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Recurrence of deformity in Blount's disease after surgical correction

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Recurrence of deformity in Blount's disease after surgical correction



Eduard van Greunen

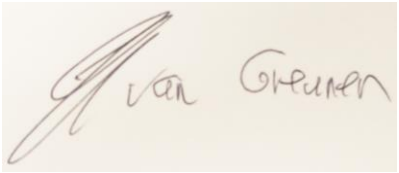
Student number: 1032828

A research report submitted to the Faculty of Health Sciences, University of the Witwatersrand, in partial fulfilment of the requirements for the degree of Master of Medicine

Johannesburg, 2019

DECLARATION

I, Eduard van Greunen, declare that this Research Report is my own, unaided work. It is being submitted for the Degree of Master of Medicine in the branch of Orthopaedic Surgery at the University of the Witwatersrand, Johannesburg. It has not been submitted before for any degree or examination at any other University.



25th day of February 2019 in Johannesburg

DEDICATION

To my loving wife and family.

PRESENTATIONS ARISING FROM RESEARCH PROJECT

Preliminary results presented at South African Orthopaedic Association (SAOA) Congress 4 September 2017 Port Elizabeth as a podium presentation.

ABSTRACT

Introduction: Blount's disease may recur after surgical correction of the deformity. Vastly different recurrence rates have been reported in the literature. The aim of this study was to investigate the recurrence rate of infantile and juvenile Blount's disease following high tibial and fibular valgus osteotomy at the Chris Hani Baragwanath Academic hospital.

Method: This was a retrospective cohort comparative study. We analysed the files, theatre records and X-rays of 44 patients who presented with infantile or juvenile Blount's disease at Chris Hani Baragwanath Academic Hospital from 1 January 2009 until 31 August 2018. Demographics, family history, body mass index, walking age and age at surgery were determined. Pre-, post- and follow up anatomic femorotibial angles, were measured from X-rays. The limbs were divided into Langenskiöld stages based on pre-operative X-rays.

Results: The study found an overall recurrence rate of 63.3% in limbs with Blount's disease. In the infantile group 59.2% of limbs had recurrence while in the juvenile group there was a limb recurrence rate of 72.2%. Surgery after the age of four years was identified as a risk factor for recurrence in infantile Blount's disease. Other factors identified as significant for recurrence were advanced Langenskiöld stages and overcorrection of less than 15 degrees of valgus.

Obesity, early walking, gender and the pre-operative anatomical femorotibial angle were not found to be risk factors for recurrence in this study.

Conclusion: The recurrence rate for high tibial valgus osteotomies at Chris Hani Baragwanath Academic Hospital for infantile Blount's disease was 59.2% and 72.2% in the study period for infantile and juvenile types, respectively. Surgery after the age of four years, late presentation, advanced Langenskiöld stages and overcorrection of less than 15 degrees of valgus were noted to increase the recurrence rate. Using results from the literature and the findings in this study it is recommended to perform a high tibial valgus osteotomy with overcorrection before the age of four years in infantile Blount's disease. Early referral of tibial bowing should be encouraged to prevent higher Langenskiöld stages at time of presentation and allow for earlier surgery to reduce the recurrence of deformity.

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NOMENCLATURE

Alkaline phosphatase	(ALP)
Anatomic femorotibial angle	(aFTA)
Body mass index	(BMI)
Centers for Disease Control and prevention	(CDC)
Chris Hani Baragwanath Academic hospital	(CHBAH)
Erythrocyte sedimentation rate	(ESR)
Femorotibial angle	(FTA)
High tibial and fibular valgus osteotomy	(HTO)
Medial proximal tibial angle	(MPTA)
Metaphyseal-Diaphyseal angle	(MD angle)
Probability	(*p)*
Picture archiving and communication system	(PACS)
Standard deviation	(SD)

CHAPTER 1: INTRODUCTION AND LITERATURE REVIEW

1.1 Background

Recurrence after surgery in Blount's disease is a well-known entity. The literature is sparse on the subject and studies have shown widely conflicting results regarding recurrence rates (1-3).

Blount's disease may be classified as infantile, juvenile and adolescent according to the age of onset (4). The groups differ on age at presentation, clinical features, aetiology, X-Ray appearance and treatment (Table 1.1) (Figure 1.1) (1, 4, 5). The juvenile group can be considered as intermediate in presentation between the infantile and adolescent groups (4, 6).

Table 1.1 Infantile versus Juvenile Blount's disease (1, 6-8)

	Infantile	Juvenile
Age of onset	Less than four years	Between four and ten years
Sides affected	Likely to be bilateral	Usually bilateral
Obesity	Typically normal weight	Usually obese
Pain	Absent	May be presenting complaint rather than deformity
Prominent proximal medial metaphyseal "beaking"	Present	Less pronounced or absent
Metaphyseal depression on X-ray	Present	Absent or minimal
Physis X-ray appearance	Regular	Irregular

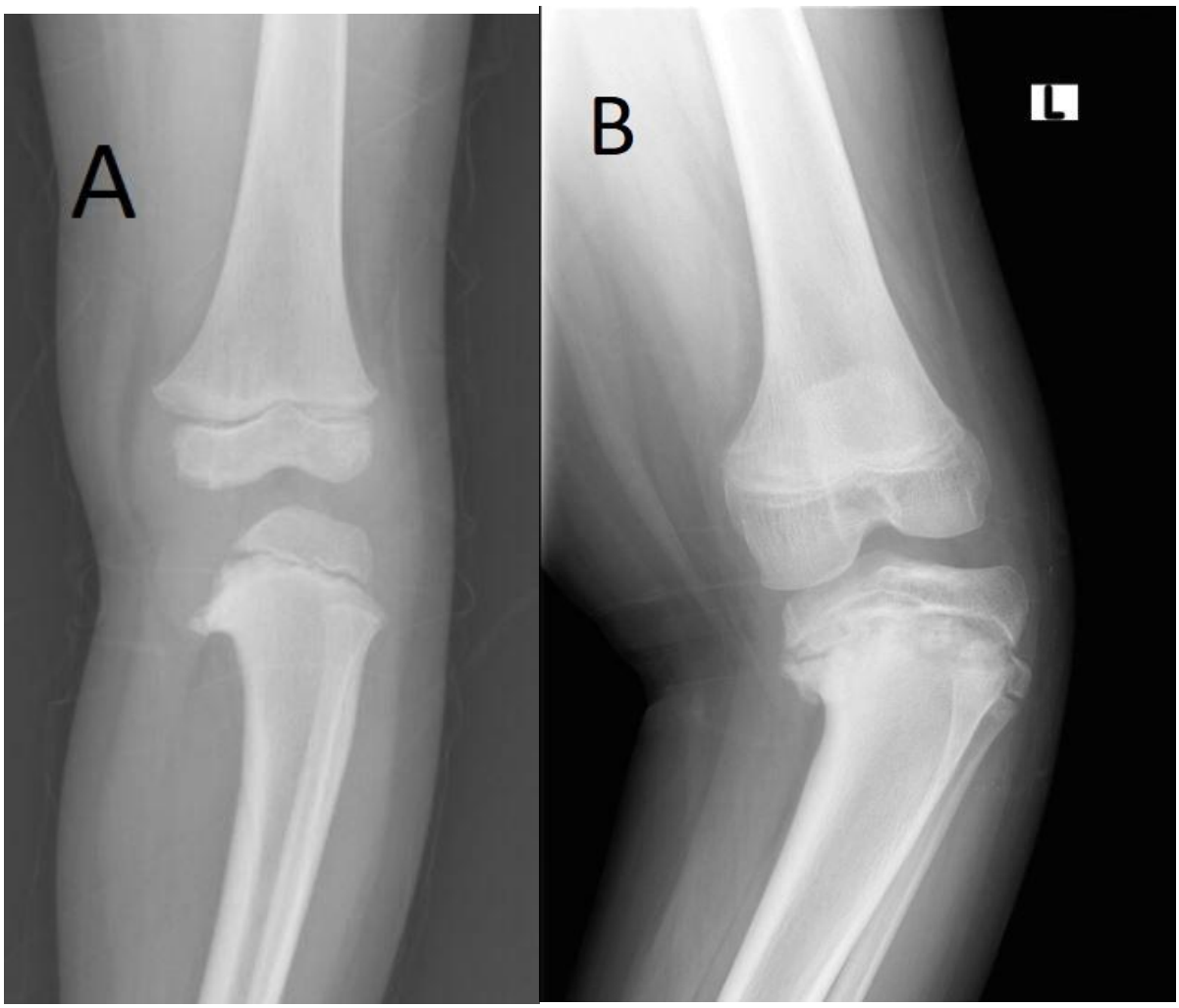


Figure 1.1. Limbs from the study illustrating: Infantile (A) versus Juvenile (B) Blount's disease X-ray appearance

Acute or gradual correction of Blount's disease has been described (1, 4, 5). In this study acute correction with high tibial and fibular valgus osteotomy (HTO) was studied since it provides pre-operative and post-operative fixed measurements and is the protocol followed in the study centre.

Infantile and juvenile Blount's diseases were studied together since both are amiable to acute correction with HTO with a long period of remaining growth and thus potential to recur.

The goal of the study is to determine the recurrence rate of infantile and juvenile Blount's disease at Chris Hani Baragwanath Academic Hospital (CHBAH) after HTO.

The records for patients that presented with Blount's disease from 01 January 2009 until 31 August 2018 were used to gather data.

1.2 Literature review

1.2.1 Historical perspective

Phillip Erlacher was the first to describe a progressive deformity in the proximal tibia of children in 1922 (4). In 1937 Walter Blount (9) published a series of 15 cases from the literature of tibia vara and added 13 cases of his own. Blount used the term "osteochondrosis deformans tibiae" (4, 9). Blount noted that the deformity was similar to Madelung's disease of the distal radius or coxa plana but situated at the proximal medial tibial epiphysis (9). Blount stated that whilst rickets may be confused with tibia vara on clinical grounds, the radiographic appearance of the conditions are distinct. The X-Ray appearance of tibia vara is similar to that of coxa plana (9). He described an infantile type which occurs in the first year or two of life which presents with a sloping epiphysis and a beaklike metaphysis (9). He also presented an adolescent type which occurs before puberty which he attributed to possible local trauma or infection (9). Blount advocated for mechanical relief of strain on the deformity and advocated for the use of an osteotomy to correct severe varus. Notably he found that patients may have recurrence after surgical correction of the deformity in his original article in 1937 (9).

Blount's disease is used as a term for the condition for several reasons: tibia vara is nonspecific, it does not describe any possible aetiology and the deformity may have a femoral component as well (5, 10). Another term used is osteochondrosis deformans tibiae which implies avascular necrosis as a cause for the condition, however no necrosis has been observed in biopsy specimens of children with Blount's disease (1, 10, 11). Finally, Blount was the first to give a detailed description to the condition which now bears his name (4, 9, 10).

Cook et al. (12) used finite element analysis to determine the effect of increasing varus on the medial proximal physis in tibiae in children. These authors found that in an obese five year old child with ten degrees of genu varum during single-limb stance; the forces generated on the proximal medial tibial plateau exceeded those needed to retard growth (12). This study supports the fact that children with Blount's disease will progress if not corrected as the forces across the medial proximal tibial growth plate will inhibit growth thus exacerbating the deformity (12).

Clinical recurrence is present when the proximal tibia has a varus deformity which is clinically unacceptable after surgical correction has been performed. Clinical recurrence is highly subjective and radiological recurrence may be a preferred metric. Radiological recurrence has been suggested when the femorotibial angle (FTA), an angle formed between lines drawn on the femoral and tibial anatomical axes, is in more than ten degrees of varus on follow up (13, 14). Using the findings of Cook et al., ten degrees will be used in this study as a value for recurrence since this amount of varus will predispose to worsening deformity (12).

Recurrent proximal tibial bowing after surgical correction may lead to cosmetic concerns which may be unacceptable to the patient or caregivers (3, 15, 16). Jones et al. (17) found that an FTA of between zero and five degrees of valgus showed the highest levels of satisfaction and the lowest levels of pain with patient self-reported outcome measures. Shortening of the affected limb can be present in cases of unilateral Blount's disease with unilateral recurrence (4, 17, 18) .

Blount's disease has been associated with clinical sequlae later in life such as early osteoarthritis of the knee and meniscal lesions (4, 18-21). Zayer (19) found that increasing varus angulation led to higher rates of osteoarthritis in adults who have Blount's disease. Ingvarsson et al. (18) found high rates of medial meniscal lesions in adults who had operative or non-operative management as children for Blount's disease. It was postulated that it may be due to hypertrophic and highly mobile medial menisci in Blount's disease as shown by Siffert and Katz (22).

1.2.2 Classification

Various classifications have been proposed to divide Blount's disease into groups.

Blount initially divided Blount's disease into an infantile and adolescent type with the infantile type occurring at one or two years and an adolescent type occurring before puberty (9).

Blount's disease can also be divided into early and late onset groups with the early group occurring before four years of age and the late group occurring after four years of age (4, 5, 23, 24).

Thompson et al. (6) classified Blount's disease as an infantile type occurring before four years of age, a juvenile type occurring between four and ten years and an adolescent type occurring after the age of ten years (4-6).

The infantile and adolescent types differ regarding age at presentation, clinical features, aetiology, X-Ray appearance and treatment, whereas the infantile and juvenile groups are more similar in their comparative findings (1, 4, 5). The juvenile group can be considered as intermediate in presentation between the infantile and adolescent groups (4, 6).

There is extreme variability in the reported recurrence rate in the literature. Recurrence rates of more than 50% have been reported by several authors (1, 5). Examples of more recent studies in the literature found lower rates which include 15.1% (25), 17% (13) and 17.6% (3). The recurrence rate is currently unknown at CHBAH.

1.2.3 Clinical presentation

Blount's disease presents as a progressively worsening multi-planar deformity of the proximal tibia in children caused by disordered ossification of the medial proximal tibial physis (5). These three dimensional deformities are varus, internal rotation and procuvatum of the tibia (5).

Patients with infantile Blount's disease present after walking has commenced with varus and internal torsion of the tibia (4). They may have a palpable medial metaphyseal beak which is also visible on X-Ray (1, 4). Children with infantile Blount's disease may have bony instability presenting as a lateral thrust (1, 4). In children with advanced infantile Blount's disease with intra-articular depression, the knee is stable in full extension when a varus force is applied but the medial femoral condyle may sublux into the depressed posteromedial plateau of the tibia at twenty degrees of flexion with a varus force – this is known as the Siffert-Katz sign (1, 15).

About 50% of cases present as a bilateral deformity. In those that are unilateral a leg length discrepancy may be present (4).

The juvenile group present between the ages of four and ten years with tibia varus deformities which are usually bilateral (4). Children with juvenile Blount's disease are usually obese and knee pain may be the presenting complaint (6).

Children with adolescent Blount's disease are typically obese with wide thighs and present with varus deformity which may be unilateral, bilateral or asymmetric (1, 4). Children with adolescent Blount's disease may present with vague pain in the knee (4). In unilateral or asymmetrical cases there may be a limb length discrepancy (1, 4). Children with adolescent Blount's disease were excluded from this study.

1.2.4 Prevalence

Blount's disease is considered rare (1, 26). Smith noted that out of 5000 patients presenting to his institution over 50 years with tibial bowing, 37 had infantile Blount's disease, less than one percent (27). An aetiological study of infantile Blount's disease by Bathfield and Beighton found an occurrence rate of three per 10 000 of children in the catchment area for CHBAH (8).

1.2.5 Aetiology and pathophysiology

The exact aetiology of infantile, juvenile and adolescent Blount's disease is unknown (1, 2, 4, 5).

A mechanical basis for the deformity in Blount's disease has been suggested since it may occur in children who are early walkers or overweight (1, 10, 23, 28). This theory may be correct since correction of the deformity which leads to offloading of the physis may lead to permanent cure, however this does not explain unilateral involvement or cases in which there was not early walking and the child was of normal weight (5, 10).

A racial predisposition has been noted in North America amongst black and Hispanic children (4). A genetic component may be present as a family history of bowed legs has been found in several studies (7, 8, 26, 29, 30). More recent studies have investigated the role of Vitamin D and Zinc in the development of Blount's disease (2, 31, 32).

Obesity

A strong correlation between Blount's disease and obesity is well known (2, 4, 33, 34). Several studies have shown that increased body mass index (BMI) correlates with varus deformity of the tibia (2, 4, 5, 8, 12). The Centers for Disease Control and prevention (CDC) defines childhood obesity as a BMI above the 95^{th} centile (35). A child is considered overweight when the BMI is between the 85^{th} and 95^{th} centiles (35).

The Hueter-Volkman law (36) states that "in the skeletally immature bone growth is relatively inhibited in areas of increased pressure and relatively stimulated in areas of decreased pressure or tension". In Blount's disease this principle suggests that increased load on the medial compartment will lead to decreased growth at the medial proximal tibial physis and the medial distal femoral physis (1, 5, 36). Cook et al. (12) showed that the compressive forces generated with ten degrees of varus in an obese child were greater than needed to retard growth in the medial tibial physis during walking (5, 12).

Another proposed theory by David et al. is that in obese children the thighs are wide and forced into a distinctive fat thigh gait (37). This fat thigh gait leads to increased medial loading of the physis and according to the Hueter-Volkman law will lead to decreased medial growth at the proximal tibial physis and distal femoral physis (1, 2, 5, 37).

Marked obesity leads to higher rates of recurrence. Ferriter and Shapiro (14) noted that obesity above the 97th centile for weight was a risk factor for recurrence. It is postulated that more stress is applied to the medial proximal tibia and medial distal femur in obese children (1, 14).

Early walking

Infantile and juvenile Blount's disease has been associated with early walking (1, 4, 28). In children who walk before one year of age the mechanical axis of the leg is medial to the knee due to normal physiological bowing which has not yet resolved (1, 4, 28). This abnormal loading of the medial compartment of the knee is thought to be a factor in the development of Blount's disease in keeping with the Hueter-Volkmann principle (1, 4, 28, 36). The worsening varus deformity further moves the mechanical axis of the leg medially thereby exacerbating the causative factor and increasing the deformity (28).

We are not aware of any published studies that suggest early walking could be a risk factor for recurrence of Blount's disease post-surgical correction.

Race and gender

According to the literature Blount's disease occurs more commonly in black and Hispanic children (1, 4, 8, 10, 25). Blount and Langenskiöld described cases of Blount's disease in Caucasian children in America and Finland, respectively (9, 23). South African studies have confirmed a racial predilection (3, 8). In 1978, Bathfield found that in his study of 110 children with Blount's disease in the Witwatersrand region of South Africa, all 110 were black (8). Another South African study done in the Western Cape region by White et al. (3) found 86% of those with Blount's disease were black patients.

Studies have shown that black children presented with higher Langenskiöld stages at an earlier age – this may lead to a higher risk of recurrence after surgical correction (4, 5).

There is uncertainty whether the condition occurs more commonly in males or females. Blount and Langenskiöld found more males in their studies (1,2). Bathfield found 55% males in his study at our institution in 1978 while White et al. found 60% females in their study in a South African population in the Western Cape Province (3, 8). The reason for this may be due to the general small population sizes in studies on Blount's disease and the mixed reporting between subgroups of Blount's disease where demographics are concerned.

Metabolic causes

Various metabolic derangements have been investigated as possible causes of Blount's disease. Low levels of Vitamin D can lead to abnormal bone and growth plate mineralisation (31). Low levels of Vitamin D have been postulated as a possible cause of Blount's disease (32). However, studies have presented conflicting evidence regarding Vitamin D deficiency as a causative factor for Blount's disease (31, 32). Lisenda et al. found, in a study done in the same centre as the current study, that 16% of patients had levels of less than 20 nanograms per millilitre of Vitamin D which is considered Vitamin D depleted. These levels were not significantly different to a cohort of matched patients without Blount's disease from the same population (31). Vitamin D levels in recurrence was not investigated by these authors (31).

Vitamin D deficiency has been suggested as a possible cause of increased recurrence rates due to the decreased mineralisation of the bone and growth plates leading to inferior strength and resistance to loads (32). Other biochemical markers that have been studied include alkaline phosphatase (ALP), serum calcium, serum phosphate, haemoglobin and erythrocyte sedimentation rate (ESR) (8, 31, 38). Bathfield and Beighton found normal levels of ALP in the same centre as the current study in 1978, while Lisenda et al. found that 38% of patients with Blount's disease had elevated levels of ALP.

Low levels of Zinc have been noted in patients with Blount's disease (38). Zinc is an antioxidant which plays a role in normal bone homeostasis (38). Malnutrition results in Zinc deficiency that may contribute to the development of Blount's disease (38).

No studies that we are aware of have proven any link between micronutrient deficiencies and recurrence of Blount's disease after surgical correction.

1.2.6 Differential diagnosis

Bowing of the legs can be physiological or pathological (1, 28, 39, 40).

There is a continuum between physiological bowing of the tibia and infantile Blount's disease and the two conditions can be difficult to distinguish on clinical grounds (4). Children with physiological bowing are often early walkers with a bilateral deformity and an intoed gait (1, 28). Radiological investigations are used to aid in decision making. The Metaphyseal-diaphyseal (MD) angle of Drennan is used to differentiate between normal physiological bowing and Blount's disease in children above the age of 18 months when there is enough ossification to measure the Metaphyseal-Diaphyseal angle (MD angle) on X-Ray (Figure 1.2.) (40). An MD angle of more than 16 degrees has a 95% chance of being Blount's disease while an MD angle of less than ten degrees has a 95% probability of being physiological bowing (4, 39). If it is unclear, it is suggested to wait six months and re-evaluate the affected limbs (1). Physiological bowing of the tibia corrects spontaneously before the age of three years and no treatment is necessary (28).

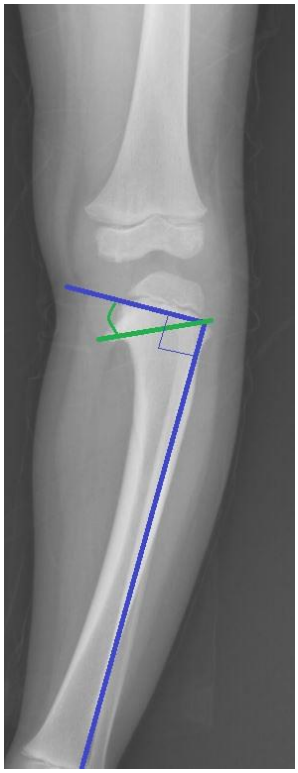


Figure 1.2 The MD angle of Drennan (in green)

Bowing is more likely to be pathological if it is unilateral, there is shortening of the affected limb, short stature, obesity and a lateral thrust (1, 28). Bowing which is present after the age of three years is pathological (1, 28). Pathological causes of bowing include: Rickets, skeletal dysplasias and damage to the physis. Rickets may present with bowed legs but the radiological appearance is different compared to Blount's disease (1, 9, 10). Likewise, skeletal dysplasias may be differentiated from Blount's disease on clinical and radiological grounds (1, 10, 28). Infection or trauma to the physis can lead to growth disturbance in the proximal tibia which again can be distinguished from Blount's disease by history and on X-Ray findings (1, 28) .

1.2.7 Radiological findings

Blount described the radiological findings in infantile Blount's disease as varus of the tibia, a widened physis, a sloping epiphysis and beaklike recurving metaphysis with cartilage islands near the beak medially (9). Sabharwal et al. found advanced skeletal age in patients with Blount's disease (41).

In the juvenile and adolescent types varus deformity with widening of the proximal medial tibial physis is observed on X-Rays of the lower limb (4). Other features noted radiologically include distal femoral physeal growth disturbances or varus. There may be a compensatory distal tibia valgus (1).

Langenskiöld classified infantile Blount's disease into six stages radiologically on X-Ray (Figure 1.3) (23). Intra-observer agreement is poor (4). Subsequent studies have questioned the prognostic value of these stages (4, 5).

The Langenskiöld classification consists of (23, 42):

***Stage I*:** Beaking of the proximal medial tibial metaphysis.

Stage II: Saucer shaped defect of medial metaphyseal beak with medial epiphyseal wedging.

Stage III: Medial saucer shaped defect deepens into metaphyseal step. Medial epiphysis becomes irregular.

Stage IV: The epiphysis extends over medial metaphyseal beak filling in the metaphyseal step.

Stage V: Appearance of a horizontal cleft called the double epiphysis sign.

Stage VI: Closure of medial epiphysis (42).

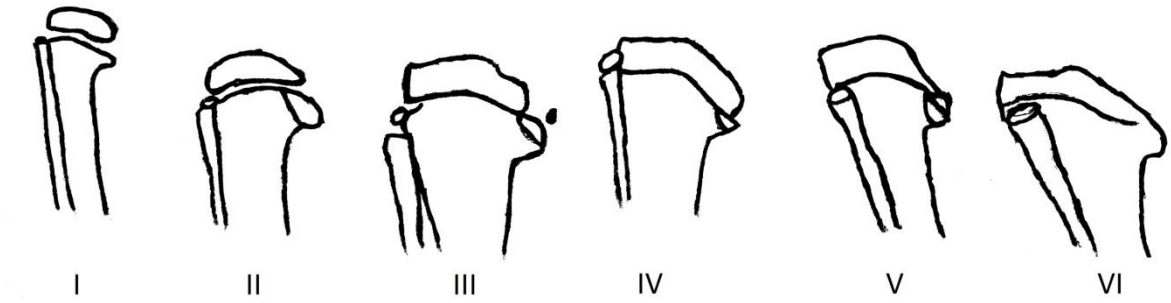


Figure 1.3 Line drawing by author of X-Rays from Langenskiöld's 1981 article illustrating the Langenskiöld stages (24)

Worsening recurrence rates are associated with increasing Langenskiöld stages due to the formation of a physeal bar which closes the growth plate (1, 2, 43). Even if correction is done in later stages, it will likely fail since the child will continue growing more on the lateral side of the proximal tibia (1, 2, 43).

1.2.8 Radiological measurements

Several measurements are used in assessing Blount's disease on an X-Ray. The gold standard for determining angulation of the knee is a full limb standing radiograph with patellae facing forwards (44). A weight bearing knee X-Ray has been shown to be comparable to full limb radiography in determining varus deformities around the knee in adults (44, 45). We are not aware of any specific studies that compare full length and focused knee X-rays in the paediatric population for determining varus deformities.

On the full limb radiograph the mechanical femorotibial angle (mFTA) can be determined by drawing a line from the centre of the femoral head to the centre of the knee (the mechanical axis of the femur), and a line from the centre of the tibia to the centre of the ankle (the mechanical axis of the tibia) (46) .

The anatomic femorotibial angle (aFTA) is measured by drawing a line from the centre of the proximal femur shaft to the centre of the knee (the femoral shaft axis), and the mechanical axis of the tibia (46) (Figure 1.4). The aFTA can be used when full length radiographs are unavailable (44-46).

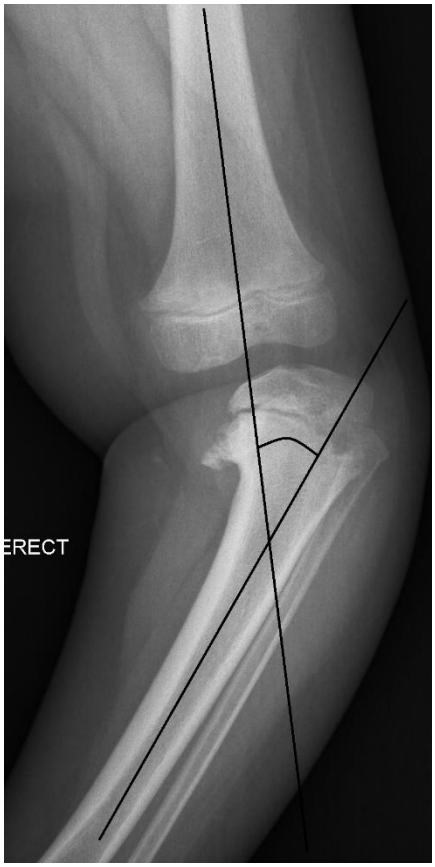


Figure 1.4 The anatomic femorotibial angle (aTFA) illustrated by main author from study limb

1.2.9 Non-operative management

Bracing has been suggested by some as a treatment in infantile Blount's disease (47-49). Other authors have suggested that early stage infantile Blount's disease may spontaneously correct without any treatment (50, 51). Richards, (48) Zionts (49) and Raney (47) while showing that bracing may be effective in children less than three years of age; did not have control groups, could not show that the bracing was worn as prescribed and did not account for the fact that infantile Blount's disease may spontaneously correct as suggested by Blount (10, 51, 52). Compliance with bracing may be difficult, especially in bilateral cases (1). Bracing is not likely to be effective in children older than three years, obese children, bilateral deformities and Langenskiöld stage three or above (1, 4, 5, 48, 53).

Bracing as a non-operative treatment of juvenile and adolescent Blount's disease has not been described (4). Bracing is unlikely to succeed since all these patients per definition will be over three years of age. Failure of bracing requires operative management (1, 10, 48). In the current study, centre bracing is not used as it is not generally available in our institution, and the patients with infantile Blount's disease tend to present to us at a later stage and older age.

1.2.10 Surgical management

Blount, Langenskiöld and Riska advocated for early surgical intervention in children who are not candidates for bracing (7, 9, 23, 52).

Blount advocated for a simple osteotomy and plaster of Paris without internal fixation to align the tibia in infantile Blount's disease in his original article (9). Langenskiöld described a curved proximal tibial and fibular osteotomy with bayonet apposition without using internal fixation in Plaster of Paris (24).

High tibial and fibular valgus osteotomy (HTO) with crossed Kirschner wires, Steinman pins, plate fixation or external fixation may be used in the infantile, juvenile or adolescent groups to achieve acute correction (1, 4, 5, 14, 54). In the infantile and juvenile groups, overcorrection into valgus is advised to decrease the chance of recurrence (4, 5, 14, 24, 30, 52, 55). Loder and Johnston recommended an overcorrection of five to ten degrees of valgus (43). In the adolescent group, correction into neutral or slight valgus is advised since there is less growth potential and a risk of having a residual valgus (4).

Surgery after the age of four years has been recognised by several authors as a risk factor for recurrence (20, 43, 55). In Chotigavanichaya et al.'s study, 46% of patients that had an HTO before four years compared to 94% of patients that had an HTO after four years had recurrence, respectively (55). Ferriter and Shapiro found a 31% recurrence rate with HTO before the age of four and a half, and a recurrence rate of 76% after the age of four and a half (14).

Temporary hemiepiphysiodesis with staples was described by Blount (56). More recently use of tension band plates to provide temporary hemiepiphysiodesis has been described in infantile, juvenile and adolescent patients (1, 4, 5, 57).

Gradual correction with external fixation has been suggested as a treatment for adolescent Blount's disease by several authors (1, 5, 58-63).

In patients with higher Langenskiöld stages, a so called double elevating osteotomy named by Gregosiewicz is another surgical option (64). The procedure itself is a misnomer since the medial tibial plateau is elevated and a HTO is then performed either acutely or with a frame to provide gradual correction and there is not a double elevation. A bone graft may be taken from the valgus osteotomy or a tricortical graft may be taken from the ilium. The consideration for doing a double elevating osteotomy is to decrease the recurrence rate in advanced Blount's disease. A lateral epiphysiodesis has been noted to decrease the rate of recurrence after a double elevating osteotomy (65).

CHAPTER 2: METHODOLOGY

2.1 Study Aim

To determine the recurrence rate of infantile and juvenile Blount's disease post high tibial osteotomy and to identify factors that influence the recurrence rate.

2.2 Study Objectives

The objectives of this study were to identify the rate of recurrence and whether the following aspects were risk factors on the recurrence rates in the study population:

- Demographics
- Family history
- Body mass index
- Amount of correction with high tibial valgus osteotomy
- Langenskiöld staging at time of surgery
- Age at time of surgery
- Role of the FTA

2.3 Research Methodology

This was a retrospective cohort comparative study.

Another study on Blount's disease ran concurrently with this study. Different aspects were researched in the same population group and some of the data were shared between the studies.

This is a single centre study incorporating patients from CHBAH.

2.4 Materials and Methods

Data collection: Files and theatre records kept in the central storage area at CHBAH were used.

X-Rays were retrieved from the picture archiving and communication system (PACS) at CHBAH or film printouts from the central storage area.

2.5 Study Sample

Inclusion criteria: All patients aged zero to ten years or were less than ten years old at the time of development of Blount's disease who had HTOs done were included. This included patients in the infantile and juvenile groups which were previously classified as infantile and juvenile onset groups but were assessed again on radiological appearance and time of onset of varus.

Exclusion criteria: Patients of the adolescent group. These patients have onset of varus above the age of ten years. Patients with inadequate records and X-Rays were excluded. Limbs with less than 12 months of follow up time after surgical correction were excluded unless recurrence occurred before 12 months.

Definition of recurrence: ten degrees of varus or more as measured on standing X-rays at final follow up was used as our definition of recurrence. If a repeat surgery was performed for a limb but no X-ray was available, the limb was excluded. If the X-ray before repeat surgery showed less than ten degrees of varus it was excluded (one limb).

Follow up: an end point (for recurrence) of follow up for each limb was defined as an X-ray follow up image of more than ten degrees of varus. Preference was given to the most adequate radiological X-ray nearest to the repeat surgery if a repeat surgery was done. No further follow up was done since recurrence had occurred according to our definition. Limbs that did not reach ten degrees of varus by the end of the study period of 31 August 2018 with at least 12 months follow up were defined as non-recurrent.

Files, theatre records and X-Rays were recovered in all patients aged zero to ten years that had HTO for infantile and juvenile Blount's disease at CHBAH between 01 January 2009 and 31 August 2018 with a minimum of 12 months follow up for non-recurring limbs.

2.6 Data Collection

Data were collected by the main investigator (EvG) and a research assistant (ME). Files and theatre records were collected from the CHBAH file storage.

X-Rays were collected from the PACS system, or photographed from the central storage centre. X-rays were evaluated for quality, if they were standing for pre-operative and follow up, if they were AP with the patella facing forward and if they were of the knee to allow the anatomic axis to be drawn. X-rays that were not adequate were excluded.

From the files and theatre records the date of birth, the date of referral, the age the deformity was first noticed, the walking age, the sex, race, type of Blount's disease, affected limb or limbs, family history, date of surgery, type of surgery and second surgery if done were captured into a Microsoft Excel 2007 spreadsheet. If a limb reached ten degrees of varus on an X-ray, it was defined as recurrence and no further follow up was done. We did not determine how many further surgeries were done on each limb.

From the X-Rays the Langenskiöld stage was determined, as well as the date of each image and the side. The data were captured into the same Microsoft Excel 2007 spreadsheet.

Using pictures of the X-Rays a freeware programme, MB-Ruler, was used to measure angles of lines drawn on the X-Rays using Microsoft Paint. The anatomic femoral axis and the anatomic tibial axis were drawn. The measured angles were captured in the same Microsoft Excel 2007 spreadsheet.

We attempted to collect all the X-Rays (pre-operative and post-operative) however, some were of poor quality and could not be used, and some were missing.

2.7 Data Analysis

Microsoft Excel 2007 was used to create the data collection sheet and capture the relevant data. Descriptive and inferential statistics were used. Statistical analysis was done in consultation with a biostatistician. Categorical variables were described using percentages. The Student t test, the Mann Whitney test and the Fishers exact test were used to determine significance where appropriate using Graphpad InStat version five, a statistical software package, in which case a p-value of <0.05 was considered significant.

2.8 Limitations

Limitations of the study were the nature of the study being retrospective - loss of several X-Rays as a result of not being filed or being misplaced by patients or their parents. We had access to most of the X-Rays done at CHBAH due to them being stored digitally.

We were unable to collect all the weights and heights (and thus body mass index) at presentation of all the patients if they were not noted in the file.

Some of the subgroups had small numbers of data points to study for example the juvenile group had eleven recurring patients and three non-recurring patients.

Lastly, the follow up time was not until skeletal maturity and the recurrence rate would have potentially been higher had the follow up time been longer.

CHAPTER 3: RESULTS

3.1 Demographics

In this study there were 52 patients in the age group of zero to ten years at time of diagnosis of Blount's disease that had high tibial osteotomies for Blount's disease between 01 January 2009 and 31 August 2018(the total group).

There were 34 patients identified in the infantile group and 18 in the juvenile group. In the study four patients were excluded from the infantile group due to inadequate follow up and four were excluded from the juvenile group due to inadequate follow up. After exclusions, a total of 44 patients were included, 30 from the infantile group and 14 from the juvenile group. There were 6 males and 38 females in total. All the patients that were identified were black Africans. In this study 60 limbs which had HTO's were identified in total. There were 42 limbs included in the infantile group and 18 in the juvenile group. Excluded from the study were five limbs which had tension band plating done as the initial treatment, four in the infantile group and one in the juvenile group (Table 3.1).

The mean age at presentation to CHBAH was 71.8 months for the total group with a range of 18 to 147 months, 61.2 months for the infantile group with a range of 18 to 147 months and 94.6 months for the juvenile group with a range of 61 to 138 months. (Tables 3.2.1, 3.2.4 and 3.2.5).

The average length of follow up for all patients was 27.88 months with a range of six months to seventy-four months in the total group. In the infantile group the mean follow up was 27.86 months with a range of six months to 73 months and in the juvenile group the mean follow up was 27.94 months with a range of six months to 74 months (Tables 3.2.1, 3.2.4 and 3.2.5). In the total group the shortest follow up time was twelve months for non-recurring limbs. The longest follow up time for a non-recurring limb was 74 months.

Table 3.1 Demographics

Variable	Infantile	Juvenile	Total
Number of patients	30	14	44
Gender Male/Female	4/26	2/12	6/38
Bilateral presentation number of patients	17	6	23
Unilateral presentation number of patients	13	8	21
Affected limb (left)	26	10	36
Affected limb (right)	16	8	24
Affected limb total	42	18	60

3.2 Recurrence

There was a limb overall recurrence rate of 63.33% at final follow up. In the infantile group 59.5% of limbs had recurrence after HTO. In the juvenile group of limbs there was a recurrence rate of 72.2%. Although there were more cases of recurrence in the juvenile group, this was not a statistically significant difference between the groups for recurrence ($p = 0.3963$). Bilateral Blount's disease occurred in 24 out of 44 of the total group of patients. There was a family history of Blount's disease in five of the 44 patients; four of these patients had recurrence which gives a rate of 12.5% family history in the recurrent group versus 8.3% in the non-recurrent group. Though there were a higher number of patients with a positive family history in the recurrent group the difference was not statistically significant ($p = 1$). BMI was not found to be statistically significant ($p =0.774925$). Developmental history was also not a statistically significant risk factor for recurrence ($p = 0.942686$) (Table 3.2). The age at presentation was not found to be significant in the total group ($p = 0.098647$).

The difference in follow up time from initial HTO to last follow up was statistically significant ($p = 0.00705$) between the recurrent and non-recurrent groups of limbs in the total group with a longer follow up time for the non-recurrent limbs. The non-recurrent limbs had a mean follow up of 35.82 months while the recurrent limbs had a mean follow up of 23.29 months (Table 3.2).

The differences in Langenskiöld stages between recurring and non-recurring limbs in the total group were found to be statistically significant ($p = 0.02382$). Dividing the Langenskiöld stages into stage four or less and five and more did show statistically significant differences between the non-recurring and recurring groups ($p = 0.0143$). Different recurrence rates were noted with the Langenskiöld stages of limbs in the total group: 0% recurrence at stage two, (zero out of three limbs) 58.8% at stage three (ten out of 17 limbs), 50% at stage four (ten out of 20 limbs) and 87.5% at stage five (14 out of 16 limbs) (Figure 3.1).

The BMI at presentation was known in 29 of the 44 of the total patients, 4% were underweight with a BMI below the 5^{th} centile, 20% were of normal or healthy weight with a BMI between the 5^{th} and 85^{th} centiles, 17% were overweight with a BMI between the 85^{th} and the 95^{th} centiles and 59% were obese with a BMI above the 95^{th} centile as defined by the CDC (Table 3.3).

There was no statistically significant difference between the recurrence rates between patients with a BMI above or below the 97^{th} centile for BMI as used by Ferriter and Shapiro (14) in their study ($p = 0.7279$) (Table 3.4).

Table 3.2 Recurrence in total group

Variable	Recurring	Non-recurring	p value
Number of patients	32	12	
Infantile / juvenile patients	21/11	9/3	0.7222 Fishers exact test
Limbs	38	22	
Infantile / juvenile limbs	25/13	17/5	0.3963 Fishers exact test
Male/Female	4/28	2/10	0.6577 Fishers exact test
Bilateral patients / unilateral patients	20/12	4/8	0.1024 Fishers exact test
If unilateral L/R	10/2	5/3	0.3473 Fishers exact test
Family History(Patients) – yes/no	4/28	1/11	1 Fishers exact test
Patient age at presentation in months (SD)	77.13 (30.70) N=32	57.67 (42.03) N=12	0.098647 Unpaired t-test
Patient BMI Mean (SD)	24.74 (8.22) N=24	23.43 (13.55) N=5	0.774925 Unpaired t-test
Weight kg (SD)	44.47(20.86) N=24	42(38.50) N=5	0.837938 Unpaired t-test
Height in meters (SD)	1.31 (0.18) N=24	1.26 (0.21) N=5	0.517693 Unpaired t-test
Walking months (SD)	12.33 (3.04) N=24	12.43 (3.10) N=7	0.942686 Unpaired t-test
Langenskiöld stage Median (Range)	4(3-5) N= 34	4 (2-5) N = 22	0.02382 Mann Whitney test *
Langenskiöld stage <3 / Langenskiöld stage ≥3	0/34	3/19	0.0556 Fishers exact test
Langenskiöld stage <4 / Langenskiöld stage ≥4	10/24	10/12	0.262 Fishers exact test
Langenskiöld stage <5 / Langenskiöld stage ≥5	20/14	20/2	0.0143 Fishers exact test *
Limbs months of follow up(SD)	23.29 (14.67) N=38	35.82 (19.87) N= 22	0.00705 Unpaired t-test **

SD = Standard deviation

* P < 0.05 = Significant

** P < 0.01 = Very significant

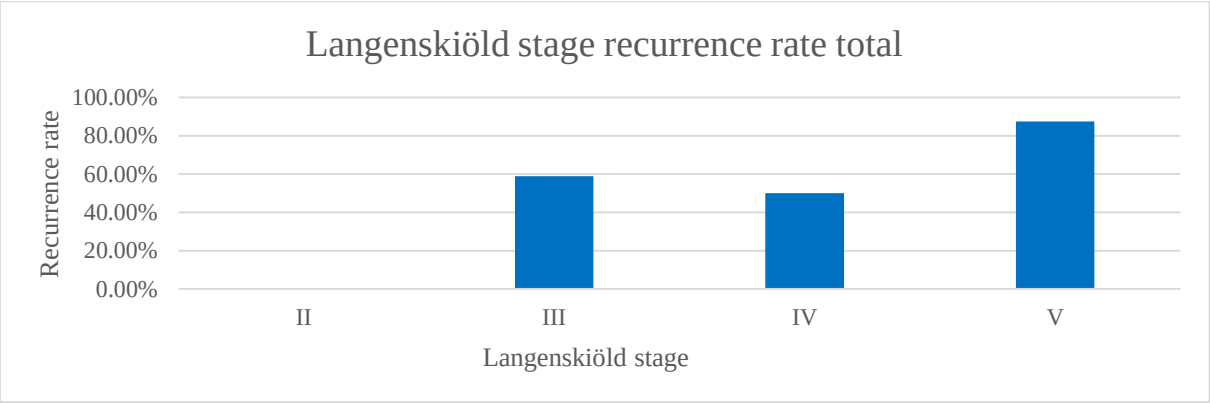


Figure 3.1 Langenskiöld stage and recurrence total group

Table 3.3 Infantile and juvenile BMI per CDC centiles for Teen and Child BMI

BMI	Underweight	Normal	Overweight	Obese
Infantile	1	5	3	11
Juvenile	0	1	2	6

Table 3.4 Infantile and juvenile BMI using the 97th centile

	Recurring	Non recurring	P value
Infantile limbs <97th/ ≥97th centile	12/10	5/3	1 Fishers exact test
Juvenile limbs <97th/ ≥97th centile	2/8	1/2	1 Fishers exact test
Total limbs <97th/ ≥97th centile	14/18	6/5	0.7279 Fishers exact test

There was recurrence in 25 out of 42 limbs in the infantile group which had an HTO giving a recurrence rate of 59.5%. Gender, the age at referral, family history, BMI and developmental history were not statistically significant factors for recurrence in the infantile group (Table 3.5).

Patients who had surgery on their limbs before or after four years had a statistically significant difference between the recurring and non-recurring infantile groups (p = 0.0449) (Table 3.5).

The follow up time was significantly longer in the non-recurrent group of infantile limbs (p = 0.003771) (Table 3.5).

The BMI was known in 20 of the 30 infantile patients, of these 45% were obese, 30% were overweight, 25% had a healthy or normal weight and 5% were underweight according to the CDC child and teen BMI centiles. There was no statistical difference between the recurrence rates between patients with a BMI above or below the 97^{th} centile for BMI ($p = 1$) . (Tables 3.22 and 3.2.3)

The Langenskiöld staging did not have a statistically significant difference between the recurring and non-recurring infantile groups as a median between the groups or grouping the stages together ($p = 0.12852$). The recurrence rate varied with the Langenskiöld stage of limbs in the infantile group: 0% recurrence at stage two (zero out of three limbs), 58.8% at stage three (ten out of 17 limbs), 53.8% at stage four (seven out of 13 limbs) and 83.3% at stage five (five out of six limbs) (Figure 3.2).

Table 3.5 Recurrence in infantile group

Variable	Recurring	Non-recurring	p value
Patients	21	9	
Limbs	25	17	
Male/Female	3/18	1/8	1.00 Fishers exact test
Bilateral / unilateral	14/7	4/5	0.4181 Fishers exact test
If unilateral L/R	7/0	4/1	0.4167 Fishers exact test
Family History – yes/no	3/18	1/8	1 Fishers exact test
Patient age at presentation in months (SD)	67.43 (31.34) N=21	46.67 (38.56) N=9	0.131711 Unpaired t-test
Patient BMI Mean (SD)	21.67 (5.78) N=16	25.09(15.05) N=4	0.459763 Unpaired t-test
Weight (SD)	35.62 (15.21) N=16	44.5(43.99) N=4	0.492608 Unpaired t-test
Height in meters (SD)	1.26 (0.18) N=16	1.23 (0.23) N=4	0.721724 Unpaired t-test
Walking months (SD)	12.5 (3.36) N=18	13.2 (3.27) N=5	0.68331 Unpaired t-test
Langenskiöld stage Median (Range)	4 (3-5) N=23	3(2-5) N=17	0.12852 Mann Whitney test
Langenskiöld stage <3 / Langenskiöld stage ≥3	0/22	3/14	0.0744 Fishers exact test
Langenskiöld stage <4 / Langenskiöld stage ≥4	10/12	10/7	0.5231 Fishers exact test
Langenskiöld stage <5 / Langenskiöld stage ≥5	17/5	16/1	0.2056 Fishers exact test
Limbs follow up in months(SD)	20.28(13.54) N=25	36.18 (20.01) N=17	0.003771 Unpaired t-test **
Surgery before 4 years (Limbs)/ Surgery after 4 years (Limbs)	2/23	6/11	0.0449 Fishers exact test *

* P < 0.05 = Significant

** P < 0.01 = Very significant

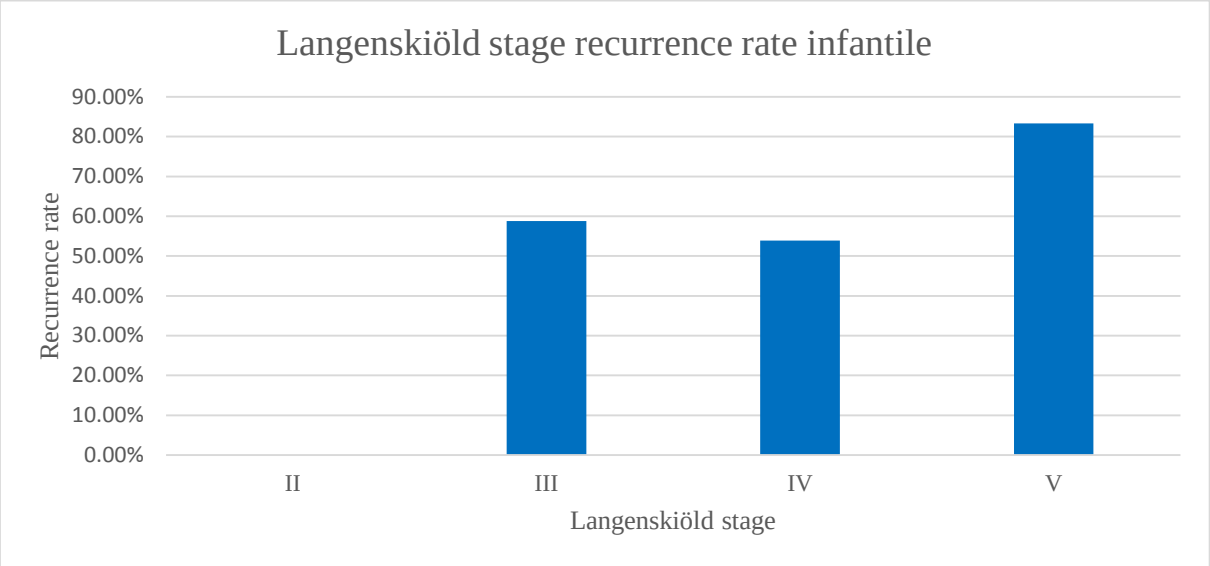


Figure 3.2 Langenskiöld stage and recurrence in the infantile group

In the juvenile group of limbs there was a recurrence rate of 72.2%. Gender, the age at referral, family history, BMI and developmental history were not statistically significant factors for recurrence in the juvenile group (Table 3.6).

The difference in follow up time from initial HTO to last follow up was not statistically significant between the recurrent and non-recurrent groups of limbs in the juvenile group with a longer follow up time in the non-recurrent group ($p = 0.471118$) (Table 3.6).

The Langenskiöld staging between the recurring and non-recurring juvenile groups as a median between the groups was not found to be statistically significant ($p = 0.09102$). The recurrence rate varied with Langenskiöld stage of limbs in the juvenile group: 42.9% at stage four (three out of seven limbs) and 90% at stage five (nine out of ten limbs) (Figure 3.3). These differences were not found to be statistically significant with the only p-value able to be calculated being between stage one to four and stages five and six ($p = 0.1007$) (Table 3.6).

The BMI was known in nine patients in the juvenile group, 67% were Obese (six patients), 22% were overweight (two patients) and 11% were normal or healthy weight (one patient) according to their CDC child and teen BMI centiles (Table 3.3). There was no statistical difference between the recurrence rates between patients with a BMI above or below the 97th centile for BMI ($p = 1$)(Table 3.4).

Table 3.6 Recurrence in the juvenile group

Variable	Recurring	Non-recurring	p value
Patients	11	3	
Limbs	13	5	
Male/Female	1/10	1/2	0.3956 Fishers exact test
Bilateral / unilateral	6/5	0/3	0.2088 Fishers exact test
If unilateral L/R	3/2	1/2	1 Fishers exact test
Family History – yes/no	1/10	0/3	1 Fishers exact test
Patient age at presentation in months (SD)	95.64 (19.59) N=11	90.666 (39.88) N=3	0.757784 Unpaired t-test
Patient BMI Mean (SD)	30.87 (9.28) N=8	16.80 (N/A) N=1	N/A
Patient Weight in kilogram (SD)	62.18 (19.92) N=8	32 (N/A) N=1	N/A
Patient Height in meters (SD)	1.42 (0.13) N=8	1.38 (N/A) N=1	N/A
Walking (SD)	11.83(1.94) N=6	10.5(2.12) N=2	0.43915 Unpaired t-test
Langenskiöld stage Median (Range)	5 (4-5)	4 (4-5)	0.09102 Mann Whitney test *
Langenskiöld stage <3 / Langenskiöld stage ≥3	0/12	0/5	N/A
Langenskiöld stage <4 / Langenskiöld stage ≥4	0/12	0/5	N/A
Langenskiöld stage <5 / Langenskiöld stage ≥5	3/9	4/1	0.1007 Fishers exact test
Limbs Follow up in months(SD)	25.85 (18.01) N=13	33.4 (23.23) N=5	0.471118 Unpaired t-test

* $P < 0.05$ = Significant

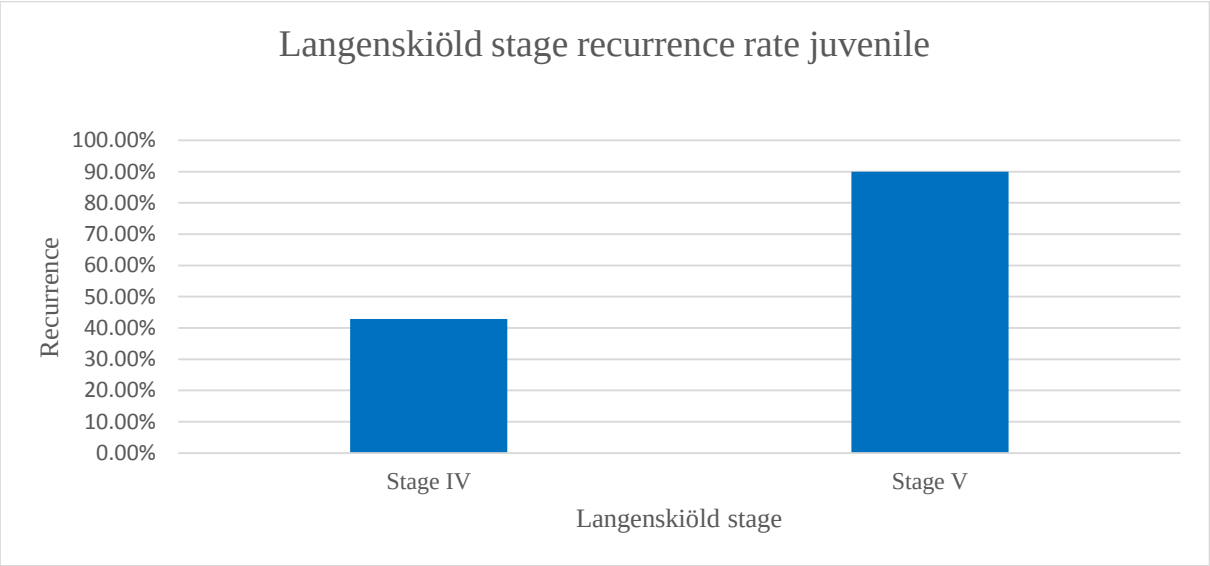


Figure 3.3 Langenskiöld stage and recurrence in the juvenile group

In the total group consisting of the infantile and juvenile groups the differences between non-recurring and recurring pre-operative FTA (p = 0.9550.4), amount of correction (p = 0.114356), and less than five degrees of correction of valgus was not significant (p = 0.6448). There was a statistically significant difference between the post-operative FTA with a higher valgus in the non-recurring group (p = 0.00063). There was a statistically significant difference between the recurring and non-recurring groups of limbs with more than 15 degrees of valgus post op with those that were corrected more than 15 degrees not recurring (p = 0.0008). The follow up in recurring and non-recurring FTA difference was statistically significant as expected (p <0.00001) (Table 3.7).

Table 3.7 Total group measurements

	Recurring	Non-recurring	P Value
Pre-operative FTA (SD)	23.81 (11.16) N=31	23.63 (7.50) N=15	0.955034 Unpaired T test
Post-operative FTA(SD)	-10.22 (5.81) N=27	-17.41 (5.98) N=14	0.00063 Unpaired T test ***
Amount of correction pre and post FTA	34.84 (11.27) N= 26	41.08(10.53) N=12	0.114356 Unpaired T test
Correction < 5° Valgus />= 5° Valgus	4/23	1/13	0.6448 Fishers exact test
Correction < 10° Valgus />= 10° Valgus	12/15	2/12	0.0838 Fishers exact test
Correction < 15° Valgus />= 15° Valgus	21/6	3/11	0.0008 Fishers exact test ***
Follow up FTA(SD)	16.72 (6.52) N= 38	-2.52 (7.95) N=22	<0.00001 Unpaired T test***

*** P < 0.001 = Extremely significant

In the infantile group the differences between non-recurring and recurring pre-operative FTA (p = 0.985751), amount of correction (p = 0.18637), and less than ten degrees of correction of valgus was not significant (p = 0.2492). There was a statistically significant difference between non-recurring and recurring post-operative FTA with a higher valgus in the non-recurring group (p = 0.006631). There was a statistically significant difference between the recurring and non-recurring groups of limbs with more than 15 degrees of valgus post op (p = 0.0143). The follow up recurring and non-recurring FTA difference was statistically significant as expected (P < 0.00001) (Table 3.8) (Figures 3.4 and 3.5).

Table 3.8 Infantile group measurements

	Recurring	Non Recurring	P value
Pre-operative FTA (SD)	23.52 (11.70) N=22	23.45 (8.05) N=12	0.985751 Unpaired T test
Post-operative FTA(SD)	-10.57 (6.20) N=21	-17.20 (6.47) N=12	0.006631 Unpaired T test **
Amount of correction pre and post FTA (SD)	34.33 (10.76) N=20	40.01 (11.30) N= 10	0.18637 Unpaired T test
Correction < 5° Valgus />= 5° Valgus	3/18	1/11	1 Fishers exact test
Correction < 10° Valgus />= 10° Valgus	9/12	2/10	0.2492 Fishers exact test
Correction < 15° Valgus />= 15° Valgus	15/6	3/9	0.0143 Fishers exact test *
Follow up FTA(SD)	16.25 (7.02) N=25	-3.75 (8.11) N=17	<0.00001 Unpaired T test ***

* P < 0.05 = Significant

** P < 0.01 = Very significant

*** P < 0.001 = Extremely significant

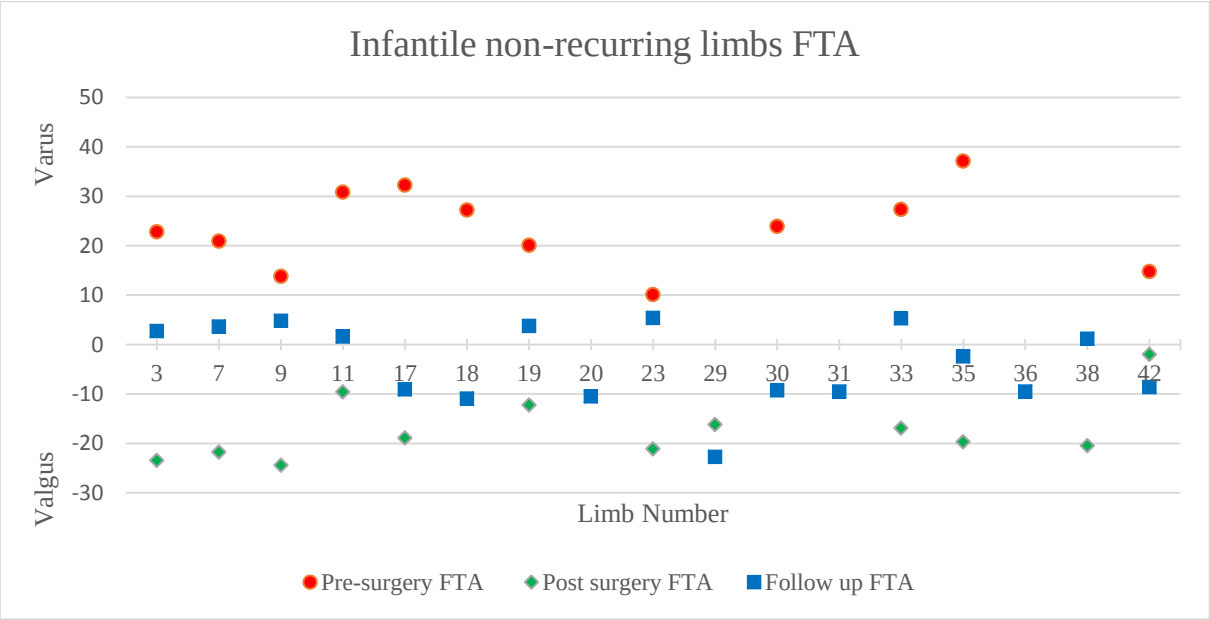


Figure 3.4 Infantile non-recurring limbs FTA by limb number

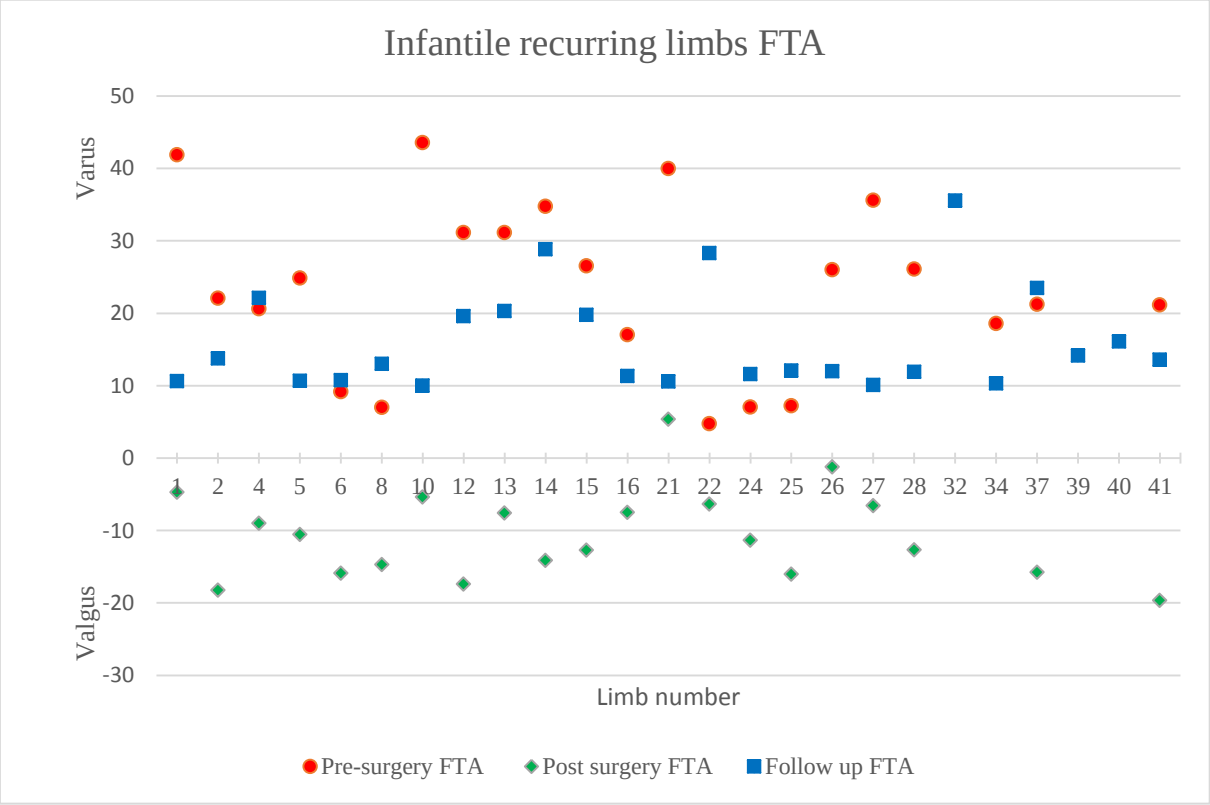


Figure 3.5 Infantile recurring limbs FTA by limb number

In the juvenile group the differences between non-recurring and recurring pre-operative FTA (p = 0.958584), amount of correction (p = 0.313727), and less than ten degrees of correction of valgus was not significant (p = 0.4643). There was a statistically significant difference between non-recurring and recurring post-operative FTA with a higher valgus in the non-recurring group (p = 0.026935). There was a statistically significant difference between the recurring and non-recurring groups of limbs with more than 15 degrees of valgus post op (p = 0.0357). The follow up recurring and non-recurring FTA difference was statistically significant as expected (p = 0.00009) (Table 3.9) (Figures 3.6 and 3.7).

Table 3.9 Juvenile group measurements

	Recurring	Non Recurring	P value
Pre-operative FTA (SD)	24.67 (9.75) N=10	24.36 (6.03) N=3	0.958584 Unpaired T test
Post-operative FTA(SD)	-9.01 (4.43) N=6	-17.8 (1.56) N=3	0.026935 Unpaired T test *
Amount of correction pre and post FTA	35.37 (13.47) N= 6	46.415 (0.90) N= 2	0.313727 Unpaired T test
Correction < 5° Valgus />= 5° Valgus	1/5	0/2	1.00 Fishers exact test
Correction < 10° Valgus />= 10° Valgus	3/3	0/2	0.4643 Fishers exact test
Correction < 15° Valgus />=15° Valgus	6/0	0/2	0.0357 Fishers exact test *
Follow up FTA(SD)	17.59 (5.64) N= 13	1.66 (6.38) N=5	0.00009 Unpaired T test ***

* P < 0.05 = Significant

*** P < 0.001 = Extremely significant

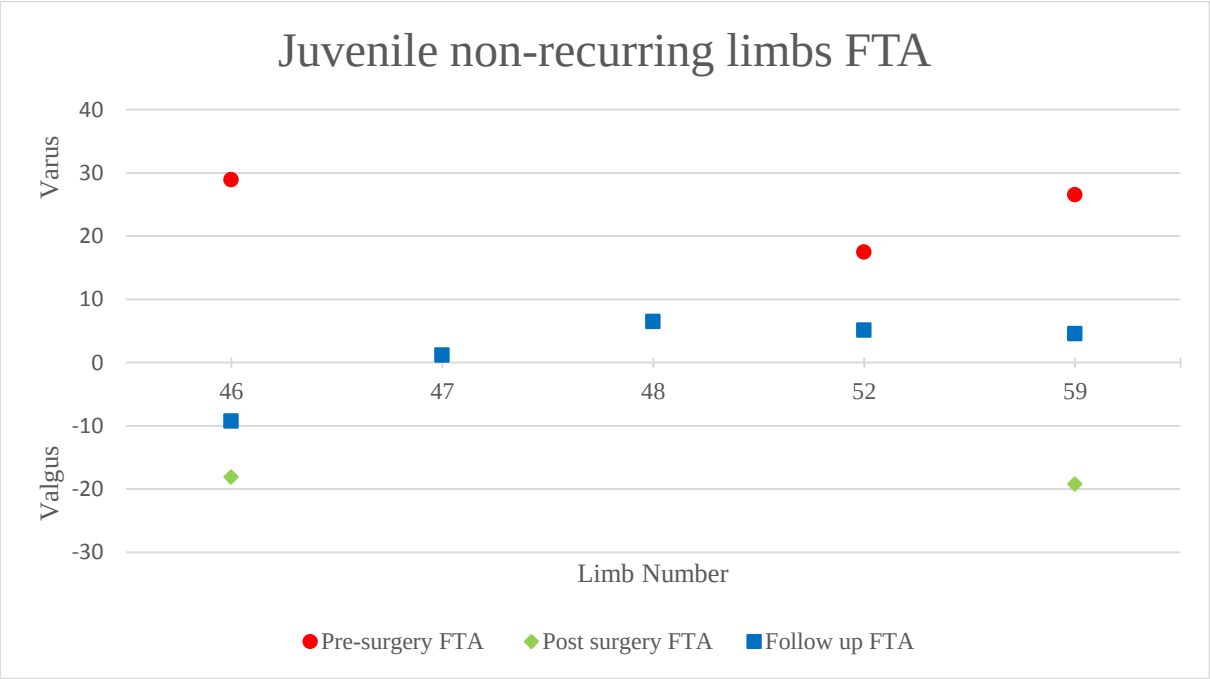


Figure 3.6 Juvenile non-recurring limbs FTA by limb number

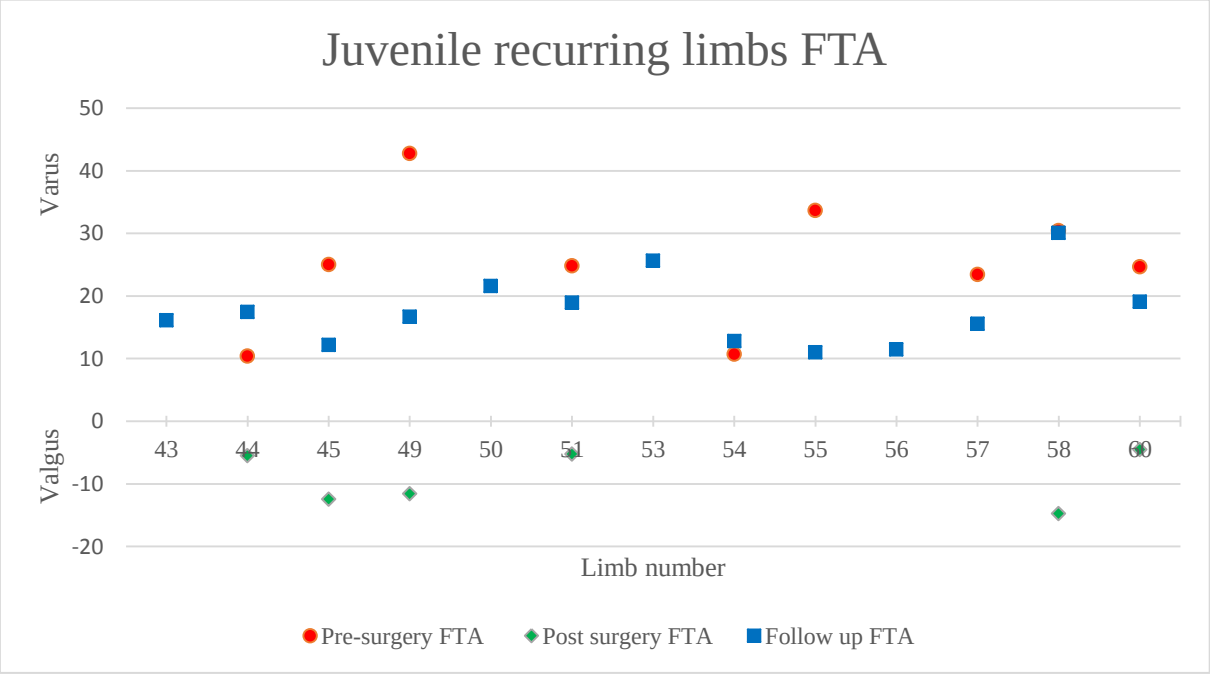


Figure 3.7 Juvenile recurring limbs FTA by limb number

CHAPTER 4: DISCUSSION

4.1 Recurrence of Blount's disease

4.1.1 Introduction

Blount's disease is considered a rare condition (1, 4, 5). Our institution has a large catchment area and previous studies done at CHBAH have found large groups of patients in comparison with international studies (8, 31).

4.1.2 Recurrence rate at CHBAH

The recurrence rate after HTO at CHBAH for infantile and juvenile Blount's disease was previously unknown. There have been previous studies on Blount's disease done at CHBAH, but none have looked at the recurrence rate in infantile and juvenile Blount's disease after HTO at CHBAH (8, 31). In the total group of limbs there was a recurrence rate of 63.3%. In the infantile group 59.5% of limbs had recurrence after HTO. In the juvenile group of limbs there was a recurrence rate of 72.2%.

4.1.3 Age of surgery as a risk factor for recurrence

In infantile Blount's disease delayed surgery is a recognized risk factor for recurrence (4, 5). Several authors have found that surgery after the age of four years is a statistically significant risk factor for recurrence of Blount's disease after HTO (20, 43, 55). Chotigavanichaya et al found 46% of limbs that had an HTO before four years and 94% of limbs that had a HTO after four years had recurrence respectively (55). Ferriter and Shapiro found that surgery after four and a half years was a statistically significant risk factor for recurrence after HTO (14). Ferriter and Shapiro found a 31% recurrence rate with HTO before the age of four and a half and a recurrence rate of 76% after the age of four and a half (14).

In the current study surgery after the age of four years was found to be statistically significant ($p = 0.0449$). In this study 25% of limbs which had an HTO before the age of four years had recurrence while 67.7% of limbs who had an HTO after the age of four years had recurrence. This recurrence rate is comparable to other studies (14, 55)

4.1.4 Length of follow up and recurrence

The mean length of follow up was 27.88 months in the total group, 27.86 months in the infantile group and 27.94 months in the juvenile group. There was a statistically longer follow up time in the total ($p = 0.00705$) and infantile non-recurrent groups ($p = 0.0449$) which is to be expected to try and ensure that the recurrence rate actually reflects all those that will recur.

In the non-recurring group mean follow up was 35.82 months in the total group (range 12 - 74), 36.18 months in the infantile group (range 12-73) and 33.4 months in the juvenile group (range 15-74).

In the recurring group mean follow up was 23.29 months in the total group (range 6- 63), 20.28 months in the infantile group (range 6 - 63) and 28.58 months in the juvenile group (range 6-63).

Due to the formation of a physeal bar in later Langenskiöld stages and damage to the growth plate longer follow up usually leads to higher recurrence rates. Follow up to skeletal maturity will determine the final recurrence rate allowing for late recurrence even though the follow up time in the study is already significantly longer in the non-recurrent groups.

4.1.5 Obesity as a risk factor for recurrence

Several studies have shown that obesity is associated with development of Blount's disease (4, 33, 34, 66). Thompson et al. found obesity to be a risk factor for recurrence (6). In this study patients with a BMI above the 97^{th} centile were compared with patients lower than the 97^{th} centile for weight as in Thompson's study (6). In the total, (infantile and juvenile groups combined), infantile and the juvenile groups no statistically significant difference was found in recurrence rates between patients with a BMI above or below the 97^{th} centile.

No absolute BMI or centile was found in this study that supports obesity as a risk factor for recurrence.

4.1.6 Early walking as a risk factor for recurrence

Early walking has not been described as a risk factor for recurrence but it has been postulated as a risk factor for Blount's disease itself (4, 5). In this study walking ages were compared in the total, infantile and juvenile groups between recurring and non-recurring patients. The mean walking age was 12.33 months in the recurring group and 12.43 months in the non-recurring total groups. No statistically significant difference was found between recurring and non-recurring patients in any of the groups for early walking as a risk factor for recurrence.

4.1.7 Race and gender as a risk factor for recurrence

Blount's disease has been described as being more common in black and Hispanic children (4, 5). Studies have shown conflicting results regarding if males or females more commonly have Blount's disease (7, 9, 11, 55). We were unable to determine if race was a risk factor for recurrence since all our patients were black African children. In this study 87.5% and 83.3% of the patients were female in the recurring and nonrecurring groups, respectively. This was not a statistically significant risk factor for recurrence ($p = 0.6577$).

4.1.8 Langenskiöld stage as a risk factor for recurrence

Increased Langenskiöld stages have been shown to have a higher recurrence rate (4, 43). Ferriter and Shapiro found higher recurrence rates with Langenskiöld stages three and above (14). In our study the median Langenskiöld stage was four for the recurring and non-recurring groups however, the recurring group had a statistically significant higher Langenskiöld stage on average ($p = 0.02382$). The Langenskiöld stage was a statistically significant risk factor for recurrence in the total group when divided into less than stage five and more than stage five ($p = 0.0143$).

There were different recurrence rates between the Langenskiöld stages in the total group in this study: 0% recurrence at stage two (zero out of three limbs), 58.8% at stage three (ten out of 17 limbs), 50% at stage four (ten out of 20 limbs) and 87.5% at stage five (14 out of 16 limbs) (Figure 3.1). These differences were found to be statistically significant ($p = 0.002382$). Similarly in the infantile group there was 0% recurrence at stage two (zero out of three limbs), 58.8% at stage three (ten out of 17 limbs), 53.9% at stage four (seven out of 13 limbs) and 83.3% at stage five (five out of six limbs) (Figure 3.2) These differences were not found to be statistically significant ($p = 0.12852$).

Ferriter et al. in their study on infantile Blount's disease (14) found 0% recurrence at stage one (zero out of nine limbs), 50% at stage two (four out of eight limbs), 66.6% at stage three (four out of six limbs), 80% at stage four (four out of five limbs) and 100% at stages five (eight out of eight limbs) and six (one out of one limb) (14).

There were no low grade Langenskiöld stage limbs in the juvenile group. There was a recurrence rate of 42.9% at stage four (three out of seven limbs) and 90% at stage five (nine out of ten limbs) (Figure 3.3).

The juvenile group had higher Langenskiöld scores compared to the infantile group – this may explain the higher rates of recurrence in the juvenile group. Due to the fact that the vast majority of limbs had a Langenskiöld stage of three or above in this study and there was a small group of limbs to compare it to the statistics may not be representative of the risk of higher Langenskiöld stages and recurrence.

4.1.9 FTA as a risk factor for recurrence

There is no literature to support that the pre-operative FTA is a risk factor for recurrence in Blount's disease. In this study the preoperative FTA was found to be higher in the recurring limbs in the total, infantile and juvenile groups but the difference was not statistically significant. The post-operative FTA in the total ($p = 0.00063$), infantile ($p = 0.006631$) and juvenile ($p = 0.026935$) groups was found to be in significantly more valgus in the non-recurring groups.

There was a statistically significant difference between the recurring and non-recurring limbs on follow up FTA, the recurring limbs were in more varus than the non-recurring limbs, this is self-explanatory and to be expected since a varus deformity signifies that recurrence has happened.

4.1.10 Under-correction as a risk factor for recurrence

Overcorrection of the limb with HTO into five to ten degrees of valgus has been recommended to decrease recurrence rates (4, 14). In our institution we follow these recommendations and aim for five to ten degrees of overcorrection for infantile and juvenile Blount's disease.

In this study one limb was still in varus with an FTA of 5.35 degrees of varus after HTO (Figure 3.5 Limb 21) this limb was noted to have recurred. Four limbs were in less than five degrees of valgus. Overcorrection was achieved in 36 limbs with a median FTA of 14.24 degrees valgus and a range of between 5 and 24 degrees, 17 of these had valgus of more than 15 degrees (Figures 3.4 to 3.7). Overcorrection into more than 15 degrees of valgus was found to be statistically significant in the total ($p = 0.0008$), infantile ($p = 0.0143$) and juvenile groups ($p = 0.0357$) A longer follow up study will be necessary to determine if these limbs that were overcorrected into large amount of valgus have a residual valgus deformity at skeletal maturity.

4.2 Recommendations

4.2.1 Recommendations for clinical practice

Based on the results from this study the following recommendations are made:

In children who present with confirmed infantile Blount's disease HTO should be performed before the age of four years as described by Chotigavanichaya et al. (55). Those patients with infantile Blount's disease that present after the age of four should have an HTO as soon as possible to minimise the risk of recurrence. The treating surgeon should endeavour to treat infantile Blount's disease that needs an HTO as soon as reasonably possible. Our data was statistically significant after the age of four years for recurrence. This study strengthens the current literature and we recommend that the HTO in infantile Blount's should be done before the age of four years. Parents should be warned regarding the high recurrence rate of this condition after HTO which often requires revision surgery.

Earlier referral of suspected pathological bowing of which infantile Blount's disease is a potential cause should be done by local clinics and general practitioners. A screening programme may be implemented in the normal road to health visits done for all children.

Overcorrection is recommended to between five- and ten-degrees valgus. Longer follow up is needed to determine if overcorrection of more than 15 degrees leads to residual valgus or needed corrective surgery.

4.2.2 Recommendations for research

Future research on Blount's disease in our institution should have follow up of patients with Blount's disease until skeletal maturity to assess total recurrence and possible residual valgus from overcorrection. An electronic patient database may help with future research.

CHAPTER 5: CONCLUSION

This study investigated the recurrence rate of infantile and juvenile Blount's disease at the Chris Hani Baragwanath Academic Hospital after high tibial osteotomy for the period of 1 January 2009 until 31 August 2018. The study found an overall recurrence rate of 63.3% in limbs with Blount's disease. In the infantile group 59.5% of limbs had recurrence after high tibial osteotomy. In the juvenile group of limbs there was a recurrence rate of 72.2%.

Surgery after the age of four years was identified as a risk factor for recurrence in infantile Blount's disease.

Obesity, early walking, gender and pre-operative anatomical femorotibial angle were not found to be risk factors for recurrence in this study.

The post-operative FTA was significantly higher in the non-recurring group. Overcorrection into more than 15 degrees of valgus was found to significantly decrease the recurrence rate.

The vast majority of limbs in this study had a Langenskiöld stage of three or above which is a known risk factor for recurrence.

Using results from the literature and the findings in this study it is recommended to perform high tibial osteotomy before the age of four years in infantile Blount's disease. Overcorrection into valgus is recommended for infantile and juvenile Blount's disease.

APPENDICES

Appendix A: Examples from datasheet

Table A1: Data collection sheet: demographics, referral age, family history, Langenskiöld score, BMI and walking age

Patient number	Limb number	DOB	Sex	Race	Age Noticed	Referral date	Ref Age in months	Blount's type	Sides affected	Recurrence Limb	Recurrence patient	Langenskiöld score	Family history.	Weight	Height	BMI	Walking (months)
1	1	2005/02/24	F	B	3	2013/01/13	94	Inf	L	1	1	5	0	40	1.3	23.669	11
2	2	2010/04/14	F	B	2	2012/04/18	24	Inf	B	1	1	3	0	13.8	0.894	17.266	12
2	3	2010/04/14	F	B	2	2012/04/18	24	Inf	B	0	1	3	0				
3	4	2007/06/11	F	B	2	2013/05/20	71	Inf	L	1	1	3	0				14
4	5	2008/02/28	F	B	2	2009/09/21	18	Inf	L	1	1	4	0				
5	6	2005/05/27	F	B	1	2012/02/27	81	Inf	B	1	1	3	0	28	1.24	18.21	14

Table A2 : Data collection sheet: Date of surgery, repeat surgery date, age at surgery, follow up and FTA

Patient number	Limb number	Date of surgery	Repeat surgery date	Age at surgery in months	Age at repeat surgery	Initial to Repeat	Last date of follow up	Age at follow up in months	Preop xray date	Pre FTA	Post op xray date	Post op FTA	Follow up x-ray date	Follow up FTA
1	1	2013/05/14	2018/08/17	98	161	63	2018/08/17	161	2013/05/13	41.87	2013/05/17	-4.7	2018/08/18	10,61
2	2	2014/10/02	2018/03/27	53	95	41	2018/03/27	95	2014/05/05	22.08	2014/10/09	-18.22	2018/03/26	13.76
2	3	2014/07/15		51			2018/03/26	95	2014/05/05	22.85	2014/07/17	-23.43	2018/03/26	2.76
3	4	2013/10/22	2015/02/17	76	92	15	2015/02/17	92	2013/04/15	20.64	2013/11/18	-8.98	2015/02/16	22.12
4	5	2012/07/03	2014/10/07	52	79	27	2014/10/07	79	2012/07/01	24.85	2012/09/17	-10.56	2014/10/06	10.67
5	6	2012/06/21	2015/06/09	84	120	35	2015/06/09	120	2012/02/27	9.16	2012/06/22	-15.87	2015/06/08	10.77

Appendix B Post Graduate approval of title

UNIVERSITY OF THE WITWATERSRAND, JOHANNESBURG

Private Bag 3 Wits, 2050
Fax: 027117172119
Tel: 02711 7172076

Reference: Ms Thokozile Nhlapo
E-mail: thokozile.nhlapo@wits.ac.za

11 January 2016
Person No: 1032828
PAG

Dr E Van Greunen
P.O Box 1281
Hartbeespoort
0216
South Africa

Dear Dr Van Greunen

Master of Medicine: Approval of Title

We have pleasure in advising that your proposal entitled *Recurrence of deformity in Blount's disease after surgical correction.* has been approved. Please note that any amendments to this title have to be endorsed by the Faculty's higher degrees committee and formally approved.

Yours sincerely

Mrs Sandra Benn
Faculty Registrar
Faculty of Health Sciences

Appendix C Ethics approval letter

R14/49 Dr Eduard van Greunen

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)

CLEARANCE CERTIFICATE NO. M150128

NAME: (Principal Investigator)	Dr Eduard van Greunen
DEPARTMENT:	Orthopaedic Surgery Chris Hani Baragwanath Academic Hospital Charlotte Maxeke Johannesburg AcademicHospital
PROJECT TITLE:	Recurrence of Deformity in Blount's Disease After Surgical Correction
DATE CONSIDERED:	30/01/2015
DECISION:	Approved unconditionally
CONDITIONS:	
SUPERVISOR:	Greg Firth

APPROVED BY: Professor P Cleaton-Jones, Chairperson, HREC (Medical)

DATE OF APPROVAL: 07/10/2015

This clearance certificate is valid for 5 years from date of approval. Extension may be applied for.

DECLARATION OF INVESTIGATORS

To be completed in duplicate and **ONE COPY** returned to the Secretary in Room 10004, 10th floor, Senate House, University.
I/we fully understand the conditions under which I am/we are authorized to carry out the above-mentioned research and I/we undertake to ensure compliance with these conditions. Should any departure be contemplated, from the research protocol as approved, I/we undertake to resubmit the application to the Committee. **I agree to submit a yearly progress report.**

Principal Investigator Signature

Date 14/10/2015

PLEASE QUOTE THE PROTOCOL NUMBER IN ALL ENQUIRIES

Appendix D Ethics extension letter

UNIVERSITY OF THE WITWATERSRAND, JOHANNESBURG

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)

11 April 2017

Dr Eduard van Greunen
Faculty of Health Sciences
Department of Orthopaedic Surgery
University of the Witwatersrand
Parktown
2190

Sent by email to: Evangreunen@gmail.com

Dear Dr Van Greunen

Re: **Protocol Ref No: M150128**
Protocol Title: Recurrence of Deformity in Blount's Disease after Surgical Correction
Principal Investigator: Dr Eduard van Greunen
Protocol Amendment: Request to extend study period from 31 December 2014 to 31 December 2016

This letter serves to confirm that the Chairman of the Human Research Ethics Committee (Medical) has approved the protocol amendment for the abovementioned protocol, as detailed in your letter dated 07 April 2017.

The following documents were received:

- Summary Letter dated 07 April 2017.
- HREC (Medical) Clearance Certificate dated 07 October 2015.
- Study Proposal.
- HREC (Medical) Application Form.

Thank you for keeping us informed and updated.

UNIVERSITY OF THE WITWATERSRAND
OFFICE OF THE DEPUTY VICE CHANCELLOR RESEARCH
11 APR 2017
RESEARCH OFFICE HREC (Medical)
JOHANNESBURG

Yours Sincerely,

Mr Lebohang Moeng
Administrative Assistant
Human Research Ethics Committee (Medical)

Research Office Secretariat: Faculty of Health Sciences, Phillip Tobias Building, 3rd Floor, Office 302, Corner York Road and 29 Princess of Wales Terrace, Parktown, 2193 Private Bag 3, Wits 2050 I T+27 (0)11-717-1234/2656/2700/1252 E: Lebo.Moeng@wits.ac.za I Office E HREC-Medical.ResearchOffice@wits.ac.za IWebsite: www.wits.ac.za/research/about-our-research/ethics-and-research-integrity/

Appendix E Ethics extension letter two

UNIVERSITY OF THE WITWATERSRAND, JOHANNESBURG

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)

19 September 2018

Dr E van Gruenen
School of Clinical Medicine
Department of Orthopaedic Surgery
Chris Hani Baragwanath Academic Hospital

Sent by email to: Evangruenen@gmail.com

Re: **Protocol Ref No:** M150128
Protocol Title: *Recurrence of deformity in Blount's Disease after surgical correction*
Principal Investigator: Dr G van Gruenen

Thank you for your undated letter, received here on 10 September 2018.

I confirm that we have no objection to your proposal to extend the study period to 31 August 2018, provided the CoE of CHBAH has no objection.

Thank you for keeping us informed and updated.

Yours Sincerely

.....

Mr I Burns
For the Human Research Ethics Committee (Medical)

Research Office Secretariat:

Physical address: Phillip Tobias Building, 3rd Floor, Office 302, Corner York Road and Princess of Wales Terrace, Parktown, Johannesburg 2193.
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Website:http://www.wits.ac.za/research/about-our-research/ethics-and-research-integrity/

Appendix F CEO approval letter

GAUTENG PROVINCE
HEALTH
REPUBLIC OF SOUTH AFRICA

CHRIS HANI BARAGWANATH ACADEMIC HOSPITAL

PA TO THE CEO
Enquiries: Ms. Thabile Ndlovu
Tel : (011) 933- 9145
Fax: (011) 938-1005
Email: Thabile.Ndlovu2@gauteng.gov.za

To : **Dr. E van Greunen**
M150128

From : **Dr. Sandile Mfenyana**
CEO: CHBA hospital

Date : **21 September 2015**

Re : **Application for Permission to conduct a study in Recurrence of Deformity in Blount's disease after surgical correction**

Your application for permission to conduct study in Recurrence of Deformity in Blount's Disease after Surgical Correction at Chris Hani Baragwanath Academic Hospital has been approved by the CEO: Dr. Sandile Mfenyana.

Hoping that the institution (CHBAH) will meet the requirements of the study concerned.

Wishing you well in your future endeavors

Regards,

Dr.SCB Mfenyana
CEO : CHBAH

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