Surgical management of pes planus in children with cerebral palsy: A systematic review

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Abstract

Purpose: Pes planus (or flatfoot) is the most common deformity in children with cerebral palsy. There are several surgical interventions used to treat it: single calcaneal osteotomies, extra-articular arthrodesis, double calcaneal osteotomy, calcaneo-cuboid-cuneiform osteotomy, intra-articular arthrodesis, and arthroereisis. There is currently no evidence on optimal treatment for flatfoot in children with cerebral palsy. Our purpose is to systematically review studies reporting complications, recurrence rates, and radiological outcomes of the surgical management of flatfoot in children with cerebral palsy.

Methods: Five databases were searched to identify studies published from inception until July 2021, with keywords relating to flatfoot, cerebral palsy, and surgical interventions. We included prospective, retrospective, and comparative study designs in the English language. Data was extracted and tabulated in duplicate into Excel, and analysis was conducted using Python SciPy.

Results: In total, 1220 studies were identified of which 44 met the inclusion criteria, comprising 2234 feet in 1364 patients with a mean age of 10.3 years and mean follow-up of 55.9 months. Radiographic outcomes showed improvement with all procedures; complications and recurrence rates were too poorly reported to compare. Only 6 (14%) studies were assessed as a low risk of bias. There was substantial heterogeneity of outcome measures.

Conclusion: There is a lack of high-quality, comparative studies assessing the radiological outcomes, complications, and recurrence rates of surgical alternatives to treat flatfoot in children with cerebral palsy. There is currently no clear evidence on optimal surgical treatment.

Level of evidence: IIa based on Oxford Centre for Evidence-based Medicine.

Keywords: Flatfoot, pes planovalgus, pes planus, surgery, cerebral palsy, pediatrics, orthopedics

Introduction

Pes planus (also known as flatfoot or pes planovalgus) is the most common foot deformity in children with cerebral palsy (CP).¹ The pathology develops due to the lateral displacement of the navicular, causing loss of the medial longitudinal arch, talar head uncovering, and talar prominence in the medial foot.¹ The condition can be categorized into flexible and stiff.² Flexible deformity involves preservation of the arch when sitting, extending the great toe or standing on tiptoes; stiff deformity involves a flat arch with limitation of motion during weight-bearing and non-weight-bearing, and is more difficult to treat.³ Higher functioning, ambulatory patients (Gross Motor Function Classification System (GMFCS) I–III) usually present with flexible flatfoot, whereas stiff flatfoot is more common in adolescents with lower functional ability (GMFCS IV–V).⁴ The deformity usually worsens during late childhood and can cause significant pain, pressure ulcers, and difficulty walking or wearing

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Surgical management is indicated when conservative measures have failed.

There are several surgical interventions used to treat pes planus but no guidelines on how to choose between them. Extra-articular arthrodesis (EAA) or single calcaneal osteotomies (SCO) are commonly used to treat children with milder, flexible deformities, and lower GMFCS levels. SCO includes calcaneal lateral column lengthening (LCL) and calcaneal slide (CS) with concomitant soft tissue procedures (peroneus brevis lengthening, tibialis posterior shortening, and talonavicular joint capsule reefing), and occasionally a medial cuneiform osteotomy. Double calcaneal osteotomy (DCO) and calcaneo-cuboid-cuneiform “triple C” osteotomies (TCO) have been used to treat moderate-to-severe deformities that would likely recur with SCO and EAA. Intra-articular arthrodesis (IAA) is an invasive procedure that has been reserved for children with GMFCS IV or V and/or severe, stiff deformities. Subtalar arthroereisis (SA) is a non-fusion procedure that has recently received renewed interest in the literature as an alternative to SCO and EAA.

The purpose of this study is to systematically review the literature regarding the radiological outcomes, complications, and recurrence rates of current surgical management of flatfoot in children with CP.

Methods

This systematic review was reported according to Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA 2020) checklist and the AMSTAR 2 critical appraisal tool. The protocol was prospectively registered on PROSPERO CRD420201239285. The authors declare no conflict of interest relevant to this work.

Search strategy

A literature search was conducted using the online Cochrane Library, EMBASE, MEDLINE, Web of Science, and PubMed databases, using the following terms: ((cerebral palsy)) AND (((pes planus) OR (flat foot) OR (pes planovalgus)) OR ((calcaneal) OR (calcaneus) OR (calcaneum) OR (slide) OR (double) OR (heel) AND (osteotomy)) OR ((fusion) OR (arthrodesis) OR ((arthroereisis) OR ((Grice Green) OR (Grice-Green) OR ((lateral column lengthening) OR (MOSCA)))). No limitations were placed on gender, date, or language. All results from inception until July 31, 2021 were included (Appendix 1).

Inclusion criteria

We included all prospective, retrospective, and comparative study designs (randomized controlled trials (RCTs), case studies, cohort studies, and case-controlled studies) reporting original/primary data on one or more of the outcomes of interest. A scoping review identified a significant lack of RCTs on this subject, thus including non-randomized studies was necessary for an all-encompassing review.

Exclusion criteria

We excluded duplicate articles, cost-effectiveness studies, and studies not reporting on primary data (such as review articles, editorials, discussions, commentaries, letters, and conference abstracts). We excluded studies not reporting data on radiographic outcomes, complications, and recurrence rates. Studies where data for pediatric patients with CP was not readily separable from other participants and where surgery was not the primary intervention were excluded on the grounds of not being relevant to the aims of the review.

Participants

Children with CP and symptomatic pes planus were included. Studies with a mean age of participants below 18 years of age were included. Children without CP treated for foot deformities other than pes planus were not included.

Intervention

The intervention was operative surgical management to treat symptomatic pes planus where conservative management had failed. The specific procedures identified by a scoping review included calcaneal LCL, EAA, CS, DCO, calcaneo-cuboid-cuneiform TCO, IAA, and SA. Data on variations of these procedures and any soft tissue procedures performed in conjunction was also extracted.

LCL is a procedure originally described by Evans that equalizes both columns in the foot via an osteotomy of the calcaneus bone approximately 1.5 cm proximal to the calcaneocuboid joint; as the lateral column is shorter in flatfoot, this equalization corrects forefoot abduction and restores the medial longitudinal arch. Mosca popularized the procedure by adding the soft tissue procedures of peroneus brevis lengthening, tibialis posterior shortening, and talonavicular joint reefing, and a plantar closing-wedge osteotomy of the medial cuneiform.

EAA, originally used by Green and first reported by Grice in 1952, involves the extra-articular positioning of a structural autograft (either fibula or anterior tibia) between the talus and the calcaneus. CS is the medial displacement of the posterior part of the calcaneus, thus creating a compensating deformity to improve the heel valgus and normal weight-bearing. DCO is a combination of LCL and CS. TCO is a versatile procedure that allows correction at the fore-, mid- and hindfoot by three osteotomies: a CS,
an opening-wedge cuboid osteotomy, and a plantar flexion closing-wedge osteotomy of the medial cuneiform.\textsuperscript{14} SA involves the insertion of an implant into the sinus tarsi or adjacent to it to prevent talonavicular impingement which consequently blocks and corrects excessive eversion movements of talus and calcaneus, and maintains the subtalar joint in a more neutral position.\textsuperscript{2}

Finally, IAA is a fusion of one or all of the joints of the hind- or midfoot, usually undertaken as a triple arthrodesis involving the talonavicular, subtalar, and calcaneocuboid joints.\textsuperscript{15}

**Comparators**

There is currently no gold standard for the surgical management of flatfoot in children with CP. We included papers that surgically managed flatfoot by LCL, CS, DCO, TCO, EAA, IAA, and SA using traditional or modified techniques. Non-surgical management of flatfoot was excluded.

**Outcomes**

Primary outcomes were radiographic angles, complications, and recurrence rates. The radiographic angles included were most commonly used to assess flatfoot: anterior–posterior talocalcaneal (AP TC), anterior–posterior talo-first metatarsal (AP T1MT), and talonavicular coverage (TNC) angles; and lateral talocalcaneal (Lat. TC), lateral talo-first metatarsal (Lat. T1MT), calcaneal-first metatarsal (C1MT), and calcaneal pitch (CP).\textsuperscript{2} Gait analysis and clinical outcomes were not assessed, as gait analysis is infrequently reported in studies and there is no current standardized tool for assessing clinical outcomes for each surgical procedure.

**Data extraction**

Study selection was performed in duplicate (P.M., C.G., and P.M., M.M.), and data extraction was performed in duplicate (P.M., C.G., and P.M., M.M.). Discrepancies over the inclusion of any study or data extraction were resolved by consensus or arbitration by senior authors (T.L.L. and M.K.).

For every article, the following data was extracted based on a scoping literature review:

- Article demographic details (number of authors, title, year published, level of evidence (1–5), funding sources). Patient demographic details (number of patients, number of feet operated on, gender of patients, mean age, and age range of patients; GMFCS level of disability; mean follow-up (months/years) and range of follow-up).

- Surgery details: type of surgery, indication for surgery, and concurrent procedures.

- Radiographic outcomes: AP TC, AP T1MT, and TNC angles; and Lat. TC, Lat. T1MT, C1MT, and calcaneal pitch.

- Complications and recurrence rates

Gait analysis and pedobarographic outcomes were not tabulated or synthesized due to the heterogeneity of the reporting between the studies.

**Assessment of methodological quality**

The level of evidence and methodological quality of included studies was assessed using the MINORS criteria.\textsuperscript{16} A MINORS score of 16/16 or 24/24 was deemed high quality (and low risk of bias), 10–15/16 or 15–23/24 was deemed moderate quality (and moderate risk of bias), and a score of <10/16 or <15 was deemed low quality (and at high risk of bias) based on previous studies that used these scores. The articles were independently assessed by three authors (P.M., C.G., and M.M.) with a senior author settling any disagreement (T.L.L.). P.M. recorded sources of funding for individual studies included in the review.

**Statistical analysis**

Where data was provided, weighted means of radiographic outcomes and recurrence rates of the surgical procedures were calculated. An independent \( t \)-test was used to compare the weighted means. All data analysis was conducted using Python SciPy.\textsuperscript{17} Radiographic results were considered statistically significant when reported to have a \( p \)-value of less than 0.05.

**Results**

**Literature search**

The initial search yielded 1220 articles for review after duplicates were removed as shown in Figure 1. Review of titles and abstracts identified 80 articles for full-text screening, of which 44 met the inclusion criteria. The main reasons for excluding articles at this stage were “no reporting of outcomes” (\( n = 11, 31\% \)) and “no separation of outcomes for patients with CP to patients with different etiology for pes planus (PP)” (\( n = 20, 56\% \)).

**Study and patient characteristics**

The search identified 10 comparative studies (23\%): 8 of these were retrospective comparative studies (18\%) and 1 was a prospective, randomized design (2\%). Of the remaining studies, 7 were prospective case series (16\%) and 27 were retrospective case series (63\%). The study characteristics and outcomes of the papers included can be seen in Table 1 and summarized in Table 2.
The studies included 2234 feet in 1364 patients with a mean age of 10.3 years (ranging from 3 to 30 years) and a mean follow-up of 55.9 months (ranging from 4.3 to 217.2 months). Studies included patients with a GMFCS level of I–V, with both stiff and flexible flatfoot deformities. There was a significant focus on ambulatory patients with GMFCS level I–III and a flexible flatfoot deformity ($n = 33, 75\%$).

### Outcomes

A majority of the papers ($75\%, n = 33$) reported on pre- and post-operative radiographic deformity correction outcomes. All of these papers clearly stated that the radiographs were weight-bearing. Overall, the radiographic angles showed significant improvement within normal range with the exception of the Lat. T1MT angle in LCL and the AP TC angle in IAA (Table 3).

The clinical outcomes were measured differently in all papers (Table 1). Similarly, of the 11 studies (25%) that reported on gait analysis, kinematics, and pedobarography, the heterogeneity of the measurements meant that a comparison of the data between studies was not possible.$^{19,32,38,46,47,49,50,55,59,60}$

Given the heterogeneity in outcome measures between the studies and their general poor quality, it was not possible to synthesize a meta-analysis. A formal narrative synthesis of the results is provided following the Synthesis Without Meta-analysis (SWiM) reporting guidelines.$^{62}$

### Complications and recurrence

Data regarding complication and recurrence rates was poorly reported (Table 4). There was no clear correlation between complication rates and GMFCS level or the severity of the deformity. Recurrence rates were highest in relation to LCL and CS, and lowest in relation to DCO, TCO, and SA (Table 5).

### Quality of studies included

The quality of the studies included was assessed according to the MINORS criteria (Figures 2 and 3). In total, 38
| Study                     | Procedure type | No. of patients (M: F) | Mean age in years (range) | GMFCS or level of disability | Outcomes used                                                                 |
|--------------------------|----------------|------------------------|---------------------------|-------------------------------|-----------------------------------------------------------------------------|
| Aboelenein et al.        | EAA            | 15 (5:10)              | 11.5 (8.3–14.5)           | II–III                        | AP, TMT, AP, TC, Lat. TMT, Lat. TC, CP, and Dogan’s scale                   |
| Abu-Faraj et al.         | EAA            | 12 (8:4)               | 13.1 ± 2.6                | Not stated                    | 3D gait analysis and plantar pressure measurements                           |
| Adams et al.             | EAA            | 42 (19:23)             | 9 (6.3–13.9)              | Not stated                    | Lat. TMT, AP and Lat. TMT, Lat. TNC, CP, Lawrence and Kelgren criteria for OA |
| Ahmed et al.             | EAA            | 35                    | 9                       | I–III                         | AP, TNC, AP and Lat. TMT, AP and Lat. TC, CP, talor declination angle        |
| Alman et al.             | EAA            | 29                    | 53                       | Ambulatory                    | AP and Lat. TC, AP, TNC, Clinical and radiographic assessment of ankle and subtalar alignment, brace, skin callouses |
| Aly et al.               | EAA            | 16 (9:7)               | 10.74 (6–16)              | I–III                         | AP and Lat. CP, TMT, Talar head uncovery (%)                               |
| Andreacchio et al.       | EAA            | 15                    | 9.1 (6.2–17.8)            | Ambulatory                    | Cosmesis, walking distance, walking support, pain                          |
| Bhan and Malhorrta       | EAA            | 10                    | 16                       | 6.2 (4–11)                    | Lat. TMT, AP, TNC, Clinical evaluation of ambulatory status, physical examination |
| Barrasso et al.          | EAA            | 26 (17:9)              | 10.5 (3.5–14.9)           | Ambulatory and non-ambulatory | Lat. TC and mobility, pain, pain, walking assessment, type of shoe, foot print analysis, callus formation, physical examination |
| Bourelle et al.          | EAA            | 17 (9:8)               | 5.4 (8.8–6.6)             | Ambulatory                    | AP and Lat. TC, Talar head uncovery (%)                                     |
| Cho et al.               | EAA            | 44 (27:17)             | 10.5                     | I–IV                          | Lat. TMT, AP and Lat. TMT, CP, Pain, walking assessment, foot print analysis, callus formation, physical examination |
| Costici et al.           | IAA            | 103 (64:39)            | 14.7 (12-20)              | I–IV                          | AP, TNC, AP, TNC, Costa Bertani angle                                       |
| de Morais Barros et al.  | IAA            | 21 (138)               | 16 (8–29)                | II–IV                         | Inclination of the calcaneus angle, Lat. TMT, AP, TNC, Clinical and radiographic assessment of ankle and subtalar alignment, brace, skin callouses |
| Elbarbary et al.         | SA             | 23 (16:7)              | 8.6 (6–12)                | I–III                         | Lat.-TCA, heel valgus alignment, OxFAQ-C (physical, school and play, emotional, shape of foot, shoe wear, walking ability) |
| El-Hilaly et al.         | TCO            | 12 (7:5)               | 9.7 (5.1–15.3)            | I–IV                          | Lat. TMT, TNC, AP, dorsoplantar TMT, Pedobarography                         |
| Enstrom et al.           | EAA            | 16                    | 6 (3–12)                 | Not stated                    | Subtalar stability, corrected valgus hindfoot, gait improvement, radiographic analysis of union |
| Ettif et al.             | EAA            | 19 (127)               | 8.6 (4–18)               | Ambulatory and non-ambulatory | Lat. TMT, Lat. Horizontal angle, CP, Mosca’s clinical criteria               |
| Geven et al.             | EAA            | 11 (5:6)               | 10.7 (6–15)              | I–IV                          | AP and Lat. TC, AP, and Lat. TMT, CP, Walking, pain, skin calluses, orthoses, shoes, and survey |
| Huang et al.             | IAA, Calcaneal LCL | 21 (8:13)         | 11 (4.9–16)              | II–III                        | AP, TNC, Mosca’s radiographic and clinical criteria, Yoo et al. criteria     |
| Jeray et al.             | EAA            | 28 (18:10)             | 7.4 (5–12)               | Not stated                    | Lat. TC, Survey, Gait and kinetic analysis, pedobarography                  |
| Kadhim et al.            | IAA, Calcaneal LCL | 78 (43:35)          | 11.9 (4.7–18.3)           | I–IV                          | Lat. TMT, AP, TNC, Gait and kinetic analysis, pedobarography, Gait and kinetic analysis, pedobarography |
| Kadhim et al.            | IAA, Calcaneal LCL | 24 (43)            | 11 (4.7–18.3)             | I–IV                          | Lat. TMT, AP, TNC, Gait and kinetic analysis, pedobarography, Gait and kinetic analysis, pedobarography |
| Kubo et al.              | SA             | 11                    | 9.2 (5–13)               | II–III                        | AP and Lat. TC, AP, and Lat. TMT, CP, lateral relative overlap of Os navicular and Os cuboideum |

(Continued)
| Study                         | Procedure type                                      | No. of patients (M: F) | No. of feet | Mean age in years (range) | GMFCS or level of disability | Outcomes used                                                       | Mean follow-up period in months (range) | MINORS score (quality) |
|------------------------------|-----------------------------------------------------|------------------------|-------------|--------------------------|-------------------------------|--------------------------------------------------------------------|------------------------------------------|------------------------|
| Leidinger et al.             | EAA                                                 | 35 (20:15)             | 51          | 7.8 (3.9–14.4)           | Ambulatory                   | Lat. TC, CP                                                         | 271.2 (192–387.6)                      | 10 (poor)              |
| Luo et al.                   | Calcaneal LCL                                       | 20 (14:6)              | 30          | 11.9                     | II–IV                        | AP and Lat. T1MT, CP, AP and Lat. TC, AP TNC, Lat. talo-horizontal angle; Foot pain, callus, tolerance to a foot orthosis | 30 (12–72)                           | 10 (poor)              |
| Mazis et al.                 | EAA                                                 | 11 (7:4)               | 16          | 9.7 (6.4–12.3)           | Not stated                   | AP and Lat. TC, AP and Lat. T1MT, TNC, CP, naviculo-cuboid overlap; Ambulatory | 43.2 (24–99.6)                      | 9 (poor)               |
| Poykier et al.               | SA                                                  | 15 (7:8)               | 27          | 12.1 (9.3–14.5)          | Ambulatory                   | Pan, loss of function; Radiographic outcomes not specified; Mobility, shoes, and braces | 612.2 (284–1116.6)                    | 10 (poor)              |
| Mur et al.                   | IAA                                                 | 5 (3:2)                | 9           | 14 (11–17)               | IV-V                         | Radiographic outcomes not specified; Foot pain, callosity; TNC, Lat talo-horizontal angle | 60 (52–69)                           | 8 (poor)               |
| Nakim et al.                 | Calcaneal LCL and either medial cuneiform dorsal opening-wedge osteotomy or medial cuneiform planar flexion closing-wedge osteotomy | 24 (14:10)             | 42          | 9.7 ± 3.4                | I–III                        | CP, AP and Lat. T1MT, multi-segment foot modeling (MSFM) gait analysis, physical examination | 14.4 (96–142)                      | 16 (poor)              |
| Narang et al.                | Calcaneal LCL                                       | 10                      | 17          | 8–18 (11–13)             | I–II                         | AP, TC, AP TN, Lat. CP, CSMT, Lat. T1MT, Foot pain and heel rise tests, video gait analysis | 12 (12)                              | 12 (good)              |
| Noritake et al.              | Calcaneal LCL                                       | 16 (10:6)              | 27          | 10.8 (5.8–14.5)          | Ambulatory                   | AP TN, AP and Lat T1MT, Lat. CP, Lat talo-horizontal angle; Mosca’s clinical criteria | 38.4 (24–60)                         | 9 (poor)               |
| Park et al.                  | EAA, Calcaneal LCL                                  | 47 (27:20)             | 81          | 8.1 (5.5–16.7)           | II                           | AP and Lat. T1MT, AP and Lat. TC, AP TNC, CT, Gait analysis, foot pain, callosity, foot orthosis, satisfaction questionnaire | 39 (26–61)                           | 16 (poor)              |
| Rethlefsen et al.            | CS, Calcaneal LCL                                   | 72 (43:31)             | 119         | 11.9                     | I–III                        | Gait kinematics and kinetics; Modified Yoo system for change in standing foot position, Modified Clohane-Dindo system for complications | 38.4 (24–62)                         | 14 (poor)              |
| Rhodes et al.                | Calcaneal LCL                                       | 36                      | 63          | 9.3 (4–18)               | I–V                          | AP, TC, AP and Lat. T1MT, AP TNC, CP, Gait analysis | 37.25 (212–537)                     | 16 (poor)              |
| Seiran et al.                | IAA                                                 | 138 (73:65)            | 253         | 12.7 (5–20)              | I–V                          | AP, TC, AP and Lat. T1MT, AP TNC, Physical examination | 57.6 (24–132)                        | 9 (poor)               |
| Stave et al.                 | EAA                                                 | 46 (28:18)             | 92          | 12.9 (7.8–18.4)          | II–IV                        | AP, Lat T1MT, CA, AP and Lat T1MT, AP TNC, CP, Modified Yoo system for change in standing foot position, Modified Clohane-Dindo system for complications | 55 (30–90)                           | 10 (poor)              |
| Sung et al.                  | Calcaneal LCL                                       | 75 (51:24)             | 75          | 11 (5–30)                | Not stated                   | AP, Lat T1MT, AP TNC, AP T1MT, AP TNC, AP and Lat. TC, AP TNC, CP, Gait analysis | 372 (12–101)                         | 12 (good)              |
| Turtuga et al.               | IAA                                                 | 32 (16:16)             | 59          | 13.9 (9–20)              | Ambulatory                   | AP, Lat T1MT, AP T1MT, AP TNC, AP TNC, AP and Lat. TC, AP TNC, CP, Gait analysis | 40 (183–667)                        | 7 (poor)               |
| Vdshou and Dimitriades       | EAA                                                 | 5 (3:2)                | 6           | 10.6 (9–14)              | Ambulatory                   | Lat. TC, TNC, Physical examination; Symptomatic foot | 96 (24–180)                         | 9 (poor)               |
| Vdshou and Dimitriades       | EAA                                                 | 9 (5:4)                | 12          | 11.7 (9–14)              | Ambulatory                   | Lat. TC, TNC, Evidence of Fusion; Appearance of the feet; Heel valgus alignment, local symptoms | 93.6 (48–180)                        | 9 (poor)               |
| Wen et al.                   | EAA, SA                                             | 26 (17:9)              | 44          | 8.5 (5–15)               | I–II                        | AP, Lat T1MT, AP T1MT, Lat. TC, AP T1MT, AP TNC, AP T1MT, AP TNC, AP T1MT, AP TNC, CP, Gait analysis | 30.1 (20–60)                        | 16 (poor)              |
| Yoo et al.                   | Calcaneal LCL                                       | 56                      | 92          | 9.2 (4–17)               | Ambulatory                   | Lat. T1MT, AP T1MT, CP, AP T1MT, AP T1MT, AP TNC, AP T1MT, AP TNC, CP, Gait analysis | 62.4 (24–93.6)                      | 9 (poor)               |
| Yun et al.                   | EAA                                                 | 30 (21:9)              | 50          | 9 (5–18)                 | Ambulatory                   | AP, Lat T1MT, AP and Lat. TCA, AP, CP, Lat. T1MT, AP T1MT, AP T1MT, AP TNC, AP T1MT, AP TNC, CP, Gait analysis; Heel valgus alignment | 37 (26–49)                           | 9 (poor)               |
| Zefang et al.                | Calcaneal LCL                                       | 32 (22:10)             | 46          | 11 (4–22)                | Ambulatory                   | Lat. TC, Lat. T1MT, Lat. CP, AP T1MT, Costa Bertran angle; Modified Phillips clinical score | 46 (36–108)                          | 12 (good)              |

AP TC: anterior–posterior talocalcaneal; AP T1MT: anterior–posterior talo-first metatarsal; Lat. T1MT: lateral talo-first metatarsal; Lat. TC: lateral talocalcaneal; CP: cerebral palsy; TNC: talonavicular coverage; MSFM: multi-segment foot modeling; C1MT: calcaneal-first metatarsal; GMFCS: Gross Motor Function Classification System; OA: osteoarthritis; AOFAS: American Orthopaedic Foot and Ankle Society; AOFAS-AH: American Orthopaedic Foot and Ankle Society Ankle-Hindfoot scoring system; OxFAQ-C: Oxford Ankle Foot Questionnaire for Children.
Table 2. Summar of included studies.

|                | Calcaneal LCL | CS | EAA | DCO | TCO | IAA | SA |
|----------------|---------------|----|-----|-----|-----|-----|----|
| No. of studies | 17            | 1  | 16  | 1   | 1   | 7   | 5  |
| Sample size (no. of feet) | 784          | 119 | 539 | 24  | 18  | 634 | 140 |
| GMFCS (I–V)    | I–V           | I–III | I–IV | I–III | I–IV | I–V | I–III |
| No. of comparative studies | 6           | 1  | 2   | 0   | 0   | 2   | 2  |

LCL: lateral column lengthening; CS: calcaneal slide; EAA: extra-articular arthrodesis; DCO: double calcaneal osteotomy; TCO: triple calcaneal osteotomy; IAA: intra-articular arthrodesis; SA: subtalar arthroereisis; GMFCS: Gross Motor Function Classification System.

Table 3. Radiographic outcomes summarised using the weighted mean for each procedure.

|                | AP TC | Lat. TC | AP T1MT | Lat. T1MT | CP | AP TC | Lat. TC | AP T1MT | Lat. T1MT | CP |
|----------------|-------|---------|---------|-----------|----|-------|---------|---------|-----------|----|
| IAA            | 42.9  | 48.2    | 25.7    | 22.2      | 12 | 33.9  | 31.6    | 5.3     | 8.7       | 12.8 |
| LCL            | 30    | 42.6    | 23.2    | 27.5      | 3.7 | 20.9  | 36.2    | 6       | 11        | 10.6 |
| EAA            | 38.6  | 45.9    | 28.5    | 29.5      | 11 | 25.9  | 33.3    | 7.9     | 10        | 12  |
| SA             | 34.4  | 47.2    | 26.5    | 26.5      | 5.2 | 27.5  | 31      | 5.11    | 5.5       | 9.8  |

AP TC: anterior–posterior talocalcaneal angle (normal range 15°–27°); Lat. TC: lateral talocalcaneal angle (normal range 25°–45°); AP T1MT: anterior–posterior talo-first metatarsal angle (normal range 3°–11°); Lat. T1MT: lateral talo-first metatarsal angle (normal range 2°–10°); CP: calcaneal pitch (13°–23°); IAA: intra-articular arthrodesis; LCL: lateral column lengthening; EAA: extra-articular arthrodesis; SA: subtalar arthroereisis.

Discussion

This is the first systematic review of surgical management of pes planus in children with CP, covering 2234 operations from 44 papers. Overall, we found that substantial deformity correction was achieved by each surgical intervention. Based on the evidence, however, it is not possible to show that one intervention is superior to others. There is a significant lack of studies on CS, DCO, TCO, IAA, and SA (Table 2). Most of the patients included in the studies in this review had flexible deformity with lower GMFCS levels; there is limited data to allow a proper assessment of treatment for moderate–severe flatfoot deformities. Ideally, studies would separate management of stiff flatfoot in GMFCS levels IV and V from flexible flatfoot in GMFCS levels I–III as it constitutes a different deformity. Many of the papers used levels I–IV or I–V, or described the patients as “ambulant” or “non-ambulant” making it difficult to undertake subgroup analysis as the data was not always clearly separated.

The radiographic outcomes show significant improvement is achievable by all surgical interventions. Severe deformity in patients with higher GMFCS levels is difficult to treat even with an invasive procedure such as IAA, and achieving long-term correction with LCL, EAA, CS, or SA is unlikely unless there is concomitant joint fusion. Four of the papers offered useful parameters for when a modified or more invasive procedure than LCL or EAA should be used to treat pes planus to avoid recurrence, but these papers were limited by the bias in the studies. Some studies combined techniques, such as Nahm et al., which are valid surgical options and would merit further research.

Our study has highlighted the need for a standardized method of measuring clinical outcomes. Four of the studies on LCL used either Mosca or Yoo’s clinical criteria, the latter of which was adopted by Ahmed et al. to assess the results of SA. These criteria could be combined in future and validated to compare different procedures, but could be adapted to incorporate activity levels to assess function. There was a notable lack of patient-reported outcomes in the studies which are essential to assess the effect of treatment on the patient’s quality of life. For example, relief of pain post-procedure is an important treatment outcome that could not be assessed in our review because it was either not measured at all or not in a consistent way. Standardized methods of measuring gait analysis, kinematics, and pedobarography are also needed given a general consensus in the included studies on the limited ability of radiographic outcomes to fully reflect the clinical picture.

The poor reporting of complications could be improved by the use of clearer definitions, for example, avoiding the interchangeable use of terms such as “non-union” and “pseudoarthrosis,” or “under-correction” and “recurrence.” The high recurrence rates seen in LCL and CS procedures compared to other procedures reflect the high risk of bias in the studies rather than the actual difference in recurrence rates, and other procedures reported significant complications such as hardware complications for SA. Any conclusions on the comparison between treatments in
Table 4. Complications and recurrence rates.

| Procedure Study modification to procedure | No. of feet | GMFCS ambulatory (A)/non-ambulatory (NA) | Complications (%) | Recurrence rate (%) |
|-------------------------------------------|-------------|----------------------------------------|-------------------|---------------------|
| Calcaneal LCL Aboelenen et al.18          | 22          | II–III                                 | Infection 4.5     | –                   |
| Adams et al.18                            | 61          | –                                      | Under-correction 9| –                   |
| – Group 1: Pin stabilization ATL, GR (PS)  |             | –                                      | Subluxation 86 (PS)| –                   |
| – Group 2: No stabilization ATL, GR (NS)   |             | –                                      | Subluxation 91 (NS)| –                   |
| Ahmed et al.21                            | 29          | I–III                                  | Pain 13.8         | 0                   |
| Evans, ATL, GR                            |             | –                                      | Infection 6.9     | –                   |
| – Under-correction 14                     |             | –                                      | –                 | –                   |
| Adams et al.20                            | 61          | –                                      | Subluxation 65    | –                   |
| – Group 1: Pin stabilization ATL, GR (PS)  |             | –                                      | –                 | –                   |
| – Group 2: No stabilization ATL, GR (NS)   |             | –                                      | Osteoarthritis 2.6| –                   |
| Cho et al.38                              | 28          | I–IV                                   | Infection 4       | 25                  |
| Ettl et al.34                             | 28          | A and NA                               | Subluxation 31.6  | (Group 1), 11.1 (Group 2) |
| Huang et al.40                            | 37          | II–III                                 | Staple penetration into talonavicular joint 31.6 (Group 1), 11.1 (Group 2) |
| – Group 1: CL, ATL, GR                    |             | –                                      | –                 | –                   |
| – Group 2: CL, medial column stabilization via talonavicular arthrodesis, ATL, GR | | | – | – |
| Ahmed et al.41                            | 63          | I–IV                                   | –                 | –                   |
| Kadhim et al.44                           | 15          | I–IV                                   | Under-correction 20| –                   |
| Luo et al.50                              | 30          | II–IV                                  | Hardware prominence requiring removal of hardware 47 | – |
| Nair et al.52                             | 24          | I–III                                  | Pain 53           | 0                   |
| Nair et al.52                             | 17          | I–II                                   | Under-correction 43| –                   |
| Noritake et al.48                         | 27          | I–II                                   | Under-correction 13| 64                  |
| Park et al.49                             | 37          | II                                     | Ovar-correction 13| 64                  |
| Park et al.49                             | 46          | I–III                                  | Ovar-correction 13| 64                  |
| Rhodes et al.51                           | 63          | I–V                                    | Pressure ulcer 24 (A)| 15 (A)              |
| – Group 1: Bovine xenograft (X) GR, ATL    |             | –                                      | Delayed union 2 (X)| 13 (X)              |
| – Group 2: Allograft (A) GR, ATL           |             | –                                      | Non-union 1 (X)   |                    |
| – Revision surgery 2 (X)                  |             | –                                      | –                 |                    |
| Sung et al.54                             | 75          | –                                      | Under-correction 28–40| –                   |
| Yoo et al.55                              | 92          | A                                      | –                 | –                   |
| Zheng et al.46                            | 46          | A                                      | Ovar-correction 7.6| 4.3                 |
| Evans, GR, PBL, RMC                       |             | –                                      | Ovar-correction 7.6| 4.3                 |
| CS Rethleisen et al.30                    | 73          | I–III                                  | Hematoma 23       | 15.2                |
| RMC or TNF                                |             | –                                      | Loss of correction 21| –                   |
| – Ovar-correction 9                       |             | –                                      | Ovar-correction 9 | –                   |
| – Subluxation 21                          |             | –                                      | Subluxation 21    | –                   |
| – Osteoarthritis 2                        |             | –                                      | Osteoarthritis 2  | –                   |
| – Osteoarthritis 2                        |             | –                                      | Osteoarthritis 2  | –                   |
| – Prolonged pain <23                     |             | –                                      | Osteoarthritis 2  | –                   |
| – Planter hypersensitivity <23            |             | –                                      | Osteoarthritis 2  | –                   |

(Continued)
Table 4. (Continued)

| Procedure | Study modification to procedure | No. of feet | GMFCS: ambulatory (A)/non-ambulatory (NA) | Complications (%) | Recurrence rate (%) |
|-----------|---------------------------------|------------|------------------------------------------|-------------------|---------------------|
| DCO       | Aly et al.²³                      | 24         | I–III                                    | – Under-correction 12.5 | 0                   |
|           | Mosca and medial slide; GR, PBL, TPA |        |                                          | – Heel ulcer 6.25   |                     |
|           |                                  |            |                                          | – Chronic heel pain 6.25 |                     |
| TCO       | El-Hilaly et al.²⁴               | 18         | I–IV                                     | – Under-correction 7 |                     |
|           |                                  |            |                                          | – Infection and removal of hardware 2.2 |                     |
| SA        | Ahmad et al.²¹                    | 28         | I–III                                    | – Pain 25           | 0                   |
|           |                                  |            |                                          |                     |                     |
|           | Elbarbary et al.²⁷               | 46         | I–III                                    | – Under-correction 7 | 0                   |
|           |                                  |            |                                          | – Infection and removal of hardware 2.2 |                     |
|           |                                   | 19         | II–III                                   | 0                  | 0                   |
|           | Kabo et al.²⁰                    | 27         | A                                        | – Implant dislocation 21 (IST) | 0                   |
|           |                                   |            |                                          | – Implant fracture 15 (EST) | 0                   |
|           |                                   | 20         | I–II                                     | – Pain 3            | 0                   |
| EAA       | Abu-Faraj et al.¹⁷               | 17         | A                                        | – Slen irritation 207 | 3.8                 |
|           | Alman et al.¹²                    | 53         | A                                        | – Migration of smooth fixation wire 3.4 |                     |
|           |                                   |            |                                          | – Over-correction 13.7 |                     |
|           |                                   |            |                                          | – Revision surgery 3.4 |                     |
|           |                                   |            |                                          | – Tibial fracture at graft harvest site 3.4 |                     |
|           |                                   |            |                                          | – Ankle valgus 10.3  |                     |
|           | Barrasso et al.²⁶                | 40         | A and NA                                 | – Heal ulcer 2.5    | 0                   |
|           | Dennyson-Fulford                 |            |                                          | – Pseudarthrosis (asymptomatic) 5 |                     |
|           | Bhan and Mahvash²⁹               | 16         | A                                        | – Infection 6.3     | 0                   |
|           | Dennyson-Fulford, fibular dowel and screw ATL, PBT, Steinher’s plantar release | | |                  |                     |
|           | Bourrelle et al.²⁷               | 26         | A                                        | – Hardware problems 31.2 | 0                   |
|           | Chigot and Sananes modification of Grice ATL | | |                  |                     |
|           | Engstrom et al.¹³                | 27         | –                                        | – Non-union 12      | 3.8                 |
|           | ATL                              |            |                                          | – Under-correction 9.8 | 3.92                |
|           | Goren et al.¹⁵                   | 15         | I–IV                                     | – Non-union 12      | 3.8                 |
|           | Modification of Grice using superperosteal fibular graft, GR | | |                  |                     |
|           | Jeray et al.¹⁷                   | 52         | –                                        | – Graft slippage 1.96 |                     |
|           |                                   |            |                                          | – Revision surgery 1.96 |                     |
|           | Laddi et al.¹⁴                   | 51         | A                                        | – Under-correction 9.8 |                     |
|           | ATL PTL                         |            |                                          | – Over-correction 7.84 |                     |
|           |                                   |            |                                          | – Skin bone fracture 3.92 |                     |
Table 4. (Continued)

| Procedure                      | Study/Modification to procedure | No. of feet | GMICS/ ambulatory (A)/non-ambulatory (NA) | Complications (%) | Recurrence rate (%) |
|--------------------------------|---------------------------------|-------------|------------------------------------------|-------------------|---------------------|
| Mazis et al.43                 | Chigot and Sananes modification of Grice ATL | 16          | --                                       | Non-union 18.8     | 12.5                |
| Park et al.44                  | Modified Dennyson-Fulford, GR, ATL PBL | 44          | II                                       | Graft absorption 18.8 | --                 |
| Shore et al.43,53              | Modified Dennyson-Fulford (dowel allograft) | 92          | II-IV                                    | Stable fibrous union 2.2 | 0                  |
| Vachou et al.46,57             | Batchelor-Grice                  | 6           | A                                        | --                | 0                   |
| Vachou and Dimitriadis57       | Batchelor-Grice                  | 12          | A                                        | --                | 0                   |
| Wen et al.48,50                | Dennyson-Fulford, GR, ATL        | 22          | I-III                                    | Pain 4.5          | 0                   |
| Yoon et al.49                  | Modified Dennyson-Fulford ATL GR PBL | 50          | A                                        | Screw-fracture 4.5 | 0                   |
| IAA                            | Double arthrodesis               | 175         | I-IV                                     | Infection 2.3     | --                  |
| de Moraes Barros Fucks et al.50| Talonavicular + calcaneocuboid joints, GR | 35          | II-IV                                    | Non-union 50      | --                  |
| Kadhim et al.34                | Allograft and screw fixation, GR, TL | 75          | I-IV                                     | --                | --                  |
| Kadhim et al.36                | Allograft and screw fixation, GR, ATL | 28          | I-IV                                     | Under-correction 29 | --                 |
| Murr et al.46                  | ATL                             | 9           | IV-V                                     | Hardware prominence requiring hardware removal 25 | -- |
| Senaran et al.52               | ATL GR                          | 233         | I-V                                      | Pain 11           | --                  |
| Turriago et al.13              |                                | 59          | A                                        | Non-union 11      | --                  |

LCL: lateral column lengthening; DCO: double calcaneal osteotomy; TCO: triple calcaneal osteotomy; SA: subtalar arthroereisis; IST: intra-sinus tarsi; EST: extra-sinus tarsi; EAA: extra-articular arthrodesis; IAA: intra-articular arthrodesis; PBL: peroneus brevis tendon lengthening; PLL: peroneus longus tendon lengthening; ATL: Achilles tendon lengthening; GR: gastrocnemius recession; FS: pin stabilisation; NS: nonstabilised; TATT: tibialis anterior tendon transfer; PLR: peroneus longus release; RMC: reefing medial capsule; NF: talonavicular joint fusion; TPA: tibialis posterior tendon advancement; PBT: peroneus brevis transfer; PTL: peroneal tendon lengthening.
Table 5. Weighted mean of recurrence rates for each procedure where data was provided.

| LCL | CS  | DCO | TCO | SA  | EAA | IAA |
|-----|-----|-----|-----|-----|-----|-----|
| 18% | 29% | 0%  | 0%  | 0%  | 2.9%| 1.6%|

LCL: lateral column lengthening; CS: calcaneal slide; DCO: double calcaneal osteotomy; TCO: triple calcaneal osteotomy; SA: subtalar arthroereisis; EAA: extra-articular arthrodesis; IAA: intra-articular arthrodesis.

Figure 2. Bar chart demonstrating how non-comparative studies scored on MINORS.

Figure 3. Bar chart demonstrating how comparative studies scored on MINORS.
Regarding recurrence rates and complications would be misleading given the small size of the studies, short follow-up and reporting bias which may have hidden recurrence rates and complications.

The strengths of this review are that it includes papers on multiple interventions with a large sample size and a long follow-up. The 44 studies reported on a homogeneous population with minimal loss to follow-up. The main limitation of this review is the quality of the included studies which were mostly graded as “poor” and thus had a high risk of bias. The robustness of our synthesized results is difficult to assess given that data was often missing from the studies, especially regarding complications of the procedures. Furthermore, the heterogenous complication results meant that any analysis between the procedures is difficult to undertake. The retrospective case series did not have comparator interventions, meaning a potential lack of systematic pre- and post-operative assessment, and a high risk of bias in the clinical and radiographic outcomes. P-values were often not provided by papers to demonstrate whether radiographic outcomes were statistically significant, and often not combined with clinical outcomes to make them useful. The prospective and comparative studies were weakened by small study sizes and short follow-up periods. Longer follow-up periods are needed to reliably assess whether there are any degenerative changes to adjacent joints that can occur after fusion. Degenerative changes after IAA were not reported in the six studies with a mean follow-up of 71.4 months, thus a longer follow-up may be needed to exclude this outcome.

Conclusion

Pes planus is the most common foot condition for children with CP; a more robust evidence base is needed to provide guidance to surgeons on the optimal intervention for patients. Our review has highlighted the need for multi-center, large-scale, prospective, comparative studies, using standardized radiographic, clinical, and pedobarographic outcomes. Future studies should focus on patients with severe, stiff deformities, and higher GMFCS levels, and how the addition of fusion to procedures affects these patients in the long term.

Author contributions

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Compliance with ethical standards

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Appendix I

Database: Ovid MEDLINE(R) ALL <1946 to July 31, 2021>

Search Strategy:
1  cerebral palsy.mp. [mp =ti, ab, hw, tn, ot, dm, mf, dv, fx, dq, nm, kf, ox, px, rx, ui, sy] (70342)
2  (pes planus or flatfoot or pes planovalgus).mp. [mp =ti, ab, hw, tn, ot, dm, mf, dv, kw, fx, dq, nm, kf, ox, px, rx, ui, sy] (2997)
3  (lateral column lengthening or MOSCA).mp. [mp =ti, ab, hw, tn, ot, dm, mf, dv, kw, fx, dq, nm, kf, ox, px, rx, ui, sy] (4471)
4  ((calcaneal or calcaneum or calcaneus or slide or heel or double) and osteotomy).mp. [mp =ti, ab, hw, tn, ot, dm, mf, dv, kw, fx, dq, nm, kf, ox, px, rx, ui, sy] (2997)
5  (fusion or arthrodesis).mp. [mp =ti, ab, hw, tn, ot, dm, mf, dv, kw, fx, dq, nm, kf, ox, px, rx, ui, sy] (601149)
6  Arthroereisis.mp. [mp =ti, ab, hw, tn, ot, dm, mf, dv, kw, fx, dq, nm, kf, ox, px, rx, ui, sy] (394)
7  (Grice Green or Grice-Green).mp. [mp =ti, ab, hw, tn, ot, dm, mf, dv, kw, fx, dq, nm, kf, ox, px, rx, ui, sy] (40)
8  2 or 3 or 4 or 5 or 6 (607577)
9  1 and 7 (1499)