intensity in an ED. Am J Emerg Med. 1998;16(2):132–6.
97. Miró J, Huguet A. Evaluation of reliability, validity, and preference for a pediatric pain intensity scale: the Catalan version of the faces pain scale--revised. Pain. 2004;111:59–64.
98. Stuppy DJ. The Faces Pain Scale: Reliability and validity with mature adults. Appl Nurs Res. 1998;11(2):84–9.
99. von Baeyer CL. Children's self-reports of pain intensity: Scale selection, limitations and interpretation. Pain Res Manag. 2006;11(3):157–62.
100. Wearing S, Hennig E, Byrne N. Musculoskeletal disorders associated with obesity: a biomechanical perspective. Obes … [Internet]. 2006;7:239–50. Available from: http://onlinelibrary.wiley.com/doi/10.1111/j.1467-789X.2006.00251.x/full
101. Smith TO, Davies L, Hing CB. A systematic review to determine the reliability of knee joint position sense assessment measures. Knee [Internet]. Elsevier B.V.; 2013;20(3):162–9. Available from: http://dx.doi.org/10.1016/j.knee.2012.06.010
102. Scheper MC, Engelbert RHH, Rameckers EAA, Verbunt J, Remvig L, Juul-Kristensen B. Children with generalised joint hypermobility and musculoskeletal complaints: State of the art on diagnostics, clinical characteristics, and treatment. Biomed Res Int. 2013;2013:1–13.
103. Proske U, Gandevia SC. The kinaesthetic senses. J Physiol. 2009;587(Pt 17):4139–46.
104. Hardin A, Tippett S. The Effects of Muscle Fatigue on and the Relationship of Arm dominance to Shoulder Proprioception. J Orthop Sport Phys Ther. 1996;23(6):348–52.
105. Michelson JD, Hutchins C. Mechanoreceptors in human ankle ligaments. J Bone Joint Surg Br. 1995;77(2):219–24.
106. Bullock-Saxton JE, Wong WJ, Hogan N. The influence of age on weight-bearing joint reposition sense of the knee. Exp Brain Res. 2001;136:400–6.
107. Wycherley AS, Helliwell PS, Bird HA. A novel device for the measurement of proprioception in the hand. Rheumatology. 2005;44:638–41.
108. Mishra P, Sharma A, Bhat L, Narwal R. Analysing age related changes in Proprioceptive and Kinesthetic sensations in community dwelling elderly subjects . Asian J Multidiscip Stud. 2013;1(2):1–8.
109. Faigenbaum AD, Myer GD, Fernandez IP, Carrasco EG, Bates N, Farrell A, et al. Feasibility and reliability of dynamic postural control measures in children in first through fifth grades. Int J Sports Phys Ther [Internet]. 2014;9(2):140–8. Available

from:
http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=4004119&tool=pmcentrez&rendertype=abstract

110. Hartman JG, Looney M. Considerations for Normalizing Measures of the Star Excursion Balance Test. Meas Phys Educ Exerc Sci. 2003;72(2):89–100.
111. Kobesova A, Kolar P. Developmental kinesiology: Three levels of motor control in the assessment and treatment of the motor system. J Bodyw Mov Ther [Internet]. 2014;18:23–33. Available from: http://dx.doi.org/10.1016/j.jbmt.2013.04.002
112. Rombaut L, Malfait F, De Wandele I, Taes Y, Thijs Y, De Paepe A, et al. Muscle mass, muscle strength, functional performance, and physical impairment in women with the hypermobility type of ehlers-danlos syndrome. Arthritis Care Res. 2012;64(10):1584–92.
113. Takken T, Terlingen HC, Helders PJM, Pruijs H, Van Der Ent CK, Engelbert RHH. Cardiopulmonary fitness and muscle strength in patients with osteogenesis imperfecta type I. J Pediatr. 2004;145(6):813–8.
114. Pacey V, Adams RD, Tofts L, Munns CF, Nicholson LL. Proprioceptive acuity into knee hypermobile range in children with joint hypermobility syndrome. Pediatr Rheumatol Online J [Internet]. 2014;12(1):1–7. Available from: http://www.ped-rheum.com/content/12/1/40
115. Beenakker E.A.C., van der Hoeven J.H., Fock J.M. MNM. Reference vakues of maximum isometric muscle force obtained in 270 children aged 4-16 years by hand-held dynamometry. Neuromuscul Disord [Internet]. 2001;11:441–6. Available from: http://www.nlm.nih.gov/medlineplus/neuromusculardisorders.html
116. Bohannon RW. Test-retest reliability of hand-held dynamometry during a single session of strength assessment. Phys Ther. 1986;66(2):206–9.
117. Ghibellini G, Brancati F, Castori M. Neurodevelopmental Attributes of Joint Hypermobility Syndrome / Ehlers – Danlos Syndrome , Hypermobility Type : Update and Perspectives. Am J Med Genet. 2015;10:1–10.
118. Clark CJ, Khattab AD. Association between Joint Hypermobility Syndrome and Developmental Coordination Disorder – A Review. Sport Med Doping Stud [Internet]. 2012;S4(1):1–6. Available from: http://dx.doi.org/10.4172/2161-0673.S4-001
119. Jaffe M, Tirosh E, Cohen A, Taub Y. Joint Mobility and Motor Development. Arch Dis Child. 1988;63:158–61.
120. Nadia Christini Valentini, Coutinho MTC, Pansera SM, San VA p dos. Prevalence of

motor deficits and developmental coordination disorders in children from South Brazil. Rev Paul Pediatr. 2012;30(3):377–84.

121. Smits-Engelsman BCM, Niemeijer AS, van Waelvelde H. Is the Movement Assessment Battery for Children-2nd edition a reliable instrument to measure motor performance in 3 year old children? Res Dev Disabil [Internet]. Elsevier Ltd; 2011;32(4):1370–7. Available from: http://dx.doi.org/10.1016/j.ridd.2011.01.031
122. Wiart L, Darrah J. Review of four tests of gross motor development. Dev Med Child Neurol. 2001;43(4):279–85.
123. Ferguson GD, Naidoo N, Smits-Engelsman BCM. Health Promotion in a Low-income Primary School: Children with and Without DCD Benefit, but Differently. Phys Occup Ther Pediatr [Internet]. 2015;35(2):147–62. Available from: http://www.tandfonline.com/doi/full/10.3109/01942638.2015.1009230
124. WUANG Y-P, SU J-H, SU C-Y. Reliability and responsiveness of the Movement Assessment Battery for Children-Second Edition Test in children with developmental coordination disorder. Dev Med Child Neurol [Internet]. 2012;54(2):160–5. Available from: http://doi.wiley.com/10.1111/j.1469-8749.2011.04177.x
125. Lingam R, Hunt L, Golding J, Jongmans M, Emond A. Prevalence of developmental coordination disorder using the DSM-IV at 7 years of age: a UK population-based study. Pediatrics [Internet]. 2009 Apr [cited 2014 Feb 5];123(4):e693-700. Available from: http://www.ncbi.nlm.nih.gov/pubmed/19336359
126. Smits-Engelsman BCM, Van Roon D, Caeyenberghs K, Swinnen SP. Development of feedforward control in a dynamic manual tracking task. Child Dev [Internet]. 2008;79(4):852–65. Available from: http://www.ncbi.nlm.nih.gov/pubmed/18717894
127. Barnett AL. Motor Assessment in Developmental Coordination Disorder: From Identification to Intervention. Int J Disabil Dev Educ [Internet]. 2008 Jun [cited 2014 Feb 5];55(2):113–29. Available from: http://www.tandfonline.com/doi/abs/10.1080/10349120802033436
128. Van Waelvelde H, Peersman W, Lenoir M, Smits Engelsman BCM. The reliability of the Movement Assessment Battery for Children for preschool children with mild to moderate motor impairment. Clin Rehabil [Internet]. 2007 May [cited 2014 Feb 5];21(5):465–70. Available from: http://www.ncbi.nlm.nih.gov/pubmed/17613568
129. Butte NF, Garza C, Onis M De. Evaluation of the Feasibility of International Growth Standards for School-Aged Children and Adolescents 1. J Nutr. 2007;137:153–7.
130. Chow SMK, Henderson SE. Interrater and test-retest reliability of the Movement

Assessment Battery for Chinese preschool children. Am J Occup Ther [Internet]. 2003;57(5):574–7. Available from: http://www.ncbi.nlm.nih.gov/pubmed/14527121

131. Engel-Yeger B, Rosenblum S, Josman N. Movement Assessment Battery for Children (M-ABC): establishing construct validity for Israeli children. Res Dev Disabil [Internet]. 2010 [cited 2014 Jan 31];31(1):87–96. Available from: http://www.ncbi.nlm.nih.gov/pubmed/19815375
132. Ferguson GD, Jelsma D, Jelsma J, Smits-Engelsman BCM. The efficacy of two task-orientated interventions for children with Developmental Coordination Disorder: Neuromotor Task Training and Nintendo Wii Fit Training. Res Dev Disabil [Internet]. Elsevier Ltd; 2013 Sep [cited 2014 Jan 30];34(9):2449–61. Available from: http://www.ncbi.nlm.nih.gov/pubmed/23747936
133. Andrews AW, Thomas MW, Bohannon RW. Normative values for isometric muscle force measurements obtained with hand-held dynamometers. Phys Ther [Internet]. 1996;76(3):248–59. Available from: http://www.ncbi.nlm.nih.gov/pubmed/8602410
134. Jaric S. Muscle Strength Testing. Sport Med. 2002;32(10):615–31.
135. Gribble P a, Hertel J, Plisky P. Using the Star Excursion Balance Test to assess dynamic postural-control deficits and outcomes in lower extremity injury: a literature and systematic review. J Athl Train [Internet]. 2012 [cited 2014 Dec 18];47(3):339–57. Available from: http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=3392165&tool=pmcentrez&rendertype=abstract
136. Plisky PJ, Rauh MJ, Kaminski TW, Underwood FB. Star Excursion Balance Test as a predictor of lower extremity injury in high school basketball players. J Orthop Sports Phys Ther [Internet]. 2006 Dec [cited 2014 Jul 12];36(12):911–9. Available from: http://www.ncbi.nlm.nih.gov/pubmed/17193868
137. Birgit Juul-Kristensen, Halkjaer Kristensen Jens, Frausing Britt, Dorte Vendelboe Jensen RH, Lars and R. Generalized Joint Hypermobility Motor Competence and Physical Activity in 8-Year-Old School Children With Motor Competence and Physical Activity in 8-Year-Old School Children With Generalized Joint Hypermobility. Pediatrics [Internet]. 2009;124(5):1380–7. Available from: http://pediatrics.aappublications.org/content/124/5/1380.full.html
138. Sohrbeck-Nøhr O, Kristensen JH, Boyle E, Remvig L, Juul-Kristensen B. Generalized joint hypermobility in childhood is a possible risk for the development of joint pain in adolescence: a cohort study. BMC Pediatr [Internet]. 2014;14:302. Available from:

http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=4305244&tool=pmcentrez&rendertype=abstract

139. De Boer RM, Van Vlimmeren LA, Scheper MC, Nijhuis-Van Der Sanden MWG, Engelbert RHH. Is Motor Performance in 5.5-Year-Old Children Associated with the Presence of Generalized Joint Hypermobility? J Pediatr [Internet]. Elsevier Inc; 2015;167(3):694–701.e1. Available from: http://dx.doi.org/10.1016/j.jpeds.2015.06.034

140. Pacey V, Adams RD, Tofts L, Munns CF, Nicholson LL. Joint hypermobility syndrome subclassification in paediatrics: a factor analytic approach. Arch Dis Child [Internet]. 2015;100(1):8–13. Available from: http://www.ncbi.nlm.nih.gov/pubmed/24861050

141. Bressel E, Yonker JC, Kras J, Heath EM. Comparison of Static and Dynamic Balance in Female Collegiate Soccer, Basketball, and Gymnastics Athletes. J Athl Train. 2007;42(1):42–6.

142. Shaffer SW, Teyhen DS, Lorenson CL, Warren RL, Koreerat CM, Straseske CA, et al. Y-balance test: a reliability study involving multiple raters. Mil Med [Internet]. 2013;178(11):1264–70. Available from: http://www.scopus.com/inward/record.url?eid=2-s2.0-84887039346&partnerID=tZOtx3y1

143. D'Hondt E, Deforche B, Vaeyens R, Vandorpe B, Vandendriessche J, Pion J, et al. Gross motor coordination in relation to weight status and age in 5- to 12-year-old boys and girls: a cross-sectional study. Int J Pediatr Obes. 2011;6(2–2):e556–64.

144. Decoster LC, Bernier JN, Lindsay RH, Vailas JC. Generalized Joint Hypermobility and Its Relationship to Injury Patterns Among NCAA Lacrosse Players. J Athl Trainingrnal. 1999;34(2):99–105.

145. Pacey V, Nicholson LL, Adams RD, Munn J, Munns CF. Generalised Joint Hypermobility and risk of lower limb joint injury during sport: A systematic Review with Meta-Analysis. Am J Sports Med. 2010;38(7):1487–97.

146. Maillard S. An update of the managment of hypermobility in children. 2014;12(Suppl 1):2014.

147. Greenwood NL, Duffell LD, Alexander CM, McGregor AH. Electromyographic activity of pelvic and lower limb muscles during postural tasks in people with benign joint hypermobility syndrome and non hypermobile people. A pilot study. Man Ther [Internet]. 2011;16(6):623–8. Available from:

http://dx.doi.org/10.1016/j.math.2011.07.005

148. Hanewinkel-van Kleef YB, Helders PJM, Takken T, Engelbert RH. Motor Performance in Children with Generalized Hypermobility: The Influence of Muscle Strength and Exercise Capacity. Pediatr Phys Ther [Internet]. 2009;21(2):194–200. Available from: http://content.wkhealth.com/linkback/openurl?sid=WKPTLP:landingpage&an=00001577-200902120-00009

149. Smyth MM, Mason UC. Use of proprioception in normal and clumsy children. Dev Med Child Neurol [Internet]. 1998 Oct;40(10):672–81. Available from: http://www.ncbi.nlm.nih.gov/pubmed/9851236

150. Celletti C, Castori M, Galli M, Rigoldi C, Grammatico P, Albertini G, et al. Evaluation of balance and improvement of proprioception by repetitive muscle vibration in a 15-year-old girl with joint hypermobility syndrome. Arthritis Care Res (Hoboken) [Internet]. 2011 May [cited 2014 Nov 25];63(5):775–9. Available from: http://www.ncbi.nlm.nih.gov/pubmed/21240965

151. Coleman R, Piek JP, Livesey DJ. A longitudinal study of motor ability and kinaesthetic acuity in young children at risk of developmental coordination disorder. Hum Mov Sci [Internet]. 2001;20(1–2):95–110. Available from: http://www.ncbi.nlm.nih.gov/pubmed/11471400

152. Fong SSM, Lee VYL, Pang MYC. Sensory organization of balance control in children with developmental coordination disorder. Res Dev Disabil [Internet]. Elsevier Ltd; 2011 [cited 2014 Jan 27];32(6):2376–82. Available from: http://www.ncbi.nlm.nih.gov/pubmed/21835590

153. Geuze RH. Postural control in children with developmental coordination disorder. Neural Plast [Internet]. 2005 Jan;12(2–3):183-96-72. Available from: http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=2565450&tool=pmcentrez&rendertype=abstract

154. Hadders-Algra M. Developmental coordination disorder: is clumsy motor behavior caused by a lesion of the brain at early age? Neural Plast. 2003;10(1):39–50.

155. Fong SSM, Tsang WWN, Ng GYF. Altered postural control strategies and sensory organization in children with developmental coordination disorder. Hum Mov Sci [Internet]. Elsevier B.V.; 2012;31(5):1317–27. Available from: http://dx.doi.org/10.1016/j.humov.2011.11.003

8 Appendices

8.1 Appendix 1: Ethics approval from University of Cape Town, Human Ethics Committee

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

Room E52-24 Old Main Building
Groote Schuur Hospital
Observatory 7925
Telephone [021] 404 7682 • **Facsimile** [021] 406 6411
Email: nosi.tsama@uct.ac.za
Website: www.health.uct.ac.za/research/humanethics/forms

17 February 2015

HREC REF: 096/2015

Dr G Ferguson
Physiotherapy
Health & Rehabilitation Sciences
Old Main Building

Dear Dr Ferguson

PROJECT TITLE: HYPERMOBILITY AND MOTOR CONTROL IN CHILDREN WITH DEVELOPMENTAL COORDINATION DISORDER (MSc candidate- Ms Oluwakemi Ituen)

Thank you for submitting your study to the Faculty of Health Sciences Human Research Ethics Committee for review.

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

Approval is granted for one year until the 29th February 2016.

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)

We acknowledge that the MSc student, Ms Oluwakemi Ituen is also involved in this study.

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

Please quote the HREC REF in all your correspondence.

Yours sincerely

Signed

PROFESSOR MARC BLOCKMAN
CHAIRPERSON, FHS HUMAN RESEARCH ETHICS COMMITTEE
Federal Wide Assurance Number: FWA00001637.
Institutional Review Board (IRB) number: IRB00001938

This serves to confirm that the University of Cape Town Human Research Ethics Committee complies to the Ethics Standards for Clinical Research with a new drug in patients, based on the Medical

8.2 Appendix 2: Ethics approval from Akwa Ibom state Hospital Management Board

GOVERNMENT OF AKWA IBOM STATE OF NIGERIA

Telephone: 085-204091
Telegram: HEALTH

Our Ref:
Your Ref: MH/PRS/99/V.V1/260

OFFICE OF THE PERMANENT SECRETARY
MINISTRY OF HEALTH
IDONGESIT NKANGA SECRETARIAT
BLOCK 8, P.M.B. 1030
UYO

2nd February 2015

Ituen Oluwakemi Adebukola
Department of Health and Rehabilitation Sciences
Faculty of Health Sciences
University of Cape Town
South Africa

RE: APPLICATION FOR ETHICAL APPROVAL

I am directed to convey the approval of the Honourable Commissioner for Health to you to carry out a research study on the topic:

HYPERMOBILITY AND MOTOR CONTROL IN CHILDREN WITH DEVELOPMENTAL COORDINATION DISORDER

This approval is conditional on you seeking and obtaining an informed consent of your participants and ensuring their confidentiality and comfort.

Moreover, while conducting your study, you must at all times restrict yourself to the approved protocol. No change to the research protocol should be made without consideration and approval by the Honourable Commissioner for Health.

Please make a copy of your study available to the Office of the Honourable Commissioner for Health on completion.

Best Wishes!

Signed

Dr. Boniface E. Ukpong, MBBCh, MPH (Leeds, England)
For: Honourable Commissioner

8.3 Appendix 3: Letter of permission from Local Government Education Department- Uyo

LOCAL GOVERNMENT EDUCATION AUTHORITY-UYO
(AKWA IBOM STATE)

Telephone:
Our Ref: LGEA/UY/AD/47/T.140
Your Ref:

OFFICE OF THE EDUCATION SECRETARY
ABAK ROAD
UYO

Date: 8TH – 01 – 2015

Mrs. Ituen Oluwakemi
University of Cape Town

UFS

The Head Teacher

RE: PERMISSION TO CONDUCT A RESEARCH AMONG PRIMARY SCHOOLS PUPILS IN UYO L. G. AREA

With reference to your letter dated 07th January, 2015 on the above subject, I wish to convey approval for you to conduct a research on Hypermobility and motor control in children with developmental coordination Disorder in St. Mary's school, Obio Offot and St. Patrick's Iboko Offot, Uyo.

The authority wishes you successfull; academic research.

Signed

Elder Udeme E. Udokpo
Education Secretary

(All communications to be addressed to the Education Secretary)

8.4 Appendix 4: Parent's information sheet and consent form

UNIVERSITY OF CAPE TOWN
Faculty of Health Sciences
Department of Health and Rehabilitation Sciences
Divisions of Communication Sciences and Disorders, Nursing and Occupational Therapy, Physiotherapy; and Disability Studies
F45 Old Main Building, Groote Schuur Hospital
Observatory, Cape Town, W Cape, 7925
Tel: +27 (0) 21 406 6628/ 6428/ 6534
Fax: +27 (0) 21 406 6323
www.dhrs.uct.ac.za
29/05/14

Dear Parent/Guardian,

I am Ituen Oluwakemi, a post-graduate student at the University of Cape Town. It is in fulfilment of the requirements to obtain my M.Sc. degree. I am investigating the effect of increased movement at the joint in children with developmental coordination disorder. I will have some of my colleagues assist me in this study.

Developmental coordination disorder is a motor disorder that presents with difficulty in the acquiring and performing motor skills. Children with DCD are slower in performing motor activities. They find it difficult dressing up independently, riding bikes, playing ballgames with their friends and writing legibly. Till date little is known about why some children are present with these difficulties.

What am I asking you as parent(s)/guardian(s) to do?
I am asking for your permission to check how well your child plans his/her motor activities like combing his hair, writing, throwing and catching ball. 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 to test your child's motor skills. A few examples: we will ask your child to throw and catch a ball, to stand and hop on one leg, to run, to write and balance on a beam. The tests will take 20-30 minutes. The joints of the elbows, knees and 5th finger will also be measured after which he perform some simple tasks with wedges and perform some balance test, which will last for another 20 minutes. We do not wish to interrupt your child's learning at all. Therefore we will negotiate with the teachers to perform the tests during physical education sessions, break times or after school. If your child chooses to participate he/she will be asked to sign a form to show that he/she has agreed. Your child may also stop the test whenever he/she wishes: it is all up to him/her.

What are the risks involved and what will ensure their safety?

Some of the tests your child will be asked to perform are physical tests, for example running and jumping. This means that your child may feel tired during the tests or after. Your child will be allowed to rest between tests to ensure their safety and they will also be provided with water. We will be present during the assessment of your child with a first aid kit. If your child is seen to show any discomfort, we will stop them immediately. We will watch your child for signs of being upset, such as whining, crying, or struggling. If your child becomes upset we will stop the session for short break of 5-10 minutes. If he or she remains upset, we will stop the sessions for that day.

What are the benefits of participating?

A report will be sent to you about your child's motor performance. If the motor performance is found to be poor you will be advised on how best to support him/her. He/she will receive physiotherapy treatment at the physiotherapy unit in their school. If you will like to seek the services and support of other professional, we will provide you with a letter that you can take to any other physiotherapist.

What happens if I do not want my child to take part in the study?

Your child's participation depends entirely on your consent. Your consent is voluntary. There will be no consequence if you withdraw your child from the study. 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 and you no longer want them to take part, they can withdraw at any stage with no consequence.

What about confidentiality and privacy?

Both the information you provide through the questionnaire and child's results from all the tests will be stored under a password controlled access. All the information I will gather about you and your child will be confidential. No names will be mentioned when I report the result of my research study. A report on your child will be given to you at the end of the study. The video that will be used to record the experimental tasks will be destroyed after the conclusion of the study.

As a researcher, I may not be able to maintain 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 we be paid for taking part in the study?

I am not offering any money to the parents or the children for taking part in this research study. However, all the children who take part in the study will receive 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 Friday 8am-12pm. Telephone +2347031181086. Email itnolu001@myuct.ac.za

If you will like to speak to someone from UCT physiotherapy department, you may contact my supervisor: Mrs Gillian Ferguson, 0214066045/0829743924, Email Gillian.ferguson@uct.ac.za.

If you will like to speak to UCT Human Research Ethics Committee, please contact Prof. Marc Blockman (Chairman of the University of Cape Town Human Ethics Research Committee telephone: 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 Kemi Ituen assess my child ☐

Signature……..

NO, I DO NOT AGREE to let Kemi Ituen assess my child ☐

Signature……..

Thank you

Please return this form to the researcher

8.5 Appendix 5: Child Information Sheet and Assent form

University of Cape Town
Faculty of Health Sciences
Department of Health and Rehabilitation Sciences
Divisions of Communication Science and Disorders, Nursing and Midwifery, Occupational Therapy, Physiotherapy; and Disability Studies
F45 Old Main Building, Groote Schuur Hospital
Observatory, Cape Town, W Cape, 7925
Tel: +27 (0) 21 406 6628/ 6428/ 6534
Fax: +27 (0) 21 406 6323
www.dhrs.uct.ac.za

Dear Learner,

My name is Ituen Kemi 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.

Motor skills are activities you perform with your arms, hands and legs. For example, you use your hands to get dressed, write and catch ball,while you use your legs to kick 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 want to find out why some children find them difficult. We will like to ask if you will help us with this study so we can learn more about why such children have difficulties with their motor skills.

What do I have to do?

We will show you pictures or demonstrate the activiites. We will ask you to

Run, jump,catch, walk on a line, stand on one leg, hop, write and draw, bend your arms and legs, reach forward and touch your toes, and put beads on a string. You will also have wedges put under one foot and you will be asked to identify the leg that is higher.

What are the risks?

These activities you will perform are like the ones you do everyday. They are not harmful and wouldn't hurt you. We will be around when you do the activities so as to make sure that you are safe.

You can stop and rest when you are tired. There will be 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 you don't want to take part in 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 decide, 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 talk to my supervisor

Mrs. Gillian Ferguson, Department of physiotherapy University of Cape Town. Tel. 0214066045/0829743924 Email gillian.ferguson@uct.ac.za or

Prof. M. Blockman at the University of Cape Town, Human Research Ethics Committee Tel. 021406338. Email marc.blockman@uct.ac.za

Assent form

I agree to show the researchers how I do these activities. 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 so that plans can be made to help me if necessary.

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.

8.6 Appendix 6: Protocol for Beighton criteria

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).

8.7 Appendix 7: Beighton score sheet

Name: I.D. No: Sex: Tester:

JOINT	RIGHT	LEFT	SCORE
Passive dorsiflexion MCP5			
Passive hyperextension of elbow			
Passive hyperextension of knee			
Passive opposition thumb to forearm	YES/NO	YES/NO	
Forward flexion of trunk	YES/NO	YES/NO	
Total			

8.8 Appendix 8: Score sheet for the lower limb strength test

Name: I.D. No: Sex: Tester:

Muscles	Trial 1 (N)	Trial 2 (N)	Trial 3 (N)	Best score (N)
Knee extension right				
Knee extension left				
Ankle Plantarflexion right				
Ankle Plantarflexion left				
Ankle Dorsiflexion right				
Ankle dorsiflexion left				

8.9 Appendix 9: Score sheet for Y-Balance test

Name: I.D. No: Sex: Tester:

	R_1 (CM)	R_2 (CM)	R_3 (CM)	BEST SCORE
Right anterior				
Left anterior				
Right posteromedial.				
Left posteromedial				
Right posterolateral				
Left posterolateral				
TOTAL				

8.10 Appendix 10: Score sheet for MABC-2

Form AB 2 (7-10 years)

Name: **Sex: male/female** **ID Number:**

School: **Class:**

Tested by: **Preferred hand (writing hand): Right/Left**

Teacher Questionnaire: Yes/No **Red/Orange/Green**

	Year	Month	Day
Test date			
Date of Birth			
Age			

Item 1 Placing Pegs	**Writing Hand**	**Other Hand**	ISS Best hand	
Trial 1	s	s	ISS Other hand	
Trial 2	s	s	**Mean ISS 1**	

Item 2 Threading Lace	**Number of sec**	**ISS 2**	
Trial 1	s		
Trial 2	s		

Item 3 Drawing Trial	**Number of mistakes**	**ISS 3**	
Trial 1			
Trial 2			

Item 4 Catching with two hands	**Total correct:**	**ISS 4**	

Item 5 Throw beanbag on mat	**Total correct:**	**ISS 5**	

Item 6 One board Balance	**Right Leg**	**Left Leg**	ISS Best item 6	
Trial 1	s	s	ISS Other 6	
Trial 2	s	s	**Mean ISS 6**	

Item 7 Walk heel to toe forwards	**# of steps**	**Whole line**	**ISS 7**	
Trial 1	#	Yes/No		
Trial 2	#	Yes/No		

Item 8 Hopping on mats	**Right Leg**	**Left Leg**	ISS Best Leg	
Trial 1	#	#	ISS Other Leg	
Trial 2	#	#	**Mean ISS 8**	

Total Test Score (ISS 1-8)	**Standard Score**	**Percentile Score**	**Red/Orange/Green**

8.11 Appendix 11: Score sheet for the wedge test

Name: I.D. No: Sex: Tester

Set 1	**Right**	**Left**	**answer**	**Response 1=correct 0=incorrect**
1	9	12	Left	
2	12	3	Right	
3	3	9	Left	
4	9	6	Right	
Set 2	**Right**	**Left**	**answer**	**Response 1=correct 0=incorrect**
1	3	12	Left	
2	12	6	Right	
3	6	3	Right	
4	3	3	Catch	
Set 3	**Right**	**Left**	**answer**	**Response 1=correct 0=incorrect**
1	6	9	Left	
2	9	9	catch	
3	9	3	Right	
4	3	9	Left	

8.12 Appendix 12: Pilot Study

Aim of study: The aim of the pilot study was to determine whether proprioception and balance was different in children with GJH and children with NM.

Specific objectives: To investigate the feasibility of assessing the children on

1. The Beighton criteria.
2. The HHD.
3. The Y-Balance test
4. The wedge test

Participants: 50 children between the ages 7-10 years were randomly selected to participate in the study. Parents who gave consent and accurate information on their wards' age were 22.

Research setting: The pilot study was carried out in an English speaking public school in Uyo local government, Akwa Ibom state.

Methodology: Research design: A cross-sectional, descriptive analytical design was used.

Permission and recruitments:

Ethics approval was obtained from the Faculty of Health Sciences, Human Ethics Research committee (Ref No: 096/2015) and the Akwa Ibom state hospital management board (MH/PRS/99/V.VI/260). Permission was also obtained from the Local Government Education Authority.

Participant's exclusion criteria:

- Children two years older than the standard age of the class.
- Acute injury or illness on the day of the assessment.
- Known diagnosis of a developmental disability known to affect motor performance as identified by a parent or teacher.

Instrumentation: The following outcome measures were used; The Beighton criteria, the HHD, the Y-Balance test**,** the MABC-2**,** and the wedges.

Procedure:

Ethical considerations: the same ethical consideration for informed consent, beneficence, non-malfeasance, confidentiality and privacy which were taken for the main study, were also taken in for the pilot study.

Data analysis:

Descriptive analysis was used to describe the demographics of the participants. T-test was used to check for differences between GJH, proprioception and balance.

Results

Demographic characteristics:

A total of 22 (12 males, ten females) children took part in the study. The pilot study mean age was 8.95 years, S.D. =0.89. The pilot study mean BMI was, 15.09 S.D. =2.37.

Joint mobility:

The pilot study had 11 participants with GJH and 11 participants with NM.

Table 8.1 Distribution of gender and joint mobility

	NM	GJH	Total
Male	8	4	12
Female	3	7	10
Total	11	11	22

Performance of the Generalised Joint Hypermobility and Normal Mobility groups on the Y-Balance test, HHD, and wedges

The GJH and NM groups were not significantly different in strength, (except at the right plantarflexion) balance and proprioception. Details of their performance are in table 8.2.

Table 8.2 Performance of the GJH and NM groups on the Y-Balance test, HHD, and wedges

VARIABLE	Mean NM	Mean GJH	t	df	p
anterior right (cm)	0.39	0.38	0.28	20	0.78
anterior left(cm)	0.40	0.39	0.64	20	0.53
posteromedial right(cm)	0.48	0.47	0.47	20	0.65
posterolateral right(cm)	0.51	0.49	0.87	20	0.40
posteromedial left(cm)	0.51	0.48	1.04	20	0.31
posterolateral left (cm)	0.50	0.48	0.76	20	0.46
knee extension right(n/kg)	0.33	0.28	1.43	20	0.17
knee extension left(N/kg)	0.32	0.28	1.36	20	0.19
Plantarflexion right(N/kg)	0.34	0.27	2.35	20	0.03
Plantarflexion left(N/kg)	0.32	0.27	1.69	20	0.11
Dorsiflexion right(N/kg)	0.30	0.25	1.49	20	0.15
Dorsiflexion left(N/kg)	0.26	0.24	0.70	20	0.49
total penalty score	2.36	3.00	0.56	20	0.57

Discussion:

The pilot study was conducted in a public school. The only challenge that was encountered inability of parents to provide information on their Childs' exact date of birth. It is probably because of their level of education. They were illiterate parents. It also shows that the birth of the children were not registered, if not there would been an official document to show their dates of birth.

Another challenge faced was the reluctance of some teachers to allow the children to be tested. We were allowed to only test the children during lunch break and after school. This slowed down the pace of the study.

We were able to use the Beighton criteria to identify children with GJH. The protocols of the HHD and experimental tasks were easy for the children to understand and perform.

Conclusion:

From the results of the feasibility, it was decided that the study should be conducted in a private school setting where the parents are usually more educated. This will reduce the problem of delayed response from parents and inaccurate information about wards from parents.

8.13 Comparison of Generalised Joint Hypermobility and Normal Mobility groups on strength, and balance

GJH was also defined with Beighton score cut off of ≥6, ≥7, and ≥8 to check if the outcome of the results will be different from when Beighton score cut off of ≥5 was used.

The two groups were not different in balance when compared on the different Beighton score cut off. Details of result is presented in table 8.3.

Table 8.3: Comparison of GJH and NM on balance using different cut off scores

Y-balance direction component	p-value if Cut off score ≥6	p-value if Cut off score ≥7	p-value if Cut off score ≥8
Right Anterior	0.39	0.83	0.99
Left Anterior	0.43	0.68	0.29
Right Posteromedial	0.59	0.84	0.55
Left Posteromedial	0.37	0.44	0.52
Right Posterolateral	0.63	0.87	0.76
Left Posterolateral Right Anterior	0.65	0.77	0.49

A difference was found between the GJH and NM groups only at the left knee extensors using a Beighton score cut off of ≥6. Details of the result is presented in table 8.4

Table 8.4: Comparison of the GJH and NM on lower limb strength test

Muscle groups	p-value if Cut off score ≥6	p-value if Cut off score ≥7	p-value if Cut off score ≥8
Right knee extensors	0.052	0.632	0.892
Left knee extensors	0.003	0.267	0.365
Right plantarflexors	0.586	0.317	0.402
Left plantarflexors	0.201	0.551	0.756
Right dorsiflexors	0.432	0.373	0.846
Left dorsiflexors	0.394	0.412	0.821