Ankle Disease in Juvenile Idiopathic Arthritis: Ultrasound Findings in Clinically Swollen Ankles

MADELEINE E. ROONEY, CATHERINE McALLISTER, and JAMES F.T. BURNS

ABSTRACT. ***Objective.*** The ankle joint is frequently involved in juvenile idiopathic arthritis (JIA), but it is unclear whether this is predominantly due to synovitis, tenosynovitis, or both. We performed clinic-based ultrasound examination to assess the prevalence of synovitis and tenosynovitis in children with JIA felt clinically to have active inflammatory disease of the ankle.

Methods. Thirty-four patients with 49 clinically swollen ankles were studied (19 polyarticular JIA, 13 oligoarticular JIA, 1 systemic JIA, 1 psoriatic JIA). All cases had at least one clinically swollen ankle joint. The children were assessed clinically and had ultrasound examination during routine clinic appointments.

Results. We found 71% of ankles had tenosynovitis and 39% had tenosynovitis alone. Only 29% of swollen ankles had a tibiotalar effusion alone. We found 33% had both tenosynovitis and a tibiotalar effusion. When results were analyzed by JIA subtype, we found 81% of oligoarticular JIA ankles had medial ankle tenosynovitis but only 19% had tibiotalar effusion alone. There was a significant difference between JIA subgroups for the frequency of occurrence of medial ankle tenosynovitis ($p = 0.048$) and lateral ankle tenosynovitis ($p = 0.001$).

Conclusion. The tibiotalar joint was not involved in 39% of the swollen ankles; and tenosynovitis, sometimes in isolation, was the dominant finding. This has implications for therapeutic intervention and also for an improved classification of children with JIA, especially with ankle involvement. (J Rheumatol First Release May 1 2009; doi:10.3899/jrheum.080508)

Key Indexing Terms:
ULTRASOUND JUVENILE IDIOPATHIC ARTHRITIS ANKLE TENOSYNOVITIS

Juvenile idiopathic arthritis (JIA) is the commonest form of arthritis in children under the age of 16 years. The current International League of Associations for Rheumatology (ILAR) classification has divided the condition into 7 clinical subgroups[1]. While a variety of extraarticular features are recognized, the presence of arthritis is common to all. For some subgroups — oligoarticular JIA (oligo-JIA), extended oligoarticular and polyarticular JIA (poly-JIA), rheumatoid factor-positive and negative — inclusion is determined by the number of involved joints with arthritis. Arthritis is defined in the criteria as "swelling within a joint, or limitation of the range of joint movement with joint pain or tenderness, which persists for at least six weeks, is observed by a physician and is not due to primarily mechanical disorders." For all children with JIA the knee joint is the most commonly affected, in 77%, followed by the ankle joint, where some 58% of children will have involvement[2].

From the School of Medicine, Queen's University; and the Belfast Hospitals Trust, Belfast, United Kingdom.

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J.F.T. Burns, MB, BCH, BAO, Belfast Hospitals Trust; M.E. Rooney, MB, BCH, BAO, MD, School of Medicine, Queen's University; C. McAllister, BSc, Belfast Hospitals Trust.

Address reprint requests to Dr. M. Rooney, Belfast Hospitals Trust, Musgrave Park Hospital, Rheumatology, Stockman's Lane, Belfast, Northern Ireland BT9 7JB. E-mail: m.rooney@qub.ac.uk

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Much has been written about knee joint disease, including imaging and response to treatment. However, the ankle joint, despite being the second most commonly involved joint, has been relatively ignored. There are few studies that describe rates of tenosynovitis at the ankle[3]. Numerous reports describe radiological findings in JIA ankle disease, although little is published on ankle tenosynovitis and the clinico-radiological correlation.

Standard management of ankle disease is the use of intraarticular corticosteroids (IAS), with or without methotrexate, depending on disease subtype. Patients with oligo-JIA tend to be managed with IAS as first-line therapy, whereas children with poly-JIA require systemic treatment.

Several investigators have studied the efficacy of IAS in the treatment of JIA. All have commented on the efficacy of this approach, particularly using triamcinolone hexacetonide in inducing longterm remission[4-6]. Most of these studies give detailed results only for knee synovitis.

We have noted the relatively poor response to IAS in ankle disease. Our observations concur with others who noted that the relative risk of recurrence of ankle synovitis following IAS was twice that observed in knees[5].

Since many of these children are young and thus require a general anesthetic for this procedure, these results are disappointing. We therefore hypothesized that the relatively poor response was due to incorrect identification of synovi-

tis in the ankle region. The ankle is anatomically and mechanically complex, with large tendon groups adjacent to the joints. These structures represent potentially important sites that could become inflamed. Ankle examination can be difficult in small children due to the distribution of fat, the lack of prominent anatomical landmarks, and on occasions, poor cooperation with physical examination, thus making the distinction between joint synovitis and tenosynovitis difficult. We therefore used ultrasound to evaluate the anatomical basis for clinically detected ankle joint swelling in JIA.

MATERIALS AND METHODS

We performed clinic-based ultrasound examination to assess the prevalence of synovitis and tenosynovitis in children with JIA felt clinically to have active inflammatory disease of the ankle. Our data were collected for the purpose of an audit to assess the effectiveness of ultrasound in our routine clinical practice. The children included in the audit were selected if they had clinically swollen ankles irrespective of their diagnosis. Children without clinically swollen ankles were excluded. The children were assessed clinically, then had ultrasound examination during the clinic by a pediatric rheumatologist trained in musculoskeletal ultrasound (MER). Scans were undertaken using a Sonosite 180 Plus (L38 5–10 MHz linear transducer) or Esaote MyLab 25 scanner (LA523E 7.5–12 MHz linear transducer). Scans of the tibiotalar joint were undertaken using longitudinal and transverse positions. The medial and lateral compartments were scanned parallel and perpendicular to the tendon groups. Synovitis and tenosynovitis (as defined by the OMERACT 7 consensus statement[7]) were identified and recorded for the tibiotalar joint, medial and lateral tendon groups. The other major tendon groups and tendon insertions of the ankle were scanned, and power Doppler imaging was used to identify areas of inflammation as part of a routine ultrasound examination, but the findings were not included in this audit. Figures 1 and 2 show examples of normal and abnormal tibiotalar joints. Figure 3 shows an example of tibialis posterior tenosynovitis with power Doppler signal (demonstrating the presence of increased capillary blood flow).

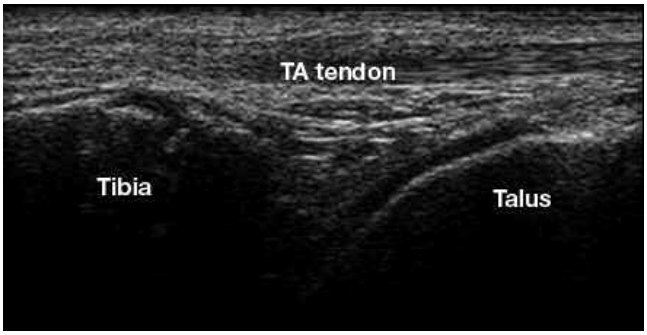

Figure 1. Normal tibiotalar joint.

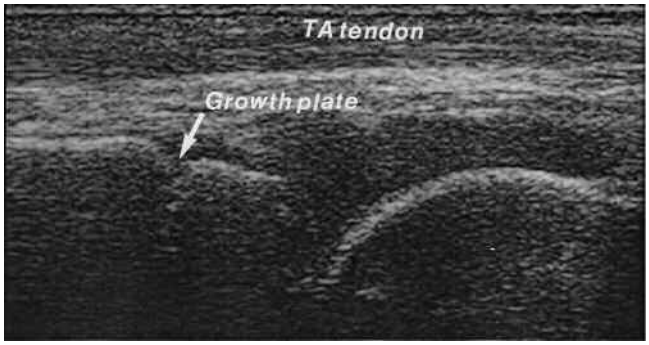

Figure 2. Tibiotalar joint with synovitis.

RESULTS

Thirty-four children with 49 swollen ankles were studied; demographic details are summarized in Table 1. Nineteen children had poly-JIA, 13 had oligo-JIA, 1 had systemic JIA, and 1 psoriatic JIA. Twenty-one of the 34 children were female. The mean age of children in the poly-JIA group on the day of examination was 11.3 years; as expected, the mean age of the oligo-JIA group was lower, 8.6 years.

Clinical findings varied from minimal swelling with pain to marked swelling around the joint. In this audit, no attempt was made to identify the site of inflammation as associated with joint or tendons. Historically, "ankle" pain and swelling have been assumed to be tibiotalar disease, with the occasional exception when tendon involvement is grossly obvious.

The combined results for all JIA subgroups are shown in Figure 4. Of the 49 clinically involved ankles, tibiotalar synovitis was observed in 30 (61%) joints. However, tibiotalar disease was present without tendon involvement in only 14 joints (29%). In contrast, tenosynovitis was a more frequent finding, observed in 35 ankles (71%). In 19 of these (39%), tenosynovitis was present in the absence of tibiotalar disease.

The findings were analyzed according to disease subtype (Table 2). The incidence of isolated tibiotalar involvement in the oligo-JIA subgroup was only 19%; it was found slightly more frequently in the poly-JIA group, 32%. However, tenosynovitis with no tibiotalar involvement was much more common in oligo-JIA (56% vs 29%, respectively). In particular, tenosynovitis of the medial ankle tendons was an extremely common finding, observed in 81% of oligo-JIA ankles, compared to 52% with poly-JIA. In contrast, lateral ankle tenosynovitis was found very rarely in oligo-JIA (6%), but was still a relatively common finding in poly-JIA (55%). There was a significant difference between JIA subgroups for the frequency of occurrence of medial ankle tenosynovitis ($p = 0.048$) and lateral ankle tenosynovitis ($p = 0.001$). Our findings indicate that tibiotalar disease in isolation is much less common than tenosynovitis alone or in conjunction with tibiotalar disease.

DISCUSSION

Our findings showed an unexpectedly high degree of tenosynovitis, which was often the only abnormality. This could be important for the diagnosis and therapy of JIA, and potentially for classification purposes. Very few publications mention the occurrence of tenosynovitis at the ankle in JIA; we therefore feel that it occurs more frequently than is clinically recognized.

Our findings raise a number of issues in relationship to JIA. First, they probably explain the poor outcome following IAS therapy for ankle disease in JIA. Historically, clini-

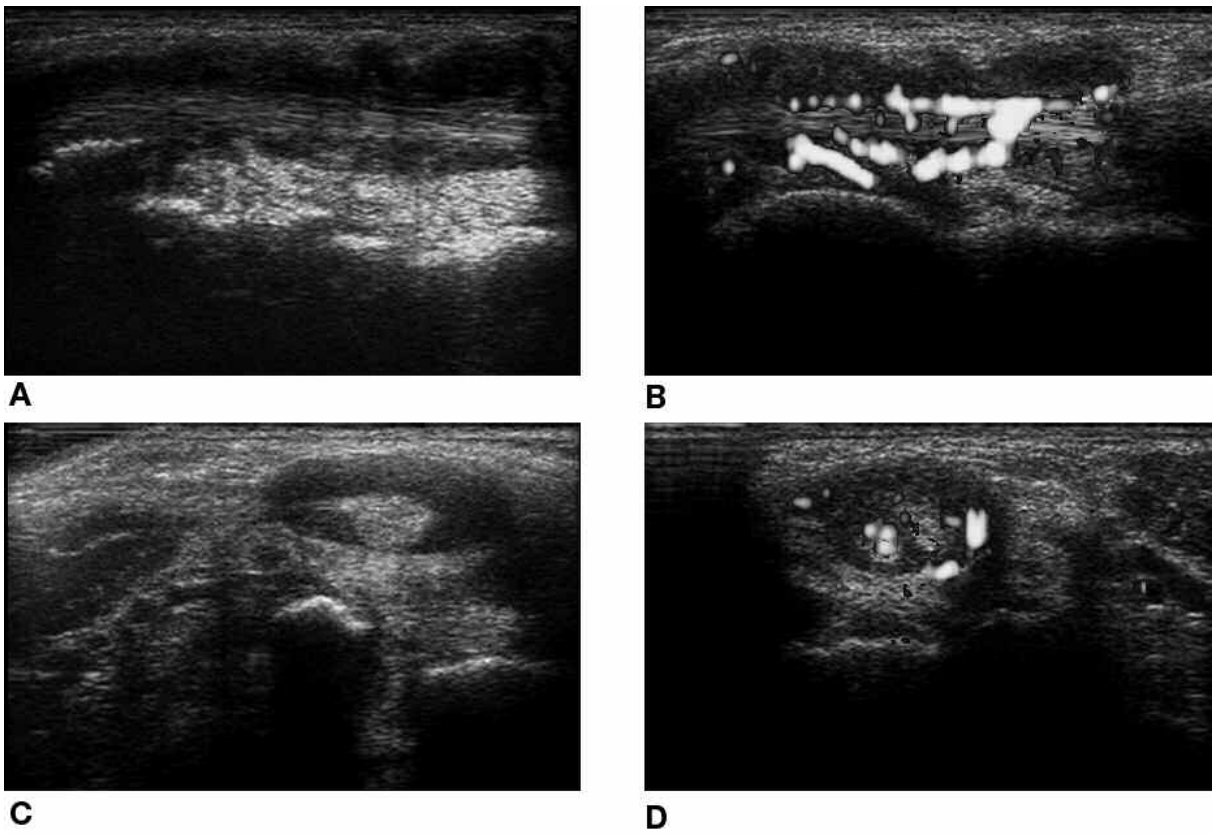

Figure 3. Tibialis posterior, longitudinal scan with tenosynovitis. B. Power Doppler image of the same tendon shows increased capillary blood flow. C. Transverse scan of tibialis posterior with tenosynovitis. D. Power Doppler image of panel C.

Table 1. Demographic data of 34 children with 49 swollen ankles.

Diagnosis	No (%)	Mean Age, yrs (range)	M/F
Poly JIA	19 (56)	11.3 (3.9–17.3)	7/12
Oligo JIA	13 (38)	8.6 (2.75–16.5)	4/9
Psoriatic JIA	1 (3)	16	1/0
Systemic JIA	1 (3)	4.5	1/0

cal practice has been to inject the tibiotalar joint when ankle swelling is present, injecting tendon sheaths only when these are thought to be involved clinically. Ultrasound and magnetic resonance imaging (MRI) scans were reserved for assessment of recurrent joint swelling or subtalar involvement. Our findings would strongly suggest that ultrasound should be performed prior to all ankle injections, as isolated tibiotalar joint disease is relatively uncommon and tenosynovitis is extremely common. Since undertaking a procedure of musculoskeletal ultrasound our clinical impression has been of improved outcomes from targeted steroid injections; we plan a prospective study to assess the response to treatment and to determine the accuracy of clinical assessment in determining the site of inflammation.

Studies have compared the clinical findings with radiological findings for ankle swelling using MRI and ultrasound in adult inflammatory arthritis[8]. Little investigation has been done on the radiological to clinical correlation in JIA[9]. Of the few studies published, none was found that

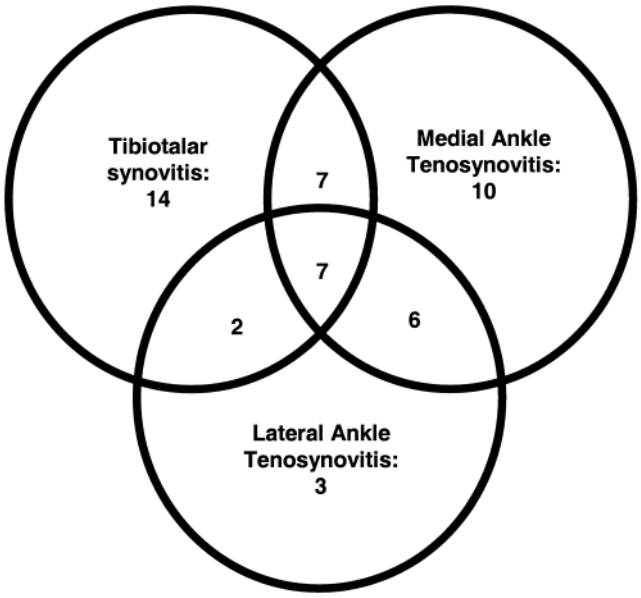

Figure 4. Venn diagram shows distribution of ultrasound findings for all JIA subgroups (n = 49).

Table 2. Results by JIA subtype.

Diagnosis	Oligo JIA, 16 Ankles, n (%)	Poly JIA, 31 Ankles, n (%)
Tibiotalar synovitis only	3 (19)	10 (32)
Tenosynovitis only	9 (56)	9 (29)
Tibiotalar synovitis and tenosynovitis	4 (25)	12 (39)
Tibiotalar synovitis total	7 (43)	22 (71)
Medial ankle tenosynovitis	13 (81)	16 (52)
Lateral ankle tenosynovitis	1 (6)	17 (55)

specifically assessed rates of tenosynovitis with ultrasound. Our findings are consistent with previous reports where clinically swollen feet and ankles in patients with JIA were evaluated by MRI; 77% of the ankles/feet had tenosynovitis identified on MRI, while synovitis was found in only 59% of cases[3]. However, ultrasound is easier to undertake in the pediatric clinic and it can be used to guide steroid injections directly.

With training and practice, musculoskeletal ultrasound skills are attainable by most clinicians. Ultrasound is well tolerated by children, avoiding the need for MRI scans in many cases. MRI scans in young children require a general anesthetic, often a considerable delay, with associated stress for both children and parents. Ultrasound obviates all this and provides real-time images of the joints and tendons. More powerful scanners provide extremely useful power Doppler signals of synovitis and enthesitis. While ultrasound evaluation needs to be standardized for detailed evaluations of pediatric joints, in its present state it provides very useful findings.

There are limitations with ultrasound; imaging of the subtalar joint is difficult and sonographers continue to debate whether we can accurately visualize it. There are few reports on ultrasound assessment of this area[10,11]. A study of adult rheumatoid arthritis hindfoot MRI imaging has shown that tibialis posterior tendon involvement occurs frequently without subtalar joint complex involvement[12].

A substantial number of children with 3 or 4 joints involved would be categorized as extended oligoarticular or polyarticular should they develop ankle disease. Our study revealed that 61% of cases had tibiotalar joint involvement. Thus almost 40% of children would be described as having ankle involvement, when in fact they have tenosynovitis alone. The ILAR classification requires joint involvement, not tendon involvement[1]. As such, we postulate that a substantial number of children may be wrongly classified as having extended oligoarticular or polyarticular disease, when they have oligoarticular disease with tenosynovitis. This might go some distance in explaining the relatively poor correlation between clinical subtype and genetic phenotype[13]. If children have been misclassified as having polyarticular JIA, they are likely to be given systemic treatment when perhaps it is not always necessary.

Our study had some limitations. All observed ankle swelling was assumed to be tibiotalar effusion for the purpose of this audit, while some may clinically have had tenosynovitis. It should be noted that we were surprised on many occasions that the site of inflammation detected with ultrasound was not where it would appear to be on clinical assessment. Also, we did not comment on the presence or absence of clinical subtalar involvement, since currently, ultrasound cannot reliably detect an effusion or synovitis in this joint. The clinician who performed the ultrasound examinations had performed the clinical examination prior to the ultrasound examination; it is possible that this could have influenced the ultrasound interpretation. In order to address this, we plan a prospective study where the ultrasound examination is performed by an ultrasonographer blinded to the clinical findings and history; prior to the ultrasound examination we will attempt to clinically define the site of inflammation.

Tenosynovitis was found very frequently in our study, with especially high rates of tibialis posterior tenosynovitis in oligo-JIA. McGonagle, *et al* have shown that fibrocartilagenous areas where tendons change direction around a bone are subject to similar compressive and shearing forces to those found at entheseal insertions[14,15]. These structures have been termed "functional entheses" and it has been suggested that this may account for the flexor tenosynovitis commonly found in psoriatic arthritis[16]. Therefore it is possible that the high frequency of ankle joint tenosynovitis is at the microanatomical level related to the seronegative arthritis spectrum. These findings therefore could provide a platform for a better anatomical-based classification of JIA.

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