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Title

Risk factors for failure of temporary hemiepiphysiodesis in Blount disease

Permalink

<https://escholarship.org/uc/item/8cn1d8ts>

Journal

Journal of Pediatric Orthopaedics B, Publish Ahead of Print(&NA;)

ISSN

1060-152X

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Publication Date

2020

DOI

10.1097/bpb.0000000000000603

Peer reviewed

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inadequate correction and unpredictable rebound growth following implant removal, resulting in loss of correction [7,8,12]. Westberry and colleagues [4,6–8] have attempted to identify risk factors associated with failure following hemiepiphyodesis in late-onset Blount disease.

Notwithstanding the amount of heterogeneous data that has amassed over time, currently, there is limited information comparing the role of temporary hemiepiphyodesis using different implants in the treatment of Blount disease. We performed a systematic review to evaluate the clinical outcome of temporary hemiepiphyodesis in children and adolescents with Blount disease and sought to identify the association of factors such as age at hemiepiphyodesis and the type of implant with clinical outcome, including need for additional unplanned surgery.

Patients and methods

Protocol

A systematic review of the treatment of Blount disease using hemiepiphyodesis with physeal implants (extra-periosteal staples or plates) and clinical outcome, including treatment failures, was performed using PRISMA (Preferred Reporting Items for Systematic reviews and Meta-Analysis) guidelines [20]. The search methodology is displayed in Fig. 1.

Eligibility criteria, information sources, and search results

Using Ovid, PubMed, Scopus, Web of Science, and CINAHL, we searched for articles using patients with Blount Disease from 1965 until 18 August 2016. Search terms included ‘(Blount OR Blount’s OR tibia vara OR genu varum OR bow legged) AND (guided growth OR hemiepiphyodesis OR osteotomy OR staples OR Phemister)’. The results were limited to studies available in the English language. Articles included in the search were either published or electronically published, ahead of print.

Study selection criteria

All abstracts were compiled into a master list and reviewed. Full-text articles related to temporary hemiepiphyodesis in Blount disease were then scrutinized for detailed patient demographic information, type of implant used, and clinical outcome. Only those articles reporting itemized individual patient data were included (Supplementary Appendix 1, Supplemental digital content 1, <http://links.lww.com/JPOB/A31>). Each article had to have listed at least two of the variables to be included in the analysis (i.e. either age at index surgery and ‘outcome’ OR infantile/adolescent Blount and ‘outcome’). For the purpose of our study, ‘outcome’ was categorized on the basis of whether the mechanical axis of the operated lower extremity was corrected to the planned alignment following the index operation. We identified cases of ‘unplanned surgery’, when implant failure (screw

break or staple back out) requiring revision or surgical site infection was reported, or an osteotomy was performed for undercorrection/overcorrection. Furthermore, we did separately extract and report available clinical information in patients who required a subsequent osteotomy.

Relevant papers that did not report individual patient data items were excluded from detailed data analysis, but saved for our review of the literature (Supplementary Appendix 2, Supplemental digital content 2, <http://links.lww.com/JPOB/A32>).

Data collection and data items

One individual (B.F.) reviewed all abstracts, read through the relevant articles including the bibliography and collected pertinent information on an electronic data collection sheet. Data items collected included the patient’s chronological age at index surgery, sex, weight/BMI, type of implant used (staples vs. plate), and whether the patient needed an ‘unplanned’ surgery on the same extremity or any other complications related to the hardware. We were unable to use the patient’s sex, weight, and BMI for analysis as there was insufficient data available. All queries were reviewed with the senior author (S.S.) and final decision regarding inclusion of data was made by consensus.

Risk of bias in individual studies

Some of the identified articles reported parameters and outcome measures that did not include demographic information that we sought, and thus we only extracted data from articles that reported individual itemized patient data that we had identified *a priori*.

Summary measures and synthesis of results

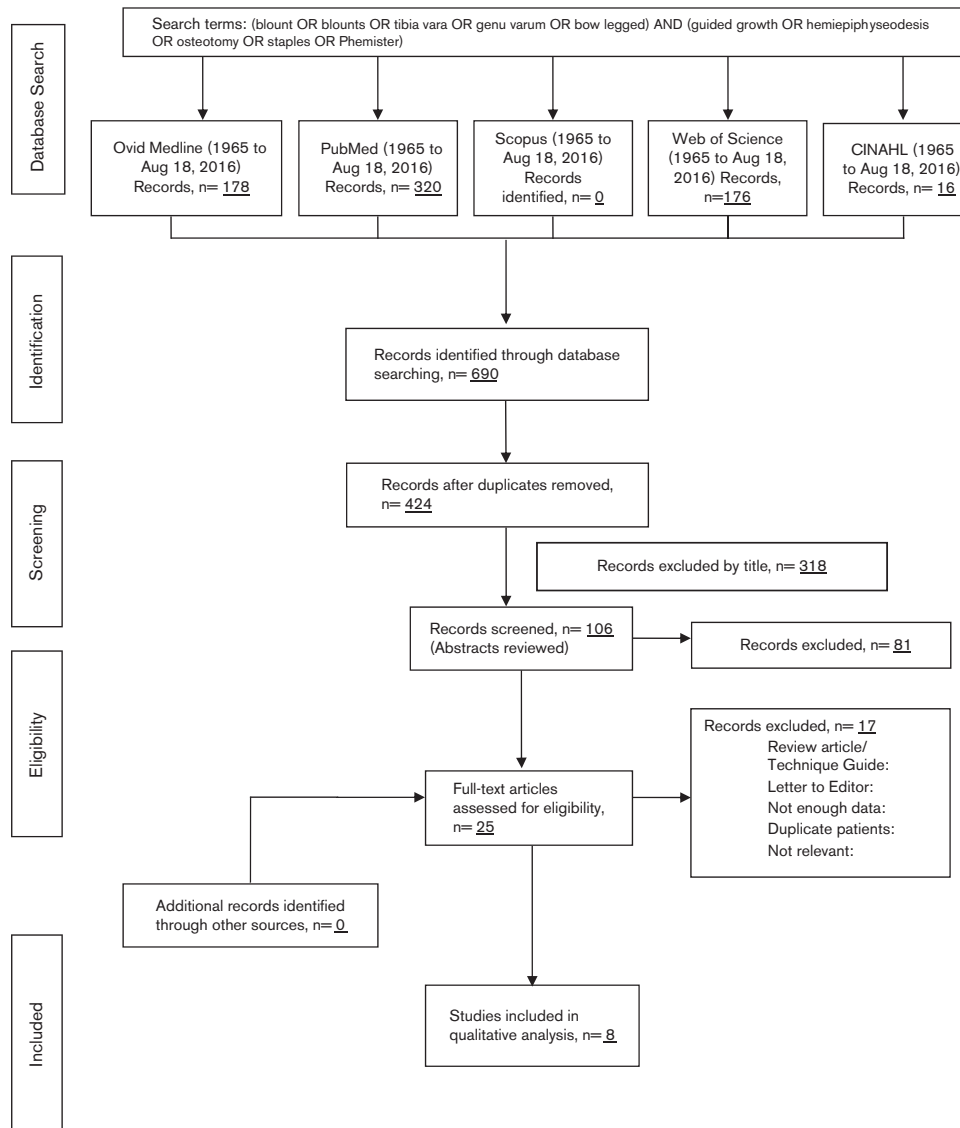
Demographic data was summarized for the entire group as well as subgroups on the basis of the implant used (staples vs. extra-periosteal plates). In our attempt to perform subgroup analysis of heterogeneous data among various studies, we used terms such as ‘expected outcome’, when the lower extremity mechanical axis was reported to be corrected to neutral alignment (or anticipated alignment, if this was mentioned in the article) following the index operation of hemiepiphyodesis, and identified cases of ‘unplanned surgery’, when implant failure (screw break or staple back out) requiring revision, infection, osteotomy, or an epiphyodesis for residual leg length discrepancy was reported.

The prevalence of clinical outcome, that is, either ‘unplanned surgery’ or ‘expected outcome’ was compared on the basis of the age at hemiepiphyodesis and implant design (staples vs. plates).

Statistical analysis

Statistical analysis was performed using SAS, version 9.3 software (SAS Institute Inc., Cary, North Carolina, USA).

Fig. 1



Detail databases searching and adapted PRISMA (Preferred Reporting Items for Systematic reviews and Meta-Analysis) flow diagram for study inclusion.

The mean differences for continuous variables was measured using unpaired *t*-test. Assessment the relationship between two categorical variables, age at surgery, and implants used, was performed using χ^2 -test. Differences were considered significant at *P* value less than 0.05.

Results

Eight papers met our inclusion criteria [9–12,14,16,17,19]. Sufficient information was available for 53 patients (Table 1) with 63 bone segments (54 tibia only, eight femur+tibia, one femur only). Chronological age at index surgery ranged from 1.8 to 14.7 years (mean: 8.8 years). Twenty-seven (43%) of the operated bone segments were in children younger than 9 years old (mean: 5 years) and 36 (57%) bone segments were in older children (≥ 9 years

Table 1 Demographics of itemized patient data

Number of limbs=63 (<i>n</i> =53 patients)	<i>n</i> (%)
Early-onset Blount	18 (29)
Adolescent Blount	9 (14)
Unspecified	36 (57)
Laterality of surgery	
Left	17 (27)
Right	10 (16)
Unspecified	36 (57)
Location of surgery	
Tibia only	54 (86)
Femur + tibia	8 (13)
Femur only	1 (1.5)

old; mean 12 years) (Table 2). To avoid multiple subgroups with overlapping disease types, we used 9 years as the cutoff for age, on the basis of the oldest age for

Table 2 Comparison of outcomes on the basis of age greater than or less than 9^a years old (all patients)

	<9 years old at surgery ^a	≥9 years old at surgery	<i>P</i> value
Number of limbs	27	36	
Age at surgery [mean (range)] (years)	5 (1.8–8.7)	12 (9–14.7)	<0.0001
Mean length follow-up (months)	19.5	19	0.89
Expected outcome [<i>n</i> (%)]	17/27 (63)	15/36 (42)	0.09
Unexpected outcome [<i>n</i> (%)]	10/27 (37)	21/36 (58)	

^aAge 9 years based on oldest age of patients in a paper studying only infantile Blount.

Table 3 Comparison of outcomes on the basis of implant used at index surgery (all patients)

	Staples	Plates	<i>P</i> value
Number of limbs	16	47	
Age at surgery [mean (range)] (years)	7.2 (3.7–13.0)	9.3 (1.8–14.7)	0.05
Mean length follow-up (months)	24	18	0.09
Expected outcome [<i>n</i> (%)]	7/16 (44)	25/47 (53)	0.51
Unexpected outcome [<i>n</i> (%)]	9/16 (56)	22/47 (47)	

hemiepiphyodesis reported in a child with confirmed infantile Blount disease in our study group. Sixteen (25%) bone segments underwent staples as index procedure (mean: 7.2 years) and 47 (77%) segments underwent plating (mean: 9.3 years) (Table 3).

Fifteen (24%) of the 63 limbs required a subsequent osteotomy. Ten of 15 limbs had an osteotomy for overcorrection/undercorrection (one limb overcorrected/nine limbs undercorrected). The average age of index hemiepiphyodesis in this subgroup was 12 years old (range: 9–14 years). Seven limbs previously had plate hemiepiphyodesis and three had staples. Five of the remaining 15 limbs had an osteotomy due to an untoward event such as implant loosening, screw breakage or, following a postoperative infection. The average age of index hemiepiphyodesis in this subgroup of five limbs was 7.4 years old (range: 3.7–13.5 years). Two limbs previously had a plate hemiepiphyodesis and three had staples.

Subgroup analysis on the basis of age at surgery

In the less than 9-year-old group (*N*=27), mean age was 5 years, with an average 19.5-month follow-up (Table 2). Ten (37%) affected bone segments underwent stapling and 17 (63%) underwent plating. Seventeen of 27 (63%) limbs had an 'expected outcome' after index hemiepiphyodesis and 15/27 (42%) had unplanned surgery. Four (15%) affected segments had hardware failure, three (11%) had infection, and two (7%) had limb length discrepancy requiring epiphyodesis.

In the 9-year-old or older group (*N*=36), mean age at index surgery was 12 years with an average 19-month follow-up (Table 2). Fifteen (42%) of 36 had an 'expected

outcome' after index hemiepiphyodesis, whereas 21/36 (58%) had unplanned surgery. Six (17%) affected segments underwent stapling and 30 (83%) underwent plating. Nine (25%) bone segments had hardware failure and 11 (31%) needed a subsequent osteotomy for undercorrection.

As expected, there was a significant difference ($P<0.0001$) in the mean age at index surgery between the less than 9-year-old group and the greater than 9-year-old group. However, there was no difference in length of follow-up ($P=0.89$). Although not statistically significant, there was a trend for the older age at index surgery group having a greater percentage of unplanned surgery (37% <9-year-old vs 58% ≥9-year-old) ($P=0.09$) (Table 2).

Subgroup analysis on the basis of type of implant

In the staple group (*N*=16), the mean age at index surgery was 7.2 years, with an average 24-month follow-up. Seven (44%) of 16 patients had an 'expected outcome' after index hemiepiphyodesis. Unplanned surgery was performed in 9/16 (66%) bone segments (Table 3, Fig. 2).

In the plate group (*N*=47), mean age at index surgery was 9.3 years, with an average 18-month follow-up (Table 3). Twenty-five (53%) of 47 patients had an 'expected outcome' after index hemiepiphyodesis. Unplanned surgery was performed in 22/47 (47%) bone segments (Table 3, Fig. 3).

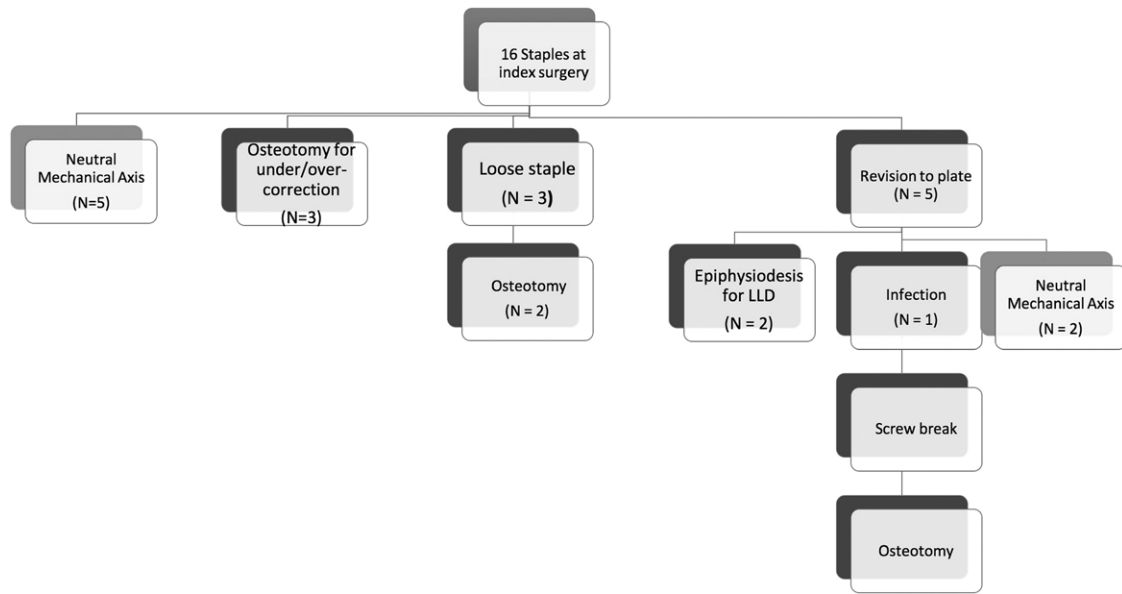
Although there was a difference in the average age at surgery between staple (7.2 years) and plate groups (9.3 years) ($P=0.05$), there was no statistically significant difference in length of follow-up ($P=0.09$) or outcome ($P=0.51$) between the treatment groups. The mean age at index surgery of patients with 'expected outcomes' was 8.2 years old (range: 1.8–14.7 years) versus 9.4 years old (range: 2.9–14.1 years) for those needing subsequent unplanned surgery ($P=0.20$).

Subgroup analysis of proximal tibia hemiepiphyodesis

To further minimize bias, we performed a subgroup analysis on patients who underwent an isolated hemiepiphyodesis of the proximal tibia, excluding any patients who had a concomitant ipsilateral distal femoral physal procedure. This excluded 18 of our 63 limbs (Tables 4 and 5). Fifteen tibias underwent staple hemiepiphyodesis (mean age: 6.8 years), and 39 underwent extraperiosteal plating (mean age: 9.9 years) (Table 4). On the basis of available data, patients undergoing plate hemiepiphyodesis were older (9.9 vs. 6.8 years; $P=0.0003$). Although a higher percentage of plate hemiepiphyodesis limbs achieved 'expected outcome' (47% staples vs 56% plates), this was not statistically significant ($P=0.52$).

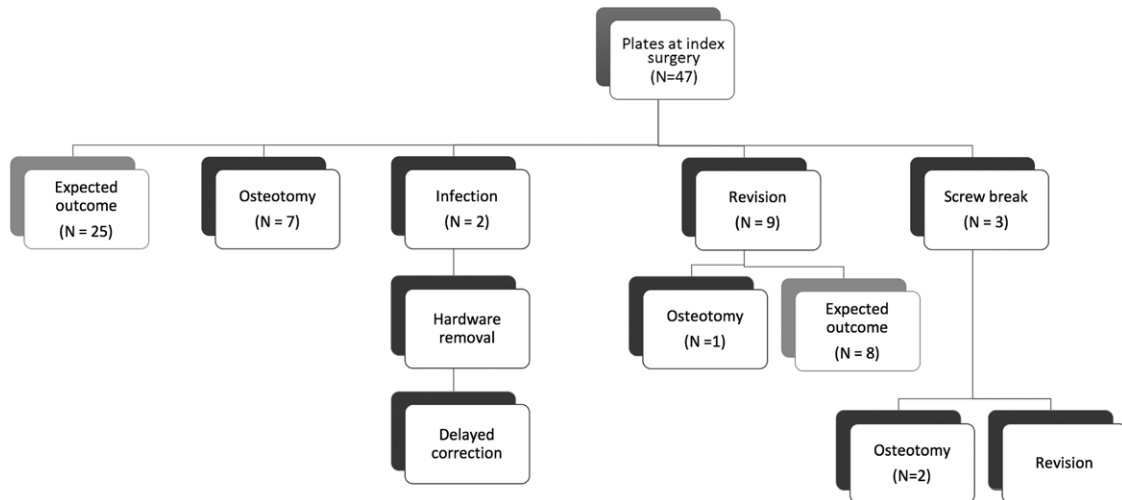
Analysis of patients having only proximal tibial hemiepiphyodesis, on the basis of age at index surgery, the

Fig. 2



Flowchart of outcomes after staple hemiepiphysiodesis at index surgery. LLD, leg length discrepancy.

Fig. 3



Flowchart of outcomes after plate hemiepiphysiodesis index surgery.

Table 4 Comparison of outcomes on the basis of implant used at index surgery (tibia only)

	Staples	Plates	P value
Number of limbs	15	39	
Age at surgery [mean (range)] (years)	6.8 (3.7–9.3)	9.9 (1.8–14.7)	0.0003
Mean length follow-up (months)	24	19	0.19
Expected outcome [n (%)]	7/15 (47)	22/39 (56)	0.52
Unexpected outcome [n (%)]	8/15 (53)	17/39 (44)	

‘expected outcome’ was similar in the two groups (67% <9 years vs. 45% >9 years; $P=0.13$) (Table 5).

Subgroup analysis on the basis of diagnosis

In our data analysis, only 45/63 (71%) limbs had a specific diagnosis (infant vs. adolescent Blount disease) noted by the original authors. Initially, we performed our data analysis with all 63 limbs on the basis of age and type of implant used. To further limit any potential bias, we then

Table 5 Comparison of outcomes on the basis of age greater than or less than 9^a years old (tibia only)

	<9 years old at surgery ^a	≥9 years old at surgery	P value
Number of limbs	21	33	
Age at surgery [mean (range)] (years)	5 (1.8–8.7)	12 (9–14.7)	<0.0001
Mean length follow-up (months)	22.3	18.2	0.24
Expected outcome [n (%)]	14/21 (67)	15/33 (45)	0.13
Unexpected outcome [n (%)]	7/21 (33)	18/33 (55)	

^aAge 9 years based on oldest age of patients in a paper studying only infantile Blount.

Table 6 Comparison of outcomes on the basis of patient diagnosis

	Infantile	Adolescent	P value
Number of limbs	18	27	
Age at surgery [mean (range)] (years)	5 (2.9–8.7)	12 (7–14.1)	<0.0001
Mean length follow-up (months)	18.6	19.0	0.93
Expected outcome [n (%)]	11/18 (61)	10/27 (37)	0.11
Unexpected outcome [n (%)]	7/18 (39)	17/27 (63)	

excluded 18 limbs with unspecified diagnosis (Table 6). 63% adolescent Blount patients underwent unplanned surgery versus 39% infantile Blount patients underwent unplanned surgery ($P=0.11$). These results remained similar to our original data set with older patients having a greater percentage of unplanned surgeries; however, the data did not reach statistical significance.

Discussion

On the basis of our analysis of the available literature pertaining to hemiepiphyseodesis for Blount disease, we can make the following observations: (i) there is no consistency among published studies regarding reporting of specific patient information, including age of onset, radiographic deformity analysis, and outcome measures, leaving us with a heterogeneous data set. (ii) There is a relatively high rate (49%) of reoperation in children with Blount disease following temporary hemiepiphyseodesis.

Why is there a high rate of reoperation among Blount patients?

The observation in our systematic review of a 49% reoperation rate following temporary hemiepiphyseodesis for children and adolescents with Blount disease was unexpectedly high. Although this may be an overestimated number due to selection and historical bias (see the Study limitations section), the higher failure rate following temporary hemiepiphyseodesis among patients with abnormal physis is well known [9,10,12,16–18]. Despite this, temporary hemiepiphyseodesis or guided growth is often the first-line treatment for angular deformity in Blount patients, with the recognition that some limbs may fail to fully correct [6,10,21]. Hemiepiphyseodesis

being a ‘smaller surgery’ allows osteotomy to be reserved as a back-up option, especially when used to correct multiplanar deformities with limb shortening [21].

A recent multinational study of guided growth (among patients with several etiologies, including Blount) found an infection rate of 1.48% among 967 physes [22], which is similar to and sometimes higher than the infection rate in the periprosthetic joint infection in a primary total hip or knee literature [23]. In our systematic review, 3/63 (5%) limbs had reoperation because of infection.

Lastly, temporary hemiepiphyseodesis in Blount patients has yielded unpredictable results because there is an abnormal proximal tibial physis. Although there are several failed attempts at temporary hemiepiphyseodesis in Blount patients in the literature, there are also some patients who achieved full corrections, independent of age and weight [18,21]. It is known that Blount patients tend to be more advanced in skeletal age compared with chronological age using bone age on a hand radiography [24], and predicting growth remaining can be more difficult. As stated in our ‘Study limitations’ section, we could not assess physal health on the basis of the severity of Blount disease for our systematic review.

Is older age a risk for temporary hemiepiphyseodesis failure?

On the basis of our analysis of a limited number of patients, whereas we did not achieve statistical significance, older age at hemiepiphyseodesis may be a risk for failure to achieve adequate correction by skeletal maturity. This may be related to the limited growth remaining in adolescents along with advanced skeletal age that has been reported in such obese children [24]. Further studies with larger number of patients would be required to fully ascertain the influence of age at surgery on subsequent angular correction in patients with Blount disease. A recent study reported on the accuracy of the ‘Multiplier Method’ in predicting temporary correction of angular deformity in general, and found it to under predict the correction by 2 months [25]. Another study found the ‘Multiplier Method’ to be unreliable in predicting coronal plane angular deformity correction [26].

Raab *et al.* [14] retrospectively evaluated 51 limbs with angular lower limb deformity secondary to various etiologies treated with staple hemiepiphyseodesis and found a higher rate of staple loosening in girls less than 9 years old and boys less than 11 years old. Three out of five Blount patients required subsequent surgery [14] (Supplementary Appendix 1, Supplemental digital content 1, <http://links.lww.com/JPOB/A31>). This may also be related to the use of smooth staples in young children with partially unossified epiphysis. Westberry *et al.* [4] concluded that age at staple or plate hemiepiphyseodesis was not statistically significant for subsequent surgery in Blount patients. Although they reported three limbs with

broken staples, the authors did not correlate clinical outcome of implant failure with age at surgery. Park *et al.* [5] retrospectively examined 33 limbs in adolescent Blount patients who underwent staple hemiepiphysiodesis and found significantly greater correction in patients younger than 10 years old. They reported a few instances of staple breakage or migration but did not correlate hardware failure with age at surgery. Bushnell *et al.* [6] correlated younger age at surgery with greater correction in adolescent Blount patients using staples. McIntosh *et al.* [7] correlated older age at permanent hemiepiphysiodesis as a risk factor for clinical failure among adolescents with Blount disease. Schroerlucke *et al.* [19] did not find a correlation between age at surgery and screw breakage following plate hemiepiphysiodesis and noted implant failure at the metaphyseal screw-bone junction in adolescents with Blount disease. Funk *et al.* [8] found that among patients with adolescent Blount disease, younger age at surgery correlated with implant failure including both staples and various types of extraperiosteal nonlocking plates.

Is implant type a risk for temporary hemiepiphysiodesis failure?

Our systematic review did not find a difference between staples and extraperiosteal plates in treatment outcome in Blount patients. Multiple studies have demonstrated similar outcomes between staples and plates [8,16,27]. In fact, one author advocated using staples rather than plates for correcting angular deformities in children because of decreased cost and similar outcomes [8]. However, it is well known that staples in younger patients can loosen and back out [9,14,27]. As a result, there is a noted trend towards using extraperiosteal plates in younger patients [27].

Because guided growth using extraperiosteal implants for Blount disease is a relatively new procedure, it was only in the past decade that the literature began suggesting stronger constructs for guided growth in obese patients with either solid screws or bigger screws or stronger materials [8,9,12,19]. In the more recently published multinational study screw breakage was only found in 3/967 (0.55%) physes; however two of the three broken screws were in patients with Blount disease [22]. Certain manufacturers offer stainless steel solid 4.5 mm which may minimize the risk for screw breakage in obese patients.

Although not fully covered in the scope of this analysis, BMI has been mentioned as a potential underlying reason for hardware failure in multiple studies examining failure of hemiepiphysiodesis in children with Blount disease [4,6–8]. However, given the study design and lack of pertinent demographic information in these case series, BMI and magnitude of deformity were not assessed as a separate risk factor in this analysis.

Study limitations

There are several limitations in our study. In this systematic review, we were limited by the lack of individual patient data in several published reports. Many studies lacked itemized data that could be extracted for analysis, for example, 18/63 (29%) of patients were not specified as adolescent or infantile Blount. As a result, we created age groups of greater or less than 9 years old, because in one study that we used [9], the oldest confirmed infantile Blount patient was 9 years old at index surgery. Although this was not a direct comparison of outcomes in adolescent Blount versus infantile Blount, we were able to assess trends in outcomes on the basis of age at index surgery. In addition, there is no consistency in the published studies regarding reporting of demographic data, for instance, some authors reported the patient's BMI, whereas others reported body weight without any information on the individual's height. Similarly, radiographic parameters for assessing limb alignment were reported by some as mechanical tibiofemoral angle [6,10,11,14], whereas others used proximal tibia articular angle [4,19], medial proximal tibial angle [5,7,8,12,17,18] lateral distal femoral angle [5,7,8,17,18], and mechanical axis zone [7,8]. One prognostic factor that we could not assess was physal health on the basis of the severity of Blount disease. We hypothesize that early-onset Blount patients with more advanced disease (Langenskiöld stages IV–VI) are more likely to fail guided growth treatment. The Langenskiöld classification for infantile Blount disease was not reported in most studies, whereas others have questioned its reliability [18]. We could not determine the amount of growth remaining at time of index hemiepiphysiodesis. Although 10 patients were 13 years old or older at time of index surgery, sex was not provided for all patients to estimate growth potential. There is some selection bias in this study. Some of the data used in our analysis focused on failure of implants used for temporary hemiepiphysiodesis. As a result, our reported rate of untoward events is likely an overestimation of the true prevalence of untoward events in this population. We also noticed a historical bias as many papers published after 2007 reported on plate hemiepiphysiodesis rather than staple hemiepiphysiodesis.

Conclusion

We performed a systematic review of temporary hemiepiphysiodesis in Blount disease to assess if age at index surgery or type of implant used affected the outcomes of surgery (unplanned surgery). The published literature contains extremely heterogeneous data that negatively impacted the number of patients that could be included in the current analysis. Our review of guided growth literature revealed that among children and adolescents with Blount disease, overall 49% of patients undergoing temporary hemiepiphysiodesis had unplanned surgeries. On the basis of our assessment of a limited number of

patients, neither age at index surgery nor type of implant used correlated with unplanned subsequent surgeries; however there were a greater percentage of unplanned surgeries in older patients with hemiepiphyseodesis.

Given the study limitations noted above, we are unable to make firm recommendations regarding the ideal age and implant of choice when contemplating temporary hemiepiphyseodesis in children with Blount disease. Although it makes intuitive sense that a threaded screw will have better purchase than a smooth pronged staple in an epiphysis that is mostly cartilaginous, as is seen in children with early-onset Blount disease, we were unable to confirm this difference on the basis of the available literature. We also noted extreme variability in the parameters used to report clinical and radiographic data in children with Blount disease. These pitfalls and potential solutions were briefly discussed in a recent study [28].

In light of the increasing popularity of guided growth implants, more robust prospective studies with consistent reporting criteria and use of itemized patient data are needed to help establish evidence-based clinical practice guidelines for the treatment of Blount disease.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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