Consultation With an Arthritis Specialist for Children With Suspected Juvenile Rheumatoid Arthritis

A Population-Based Study

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Objectives: To describe consultation with an arthritis specialist because of suspected new-onset juvenile rheumatoid arthritis (JRA) and to determine factors associated with prompt consultation.

Design: Retrospective cohort study.

Setting: Physician reimbursement administrative data were obtained for all children aged 16 years or younger in the Province of Quebec (Canada).

Participants: Suspected new-onset cases of JRA in 2000 were defined by a physician visit because of JRA, providing there had been no such claims in the preceding 3 years.

Main Exposure: First JRA diagnosis made by a non–arthritis specialist.

Main Outcome Measures: First consultation with an arthritis specialist subsequent to diagnosis by a non–arthritis specialist and time to first consultation with an arthritis specialist.

Results: Of 352 children and adolescents with suspected new-onset JRA identified by non–arthritis specialists, 159 (45.2%) were subsequently seen by an arthritis specialist. Mean (SD) time to consultation for those seen was 115.3 (213.8) days (median, 28 days). Younger children were more likely to obtain care from an arthritis specialist compared with those having JRA first diagnosed by a general practitioner.

Conclusion: Most patients with suspected new-onset JRA do not obtain prompt care from an arthritis specialist. Further research and action should focus on this issue so that outcomes may be optimized.

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Juvenile rheumatoid arthritis (JRA) is one of the most common chronic diseases of childhood. It is often associated with severe joint destruction and disability and frequently extends past adolescence into adulthood. More than one-third of patients with JRA develop joint destruction and other complications within 6 years after diagnosis. Most of this damage occurs early in the course of the disease; thus, early aggressive therapy may be especially important. It has also been suggested that, if remission does not occur within 10 years after onset of JRA, the disease will continue to be active.

Management of JRA involves a multidisciplinary approach including pharmacologic treatment, physical therapy, and other aids. New advances in pharmacologic treatments have been promising, and early intervention with disease-modifying antirheumatic drugs may help minimize joint damage and increase remission rates. However, use of such drugs can be complex, particularly insofar as appropriate dosing in children and monitoring for possible adverse effects. In addition, the long-term effects of these medications in children are unknown. Because rheumatologists have the most experience with such agents, early consultation with a rheumatologist or pediatric arthritis specialist is important.

To our knowledge, there have been no population-based studies of consultation with arthritis specialists for children and adolescents with suspected JRA. The objectives of this study were to describe consultation with an arthritis specialist because of suspected JRA and to determine factors associated with prompt consultation.
DATA SOURCE AND STUDY POPULATION

Data were obtained from a physician claims administrative database including all residents of the Province of Quebec (the Régie de l’Assurance Maladie du Québec). We obtained permission from the Access to Information Commission of the Province of Quebec before obtaining these data. Our study population consisted of all children and adolescents aged 16 years or younger who had visited a physician in 2000 because of JRA (International Classification of Diseases, Ninth Revision, Clinical Modification [ICD-9-CM] code 714). The cutoff of 16 years was selected because this is one of the diagnostic criteria for JRA. Data were available for these children and adolescents for the period between January 1, 1997, and June 30, 2003. Although the terminology has changed in the last decade and JRA is considered specifically, of the 159 patients, 71 (44.7%) were seen by the arthritis specialist (45.2%) visited an arthritis specialist and by another specialist in 65 cases (18.2%). Only 159 cases according to physician specialty at the first recorded JRA visit were classified according to the density of primary and secondary care facilities and was described as high, moderate, or low, depending on whether both, 1, or no primary and secondary services were available. Physician-related factors assessed included sex,3 years since graduation, and specialty (pediatrician or other specialist vs general practitioner).

STATISTICAL ANALYSIS

We described the characteristics of all children and adolescents having a diagnosis of JRA in 2000 and categorized the cases according to physician specialty at the first recorded JRA visit. We then described contact with an arthritis specialist for both incident cases and non–incident cases.

We analyzed the data for those 352 incident cases whose first JRA visit was to a non–arthritis specialist. In initial analyses, we compared those who consulted with an arthritis specialist any time during follow-up with those who did not, using multiple logistic regression. Our primary analyses then focused on factors associated with time to first consultation with an arthritis specialist. The incident JRA visit was defined as “event zero,” and the time to the first visit to an arthritis specialist was defined as “event time.” Patients who did not consult with an arthritis specialist by the end of follow-up (end of the study [June 30, 2003] or death) were censored at that time. Multivariate Cox proportional hazards regression14 was used to estimate the independent effects of patient sex, age (dichotomized at the mean), SES, and service availability, and the characteristics of the physician visited at the time of the initial JRA diagnosis (sex, years since graduation, and specialist vs general practitioner). At regression analyses, all hypotheses were tested using the 2-tailed Wald test, with P < .05 considered statistically significant.

RESULTS

Of the 842 patients with JRA recorded in Quebec in 2000, 557 (66.2%) had contact with an arthritis specialist some time between January 1, 1997, and June 30, 2003. Of these 842 patients, 523 had incident JRA. Their characteristics are given in Table 1 and compared with those of the 319 patients with nonincident JRA, that is, those who had a JRA visit previous to the first visit in 2000. The mean (SD) age for the entire group was 10.0 (4.4) years.

Among the 523 incident cases, 352 were first assigned a JRA diagnosis by a non–arthritis specialist. For the 352 patients, the diagnosis was made by a family physician in 154 cases (43.8%), by a pediatrician in 134 cases (38.1%), and by another specialist in 65 cases (18.2%). Only 159 of these 352 patients with JRA initially diagnosed by a non–arthritis specialist (45.2%) visited an arthritis specialist within the subsequent 3-year follow-up. Of these 159 patients, 22 (13.8%) were diagnosed by the arthritis specialist as having JRA, 36 (22.6%) as having another type of arthritis (ICD-9-CM codes 710-720, excluding code 714 for JRA), and 101 (63.6%) as having a noninflammatory or nonrheumatologic disorder. Consultation patterns are shown in Figure 1. In the 159 patients who consulted an arthritis specialist, the mean (SD) time between the first diagnostic visit with the non–arthritis specialist and consultation with the arthritis specialist was 115.3 (213.8) days (median, 28.0 days; interquartile range, 2–105 days). Specifically, of the 159 patients, 71 (44.7%) were seen by the
Because children with musculoskeletal signs or symptoms often see orthopedic surgeons because of these problems, we determined that 63 of the 159 children or adolescents who consulted an arthritis specialist (39.6%) saw an orthopedist first.

Using the algorithm of MacLean et al (ie, 2 coded JRA visits by a non–arthritis specialist at least 2 months apart within 2 years), we identified 43 incident JRA cases, 31 of whom (72.1%) were subsequently seen by an arthritis specialist. The median time from the first diagnostic JRA visit to consultation with an arthritis specialist for these 31 children was long: 286 days (interquartile range, 167-394 days).

Univariate analyses revealed that more female patients consulted arthritis specialists (P = .02), as did children and adolescents who lived in areas with higher service availability (P = .02) and who had their first JRA diagnosis posed by a non–arthritis specialist or a pediatrician as opposed to a general practitioner (P < .001).

Results of multivariate logistic regression analysis are given in Table 2. Girls and those in whom JRA was first diagnosed by a pediatrician or another specialist as opposed to a general practitioner were more likely to have consulted with an arthritis specialist. Children and adolescents living in areas with medium service availability...
Our primary analyses indicated that only 45% of children and youth with incident JRA or suspected JRA (all subtypes) who consulted an arthritis specialist first consulted an orthopedic surgeon. Consultation with a pediatric arthritis specialist is most often because of musculoskeletal pain, which is found to be inflammatory disease in a minority of patients. The effects of first referral to an orthopedic surgeon and possibly overreferral of a child with noninflammatory disease to an arthritis specialist may result in delay in consultation with an arthritis specialist.

Younger children and female patients tend to consult an arthritis specialist sooner. Cuesta et al15 reported that younger children with pauciarticular JRA who consulted pediatric rheumatologists tend to be referred first to orthopedists. The differential diagnosis of pauciarticular JRA is more likely to include orthopedic problems as opposed to the differential diagnosis of polyarticular or systemic-onset arthritis. Moreover, these authors did not consider all children and adolescents having a diagnosis of JRA but only those who consulted a rheumatologist, and they did not indicate whether the time to consultation was shorter for older children. Physicians may be more likely to refer female patients with suspected JRA because they may be more certain of the diagnosis inasmuch as JRA occurs more frequently in girls.6 The finding that patients diagnosed by other specialists were more likely to consult an arthritis specialist may be because specialists are more likely to refer to other specialists when they recognize that a patient’s disorder is not within their domain of expertise, whereas a general practitioner may continue to see the patient, order tests, and observe the patient over time.17 Moreover, this concurs with the findings of McGhee et al,6 who found that orthopedic surgeons are more likely to refer children with JRA to rheumatologists than are primary care physicians and other specialists. Our finding that patients who were first diagnosed by a pediatrician are more likely to consult an arthritis specialist than are those who were first diagnosed by a family practitioner agrees with the findings of Freed et al,18 who reported that 42% of pediatricians and 32% of family practitioners claim to refer all of their patients with JRA.

Unlike findings in adult rheumatoid arthritis,19 we did not find an association between SES and consultation with an arthritis specialist. This is encouraging, especially in view of recent findings of worse outcomes for patients with JRA with lower SES.20 Studies based on administrative data, such as ours, have inherent limitations including lack of quality-control of diagnostic coding and difficulty in capturing disease severity. Based on our data, there were 193 incident cases...
of JRA: 171 first diagnosed by an arthritis specialist and another 22 of the 159 initially diagnosed by a nonspecialist and later confirmed by an arthritis specialist. Based on the Quebec population of approximately 1.5 million persons aged 0 to 16 years, the cumulative incidence for 2000 in our study would be 12.9 per 100 000 persons, which is slightly higher than the 11.7 per 100 000 persons estimated in Rochester, Minnesota,24 and lower than the 19.5 per 100 000 persons estimated in Finland.26 In this study, we did not insist on high sensitivity of the diagnosis. If the diagnosing physician coded a diagnosis as JRA, we assumed that he or she believed that the patient had JRA or, perhaps, a related condition. Diagnoses can change with time; for example, it may be reasonable for a physician to observe a patient with suspected post–viral arthritis to determine whether symptoms resolve. Nevertheless, we contend that, if a physician suspected JRA, in view of recommendations for early treatment, consultation with an arthritis specialist may be important. When using the more stringent algorithm of MacLean et al,31 the consultation rate was much higher (72%) but still indicated that more than one-fourth of these children and adolescents did not consult an arthritis specialist because of JRA. Moreover, the time to consultation was long. We could not differentiate types of JRA from the database codes. Disease severity may affect perceived need for consultation because, we presume, more severe disease, such as systemic disease, may be diagnosed sooner and specialized treatment may be considered more urgent.27 Nevertheless, early treatment of JRA is thought to be beneficial regardless of severity.8,9

We did not include in our analysis those patients in whom JRA was initially diagnosed by an arthritis specialist. These patients were most probably referred to an arthritis specialist by another physician at some time. Some of these patients may have had disease that had been in remission for more than 3 years and, therefore, were not incident cases. Others may have been referred with a diagnosis other than JRA. It is impossible to identify a priori all patients having other diagnoses who would possibly be referred to an arthritis specialist, especially inasmuch as they may have been referred but the arthritis specialist may not have confirmed a diagnosis of JRA. Despite these limitations, we believe that our analysis depicts consultation patterns for patients with suspected JRA, defined by a coded JRA visit to a non–arthritis specialist.

Another consideration is that administrative databases describe actual consultations with specialists. We cannot know whether these constitute all referrals because some referrals may not be acted on. In addition, we were unable to address other issues related to accessibility to specialists such as waiting times, transportation difficulties, family problems, or mental health issues. We classified patients as having suspected new-onset JRA if they did not visit a physician because of JRA in the previous 3 years. In some patients, JRA may have been in remission for several years and may have been erroneously included as new-onset JRA. However, one would expect that these patients would have been more, not less, likely to consult an arthritis specialist because they may have done so in the past. Thus, if such patients were included in our incident group, then the rate of consultation for new-onset JRA may be even lower than what we report.

In conclusion, our data show a low rate of rheumatology consultation because of suspected new-onset JRA. Of possible concern is that this reflects a shortage of pediatric rheumatologists in the Province of Quebec, as has been reported in North America in general.28 This would compound the difficulty primary care physicians have insofar as the diagnosis and treatment of JRA. Potential solutions include both improved medical education and the implementation of good screening techniques to help primary care physicians identify probable JRA. In addition, triaging rheumatology consultations according to the degree of concern on the part of the referring physician may enable patients with inflammatory arthritis to receive prompt, appropriate management and, thus, maximize outcomes for this potentially disabling condition. Also, increasing the number of pediatric rheumatologists may improve the situation.

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