Calcaneal cuboid joint motion and osteotomy stability in children one year after calcaneal lengthening osteotomy

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ABSTRACT

Introduction: Flexible pes planovalgus in children may be corrected with a calcaneal lengthening osteotomy (CLO). However, CLO surgery may displace the distal calcaneal fragment dorsally and affect the motion in the calcaneo-cuboid joint (CCJ). We used radiostereometric analysis (RSA) to evaluate CCJ motion and CLO stability in children one year after CLO.

Methods: Ten patients (10 feet) with symptomatic flexible pes planovalgus were investigated one year after CLO. Mean age was 11.5 years (range 8.2–14.2). Standardised RSA measurements of the foot were obtained one year after surgery without and with weight-bearing (single leg stance). Tantalum markers inserted during surgery, were used to describe the CCJ motion as cuboid bone motion with respect to the distal calcaneus, and the CLO stability as distal calcaneal migration with respect to the proximal calcaneus.

Results: One year after surgery the CLO was stable. The motion in the CCJ upon full weight-bearing was mean -1.04 mm (CI95% -1.40 ; -0.67) joint distraction, mean 2.27 mm (CI95% 1.57 ; 2.96) cuboid dorsal translation, mean -1.94 mm (CI 95% -2.68 ; -1.20) cuboid medial translation, and mean 7.43° (CI 95% 3.97 ; 10.88) adduction.

Conclusion: The motion in the CCJ of children with CLO corrections for pes planovalgus is similar to that of adults during stance load with a normal foot posture and the patients were asymptomatic. Marker-based RSA may be used to evaluate causalities of foot symptoms after CLO surgery.

1. Introduction

Flexible pes planovalgus in paediatric patients may be corrected with a calcaneal lengthening osteotomy (CLO).

During distraction of a calcaneal lengthening osteotomy, the distal calcaneal fragment tends to displace dorsally causing a subluxation of the calcaneo-cuboid joint (CCJ), which may affect the motion and stability of the CCJ. Limited data suggests, that the subluxation may partially resolve,¹ however authors have been concerned with a risk of secondary osteoarthritis in the CCJ,^{1–4} but the potential clinical implications remain unresolved.

In the literature, the assessment of the distal calcaneal fragment displacement is typically based on measurements obtained from weight-bearing lateral projections of conventional X-rays. However, the CCJ morphology ranges from flat to a concave-convex joint with inter-individual variations,⁵ and thus a one-dimensional description of the

distal calcaneal displacement seems incomplete.

Recently, in an experimental study, marker-based radiostereometric analysis (RSA) was proven feasible and precise in measurements of stability in the CCJ with a precision below 0.5 mm for translations and below 2° for rotations of the calcaneal and the cuboid bones.⁶ We used radiostereometric analysis (RSA) without and with weight-bearing during one-legged stance to evaluate CCJ motion and CLO stability in paediatric patients one year after CLO for pes planovalgus.

2. Methods and patients

2.1. Design

The study evaluated 10 paediatric patients (10 feet) at a planned one-visit follow-up one year after surgery with a CLO. The study was nested within the RCT AUTO/HATCP study (ClinicalTrials.gov

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Table 1

Patient demographics.

Patients (n)	10
Age (years; range)	11.5 (8.2–14.2)
Male/female	3/7
Height (cm)	149.8 (132–164)
Weight	42.3 (32–65)
Right/left	4/6
HATCP/AUTO	5/5
Underlying pathology	Idiopathic juvenile psoriatic arthritis (1), cerebral palsy* (2), delayed motoric development (1)

*GMFCS level 1.

NCT01770574)⁷. The study was registered within the Danish Data Protection Agency (J.No 1-16-02-86-12) and ethical approval was obtained from the Central Denmark Region Committees on Biomedical Research Ethics (Record No. 1-1072-250-12, Appendix No. 43345).

2.2. Patients

Patient demographics can be found in Table 1. In all patients the indication for surgery was painful flexible pes planovalgus. Ambulatory function was intact and none had major cognitive impairments. The validated Oxford Ankle Foot Questionnaire (OxAFQ)⁸ was completed before surgery and at one year follow-up (Table 1. Patient demographics.).

2.3. Operative procedure and bead marking

All CLOs were performed in a standardised manner by one of two surgeons (OR, BMM). A tourniquet was used, and then a lateral longitudinal skin incision was made distally to the CCJ expanding proximally and plantarly. The peroneal tendons were identified and retracted. The extensor brevis muscle was incised subperiostally and elevated proximally off the calcaneal bone. Percutaneous elongation of the Achilles tendon (PETA) was performed in four cases. The calcaneal osteotomy was performed about one cm proximal and parallel to the CCJ with an oscillating saw. By use of a laminar spreader the osteotomy was distracted until sufficient lateral calcaneal length was achieved to correct the pes planovalgus deformity, as judged by the surgeon.

Intraoperatively, patients were randomised to either hydroxyapatite-tricalcium phosphate (HATCP) graft or iliac crest bone graft (AUTO). Prior to graft insertion, in preparation for RSA, the cuboid bone, the distal calcaneal bone fragment (dCF) and the proximal calcaneal bone fragment (pCF) were marked with tantalum beads by use of bead injectors (Kulkanon, Wennbergs Finmek AB, Sweden). Bead-marking of each bone/bone fragment was done with 4–5 beads, as widely distributed as possible. The osteotomy was used as entry-point to access the dCF (0.8 mm beads) and pCF (1.0 mm beads). The cuboid was accessed through a distally expanded lateral skin incision, carefully avoiding the capsule, and 1.0 mm tantalum beads were inserted through a small

drillhole in the lateral cortex. The graft (AUTO or HATCP) was customised into a trapezoid-shape. Following the bead and graft insertion, the position of each was controlled by fluoroscopy.

The foot was immobilized in a below-knee X-lite circular cast, applied with the ankle in neutral and the patients followed a non weight-bearing regime for six weeks.

2.4. RSA set-up and examinations

A standard set-up with a complete AdoraRSA suite (Nordic Roentgen Technique, Hasselager, Denmark) was used including high quality DR imaging technology with a resolution of 4 lp/mm (Canon CXDI 70C detectors for supine positions and 50RF detectors for standing positions). A focus-grid uniplanar carbon calibration box was used (Box 24, Medis Special, Leiden, The Netherlands) (Fig. 3). The exposure was set to approximately 70 kV, 10 mAs, fine focus. The calibration box defined the position and orientation of the global coordinate system. Each patient was first positioned supine with the operated foot oriented parallel to the calibration cage, the lateral side of the foot facing the calibration cage, the medial side facing upwards, and emphasizing a neutral position of the ankle (Fig. 2). The focus was centred on the CCJ. Next, loaded stereoradiographs were obtained with the patient standing on the operated foot (single leg stance on a box with balance support obtained from a rack), again with the medial side of the foot facing the X-ray tubes, and the lateral side facing the calibration cage (Fig. 1) and the ankle in neutral. Patients were instructed to step down and then step back up on the box a second time within a double-examination. The stereoradiographic set-up (tube position, distances, angulations) and the calibration box were similar in the supine and standing set-up.

2.5. RSA analysis and bone marker-models

The stereoradiographs were analysed by one observer (PM) using the marker-based analysis tool in the software Model-based RSA 3.32 (RSACore, Leiden, The Netherlands). Three marker-models (pCF, dCF and cuboid) were created (Fig. 3). All migration results are presented with reference to a right foot coordinate system as aligned with the global coordinate system in the calibration box (Fig. 2). For evaluation of stability in the CLO, the dCF marker-model was defined as the migrating object with reference to the pCF marker-model (rigid object). For evaluation of motion in the CCJ the cuboid was defined as the migrating object with reference to the dCF (rigid object). Interpretation of migration in the CCJ is described by the migration of the cuboid along and about the 3 orthogonal axes of the calibration cage as follows: x-translation (- CCJ distraction/+ CCJ compression), y-translation (+dorsal cuboid translation/-plantar cuboid translation), z-translation (+cuboid medial translation/-cuboid lateral translation), x-rotation (+pronation/-supination, y-rotation (+adduction/-abduction), z-rotation (+plantar flexion/- extension) (Fig. 2).

We assumed that the osteotomy was stable one year after surgery; however we provide RSA data from the osteotomy as verification of no

Fig. 1. The supine (center) and single-leg stance (right) RSA set-up (left).

Fig. 2. An illustration of a right foot viewed from the medial side in the position in the RSA set-up. Four tantalum markers are inserted in the cuboid, the dCF fragment and the pCF fragment for RSA analyses. The coordinate system is defined by the calibration box and used to describe the CLO stability and the CCJ motions.

Fig. 3. Example of a loaded RSA examination of a left foot with the three bone models (red: cuboid, blue: dCF and magenta: pCF). The bone marker-models can be easily distinguished from each other.

inducible displacement upon weight-bearing, and thus document that any recorded motion in the CCJ was not due to instability in the osteotomy, but occurred within the CCJ.

The analysis criteria included a condition number of the marker-models of maximum 350^{6,7}, a minimum of three markers within each marker-model throughout all scenes, and a rigid body error threshold of maximum 0.5 mm according to the standard settings in the analysis software. Repeatability/precision of the RSA measurements, assessed by double-examinations, is expressed as a coefficient of repeatability, $CR = \pm 1.96 \cdot SD_{diff}$. The systematic error of the RSA system should optimally be zero, and is expressed as the mean difference ($mean_{diff}$) within a double-examination.

2.6. Statistics

We did not perform sample size calculations due to the exploratory nature of the study. Paired t-tests were applied and assumptions of normality and equal variations were checked with Shapiro-Wilk test and f-test respectively. Correlations between patient reported symptoms and CCJ motion were examined ad modum Spearman. Statistical computations were performed in Stata/IC 12.1® for Mac (StataCorp, USA). Statistical significance was assumed at $p < 0.05$.

3. Results

Ten patients (4 right feet and 6 left feet) one year after CLO were investigated in this study. There were 7 females and 3 males, with a

Table 2

Precision/repeatability of translations within the calcaneal osteotomy (n = 10) and the CCJ (n = 10).

Osteotomy	Tx	Ty	Tz	Rx	Ry	Rz
^a Mean _{diff}	0.00	−0.01	−0.03	0.33	−0.30	−0.13
^b SD _{diff}	0.04	0.11	0.18	0.67	0.67	0.50
^c CR	0.08	0.21	0.35	1.31	1.31	0.98
^d 95%CI	−0.01;	−0.03;	−0.07;	0.18;	−0.45;-	−0.24;-
	0.01	0.02	0.01	0.48	0.15	0.02
CCJ	Tx	Ty	Tz	Rx	Ry	Rz
Mean _{diff}	−0.03	0.09	−0.02	−0.02	−0.07	−0.50
SD _{diff}	0.08	0.11	0.25	0.95	1.27	1.85
CR	0.15	0.21	0.50	1.87	2.49	3.63
95%CI	−0.04;-	0.07;	−0.08;	−0.23;	−0.35;	−0.91;-
	0.01	0.12	0.04	0.19	0.21	0.08

^a Mean_{diff}: Difference of means between two scenes in a double-examination.

^b SD_{diff}: Standard deviation of the Mean_{diff}.

^c CR: Coefficient of repeatability, $\pm 1.96 \times \text{SD}_{\text{diff}}$.

^d 95%CI: 95% confidence interval of the Mean_{diff}.

mean age of 11.5 years (range 8.2–14.2). Mean height and weight were 150 cm (range 132–164) and 42 kg (range 28–65).

Precision/repeatability of the RSA measurements of the single-leg stance was below 0.5 mm for translations in the osteotomy and the CCJ. Precision for rotations was below 2° for the osteotomy and below 4° in the CCJ. (Table 2).

The osteotomy (Fig. 4 a-b, left) was considered stable upon loading at one year follow-up.

The motion in the CCJ upon full weight-bearing in a single leg stance was mean −1.04 mm (CI95% −1.40; −0.67) joint distraction/widening,

mean 2.27 mm (CI95% 1.57; 2.96) cuboid dorsal translation, mean −1.94 mm (CI 95% −2.68; −1.20) cuboid medial translation, and mean 7.43° (CI 95% 3.97; 10.88) adduction. Other rotations were very small (Fig. 4b).

Generally, the OxAFQ item scores were high at follow-up, suggesting the children were not affected in their physical function and participation in every day activities (Appendix 1). There was no correlation between any of the OxAFQ item scores on pain during weight-bearing at 1 year and the CCJ motion.

4. Discussion

We used marker-based RSA to assess motion in the calcaneal-cuboid joint in paediatric patients one year after CLO for symptomatic flexible pes planovalgus. We found the osteotomy to be stable at one year, and we found translations and rotations to occur in the CCJ upon loading. There was a joint distraction of 1 mm, the cuboid translated dorsally and laterally by approximately 2 mm and rotated medially (adduction) by 7° during single leg stance.

Anatomical cadaver studies⁵ have shown that the CCJ has inter-individual variation being a more or less reciprocal concave-convex joint with an infero-lateral extension of the cuboid bone under the calcaneal surface acting as a pivot. The primary movements are medial-lateral rotations and small gliding movements. Bojsen-Møller also found that the cuboid varied in shape, from a flat to a concave surface. He found the flat cuboid to be more mobile, but he was not able to determine whether there was an association between flat cuboid and flat feet.⁵ In a cadaver study by DiNucci et al.¹⁰ the CCJ capsule was found to be pouch-like on the dorsal facet, with no restriction on dorsal

Fig. 4. Boxplots of xyz-translations (mm) (Figure 4a) and xyz-rotations (degrees) (Figure 4b) within the osteotomy and the CCJ one year after CLO in ten paediatric patients. The plots show the stability within the osteotomy and the motion within the CCJ from neutral unloaded supine position to a neutral single limb stance (loaded position).

and distal motion between the distal calcaneal fragment and the cuboid bone.

During CLO the distal calcaneal fragment tends to dislocate dorsally and result in subluxation in the CCJ. On weight-bearing lateral radiographs, Adams et al. calculated the “dorsal height of the articular surface above the cuboid as a percentage of the total length of the articular surface of the calcaneus at the CCJ” before, during and after surgery with final follow up ranging between 41 and 102 months. They found that dorsal displacement of the distal calcaneal fragment occurs even if temporary k-wire stabilisation is attempted. Dunn et al. (Dunn & Meyer 2011) reviewed lateral projections of standing X-rays on 50 feet with the objective to investigate the displacement of the anterior process of the calcaneus following CLO. They found the distal calcaneal fragment to displace dorsally with relation to the cuboid bone in the early post-operative period, but to resolve at final follow-up (ranging from 8 to 107 weeks). This is in line with our previous findings in this study population. Dorsal motion of the distal calcaneal fragment in relation to the proximal calcaneal fragment was noticed up till 6 months after surgery (supine stereoradiographs), and after one year it had resolved.⁷ However, these are all static descriptions, and not during motion.

In a case study on an 11-year-old ballet dancer, ultrasound was used to analyse motion of the CCJ.¹¹ The authors found plantar translation of the cuboids, during ankle plantar-flexion and dorsal translation during dorsi-flexion on the ballet dancer's feet. Similarly, our study showed dorsal translation of the cuboids with full weight-loading on the foot in neutral position.

The ultrasound technique is examiner-dependent and the precision was not reported in the study.¹¹ X-ray precision in relation to cuboid height was examined by Hastings et al. 2011.¹² It was measured as the “perpendicular distance from the plantar aspect of the cuboid to a line drawn from the plantar surface of the calcaneal tuberosity to the plantar aspect of the 5th metatarsal head” in supine and weight-bearing lateral X-rays using iSite PACS software. They reported the SD of differences to be 2.4 mm (intra-rater repeatability) for cuboid height, which should be relatively easy to measure. Acknowledging that the variance is based on measurements of the cuboid, not the anterior calcaneal process, it is fair to assume that any difference below 2.4 mm is due to measurement error with this method. In comparison the clinical precision of a similar CCJ-motion measured with the marker-based RSA method in our study is 0.1 mm.

Some limitations and strengths should be mentioned. Marker-based RSA measurements use the center of a marker configuration model, and not anatomical landmarks as reference points. This enables assessment of relative motion between bones, and not absolute displacements between anatomical landmarks. Further, marker-models do not use the kinematic coordinate system of the tarsal bones, but refer measured motions between bones to the coordinate system of the calibration box. The RSA set-up and foot position should therefore be correctly designed and aligned with the joint surface, in order to obtain meaningful joint motion estimates. RSA has several well-established advantages over conventional X-rays: The required radiation dose is smaller. It allows for measurements of three-dimensional motions between regions of interest with high precision and accuracy, and the motions can be measured in static as well as dynamic settings, and followed over time.

In this study the primary aim was to investigate the motion in the CCJ in a paediatric population after healing of the CLO. As the markers were inserted during surgery, it was not possible to obtain preoperative stereoradiographs, and thus we cannot arbitrate whether CLO surgery affected the motions during stance load in the CCJ. We set an *a priori* condition number limit of 350, which is higher than the condition number of 300 used to analyse the osteotomy in the clinical trial and the cadaver study previously performed by the same research team. As the focus of analysis was the CCJ, we accepted that the spread of the tantalum markers in all three dimensions within the paediatric cuboid bones was challenging due to the small size of the feet and the fact that it was imperative to leave the CCJ untouched. Thus, we accepted a slightly

higher limit for the condition number, which could have affected the measured rotations, but not the translations. Weight-bearing conventional X-rays were not routinely obtained prior to surgery in our department, and therefore these are not given further attention in the present study. We investigated a population with flatfeet, and unfortunately we do not have a normal population cohort to which CCJ motion can be compared, and it would not be ethically sound to place bead markers in the bone of healthy children for this purpose. Perhaps the closest comparison of CCJ motion, is the study of Lundgren et al.⁹ The authors used an invasive pin-tracking technique, by surgically anchoring external markers on the lower leg and foot bones, in healthy male adults to describe the kinematics of the respective bones. In this normal population they found, similar to our study, that during stance phase, the motion of the cuboid relative to the calcaneal bone follows a pattern of dorsal flexion, eversion and slight motion abduction-adduction. They provided the mean (SD) total range of motion in the sagittal, frontal and transverse planes to be 9.7° (5.2°), 11.3 (3.9°) and 8.1° (2.0°). Novel RSA concepts using three-dimensional volume models of bones from CT scans has the potential for future non-invasive examinations of the tarsal bones both before and after i.e. CLO surgery.¹³ This would further allow for a full kinematic analysis of tarsal bone interactions during loading, toe-stance, or even gait cycles. Recently, dynamic RSA with CT bone models has been used to evaluate wrist, hip and knee joint pathomechanics in cadavers.^{14–17} Other non-invasive methods such as magnetic resonance imaging (MRI)^{18,19} have been shown feasible in quantifying merged tarsal bone motion, e.g. thus not looking separately at the CCJ but considering the Chopart joint. The inherent advantage of MRI is the elimination of radiation, and not only does it allow for mapping of the joint kinematics, but also allows to obtain qualitative information of soft tissue structures, comparing tarsal ligament complexes before and after surgery. The future holds several possibilities for exploring preoperative foot bone kinematics and developing individualised surgical planning in order to provide the patient with the most optimal surgical solution.

In conclusion, we found marker-based RSA to be precise and feasible for assessment of CCJ motion and CLO stability during stance load, and the method may be used to evaluate causalities of foot symptoms after CLO surgery. One year after surgery, upon full weight-bearing the CCJ exhibited a mean 1.04 mm joint distraction/widening, 2.27 mm cuboid dorsal translation, 1.94 mm cuboid medial translation and adduction 7.43° adduction. In the future, non-invasive RSA with CT bone models could option a comparison of CCJ kinematics in case-control subjects and further extend our understanding of possible clinical implications of CCJ pathomechanics.

Contributions

Polina Martinkevich: Study design, study conduction, statistical calculations, interpretation of data, writing paper. Ole Rahbek: Study design, study conduction, revising paper. Bjarne Møller-Madsen: Study design, study conduction, revising paper. Maiken Stilling: Study design, study conduction, interpretation of data, revising paper.

Conflict of interest

PM, OR, BMM, MS declare that they have no conflict of interest.

Declaration of competing interest

There are no conflicts of interest.

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Appendix 1

Appendix 1

Item mean scores in the OxAFQ one year after surgery as reported by children (self report) and their parent(s) (proxy report). Highest (best) item score is 4 and lowest (worst) item score is 0. Cut-offs for severity of symptoms were defined as severe (0–1) moderate (2) mild (3) and none (4).

Items in the OxAFQ	Children's report (n = 8)	Parent's report (n = 10)
	mean ± sd (range)	
Q1 Have you found walking difficult because of your foot and ankle?	3 ± 1 (2–4)	3 ± 1 (2–4)
Q2 Have you found it difficult to run because of your foot and ankle?	3 ± 1 (1–4)	3 ± 1 (1–4)
Q3 Has it been difficult to stand up for long periods?	3 ± 1 (2–4)	3 ± 1 (2–4)
Q4 Have you had pain in your foot and ankle?	3 ± 1 (2–4)	3 ± 1.1 (2–4)
Q5 Have your legs been sore or ached after walking or running?	3 ± 1 (1–4)	3 ± 1 (1–4)
Q6 Have you felt tired because of your foot and ankle?	3 ± 1 (3–4)	3 ± 1 (2–4)
Q7 Has your foot or ankle stopped you joining in with other in the playground?	4 ± 1 (2–4)	4 ± 1 (1–4)
Q8 Has your foot or ankle stopped you playing in the park or outside?	3 ± 1 (2–4)	3 ± 1 (2–4)
Q9 Has your foot or ankle stopped you in taking part in PE lessons?	3 ± 2 (0–4)	3 ± 1 (0–4)
Q10 Has your foot or ankle stopped you in taking part in any other lessons at school?	3 ± 1 (2–4)	4 ± 0 (3–4)
Q11 Have you been bothered by how your foot or ankle looks?	3 ± 1 (1–4)	4 ± 1 (0–4)
Q12 Has the way you walked bothered you?	3 ± 1 (1–4)	3 ± 1 (0–4)
Q13 Have you been embarrassed because of your foot or ankle?	4 ± 1 (1–4)	3 ± 1 (1–4)
Q14 Has anyone been unkind to you because of your foot or ankle?	4 ± 0 (4–4)	4 ± 1 (2–4)
Q15 Has your foot or ankle stopped you wearing any shoes you wanted to wear?	2 ± 1 (0–4)	3 ± 1 (0–4)

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