

Clinical science

Comprehensive and reliable sonographic assessment and scoring system for inflammatory lesions of the paediatric ankle

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Abstract

Objective: The clinical decision-making process in paediatric arthritis lacks an objective, reliable bedside imaging tool. The aim of this study was to develop a US scanning protocol and assess the reliability of B-mode and Doppler scoring systems for inflammatory lesions of the paediatric ankle.

Methods: As part of the Childhood Arthritis and Rheumatology Research Alliance (CARRA) US group, 19 paediatric rheumatologists through a comprehensive literature review developed a set of standardized views and scoring systems to assess inflammatory lesions of the synovial recesses as well as tendons of the paediatric ankle. Three rounds of scoring of still images were followed by one practical exercise. Agreement among raters was assessed using two-way single score intraclass correlation coefficients (ICC).

Results: Of the 37 initially identified views to assess the presence of ankle synovitis and tenosynovitis, nine views were chosen for each B-mode and Doppler mode semi-quantitative evaluation. Several scoring exercises and iterative modifications resulted in a final highly reliable scoring system: anterior tibiotalar joint ICC: 0.93 (95% CI 0.92, 0.94), talonavicular joint ICC: 0.86 (95% CI 0.81, 0.90), subtalar joint ICC: 0.91 (95% CI 0.88, 0.93) and tendons ICC: 0.96 (95% CI 0.95, 0.97).

Conclusion: A comprehensive and reliable paediatric ankle US scanning protocol and scoring system for the assessment of synovitis and tenosynovitis were successfully developed. Further validation of this scoring system may allow its use as an outcome measure for both clinical and research applications.

Keywords: JIA, ankle, diagnostic imaging, musculoskeletal ultrasound

Rheumatology key messages

- A comprehensive and feasible sonographic scanning protocol to evaluate the paediatric ankle was developed.
- A semi-quantitative scoring system for each view was developed and showed good reliability.
- Paediatric sonographic scoring systems may provide objective assessment tools and represent important outcome measures.

Introduction

JIA is the most common inflammatory arthropathy of childhood characterized by inflammation of joints, tendon sheaths and entheses [1]. The number and type of affected joints vary according to the category of disease; however, it has been recognized that the ankle is one of the more commonly involved joints across all categories [2]. There are several unique challenges related to the assessment of the paediatric ankle joint. First, the ankle consists of several joints, including the tibiotalar joint (TTJ) and the subtalar joint (STJ). The STJ is further divided into an anterior STJ recess which includes the talonavicular joint (TNJ) and a posterior STJ recess. There are also several tendon groups in the ankle that are important to evaluate in JIA. Accurate detection and localization of pathology of these individual structures is not possible by physical examination and requires the use of imaging methods [3, 4]. Second, common developmental and anatomic conditions such as joint hypermobility, pes planus and misalignment create additional challenges in the assessment of ankle joint mobility and function [5]. Third, delays in the identification and treatment of ankle pathology may result in severe long-term functional and physical defects that can negatively impact the patient's quality of life [6–8]. Fourth, ankle involvement in JIA has been associated with worse long-term outcomes [9], especially if present within the first year of disease onset.

Imaging is an important component of the joint evaluation in children with JIA. Modalities include conventional radiographs, MRI and musculoskeletal US (MSUS). Conventional radiographs detect late complications of joint inflammation. In contrast, MSUS and MRI are more sensitive in identifying active inflammation [10]. Contrast-enhanced MRI in comparison with MSUS is more time consuming, costly, invasive (often requiring i.v. contrast and/or i.v. sedation) and requires resources that are not available for regular assessments. MSUS, on the other hand, can be performed at the bedside, without the need of i.v. contrast material or sedation, and allows for evaluation of the key structures affected by JIA including joints, cartilage, tendons, entheses and soft tissues. Studies comparing clinical examination and sonographic findings of the ankle in children with JIA have found poor correlation between the two when assessing the TTJ, STJ and ankle tendons [11, 12].

A major limitation to the use of MSUS in the care of children with JIA and ankle involvement is the lack of standardized and validated paediatric-specific scanning protocols and scoring systems. To overcome this limitation, the Childhood Arthritis and Rheumatology Research Alliance (CARRA) JIA-US subgroup has focused on establishing reliable scanning protocols and scoring systems to be used in JIA. In 2019, our group described a new reliable and reproducible US image acquisition protocol and scoring system for the paediatric knee [13]. A preliminary scanning and scoring system for the

assessment of arthritis in the paediatric shoulder, elbow, wrist, finger, hip and ankle joints proposed by this group revealed excellent reliability for all joints but the ankle [14]. Given the clinical importance of this joint, the aim of this project was to establish a feasible and reliable sonographic scanning protocol and scoring system for the assessment of arthritis and tenosynovitis in the paediatric ankle.

Patients and methods**Scanning protocol**

The OMERACT stepwise approach to select and develop imaging outcome measurement instruments was followed for the development of the proposed protocols [15] (Supplementary Fig. S1, available at *Rheumatology* online). In the absence of a comprehensive ankle-specific paediatric MSUS scanning protocol, a literature review was done for the time frame 1980–2018. Due to scarce information on paediatric joints, we also referred to protocols used in adults with arthritis and haemophilia (where US is used to determine joint involvement) (Supplementary Table S1, available at *Rheumatology* online [11, 12, 16–42]).

Subsequent steps were conducted through a consensus process requiring 100% agreement among participants for all components of the scanning protocol. All participants were paediatric rheumatology providers and members of the CARRA JIA-US subgroup. All participants successfully completed an 8-month in-depth MSUS training offered by the Ultrasound School of North American Rheumatologists (USSONAR) and/or completed a 1-year full-time US fellowship and/or hold the Musculoskeletal Ultrasound Certification in RheumatologyTM (RhMSUSTM), a certification endorsed by the ACR that demonstrates competency in MSUS. All participants had at least 1 and up to 14 years of experience in paediatric MSUS after training and/or certification (median time of paediatric MSUS experience: 4.5 years). The study was approved by the Cincinnati Children's Hospital Medical Center Institutional Review Board and subjects recruited provided written assent and consent to participate.

As a first step, 12 members of the CARRA JIA-US subgroup developed a preliminary scanning protocol through virtual meetings. This preliminary scanning protocol was based on findings from the literature review, addressing the optimal assessment of synovitis in the TTJ, STJ and TNJ, as well as tenosynovitis of the anterior, medial and lateral tendon compartments of the ankle.

The second step took place during the 2018 CARRA Annual Scientific Meeting, where content and face validity, feasibility and applicability of the preliminary scanning protocol were assessed by 18 members (including the aforementioned 12 members). The group first reviewed the protocol for content validity to ensure that all relevant structures are imaged in sufficient detail and with clear anatomical

landmarks to document normal findings or capture pathology of interest. For face validity the reviewers ensured that all relevant aspects are represented when looking at the protocol.

Two practical exercises were conducted during the same meeting on a total of eight volunteers including five subjects with JIA diagnosed based on ILAR criteria (ages 12–18 years) and three healthy individuals (ages 5–15 years). In addition to providing further verification of content validity of the preliminary protocols, these practical exercises assessed the feasibility (time needed to complete the required views) and applicability (technical feasibility of required image acquisition). Participants were assigned to four groups to facilitate standardized scanning of the volunteers by all participants. For each group a leader was chosen to monitor and record patient positioning and machine settings for each of the recommended views. In general, B-mode settings included a frequency of 9–12 MHz and Power Doppler signal included a low-flow setting (pulse repetition frequency 0.4–1.0, low wall filter), frequency was adjusted to obtain maximum sensitivity and the gain was adjusted to just below artefact levels. Based on the practical exercises, modifications were made by the group to improve feasibility and applicability, and further increase content validity.

In a third step, participants were asked to scan and submit images for at least three JIA subjects aged 2–16 years with ankle pathology. These images were evaluated during a total of four meetings (one in-person and three teleconference) to ensure each view was feasible across the age range, allowed visualization of the stated anatomical landmarks, and showed the extent of anticipated pathology. Images were then stored to create an image library for future exercises.

In a fourth step, the MSUS scanning protocol for the paediatric ankle was finalized during the 2019 CARRA Annual Scientific Meeting, using a nominal group technique.

Scoring system

US abnormalities were defined according to the OMERACT standardized definitions for US pathology [43, 44]. For each of the views chosen in the final scanning protocol a preliminary semi-quantitative scoring system was developed using a consensus process. A literature review on US scores provided the basis for the consensus process. Reliability of the scoring system was assessed using de-identified images reflecting different degrees of pathology at different ages. During two rounds of scoring exercises, modifications were made until excellent inter-rater reliability was met. Modifications of the preliminary scoring system were related to the terminology that was used to describe the extent of the pathology and the anatomic landmarks used as a reference. Excellent level of reliability was defined by an intraclass correlation coefficient (ICC) >0.75.

Statistical analysis

A nominal group technique was used to determine the views that would be included in the final scanning protocol. For this step, at least 80% agreement was required among participants as recommended by the OMERACT stepwise approach [15]. Inter-reader reliability of the scoring system was determined using the two-way single-score (ICC) method. The 95% CI are also provided. Established ICC ratings are excellent 0.75–1.00, good 0.60–0.74, fair 0.40–0.59 and poor <0.4 [45]. Data were analysed using SAS v9.4©, Cary, NC, USA.

Results

Literature review

Seventy-eight articles were identified as relevant. Of these articles, 29 were selected by the working group to provide the basis for the consensus process that was used to develop the scanning protocol and scoring system (Supplementary Table S1, available at *Rheumatology* online).

Scanning protocol

In step 1, based on the literature review and clinical considerations, a total of 38 possible views were identified to assess the presence of ankle synovitis and tenosynovitis in children (Supplementary Table S2, available at *Rheumatology* online). Consideration was given to both the positioning of the ankle including neutral, mild and maximum plantarflexion of the TTJ and inversion/eversion of the STJ. The use of both static images and dynamic videos to evaluate the presence and severity of abnormal findings was also considered.

Initially, we proposed obtaining short duration videos capturing (i) medial to lateral sweep of the TTJ while holding the ankle in neutral and plantarflexed positions, (ii) proximal to distal sweep of the TTJ, flexor tendons and TNJ, (iii) neutral to plantarflexion movement of the TTJ and (iv) dynamic inversion/eversion of the medial and lateral aspects of the STJ. In step 2 (see Patients and methods) it became apparent that the incorporation of videos into this exercise proved challenging due to the shape of the ankle and its bony prominences which resulted in poor definitions of key structures (i.e. tendons) and anatomical landmarks. The videos did not yield additional information to that obtained using the static images alone. Some patients were unable to maintain a neutral and/or maximum plantarflexed position due to active arthritis (of the hip, knee and/or ankle) or other limitations such as tight muscles and/or tendons. Consequently, videos were eliminated, and the scanning protocol was based on standardized static views. Other views were also eliminated as they did not add to content and face validity. Therefore, the original 38 views in the scanning protocol were reduced to 11 views.

At the 2019 CARRA Annual Scientific Meeting, the scanning protocol of 11 views was amended further using the nominal group technique and the final ankle scanning protocol was established. It included a total of nine views for each B-mode and Power Doppler-mode settings (Table 1, Supplementary Figs S2–S6, available at *Rheumatology* online). This scanning protocol was deemed feasible with a scanning time of 16–25 min. While orthogonal views are necessary to confirm pathology, in the context of establishing a scanning protocol and scoring system, one plane that would enable the visualization of the entire range of pathology in the most reliable way was chosen. It is important to note that the STJ has an anterior and a posterior part. In our protocol the STJ is assessed in a medial and lateral view. The medial view captures the anterior STJ as this is most superficial in this view, and the lateral view captures the posterior STJ.

In general, when applying the novel ankle US scanning protocol, it is recommended to:

- i) Have a thick layer of gel when obtaining images.
- ii) Perform a dynamic examination to facilitate differentiation of cartilage from an effusion and recognition of pathology. Manoeuvres include, but are not limited, to

Table 1. Final US scanning protocol for the paediatric ankle^a

View	Position of the probe	Structure evaluated	Position of the leg/foot
1. Anterior midline long	Longitudinal in midline, visualization of the distal tibia, talar dome and talar neck	Tibiotalar joint	Plantar flexion ^b
2. Anteromedial long	Longitudinal medial, distal medial aspect of tibia and entire length of talus		
3. Anterolateral long	Longitudinal lateral, distal lateral aspect of tibia and entire length of talus		
4. Talonavicular midline	Longitudinal with the proximal end on the talus and the distal end over the navicular bone	Talonavicular	Plantar flexion ^b
5. Medial view of STJ for anterior STJ recess	Proximal end of the probe anterior to the medial malleolus showing the talus and distal end pointing towards the heel of the foot with the ST (calcaneus) in view. The anterior STJ recess is visible from medial between talus and ST	Anterior and posterior STJ	Neutral ^c
6. Lateral view of STJ for posterior STJ recess	Perpendicular to the sole over the sinus tarsi. Slide the probe posteriorly following the posterior STJ viewed from laterally		
7. Anterior tendon compartment transverse	At the level of talar dome in transverse. Move the probe proximally and distally to find the area of maximal distension of each abnormal tendon sheath. If tendons are normal, obtain an image at the level of the talar dome	Anterior tendon compartment	Neutral ^c
8. Medial tendon compartment transverse	Transverse retro-malleolar medial. Move distally along the tendons to find the area of maximal distension of each abnormal tendon sheath and obtain an image. If tendons are normal, obtain an image at the level of the ST	Medial tendon compartment	Neutral ^c
9. Lateral tendon compartment transverse	Transverse retro-malleolar lateral. Move distally along the tendons to find the area of maximal distension of each abnormal tendon sheath and obtain an image. If tendons are normal, obtain an image at the level of the lateral view of the posterior STJ	Lateral tendon compartment	Neutral ^c

^a For all views recommend performing a full scan of the target structure and to obtain B-mode and Power Doppler–mode static images at the level of greatest abnormality. Always confirm pathology in a second plane. Dynamic examination is encouraged to facilitate recognition of pathology.

^b Plantar flexion: supine, knee flexed at 90°, foot flat on table but if patient unable to hold this position, place the leg straight on the table.

^c Neutral: supine, leg straight and foot relaxed; for medial view of STJ and medial tendon compartment consider slight eversion, for lateral view of STJ and lateral tendon compartment consider slight inversion. STJ: subtalar joint; ST: sustentaculum tali.

dorsiflexion/plantarflexion, eversion/inversion, compression and probe heel–toe movement.

- iii) Scanning the entire area of interest is recommended.
- iv) Follow the course of the tendons while avoiding anisotropy.
- v) Include the bony landmarks and overlying cartilage as recommended per protocol.
- vi) Obtain the images representing pathology at the site of maximal pathology.

Scoring protocols

A semi-quantitative scoring model for the joints in B-mode ranging from 0 = absence, 1 = mild, 2 = moderate to 3 = marked synovitis was adopted with specific definitions for each of the grades depending on the joint and view. For the tendons a binary score with tenosynovitis 0 = absent or 1 = present was developed (Table 2, Supplementary Figs S2–S6, available at *Rheumatology* online). For Power Doppler, a semi-quantitative system ranging from 0 = no abnormal synovial flow to 3 = marked synovial flow was chosen, and a binary system of 1 = presence or 0 = absence of Doppler signal was used for the tendon sheaths. The Power Doppler scoring system was adopted from a previously published paediatric-specific scoring system

[13] and applied to the Doppler signals within the synovial recess and synovial hypertrophy only.

The initial scoring exercise included a total of 124 images obtained in children aged 2–18 years, distributed equally across this age range, and including a similar number of images for each of the grade 0–3 categories (Supplementary Table S3, available at *Rheumatology* online). Excellent agreement for B-mode on the midline view of the anterior TTJ (0.75), good agreement for the views assessing the anterior STJ from medial (0.67) and fair reliability for views assessing the TNJ (0.51), the posterior STJ from lateral (0.51) and the tendons (0.49) were found (Table 3). Detailed discussion of the views and scoring system via five teleconferences resulted in further modification of the scoring systems. One recurring limitation when evaluating the scoring systems was the variability among scorers delineating the extent of the ‘full potential area of the synovial recess’. The space enclosed by the joint capsule was agreed upon as the anatomical landmark to define how far the synovial recess may extend and to delineate the maximum area relative to which the scoring in percent would be done. It was acknowledged that occasionally pathology may extend beyond this landmark. The use of non-specific terms when describing pathology such as ‘convex

Table 2. Final US scoring system for the paediatric ankle

Joint	Grade	Definition for B-mode images ^a
TTJ	0	Absent synovitis, normal fat into the joint space or physiologic minimal distension of the distal talar recess
	1	TTJ recess is mildly distended filling <25% of the expected maximum area of the recess
	2	TTJ recess is moderately distended filling 25–50% of the expected maximum area of the recess
	3	TTJ recess is significantly distended filling >50% of the expected maximum area of the recess
Talonavicular joint	0	A normal joint recess
	1	Distension of the joint recess between the head talus and the navicular bone that leads to a change from the angle/V-shaped recess to a minimally distended recess
	2	Convex distension of the joint recess extending no more than 50% over either the head of the talus or the navicular bone
	3	Convex distension of the joint recess extending more than 50% over either the head of the talus or the navicular bone
STJ	0	Normal fat into the joint space leading to an angle/V-shaped recess
	1	Distension of the joint recess between talus and calcaneus, that leads to change from the angle/V-shaped recess to a minimally distended recess
	2	Convex distension of the joint recess extending no more than 50% over either of the visible portion of the bony landmarks ^b
	3	Convex distension of the joint recess that extends over more than 50% of either of the visible portion of the bony landmarks ^b
Tendon	0	Tendon sheath adjacent to the tendon fibres with no abnormal distention
	1	Presence of near or completely circumferential (to differentiate from physiologic localized pocket of fluid) hypochoic or anechoic thickened tissue with or without fluid within the tendon sheath that is seen in two perpendicular planes

^a The Power Doppler scoring system was adopted from Ting *et al.* [13] and applied to the Doppler signals within the synovial recess and SH only. Briefly, grade 0: shows the presence of no signal, grade 1: 1–3 signals within the area of SH only, grade 2: >3 signals or confluent signals present in <50% of the area of SH, grade 3: confluent signals are present in >50% of the area of SH.

^b ‘Bone profile’ instead of ‘bony landmarks’ may also be used. TTJ: tibiotalar joint; STJL: subtalar joint; SH: synovial hypertrophy.

Table 3. Inter-reader reliability of the ankle scoring systems

Area	B-mode ICC (95% CI ^a)	Power Doppler–mode ICC (95% CI ^a)
Midline TTJ	0.93 (0.92, 0.94)	0.93 (0.90, 0.95)
Lateral percent TTJ	0.92 (0.89, 0.94)	*
Medial percent TTJ	0.91 (0.88, 0.94)	*
TNJ	0.86 (0.81, 0.90)	0.88 (0.82, 0.92)
Anterior aspect STJ (from medial view)	0.91 (0.88, 0.93)	0.87 (0.80, 0.92)
Posterior aspect STJ (from lateral view)	0.91 (0.88, 0.93)	0.96 (0.94, 0.97)
Tendons	0.96 (0.95, 0.97)	0.99 (0.98, 0.99)

^a ICC was based on a two-way random effects model for a single measure. Excellent ICC was defined to be between 0.75–1.00, good 0.60–0.74, fair 0.40–0.59 and poor <0.4.

* Unable to calculate given scarce availability of US images positive for Power Doppler in this location. TTJ: tibiotalar joint; TNJ: talonavicular joint; STJ: subtalar joint; ICC: intraclass correlation coefficient.

distention’ or ‘minimal extension’ also contributed to the variability of grading the visualized pathology. Therefore, it was decided to try a percentage system as well as a system dividing the entire area into thirds to improve reliability. The presence of a significant amount of cartilage on the TTJ and STJ views of young children was noted to confound the scoring. Therefore, we determined that prior knowledge of the ankle anatomy, normal sonoanatomy in children and clear delineation of cartilage outlines are of utmost importance to support recognition of cartilage and pathology scoring.

The second scoring exercise included a total of 221 images obtained in children aged 2–18 years (Supplementary Table S3, available at *Rheumatology* online). This exercise revealed excellent agreement for all anatomic sites evaluated by our scanning protocol (Table 3). For the TTJ this preliminary scoring system included both a percentage and a division into thirds of the area system to score the degree of pathology. While both systems demonstrated excellent reliability, we chose to adopt the percentage system as it was more consistent with the scoring system we implemented for the STJ and

TNJ. Reliability for the Power Doppler view of the lateral and medial aspect of the TTJ were not assessed given the limited availability of US images positive for Power Doppler on these locations.

Discussion

Identification of ankle pathology in children has major therapeutic and prognostic implications. However, the clinical assessment of ankle arthritis in children is challenging [12]. Identifying the involved compartments is crucial to understanding the full extent of pathology, particularly when considering proper needle placement for IA injection. In this study, we developed a comprehensive and feasible paediatric ankle US scanning protocol as well as a novel and reliable scoring system for the assessment of ankle arthritis/tenosynovitis in children.

Using a standardized approach, we propose an all-inclusive and extensive scanning protocol for key ankle structures affected in JIA including the TTJ, STJ and TNJ as well as the

anterior, medial and lateral tendon compartments. A key feature of the CARRA scanning protocol is the emphasis given to dynamic US examination while providing standardization of the musculoskeletal examination. For example, scanning of the STJ requires dynamic examination starting from a neutral position followed by eversion and inversion of the ankle while directly observing the change on the STJ recess. For scoring purposes, the area of maximum pathology is identified during this dynamic process, and the sonographer obtains a static image in the area of greatest pathology. The anatomical landmarks should be visible in the chosen image. This will then permit reliable scoring while still capturing the site of maximum pathology. The final CARRA ankle scanning protocol is comprehensive, yet it can be feasibly performed in clinical practice. The STJ is a complex joint with both an anterior and a posterior component, and future studies including a comparison with MRI will need to determine whether this protocol adequately captures pathology in this joint.

Given the distinctive anatomy and the unique pathology of each of the ankle joints (TTJ, STJ, TNJ) a specific definition for the various scores was developed for each joint and view. The TTJ scoring system proved to be reliable when applied to the midline, medial or lateral aspects of the TTJ. This is important to address the different distribution of TTJ pathology among patients [39]. Because it is difficult to diagnose tenosynovitis clinically, a dedicated scanning and scoring protocol for each ankle tendon compartment was created. This additional tendon scoring did not add a significant amount of time to the live exercises, as the tendons are often visible in certain views of the joint with only minimal adjustments necessary if they are not readily visible.

Essential work on joint-specific scanning protocols and scoring systems for the assessment of arthritis in the paediatric joints, including the ankle, was recently published [14, 46]. These scoring systems showed excellent to good inter-rater reliability for the ankle joints when applied by a limited number of scorers and in a limited number of views. The proposed CARRA ankle scanning protocol and scoring system has several advantages. First, the reliability data of the CARRA ankle scoring system was acquired in a larger group of participants ($n = 16$ participants) with varying levels of expertise (1–14 years of experience in MSUS), who are affiliated with 11 North American academic institutions, thus demonstrating good external validity. All participants had undergone standard and comprehensive training and even when in practice for a shorter time afterwards already had accumulated significant experience. These prerequisites ensured a standardized level of knowledge among the participants. All of the participants in this study were paediatric rheumatology providers. No radiologists participated in this work. This was largely due to the fact that CARRA membership and the US subgroup through which this work was organized mainly consists of paediatric rheumatologists. We did not think the limited nature of the participant's medical subspecialties was a weakness, as all members had MSUS scanning expertise in the ankle. Furthermore, the clinical perspective of a paediatric rheumatology provider may be helpful in the clinical application of scanning and scoring protocols. It is possible that participants with more scanning experience may have influenced the less experienced participants. On the other hand, limiting the participants to those with more scanning experience may create a system geared more towards an expert level, which would limit its applicability in general clinical practice. In a relatively large group the dominance of single experts may also be less significant.

Overall, we found that the heterogeneity of the group ensured a solid minimum standard, which we think is an advantage. It is expected that the CARRA ankle scanning protocol and scoring system can easily be adopted in clinical practice to facilitate an inclusive and systematic evaluation of the ankle. The protocol can be applied to all paediatric age groups as we assessed its reliability in a total of 345 ankle images acquired in children aged 2–16 years. This is important given the variable appearance of the cartilage and bony contours in children of different skeletal maturation. In addition to examining the reliability of the MSUS scoring system for the TTJ, TNJ and STJ, the CARRA scoring system also evaluated the inter-rater reliability for the tendon scoring system. Although a single view was used for scoring, pathology always needed to be confirmed in a second plane. For example, we used the longitudinal view in the TTJ because it allowed evaluation of the complete synovial recess no matter how large the distension is. This would be more difficult in the transverse view. A limitation for this study is that we did not include the posterior aspect of the ankle or the entheses in our evaluation. It is not clear at this point whether the assessment of the posterior aspect of the ankle provides additional information that cannot be obtained from the views proposed, but this will need to be explored further. Data for US enthesitis in children is limited and no paediatric-specific validated definitions for pathology are available currently. Future studies comparing all the recently described ankle scoring protocols will have the potential to strengthen a scoring system that can be used universally.

Studies assessing the correlation of the scoring systems with clinical, imaging and biologic markers of disease activity are needed to further validate these paediatric-specific scoring systems. Validation studies will support the systematic use of MSUS as a complementary tool to clinical assessment of arthritis, in terms of allowing earlier diagnosis and more careful monitoring of disease activity in JIA. The CARRA ankle MSUS scanning protocols and scoring systems may serve as an assessment tool and outcome measure for both clinical and research applications seeking to improve the outcomes of children with JIA.

Supplementary data

Supplementary data are available at *Rheumatology* online.

Data availability statement

The data underlying this article will be shared on reasonable request to the corresponding author.

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References

- Ravelli A, Martini A. Juvenile idiopathic arthritis. *Lancet* 2007; 369:767–78.
- Hemke R, Nusman CM, van der Heijde DM *et al.* Frequency of joint involvement in juvenile idiopathic arthritis during a 5-year follow-up of newly diagnosed patients: implications for MR imaging as outcome measure. *Rheumatol Int* 2015;35:351–7.
- Javadi S, Kan JH, Orth RC, DeGuzman M. Wrist and ankle MRI of patients with juvenile idiopathic arthritis: identification of unsuspected multicompartmental tenosynovitis and arthritis. *AJR Am J Roentgenol* 2014;202:413–7.
- Lanni S, Marafon DP, Civino A *et al.* Comparison between clinical and ultrasound assessment of the ankle region in juvenile idiopathic arthritis. *Arthritis Care Res (Hoboken)* 2021;73:1180–6.
- Romeo DM, Lucibello S, Musto E *et al.* Assessing joint hypermobility in preschool-aged children. *J Pediatr* 2016;176:162–6.
- Woolnough L, Pomputius A, Vincent HK. Juvenile idiopathic arthritis, gait characteristics and relation to function. *Gait Posture* 2021;85:38–54.
- Kuntze G, Nesbitt C, Nettel-Aguirre A *et al.* Gait adaptations in youth with juvenile idiopathic arthritis. *Arthritis Care Res (Hoboken)* 2020;72:917–24.
- Hendry GJ, Shoop-Worrall SJ, Riskowski JL *et al.* Prevalence and course of lower limb disease activity and walking disability over the first 5 years of juvenile idiopathic arthritis: results from the childhood arthritis prospective study. *Rheumatol Adv Pract* 2018;2:rky039.
- Esbjornsson AC, Aalto K, Brostrom EW *et al.*; Nordic Study Group of Paediatric Rheumatology (NoSPeR). Ankle arthritis predicts polyarticular disease course and unfavourable outcome in children with juvenile idiopathic arthritis. *Clin Exp Rheumatol* 2015;33:751–7.
- Colebatch-Bourn AN, Edwards CJ, Collado P *et al.* EULAR-PRES points to consider for the use of imaging in the diagnosis and management of juvenile idiopathic arthritis in clinical practice. *Ann Rheum Dis* 2015;74:1946–57.
- Pascoli L, Wright S, McAllister C, Rooney M. Prospective evaluation of clinical and ultrasound findings in ankle disease in juvenile idiopathic arthritis: importance of ankle ultrasound. *J Rheumatol* 2010;37:2409–14.
- Rooney ME, McAllister C, Burns JF. Ankle disease in juvenile idiopathic arthritis: ultrasound findings in clinically swollen ankles. *J Rheumatol* 2009;36:1725–9.
- Ting TV, Vega-Fernandez P, Oberle EJ *et al.*; Childhood Arthritis and Rheumatology Research Alliance Juvenile Idiopathic Arthritis Ultrasound Workgroup. Novel ultrasound image acquisition protocol and scoring system for the pediatric knee. *Arthritis Care Res (Hoboken)* 2019;71:977–85.
- Vega-Fernandez P, Ting TV, Oberle EJ *et al.* CARRA musculoskeletal ultrasound workgroup. Musculoskeletal ultrasound in childhood arthritis limited examination: A comprehensive, reliable, time-efficient assessment of synovitis. *Arthritis Care Res (Hoboken)* 2021; <https://doi.org/10.1002/acr.24759>.
- Terslev L, Naredo E, Keen HI *et al.* The OMERACT stepwise approach to select and develop imaging outcome measurement instruments: the musculoskeletal ultrasound example. *J Rheumatol* 2019;46:1394–400.
- Towbin R, Dunbar JS, Towbin J, Clark R. Teardrop sign: plain film recognition of ankle effusion. *AJR Am J Roentgenol* 1980;134:985–90.
- Fornage BD. Achilles tendon: US examination. *Radiology* 1986; 159:759–64.
- Fornage BD, Rifkin MD. Ultrasound examination of the hand and foot. *Radiol Clin North Am* 1988;26:109–29.
- Nazarian LN, Rawool NM, Martin CE, Schweitzer ME. Synovial fluid in the hindfoot and ankle: detection of amount and distribution with US. *Radiology* 1995;197:275–8.
- Lehtinen A, Paimela L, Kreula J, Leirisalo-Repo M, Taavitsainen M. Painful ankle region in rheumatoid arthritis. Analysis of soft-tissue changes with ultrasonography and MR imaging. *Acta Radiol* 1996;37:572–7.
- Fessell DP, Vanderschueren GM, Jacobson JA *et al.* US of the ankle: technique, anatomy, and diagnosis of pathologic conditions. *Radiographics* 1998;18:325–40.
- Wang SC, Chhem RK, Cardinal E, Cho KH. Joint sonography. *Radiol Clin North Am* 1999;37:653–68.
- Galluzzo E, Lischi DM, Taglione E *et al.* Sonographic analysis of the ankle in patients with psoriatic arthritis. *Scand J Rheumatol* 2000;29:52–5.
- Rawool NM, Nazarian LN. Ultrasound of the ankle and foot. *Semin Ultrasound CT MR* 2000;21:275–84.
- Backhaus M, Burmester GR, Gerber T *et al.*; Working Group for Musculoskeletal Ultrasound in the EULAR Standing Committee on International Clinical Studies including Therapeutic Trials. Guidelines for musculoskeletal ultrasound in rheumatology. *Ann Rheum Dis* 2001;60:641–9.
- Farley FA, Kuhns L, Jacobson JA, DiPietro M. Ultrasound examination of ankle injuries in children. *J Pediatr Orthop* 2001;21:604–7.
- Schmidt WA, Schmidt H, Schicke B, Gromnica-Ihle E. Standard reference values for musculoskeletal ultrasonography. *Ann Rheum Dis* 2004;63:988–94.
- Bianchi S, Martinoli C, Gagnot C, De Gautard R, Meyer JM. Ultrasound of the ankle: anatomy of the tendons, bursae, and ligaments. *Semin Musculoskelet Radiol* 2005;9:243–59.
- Riente L, Delle Sedie A, Iagnocco A *et al.* Ultrasound imaging for the rheumatologist V. Ultrasonography of the ankle and foot. *Clin Exp Rheumatol* 2006;24:493–8.
- Zukotynski K, Jarrin J, Babyn PS *et al.* Sonography for assessment of haemophilic arthropathy in children: a systematic protocol. *Haemophilia* 2007;13:293–304.
- Fessell DP, Jacobson JA. Ultrasound of the hindfoot and midfoot. *Radiol Clin North Am* 2008;46:1027–43, vi.
- Wakefield RJ, Freeston JE, O'Connor P *et al.* The optimal assessment of the rheumatoid arthritis hindfoot: a comparative study of clinical examination, ultrasound and high field MRI. *Ann Rheum Dis* 2008;67:1678–82.
- Naredo E, Rodriguez M, Campos C *et al.*; Ultrasound Group of The Spanish Society of Rheumatology. Validity, reproducibility, and responsiveness of a twelve-joint simplified power doppler ultrasonographic assessment of joint inflammation in rheumatoid arthritis. *Arthritis Rheum* 2008;59:515–22.
- Keshava S, Gibikote S, Mohanta A, Doria AS. Refinement of a sonographic protocol for assessment of haemophilic arthropathy. *Haemophilia* 2009;15:1168–71.
- Suzuki T, Tohda E, Ishihara K. Power Doppler ultrasonography of symptomatic rheumatoid arthritis ankles revealed a positive association between tenosynovitis and rheumatoid factor. *Mod Rheumatol* 2009;19:235–44.
- Magni-Manzoni S, Epis O, Ravelli A *et al.* Comparison of clinical versus ultrasound-determined synovitis in juvenile idiopathic arthritis. *Arthritis Rheum* 2009;61:1497–504.

37. Collado P, Naredo E, Calvo C *et al.*; ECO-JIA Study Group. Reduced joint assessment vs comprehensive assessment for ultrasound detection of synovitis in juvenile idiopathic arthritis. *Rheumatology (Oxford)* 2013;52:1477–84.
38. Martinoli C, Della Casa Alberighi O, Di Minno G *et al.* Development and definition of a simplified scanning procedure and scoring method for Haemophilia Early Arthropathy Detection with Ultrasound (HEAD-US). *Thromb Haemost* 2013;109:1170–9.
39. Suzuki T. Power Doppler ultrasonographic assessment of the ankle in patients with inflammatory rheumatic diseases. *World J Orthop* 2014;5:574–84.
40. Alsuwaidi M, Ehrenstein B, Fleck M, Hartung W. Asymptomatic versus symptomatic ankle joints in rheumatoid arthritis: a high-resolution B-mode and power Doppler ultrasound study. *Arthritis Care Res (Hoboken)* 2016;68:861–4.
41. Lanni S, Bovis F, Ravelli A *et al.* Delineating the application of ultrasound in detecting synovial abnormalities of the subtalar joint in Juvenile Idiopathic Arthritis. *Arthritis Care Res (Hoboken)* 2016; 68:1346–53.
42. Moller I, Janta I, Backhaus M *et al.* The 2017 EULAR standardised procedures for ultrasound imaging in rheumatology. *Ann Rheum Dis* 2017;76:1974–9.
43. Roth J, Ravagnani V, Backhaus M *et al.*; OMERACT Ultrasound Group. Preliminary definitions for the sonographic features of synovitis in Children. *Arthritis Care Res (Hoboken)* 2017;69: 1217–23.
44. Collado P, Windschall D, Vojinovic J *et al.*; OMERACT Ultrasound Subtask Force on Pediatric. Amendment of the OMERACT ultrasound definitions of joints' features in healthy children when using the DOPPLER technique. *Pediatr Rheumatol* 2018;16:23.
45. Cicchetti DV. Multiple comparison methods: establishing guidelines for their valid application in neuropsychological research. *J Clin Exp Neuropsychol* 1994;16:155–61.
46. Sande NK, Bøyesen P, Aga AB *et al.* Development and reliability of a novel ultrasonographic joint-specific scoring system for synovitis with reference atlas for patients with juvenile idiopathic arthritis. *RMD Open* 2021;7:e001581.