

Does idiopathic congenital talipes equinovarus have an impact on attainment of developmental milestones? A multicentre international study

K. Hughes¹
Y. Gelfer^{1,2}
M. Cokljat¹
S. Wientroub³
A. Yavor³
Y. Hemo³
M. Dunkley^{4,5}
D. M. Eastwood⁴

Abstract

Purpose The Ponseti method is a well-established approach to treating clubfoot. Potentially, both the underlying pathology and adherence to post-correction bracing can affect lower limb function and age of independent standing and walking. This cohort study investigates the age at which infants with idiopathic clubfoot treated using the Ponseti method achieved three selected developmental milestones and whether or not this correlated with treatment compliance.

Methods A prospectively collected database from four centres was visited. Inclusion criteria were patients with idiopathic clubfoot with no comorbidities or prior treatment. Age at attainment of independent standing, walking, nocturnal continence was compared across three groups: I) congenital talipes equinovarus (CTEV) children compliant with treatment; II) CTEV children non-compliant with treatment; and III) typically-developed siblings. Minimum follow-up was five years.

Results In all, 130 patients (198 feet) fitted the inclusion criteria: 43:87 (F:M). Standing was achieved by a mean 12.0 months in group I (SD 2.50); 12.0 months (SD 2.0) in II and ten months (SD 3.0) in III. Walking was achieved by a mean 15 months (SD 4.0) in group I, 14 months (SD 1.75) in II and 12 months (SD 3) in III, respectively. Both the compliant and non-compliant CTEV children were significantly slower at

achieving standing and walking compared to sibling controls ($p < 0.0001$). There was no significant difference between age of nocturnal continence between the three groups.

Conclusion Infants with idiopathic clubfoot treated according to the Ponseti method achieve independent standing and walking approximately two months later than their typically-developed siblings. The delay is not related to the use of the foot abduction brace.

Level of evidence: III

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Introduction

Congenital talipes equinovarus (CTEV) is a common congenital foot abnormality, with an incidence of approximately one in 1000 live births. The Ponseti method has become the benchmark approach to treating idiopathic clubfoot.¹ This method has reduced the need for radical surgery^{2,3} due to a reported primary correction rate of 95% or more.²

However, non-compliance with foot abduction brace (FAB) use post-casting has been recognized as a significant risk factor for the recurrence of clubfoot after primary correction.⁴ FAB use poses challenges along the treatment path with variable compliance rates. Factors such as the level of education of parents, belief in necessity of the brace and the level of information given and understanding gained about the treatment course have been shown to have a direct effect on compliance.^{1,5} In a recent study, non-compliance with the FAB was associated with parents' perception of its effect in restricting movement.⁶ Some parents fear a negative influence of the brace on the motor development of their child.¹ Others have expressed concern regarding effect on nocturnal enuresis due to difficulty in the child's ability to get out of bed independently at night. Nocturnal enuresis is defined as bedwetting more than twice per week, past the age of five years.⁷

¹ St. George's Hospital, London, UK

² St. George's University of London, London, UK

³ Dana Children's Hospital, Tel-Aviv, Israel

⁴ Great Ormond Street Hospital and University College London, UK

⁵ Royal Surrey County Hospital, Guildford, UK

Both the underlying pathology and FAB can potentially have an effect on lower limb function and age of independent standing and walking. Previous studies have found minimal developmental delay in gross motor skills in children with CTEV treated by the Ponseti method.⁸⁻¹⁰ Compared with typically-developed children who usually achieve walking between 11.7 months and 12.8 months,¹¹ children with CTEV show a delay of only one to two months in walking. However, these studies are limited by small sample sizes and a lack of a control group. Developmental milestone achievement has a range of normality that could be affected by genetic, geographic, cultural and behavioural factors.¹²⁻¹⁴ As such, no previous studies have compared milestones in children with clubfoot to their typically-developed siblings that have potentially less bias as controls.

Objectives

This study aims to assess age of attainment of gross motor developmental milestones in children with idiopathic CTEV, compare this with their typically-developed siblings and investigate whether there is any correlation with FAB use. To our knowledge this has not previously been reported in the literature.

Materials and methods

Design and setting

This was a multicentre cohort study. Four prospectively collected databases from four hospitals (St. George's Hospital, London (SGH), Great Ormond Street Hospital, London (GOSH), Royal Surrey County Hospital, Guildford (RSCH) and Dana Children's Hospital, Tel-Aviv (DCH)) were visited. The databases included all children who had been treated according to the Ponseti method for idiopathic CTEV since 2010. The Ponseti protocol used was comparable across all sites. A retrospective cohort study was then performed using typically-developed siblings as controls.

Patients

Consecutive children treated for idiopathic CTEV under our respective Ponseti services during the study period (21 December 2011 to 16 December 2013) were included (Appendix 1). All typically-developed siblings were included as controls. When data for more than one sibling was provided, one was selected randomly. Children with non-idiopathic CTEV and those who were treated primarily elsewhere were excluded. Minimum follow-up was five years.

Main outcome measures

Data extracted included demographics, Pirani score at start of treatment, Achilles tenotomy rate and compliance

with FAB. Data were extracted from the physiotherapy notes where milestones had been reported by parents and verified by the physiotherapist. This included independent standing, walking and nocturnal continence age. Nocturnal continence was defined as five or more dry nights per week. Compliance with FAB was assessed using a combination of parental reporting and the physiotherapist's assessment of the amount of FAB wear. This generated a binary value of either 'yes' or 'no' for compliance. All four units have worked together closely over the last five years and their assessment methods for FAB compliance are similar and repeatable.¹⁵⁻¹⁷ Age at which nocturnal continence was achieved and sibling developmental milestones (standing, walking and nocturnal continence ages) were either collected during the physiotherapy follow-up appointment or from the parents via telephone call.

Statistical analysis

Comparison of developmental milestones between hospital sites was performed using analysis of variance and Tukey's test for multiple comparisons. Analyzed data is presented as mean \pm SD. Pearson's correlation coefficient was calculated to assess correlations between developmental milestones and the Pirani score. Simple linear regressions were performed in order to visualize lines of best fit. Comparison of developmental milestones between CTEV children compliant with FAB, CTEV children non-compliant with FAB and controls was performed using the Kruskal-Wallis test and the pairwise Wilcoxon post hoc test for multiple comparisons. This was due to the distribution of the non-compliant group. Analyzed data was presented as median \pm interquartile range.

Statistical analyses were performed using RStudio 1.1.463 (Boston, Massachusetts) and GraphPad Prism 6 (San Diego, California). Significance was defined as $p < 0.05$.

This study was approved by the St. George's University and Dana Children's Hospital institutional review boards and National Health Service Health Research Authority (REC Reference: 19/HRA/0512).

Results

A total of 130 children with 198 clubfeet were included. In all, 130 sibling controls were included. The flowchart of the patient selection is presented in the supplementary material. Table 1 presents the demographics and treatment data.

Figure 1 presents the age of achieving the selected developmental milestones in CTEV patients compliant with the FAB (Group I), non-compliant CTEV patients (Group II) and the typically-developed sibling controls (Group III).

Table 1 Study demographics

| Category | Characteristic | n (%/sd) |
|---|-------------------------------------|--------------|
| Hospital site | St. George's Hospital (SGH) | 41 |
| | Great Ormond Street Hospital (GOSH) | 21 |
| | Royal Surrey County Hospital (RSH) | 16 |
| | Dana Children's Hospital (DCH) | 52 |
| | Total number | 130 |
| Sex | Male | 87 (66.9) |
| | Female | 43 (33.1) |
| Affected foot | Left only | 29 (22.3) |
| | Right only | 33 (25.4) |
| | Bilateral | 68 (52.3) |
| | Total number of feet | 198 |
| Mean Pirani score at start of treatment | SGH | 4.8 (sd 1.4) |
| | GOSH | 5.1 (sd 1.3) |
| | RSH | 4.7 (sd 1.3) |
| | DCH | 5.1 (sd 1.0) |
| Achilles tenotomy | All sites | 4.9 (sd 1.3) |
| | Yes | 114 (87.7) |
| | No | 15 (11.5) |
| Compliance | Unknown | 1 (0.8) |
| | Yes | 110 (84.6) |
| | No | 17 (13.1) |
| | Unknown | 3 (2.3) |

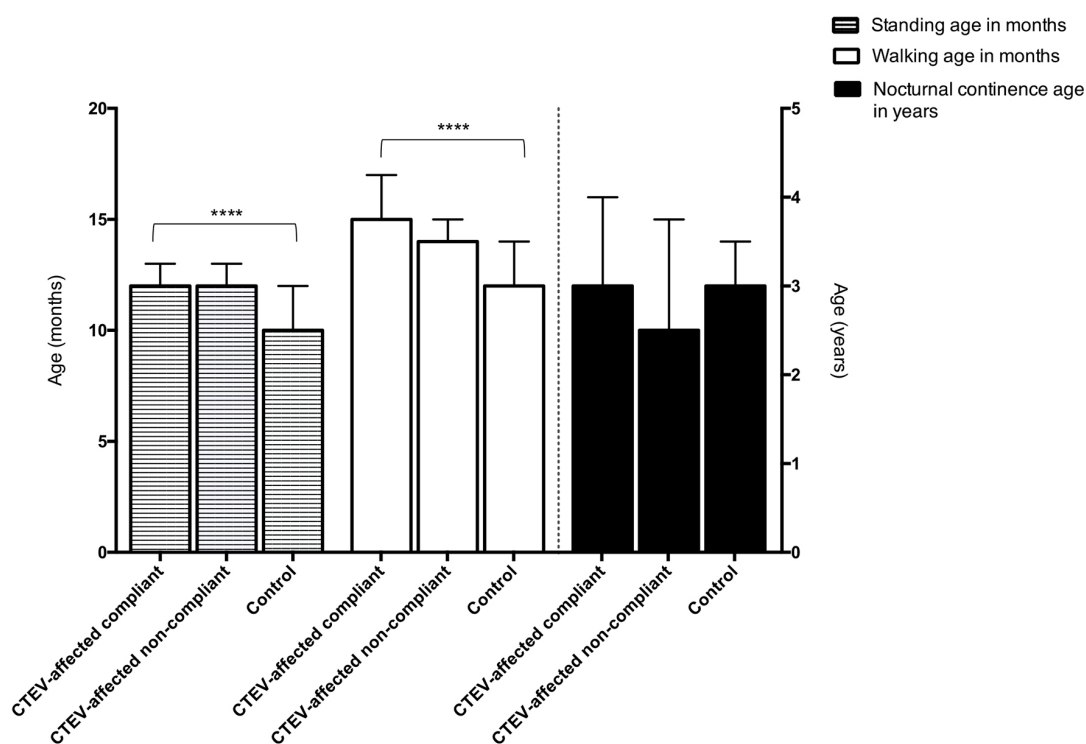


Fig. 1 Standing, walking and nocturnal continence age in congenital talipes equinovarus (CTEV) patients compliant to the foot abduction brace (FAB), CTEV patients non-compliant to the FAB, compared with their typically developed siblings. Bars show median and upper quartile.

Standing

The median age by which standing was achieved for the three groups was 12.0 months (± 2.50); 12.0 months (± 2.0) and ten months (± 3.0), respectively (Fig. 1). Both the compliant and non-compliant CTEV children were significantly slower at achieving standing, compared with sibling controls ($p < 0.0001$).

Walking

The median age at which walking was achieved in the three groups was 15 months (± 4.0), 14 months (± 1.75) and 12 months (± 3), respectively (Fig. 1). Both the compliant and non-compliant CTEV children were significantly slower at achieving walking, compared with sibling controls ($p < 0.0001$). In all, 93% (121) of CTEV children

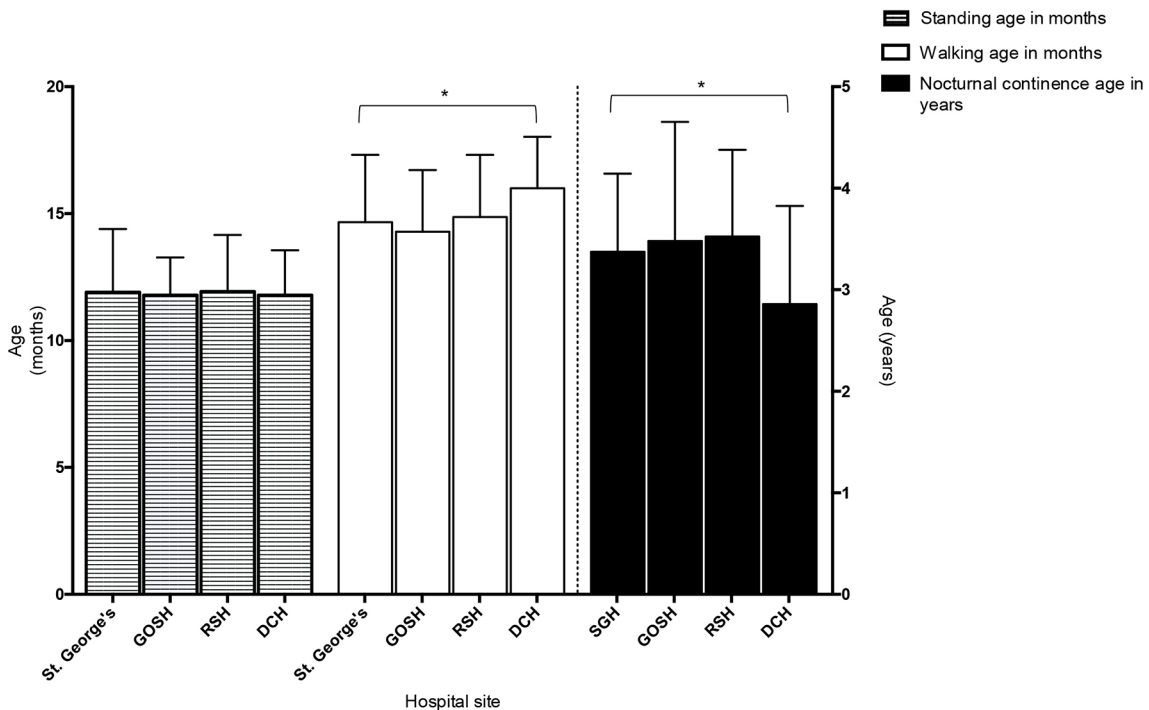


Fig. 2 Standing, walking and nocturnal continence age in congenital talipes equinovarus (CTEV) patients compared across all four hospital sites. Bars show mean and standard deviation (SGH, St. George's Hospital; GOSH, Great Ormond Street Hospital; RSH, Royal Surrey County Hospital; DCH, Dana Children's Hospital).

achieved walking before or at 18 months of age, regardless of compliance.

Nocturnal continence

The median age at which nocturnal continence was achieved in the three groups was 3.0 years (± 1.5), 2.5 years (± 1.75) and 3.0 years (± 1.0), respectively (Fig. 1). These differences were not significant ($p = 0.151$). In all, 95% (124) of children with CTEV achieved nocturnal continence by age five years or earlier.

Comparison across sites

The developmental milestones in the study group were compared across each study site (Fig. 2). There was no significant difference in standing age between the four sites ($p = 0.99$). Children from DCH achieved walking a month later than children from the other three sites ($p = 0.011$). Children from DCH achieved continence at night significantly earlier than children from the other three sites ($p = 0.02$). A total of 17 children (13.1%) were non-compliant with FAB use. Six of these children (4.6%) had been treated at SGH and 11 (8.5%) at DCH. Compliance data was not available for three children (2.3%).

Correlation with Pirani score

Developmental milestones were correlated against Pirani score to assess the effect of CTEV severity on development

(Fig. 3). If two feet were affected, the higher-scoring foot was selected for analysis, as we hypothesize that a more severely affected foot would have a greater impact on development. There was no correlation between the CTEV severity and standing, walking and nocturnal continence ages (Table 2).

Discussion

Idiopathic CTEV is a common congenital foot abnormality with the majority of the patients achieving primary correction using the Ponseti method. However, little is known about its impact on attainment of developmental milestones. No previous studies have compared attainment of developmental milestones in children with CTEV treated with the Ponseti method to typically-developed siblings. In this study, we have found a statistically significant delay in attainment of standing and walking in children with CTEV, regardless of whether they were compliant with the FAB, compared with their typically-developed sibling controls. This age of attainment appears to be delayed also compared with studies of otherwise healthy children, where the mean age range of walking reported is between 11.7 months and 12.8 months.¹¹ The extent of the delay in walking in this study is consistent with previous studies,¹⁰ which found that the mean age of walking achieved was 14.5 months.⁹ However, whilst this difference is statistically significant, it is unlikely to be clinically meaningful

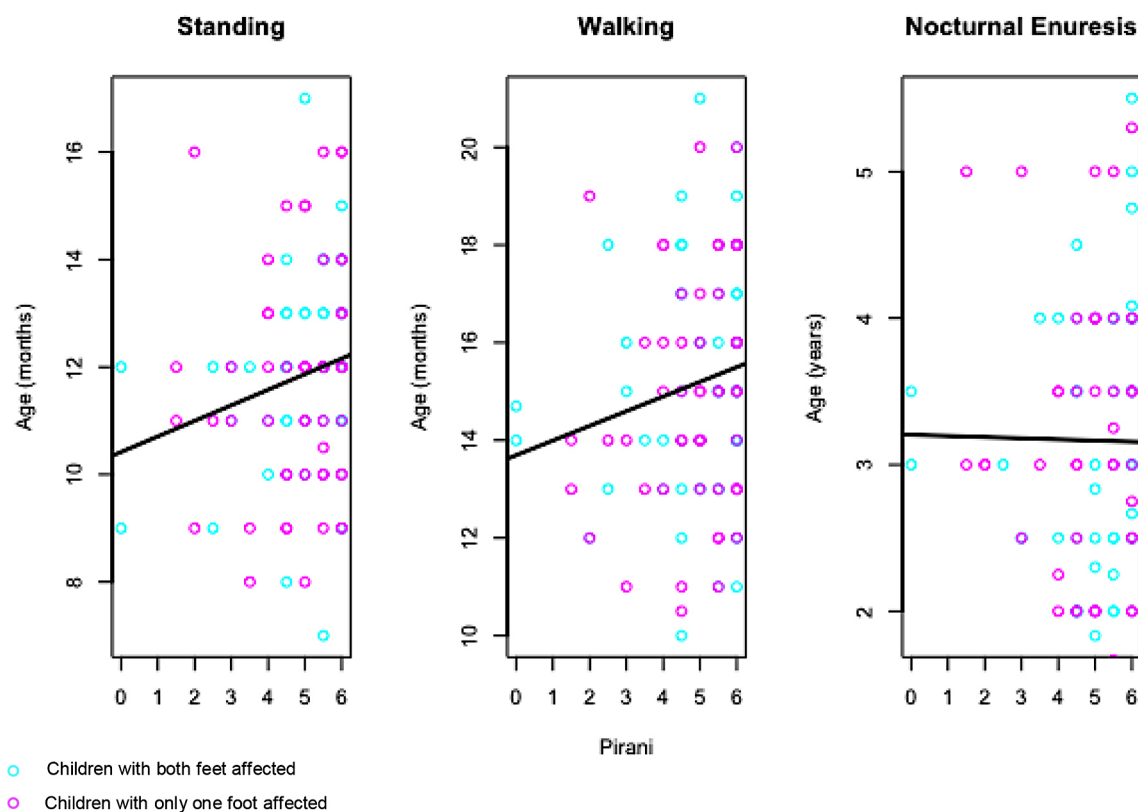


Fig. 3 The association between the severity of congenital talipes equinovarus, as measured by a Pirani score, and standing, walking and nocturnal continence ages. Line depicts best fit. Red circles represent children with only one foot affected; blue circles represent children with both feet affected.

and this delay should not cause a lasting effect on development. The majority of children with CTEV have comparable motor abilities by the time they reach nine years of age and can participate in sports such as dancing and skiing without impairment.³ Moreover, many typically-developed children do not attain standing and walking by the mean age of the population; indeed, the control group within this study achieved walking by a mean age of 13.3 months, which is still later than the theoretical population mean. Arguably, it is better to look at limit ages, which denotes the age by which all children should have achieved a developmental milestone. The commonly accepted limit age for the attainment of walking is 18 months.¹⁸ In all, 93% of the CTEV-affected children in this study achieved walking by or at 18 months. Therefore, whilst affected children may be slightly delayed compared with their non-affected siblings, they will still achieve their motor developmental milestones well within the age limit of the healthy population.⁹

The effect of CTEV and the Ponseti method on nocturnal continence was also studied. Many parents express concerns over whether the FAB increases the risk of nocturnal enuresis by hindering the child from getting out of bed during the night. No previous studies have assessed whether there is a true effect. The difference in age at

which night-time continence was achieved between children compliant with the FAB, non-compliant children and the control group was not significant. Only six children met the criteria for nocturnal enuresis,⁷ suggesting that most children with CTEV, whether or not compliant with the FAB, become continent at night well within the normal distribution of the population. Therefore, it is unlikely that the FAB has a significant clinical impact on the nocturnal diaper weaning of a child.

We attempted to ascertain whether the delay in reaching developmental milestones is a result of the Ponseti method or due to the underlying CTEV pathology. Garcia et al⁸ suggest that the use of casting and splinting early in life may impair the balance and strength of the muscles involved, therefore delaying motor development. Other studies also suggest that the presence of a clubfoot is a marker for a more global underlying motor dysfunction.¹⁹ This is demonstrated by the muscular atrophy seen in the lower leg in clubfoot that persists even after correction. No correlation was found between severity of CTEV, as assessed by the Pirani score prior to treatment and the age by which the developmental milestones were reached. This applied for children with both unilateral and bilateral CTEV. This implies that the Pirani score at presentation has no bearing on any subsequent motor delay. This is

Table 2 Pearson's correlation coefficients and R² values of correlation between congenital talipes equinovarus severity and age of attainment of developmental milestones

| | Standing age | Walking age | Nocturnal continence age |
|------------------------------------|--------------|-------------|--------------------------|
| Pearson's correlation coefficients | 0.181 | 0.157 | -0.0104 |
| R ² values | 0.0326 | 0.0246 | 0.000109 |

consistent with previous studies that have shown that the Pirani score at presentation does not affect treatment outcomes.^{15,20}

Additionally, we compared the age of attainment of the three developmental milestones between children with CTEV who were compliant with the FAB with those who were not. We found no significant differences between these groups. Therefore, it is unlikely that compliance with FAB had any clinical impact on developmental milestones through persistent immobility. It is much more likely that the mild delay seen in children with CTEV is multi-factorial, due to a combination of primary pathological factors, factors secondary to treatment and cultural and behavioural factors. It is important to acknowledge that whilst FAB use does not appear to affect developmental milestones at the time of primary correction, compliance with FAB is an important predictor of relapse at a later date.⁴ This can have its own impact on developmental milestones through a recurrent foot deformity.

We assessed whether the attainment of milestones was consistent across the different study sites. Whilst there was no difference in standing age between the children from different sites, the children from DCH were slower to develop independent walking but faster at achieving nocturnal continence compared with the United Kingdom-based hospitals. The reason for this effect is unclear. As the children from DCH were from a different country than the other three centres it is possible that this difference is confounded by both genetic and environmental factors amongst the DCH children. In reality, the relative impact of these differences is minor, and may fall within the natural variation for all children.

This study is subject to some limitations. Only 17 (13.1%) of the children were non-compliant with the FAB which resulted in a skew in data. It is also possible that a proportion of non-compliant children were excluded as they did not have a sibling to compare with. The results of the non-parametric analysis of this question were significant and, therefore, meaningful. The sibling developmental milestone data relied on parental recall, which could be subject to inaccuracy. To assess for this, we conducted a reliability study on 30 patients before the study commenced and found excellent accuracy in parents'

reporting of developmental milestones compared with documented milestones in the physiotherapy notes. Previous data also suggests that parents' recall of their child's walking age is often accurate and correlates well with true walking age. One study found that at three years old, there was greater than 70% correlation between parental recall and true walking ages.²¹ We hypothesize that attainment of developmental milestones may be even more significant in families of children with CTEV, therefore, caregivers are likely to be even more vigilant in monitoring their child's development.

The major strength of this paper is the ability to compare children with CTEV to their typically-developed siblings which has not been previously reported in the literature. Comparison with siblings may be able to overcome bias due to geographic, cultural and genetic diversity which gives a more accurate comparison of the milestones achieved.¹²⁻¹⁴

Conclusion

There is a mild developmental delay in the attainment of motor milestones in children with CTEV treated with the Ponseti method. However, it is unlikely that this delay is clinically significant. There is no delay in the attainment of nocturnal continence as a result of the use of the Ponseti method. This allows us to reassure parents regarding the FAB use as part of the Ponseti treatment programme and to emphasize its importance for the prevention of relapse.

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COMPLIANCE WITH ETHICAL STANDARDS

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OA LICENCE TEXT

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ETHICAL STATEMENT

Ethical approval: This research did not directly involve human participants and/or animals. This study was approved by the St. George's University and Dana Children's Hospital Institutional Review Boards and National Health Service Health Research Authority (REC Reference: 19/HRA/0512).

Informed consent: This project used retrospective data extracted from patient notes so informed consent was not required.

ICMJE CONFLICT OF INTEREST STATEMENT

The authors have no conflicts of interest relevant to this article to disclose.

AUTHOR CONTRIBUTIONS

KH: Designed the data collection instruments, Collected data, Contributed to the data analyses and to the initial manuscript, Reviewed and revised the manuscript.

YG: Conceptualized and designed the study, Contributed to the initial manuscript, Reviewed and revised the manuscript.

MC: Contributed to the initial analyses and the initial manuscript, Reviewed and revised the manuscript.

SW: Conceptualized and designed the study, Reviewed and revised the manuscript.

AY: Contributed to study design and data collection, Reviewed and revised the manuscript.

YH: Contributed to study design and data collection, Reviewed and revised the manuscript.

MD: Contributed to study design and data collection, Reviewed and revised the manuscript.

DME: Contributed to study design, Reviewed and revised the manuscript.

All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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Appendix 1

Flowchart of the patient selection process.

