Physicians' and Parents' Ratings of Inactive Disease Are Frequently Discordant in Juvenile Idiopathic Arthritis

ALESSANDRO CONSOLARO, ROSA VITALE, ANGELA PISTORIO, BIANCA LATTANZI, NICOLINO RUPERTO, CLARA MALATTIA, GIOVANNI FILOCAMO, STEFANIA VIOLA, ALBERTO MARTINI, and ANGELO RAVELLI

ABSTRACT. ***Objective.*** To investigate discrepancies between physicians' and parents' ratings of inactive disease in children with juvenile idiopathic arthritis (JIA) and the determinants of the discrepancy.

Methods. Study data were obtained from the clinical database generated at the study unit. Each patient visit included a standardized assessment of JIA outcome measures. One visit for each patient was selected for analysis. Three definitions of inactive disease were applied to the data: a physician-based definition (physician global assessment = 0); a parent-based definition (parent global assessment = 0); and a formal definition, based on fulfillment of newly developed criteria for inactive disease in JIA.

Results. Of 1237 visits made by 537 patients that included both physician and parent global assessments, 265 fulfilled the physician-based definition and/or the parent-based definition of inactive disease. Concordance between physicians and parents in rating the disease as inactive was seen in 40% of the visits, whereas in 60% of visits the 2 assessments were discordant. Parents tended to disagree with physicians in rating the disease as inactive if the child had pain or functional impairment, whereas physicians tended to disagree with parents in the presence of active joint symptoms. Only 2/3 of the 79 visits that fulfilled the formal definition of inactive disease also met the parent-based definition of inactive disease.

Conclusion. We found frequent discordance between physicians' and parents' ratings of inactive disease in children with JIA, which suggests that the parent's rating of a child's disease activity should be considered for inclusion in the definition of clinical remission for JIA. (First Release July 1 2007; J Rheumatol 2007;34:1773–6)

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There has been a lack of standardized and widely accepted criteria for defining remission in juvenile idiopathic arthritis (JIA). Recently, preliminary criteria for clinical remission in this disease have been developed through an international collaborative effort (Table 1)[1]. These criteria require for a patient to be classified as having inactive disease that he/she has no joints with active arthritis, no systemic manifestations attributable to JIA, no active uveitis, normal acute-phase reactants, and a physician's global assessment of disease activity indicating no disease activity. The criteria for inactive disease must be met for a minimum of 6 continuous months while taking medication in order for the patient to be considered to be in a state of clinical remission with medication. When the same criteria are met for more than 12 continuous months while no longer taking medication, the patient can be classified as being in a state of clinical remission without medication. The criteria have undergone a thorough validation process[2], have been applied retrospectively in large series of patients with JIA[3], and are now scrutinized in prospective analyses[4].

As recommended by the investigators who guided the development process, the preliminary criteria represent the first step of a work in progress, and several issues need to be addressed before they gain widespread use[1]. One problem with the criteria is that they are based only on physician-centered measures and an acute-phase reactant, whereas patient self-reported and parent proxy-reported measures are not considered[4]. Although it is unclear whether and to what extent physicians and patients/parents agree in defining remission in JIA, a number of studies have shown that they often disagree in assessing different domains of disease status, such as pain or functional ability[5-7]. Analyses of correlation with

From the Istituto di Ricovero e Cura a Carattere Scientifico G. Gaslini, Genova; and Università degli Studi di Genova, Genova, Italy.

A. Consolaro, MD, Pediatric Resident; R. Vitale, MD, Pediatric Resident; B. Lattanzi, MD, Pediatric Resident; N. Ruperto, MD, Dirigente Medico; C. Malattia, MD, Research Fellow; G. Filocamo, MD, Research Fellow; S. Viola, MD, Dirigente Medico, Pediatria II, Istituto di Ricovero e Cura a Carattere Scientifico G. Gaslini; A. Pistorio, MD, PhD, Dirigente Medico, Servizio di Epidemiologia e Biostatistica, Direzione Scientifica, Istituto di Ricovero e Cura a Carattere Scientifico G. Gaslini; A. Martini, MD, Professor of Pediatrics; A. Ravelli, MD, Associate Professor of Pediatrics, Pediatria II, Istituto di Ricovero e Cura a Carattere Scientifico G. Gaslini, and Dipartimento di Pediatria, Università degli Studi di Genova.

Address reprint requests to Dr. A. Ravelli, Pediatria II, Istituto G. Gaslini, Largo G. Gaslini 5, 16147 Genova, Italy.
E-mail: angeloravelli@ospedale-gaslini.ge.it

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Table 1. Preliminary criteria for inactive disease and clinical remission of JIA.

Inactive disease

1. No joints with active arthritis*†
2. No fever, rash, serositis, splenomegaly, or generalized lymphadenopathy attributable to JIA
3. No active uveitis (to be defined)
4. Normal ESR or CRP (if both are tested, both must be normal)
5. Physician's global assessment of disease activity indicates no disease activity (i.e., best score attainable on the scale used)

Clinical remission

Two types of clinical remission are proposed:

1. Clinical remission on medication. The criteria for inactive disease must be met for a minimum of 6 consecutive months while the patient is on medication
2. Clinical remission off medication. The criteria for inactive disease must be met for a minimum of 12 consecutive months while off all antiarthritis medication

*As defined by American College of Rheumatology: A joint with swelling not due to bony enlargement or, if no swelling is present, limitation of motion accompanied either by pain on motion and/or tenderness. †Isolated finding of pain on motion, tenderness, or limitation of motion on joint examination may be present only if explained by either prior damage attributable to arthritis that is now considered inactive, or nonrheumatological reasons such as trauma. From Wallace, *et al.* J Rheumatol 2004;31:2290-4 (reference 4), with permission.

patient/parent-centered measures, such as overall well-being, pain, and health-related quality of life scales would add considerably to the overall construct validity of the criteria. These analyses will help verify whether the term clinical remission can have a clearly understood meaning for our patients with JIA and their parents.

We investigated the discrepancy between the physicians' and parents' ratings of inactive disease in children with JIA and attempted to identify factors explaining it.

MATERIALS AND METHODS

Study data sets. Study data were obtained from the clinical database generated at the study unit that included visits carried out on an in-hospital or outpatient basis between January 1992 and December 2006 in children fulfilling the International League of Associations for Rheumatology (ILAR) criteria for JIA[8]. Each visit included a standardized clinical assessment of the JIA outcome measures listed below.

Clinical assessment. At each visit, the following clinical assessments were made by the attending pediatric rheumatologist: physician's global assessment of overall disease activity (physician global assessment) measured on a 10 cm visual analog scale (VAS; 0 = no activity, 10 = maximum activity); count of swollen joints; count of joints with pain upon movement/tenderness (tender joints); count of joints with limited range of motion (restricted joints); and count of joints with active disease (defined as the count of joints with swelling or, if no swelling was present, with limitation of movement and either pain upon movement or tenderness). Joint counts were assessed in a total of 71 joints following a standardized methodology[9]. A parent of each child was asked to make a global assessment of the child's overall well-being (parent global assessment) on a 10 cm VAS (0 = very good, 10 = very poor), to rate the intensity of the child's pain (parent pain assessment) on a 10 cm VAS (0 = no pain, 10 = very severe pain), and to complete the Childhood Health Assessment Questionnaire (C-HAQ), Italian version[10] (0 = best, 3 = worst). Acute-phase reactants included the erythrocyte sedimentation rate (ESR) determined with the Westergren method and C-reactive protein (CRP) determined by nephelometry.

Definitions of inactive disease. Three possible definitions of inactive disease were applied to the data: (1) a physician-based definition, established when the physician global assessment was = 0; (2) a parent-based definition, when the parent global assessment was = 0; and (3) a formal definition, when the newly developed criteria for inactive disease in JIA (Table 1)[1] were fulfilled.

Statistics. All outcome measures were examined as categorical variables (i.e., as = 0 or > 0, negative/positive, or normal/abnormal), and were thus reported in terms of absolute frequencies and percentages. Comparison of categorical data was performed by means of the chi-square test, or Fisher's exact test in case of expected frequencies < 5. Bonferroni's adjustment was applied as a correction for multiple comparisons to explore post-hoc differences between pairs of patient groups. All statistical tests were 2-sided; a p value < 0.01 was considered statistically significant.

RESULTS

A total of 1818 visits made by 636 patients were identified. These visits/patients were representative of the whole spectrum of severity and duration of JIA. For 1237 visits made by 537 patients, both the physician global assessment and the parent global assessment were available. All visits were examined to identify those that fulfilled the physician-based definition of inactive disease (physician global assessment = 0), the parent-based definition of inactive disease (parent global assessment = 0), and the formal definition of inactive disease[1]. In case a patient had more than one visit that fulfilled the criteria, only one visit was selected for the analysis. We chose the first visit or, if the first visit had insufficient data, the subsequent visit for which more data were available.

Because we were interested in evaluating the concordance between physicians and the parents in rating the disease as inactive through their individual global assessment, we investigated all visits that fulfilled the physician-based definition of inactive disease (physician global assessment = 0) and/or the parent-based definition of inactive disease (parent global assessment = 0). A total of 265 visits meeting these criteria were identified. Table 2 shows the evaluation of the concordance between the physician and parent global assessments in these visits. Concordance between physicians and parents in rating the disease as inactive was seen in only 40% of the visits, whereas in 60% of the visits the 2 assessments were discordant. Among discordant visits, visits in which physician global assessment was > 0 and parent global assessment was = 0 were observed more frequently than visits in which physician global assessment was = 0 and parent global assessment was > 0 (35.5% vs 24.5%). Similar results were obtained when all sets of visits (including duplicate visits, n = 367) fulfilling the above criteria were examined (data not shown).

The evaluation of factors (variables) that could potentially affect concordance between parents and physicians in defining the disease as inactive is presented in Table 3. To enable consistency in the analyses, joint counts, parent pain assessment, C-HAQ, and morning stiffness (in minutes) were dichotomized as = 0 or > 0, ESR as normal/abnormal, and CRP as negative/positive. This analysis showed that when

Table 2. Concordance between physician and parent-based definition of inactive disease (N = 265).

	No. Positive	%
No. of visits with physician and parent global assessment = 0	106	40.0
No. of visits with physician global assessment = 0 and parent global assessment > 0	65	25.5
No. of visits with physician global assessment > 0 and parent global assessment = 0	94	35.5

Table 3. Factors affecting concordance/discordance between physician-based and patient-based definition of inactive disease (N = 265).

	Concordance, Physician and Parent Global = 0			Discordance 1, Physician Global = 0 and Parent Global > 0			Discordance 2, Physician Global > 0 and Parent Global = 0		
	No. Tested	No. Positive	%	No. Tested	No. Positive	%	No. Tested	No. Positive	%
No. swollen joints = 0*§	105	97	92.4	65	61	93.8	94	11	11.7
No. tender joints = 0*§	106	95	89.6	65	58	89.2	94	40	42.6
No. restricted joints = 0*§	106	94	88.7	65	47	72.3	94	35	37.2
No. active joints = 0*§	106	96	90.6	65	57	87.7	94	5	5.3
Parent pain assessment = 0§†	104	97	93.3	64	13	20.3	94	82	87.2
C-HAQ = 0§†	104	86	82.7	64	32	50.0	93	74	79.6
Morning stiffness = 0 min	66	64	97.0	35	32	91.4	51	45	88.2
ESR < 20 mm/h	78	66	84.6	51	40	78.4	70	45	64.3
CRP negative	76	66	86.8	50	40	80.0	65	45	69.2

Symbols indicate significant comparisons ($p < 0.01$) as follows: * Concordance vs Discordance 2; § Discordance 1 vs Discordance 2; † Concordance vs Discordance 1.

physicians and parents agreed in providing a global assessment = 0, all other outcome measures also indicated inactive disease (i.e., they were = 0, normal, or negative) in the majority (82% or more) of instances. Among discordant visits, the parents provided a global rating > 0 in contrast with a physician global rating = 0 in association with a parent pain assessment > 0 and, to a lesser extent, with a C-HAQ score > 0. The physicians provided a global rating > 0 in contrast with a parent global rating = 0 in association with an active and swollen joint count > 0 and, to a lesser extent, with a restricted and tender joint count > 0. This means that parents tended to disagree with physicians in rating the disease as inactive if the child had pain or functional impairment, whereas physicians tended to disagree with parents in rating the disease as inactive in the presence of active joint symptoms.

We then evaluated the concordance between the parent-based definition of inactive disease and the formal definition of inactive disease[1] in the visits that included the parent global assessment and all measures that are part of the formal definition of inactive disease. The parent global assessment was = 0 in only 64.6% of the 79 visits that fulfilled the formal definition of inactive disease. This means that in one-third of the instances in which a patient was classified as being in an inactive disease state by the newly developed criteria for clinical remission in JIA the parent did not provide a global assessment = 0. The results were comparable when all sets of visits (including duplicate visits, n = 95) fulfilling the criteria for this specific analysis were examined (data not shown).

DISCUSSION

Agreement on defining the disease as inactive is an important aspect of physician-parent interaction, because the achievement of this disease status usually prompts the physician to start to decrease or even to discontinue the therapeutic regimen. Since these therapeutic decisions are of foremost importance to the parent and the patient, and the achievement of an inactive disease status may have major prognostic implications, it is important to ascertain whether parents' and clinicians' opinions converge or diverge and to identify the factors that may explain the discrepancy.

Substantial disagreement between parents and physicians over disease remission can lead to difficulty in assessing the efficacy of treatments.

In all chronic conditions, the parents' and patients' expectations and definition of improvement do not often coincide with those of the professionals caring for the children. We previously found that a sizable proportion of parents either under- or overestimate the degree of their child's functional ability, as measured with the C-HAQ, when compared with the objective physician's assessment[5]. In another analysis, we observed only a moderate agreement between parents and physicians in rating the intensity of children's pain[6]. We recently investigated the discrepancy between physicians' and parents' global assessments of disease status and found that physicians and parents may perceive the health status of children with JIA differently, with parents more frequently providing better ratings[7].

We have evaluated the concordance between physicians and parents in rating disease as inactive in children with JIA.

Further, we investigated concordance between the newly developed definition of inactive disease and the parent's global assessment of the child's well-being. When we examined all visits in our clinical database that fulfilled the physician-based definition of inactive disease (physician global assessment = 0) and/or the parent-based definition of inactive disease (parent global assessment = 0), we found concordance between physicians and parents in rating the disease as inactive (i.e., both global assessments = 0) in only 40% of the visits, whereas in 60% of the visits the 2 assessments were discordant. In keeping with our previous observation that parents more commonly provide better ratings than physicians[7], visits in which the physician global assessment was > 0 and the parent global assessment was = 0 were encountered more frequently than visits in which the physician global assessment was = 0 and the parent global assessment was > 0. Parents tended to rate the disease as still active, as opposed to physicians, if the child had pain or functional limitation, whereas physicians tended to rate the disease as still active as opposed to parents in the presence of active joint symptoms. In one-third of the instances in which a patient was classified as having inactive disease status through the application of the newly developed criteria for clinical remission in JIA the parent did not provide a global assessment = 0. Together, these results indicate that in a considerable proportion of visits in which the disease was rated as inactive by the subjective assessment of the physician or the new definition of inactive disease[1], the parent did not rate the child's health status as optimal. This suggests that a number of instances of physician-defined inactive disease may not be agreed upon by the parents.

We must acknowledge that by describing the parent-physician discordance in rating the disease status of children with JIA, we cannot imply that the physician's assessment is the right one. It is well known that parents and doctors may have widely different perspectives relating to their beliefs about health and illness, their expectations of medical care, their priorities for treatment, and the ways in which they interpret information about a child's disease. We should also recognize that the physician was asked to rate the overall level of disease activity, whereas the parent provided a more general assessment of the child's overall well-being. We previously noted that the parent global assessment of the child's well-being is an imperfect measure of disease activity, because it is heavily affected by the presence of pain and functional damage[11], a finding confirmed in the present analysis. It is likely that many other factors not primarily related to disease activity, such as psychosocial issues, may have a major influence on the parent's perception of the child's well-being. Thus, when assessing disease activity (and therapeutic response) the parent should be asked, as the physician is, to rate the level of the child's disease activity, besides the level of the child's well-being. We asked mothers to rate the health status of their children, but did not obtain information on children's self-reporting. However, using only parents' proxy reports instead of both parents' and patients' self-reports would fail to capture that parents and children may differ in their perceptions of health[6,12].

We found a frequent discrepancy between physicians' and parents' ratings of inactive disease in children with JIA. This finding suggests that the parent's global assessment of the child's well-being or, more appropriately, disease activity should be considered for inclusion in a future revision of the definition of clinical remission for JIA.

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