Short communication

Juvenile idiopathic oligoarthritis: analysis of 42 cases in Jordan

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التهاب المفاصل القليلة المجهول السبب في اليافعين: تحليل 42 حالة في الأردن

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الخلاصة: قام الباحثون بمراجعة استعادية لملفّات 42 مريضاً منهم 9 ذكور، و33 أنثى، مـمَّن شُخِّصَ التهاب المفاصل القليلة اليَفَعي المجهول السبب لديهم في عيادة روماتيزم طب الأطفال في مركز الملك حسين الطبي في ما بين تموز/يوليو 1995 وتشرين الأول/أكتوبر 2004. وتراوَحَتْ أعمار هؤلاء المرضى بين 1.2 و15.2 سنة، بمتوسط 3.6 سنة (الانحراف المعياري 2.4). وقد مثّلت الإناث 80٪ بين من هم دون سن السادسة. وكان معظم المرضى (64٪) يعانون من التهاب المفاصل الأحادي، حيث كانت الركبة أكثر المفاصل تأثُّراً (71٪). كما ظهر التهاب المفاصل القليلة الممتد لدى اثنَيْن من المرضى، والتهاب العنبية العديم الأعراض لدى تسعة مرضى (21٪): ستة منهم من الإناث (P = 0.350)، وسبعة (77٪) دون سن الثالثة (P = 0.301)، وسبعة (77٪) لديهم أضداد النوى (P = 0.071). وكانت 18 أنثى (43٪) إيجابيات لأضداد النوى. وقد خضع معظم المرضى لإجراءات تشخيصية وعلاجية ومعالجة مطوّلة ولا داعي لها قبل التشخيص الفعلي.

ABSTRACT We conducted a retrospective chart review for all 42 patients, 9 males and 33 females, diagnosed with juvenile idiopathic oligoarthritis between July 1995 and October 2004 at the paediatric rheumatology clinic in King Hussein Medical Centre. Age range was 1.2–15.2 years, mean 3.6 (standard deviation 2.4) years. Of those < 6 years, 80% were females. Most patients (64%) had monoarthritis; the knee was the most common joint affected (71%). Two patients had developed extended oligoarthritis. Nine (21%) had asymptomatic uveitis: 6 females (*P* = 0.350), 7 (77%) < 3 years of age (*P* = 0.301) and 7 (77%) with antinuclear antibodies (ANAs) (*P* = 0.071). Eighteen (43%) females were positive for ANAs. Most patients had undergone unnecessary, lengthy work-up and treatment before diagnosis.

Arthrite juvénile idiopathique avec forme oligoarticulaire : analyse de 42 cas en Jordanie

RÉSUMÉ Nous avons effectué une analyse rétrospective des dossiers de l'ensemble des 42 patients, 9 garçons et 33 filles, chez lesquels avait été diagnostiquée une forme oligoarticulaire de l'arthrite juvénile idiopathique entre juillet 1995 et octobre 2004 dans le service de rhumatologie pédiatrique du Centre médical Roi Hussein. Les patients étaient âgés de 1,2 à 15,2 ans, âge moyen : 3,6 ans (écart type : 2,4 ans) et 80 % des moins de 6 ans étaient de sexe féminin. La plupart des patients (64 %) présentaient une atteinte monoarticulaire, le genou étant l'articulation la plus touchée (71 %). Deux patients avaient développé une forme oligoarticulaire étendue. Nous avons compté 9 cas (21 %) d'uvéite asymptomatique, 6 d'entre eux étant de sexe féminin (p = 0,350), 7 (77 %) âgés de moins de 3 ans (p = 0,301) et 7 (77 %) porteurs d'anticorps antinucléaires (ANA) (p = 0,071). Dix-huit (43 %) filles étaient ANA-positives. La majorité des patients a subi des examens et des traitements longs et inutiles avant que soit posé le diagnostic.

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Introduction

Juvenile idiopathic arthritis (JIA) is the most common chronic rheumatic illness in children [*1*]. The incidence ranges from 1/100 000 to 22/100 000, with a prevalence of 8/100 000 to 150/100 000 [*2*]. It is a significant cause of short-term and long-term disability [*3*]. The International League of Associations for Rheumatology (ILAR) classification of JIA includes systemic onset-JIA, oligoarticular, polyarticular rheumatoid factor (RF)-positive and RF-negative, enthesitis-related-arthritis and juvenile psoriatic arthritis [*4*].

The oligoarticular subtype is the most common; it comprises 50%–60% of cases [*5*]. It is diagnosed in patients with arthritis in less than 5 joints during the first 6 months of the disease [*6*]. Involvement of the large joints of the lower extremities such as knees and ankles is predominant [*7*]. Half of the patients with oligoarthritis has monoarticular onset affecting only the knee joint [*8*]. There is usually no associated systemic manifestation and patients function remarkably well [*9*]. Chronic uveitis, usually seen in ANA-positive oligoarthritis, is the most serious clinical problem [*10*]. Arthritis of the oligoarticular subtype might be persistent, but 50% of patients develop active arthritis of ≥ 5 joints after the first 6 months of disease and are considered to have extended oligoarticular JIA; 30% of patients with extended oligoarthritis develop this within 2 years of diagnosis, which confers a worse prognosis [*10*].

We describe the clinical profile, course and complications of juvenile idiopathic oligoarthritis in 42 children in King Hussein Medical Centre in Jordan. This will lead to improvements in improve early diagnosis, treatment and recognition of potentially serious complications such as uveitis.

Methods

A retrospective chart review for all patients who received the diagnosis of juvenile idiopathic oligoarthritis between July1995 and October 2004 at the paediatric rheumatology clinic in King Hussein Medical Centre. This is the largest referral hospital in Jordan; it serves 40% of the population, with 200 000 visits per year by children.

The series consisted of 42 children under 16 years of age. Diagnosis of juvenile idiopathic oligoarthritis was made according to the ILAR classification [*1*]. The demographic features of the patients, joint distribution at presentation, course, treatment and complications were recorded.

SPSS, version 10, was used for statistical analysis. Results were expressed as means unless otherwise stated. $P < 0.05$ was considered significant.

Results

Forty-two charts for 9 (21%) males and 33 (79%) females (female:male ratio 3.7:1) were reviewed retrospectively. The patients were aged between 1.2 years and 15.2 years (mean 3.6 years) at diagnosis; 23 (55%) were under 3 years of age. Females had younger age of onset: 80% of patients below the age of 6 years at presentation were females (Table 1). Poor compliance with treatment and physiotherapy was noted for 17 (40%) patients.

The most common joint affected was the knee 29 (69%), followed by ankle 14 (33%), elbow 9 (21%) and wrist 6 (14%). Involvement of the hip, cervical spine and small joints was not documented. At presentation, 27 (64%) patients had monoarthritis and 15 (36%) had involvement of 2–4 joints (Table 1).

Table 1 **Characteristics of 42 patients at King Hussein Medical Centre, Jordan diagnosed with juvenile idiopathic oligoarthritis, 1995–2004**

Variable	Patients	
	No.	%
Sex		
Male	9	21
Female	33	79
Age at diagnosis (years)		
< 3	23	55
≥ 3	19	45
Joints affected at presentation		
1	27	64
2–4	15	36
Knee	29	69
Ankle	14	33
Elbow	9	21
Wrist	6	14
Asymptomatic uveitis (n = 9)		
Males	3	33
Females	6	67[a]
< 3 years old	7	78[a]
ANAs	7	78[a]
≥ 1 complications	9	100

[a]*P > 0.05.*
ANAs = antinuclear antibodies.

The median delay in diagnosis was 26 months; patients presenting with monoarthritis generally experienced greater delay. All of those with monoarthritis were initially evaluated by an orthopaedic surgeon as acute septic arthritis and all joints were aspirated and treated with antibiotics for 3–8 weeks and were only referred if they did not respond, the condition recurred or more joints became involved.

Patients who presented with > 1 joint were treated as poststreptococcal reactive arthritis or acute rheumatic fever for extended periods (mean 13 weeks) before being referred to the paediatric rheumatology clinic.

Over a mean follow-up period of 4.2 years, 2 (5%) patients developed extended oligoarthritis. We found 18 girls (18%) were positive for antinuclear antibodies (ANAs) compared with 3 (33%) boys.

Intra-articular corticosteroid injection was prescribed for 24 (57%) patients; methotrexate was given to 2 (5%); 17 patients (40%) had responded to nonsteroidal anti-inflammatory drugs (NSAIDs).

Flexion deformity was documented in 2 (5%) patients; both had extended oligoarthritis.

Nine (21%) patients had asymptomatic uveitis: 6 (67%) were females ($P = 0.350$), 7 (78%) were under 3 years of age ($P = 0.301$) and 7 (78%) were positive for ANAs ($P = 0.071$). All the patients had severe inflammation at onset and presented late (mean diagnostic delay 23 months); mean duration of arthritis was 34 months. All had ≥ 1 complications of chronic uveitis: synechiae, band keratopathy, cataract and glaucoma. They showed either slow response to treatment or no response at all; 7 patients (78%) had systemic treatment, including oral prednisolone and/or weekly oral low dose of methotrexate; 4 (44%) underwent surgical treatment.

Discussion

New research suggests that oligoarticular-onset JIA is not a benign disease [*7*]. In this study, 21% of the patients with oligoarthritis had asymptomatic uveitis. This is consistent with the findings of an earlier report, which noted a prevalence of 15%–20% for asymptomatic uveitis in children with oligoarticular JIA [*8*].

The poor prognosis of uveitis in our patients could be related to early onset of uveitis (before the diagnosis of arthritis), initial severe response, chronicity (mean diagnosis delay was 23 months), high proportion of patients testing ANA-positive,

short duration of arthritis and female sex [*8–10*]. Poor compliance to treatment and follow-up and delay in initiation of systemic treatment might also have exacerbated the poor outcome of uveitis in our patients.

The risk factors for uveitis in our study were not statistically significant, most probably due to the small size of the sample. Large joints were the most frequently involved, similar to other reports [*8*]. Awareness of the general practitioners and paediatricians regarding early diagnosis of oligoarticular JIA was suboptimal, resulting in significant delays in diagnosis.

More patients in our cohort were treated with intra-articular corticosteroid injection [*11*]. Although 40% our patients were not compliant to treatment and physiotherapy, we did not encounter significant flexion deformity of joints as in other reports [*8,9*]. In the 2 patients who had flexion deformity, it was limited to a single joint.

In conclusion, we found that awareness with regard to early diagnosis of juvenile idiopathic oligoarthritis was suboptimal. Most patients had undergone lengthy and unnecessary diagnostic procedures and treatment. Oligoarticular JIA is no longer considered benign even though the outcome is favourable compared with other subtypes where uveitis is a potentially serious complication.

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