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Original article

PREVALENCE AND FACTORS OF FUNCTIONAL DISABILITY IN PATIENTS WITH JUVENILE IDIOPATHIC ARTHRITIS

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ABSTRACT

The aim of this study was to identify the prevalence of functional disability and the possible factors that may be associated with functional disability in children and adolescents with juvenile idiopathic arthritis (JIA) in Sharkia Governorate. **Methods**: This cross sectional study was carried out in Rheumatology and Rehabilitation department, Faculty of Medicine, Zagazig University Hospitals. Forty-eight consecutive patients of JIA aged from 7 to 17 years underwent assessment of socio-economic and demographic characteristics, functional ability using the childhood health assessment questionnaire (CHAQ), disease activity using the Juvenile Arthritis Disease Activity Score based on 27 joints (JADAS-27), psychological symptoms using the Children's Depression Inventory (CDI) score. **Results:** Multivariate modeling was applied to determine the factors that associated with functional disability. A total of 80 % of the patients (39 of 48) had functional disability. In multiple regression analyses, high CDI scores (OR 20.78, 95 % CI 1.64 to 262.91, P =.019), JADAS-27 (OR 17.49% CI 2.16 to 141.62, P =.007), low socioeconomic status (OR 10.43, 95 % CI 1.24 to 87.57, P =.031) were strong predictors of functional disability in JIA patients.

Conclusion: Our study on patients with JIA provides evidence suggesting that a total of 80 % of the patients had functional disability, that is associated with higher CDI scores, higher JADAS -27 and a low socioeconomic status.

Keywords: Juvenile idiopathic arthritis· Functional disability· Determinants. CHAQ

INTRODUCTION

Juvenile idiopathic arthritis (JIA) is the most common chronic pediatric rheumatic disease. It is an important cause of acquired impairment and disability in children and adolescents. It can be presented in different patterns (e.g., oligoarthritis, polyarthritis and systemic arthritis). **[1]**

Some evidence reports that pre-pubertal children with JIA are physically less active when compared with healthy age- and sex-matched control subjects, hence physical activity is essential for the social, emotional, and cognitive development of children and adolescents with JIA. **[2]**

Functional ability is an important outcome measure in understanding the impact of the disease on daily life in childhood-onset rheumatic diseases **[1]**. Patients with JIA commonly experience acute and chronic pain, decreased mobility, and joint stiffness leading to restrictions of activities and isolation from their peers **[3].**

In addition to pain and physical limitations, children with JIA may also experience high levels of stress during the course of their disease. For example, they may find performing daily classroom activities challenging during periods of symptom exacerbation. Moreover, they may feel altered body image, anxiety around social acceptance. In fact, children with JIA are prone to the problems that can delay their psychosocial development **[4].**

The aim of this study was to identify the prevalence of functional disability and the possible factors that may be associated with functional disability in children and adolescents with juvenile idiopathic arthritis (JIA) in Sharkia Governorate.

SUBJECTS AND METHODS

Study design and subjects:

This cross sectional study was conducted in the inpatient and outpatient clinics of Rheumatology and Rehabilitation Department & Pediatric Department, Faculty of Medicine, Zagazig University Hospitals. All JIA patients aged from 7 to 17 years, residing in Sharkia Governorate and diagnosed according to the International League Against Rheumatism (ILAR) classification **[5]** were recruited in this study between April 2018 and October 2018.

Exclusion criteria: Patients younger than 7 years, patients with disease duration less than 1 year and those with chronic diseases, in addition to JIA, which would influence the child's function, were excluded from this study.

Written informed consent was obtained from all participants and the study was approved by the research ethical committee of Faculty of Medicine, Zagazig University. The work has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for studies involving humans.

Demographic and socioeconomic

The parents completed the form that included child's gender, age, residency and school grade, and they completed the questionnaire that measures the socioeconomic status of children and adolescents during the same visit of the clinical assessment of the patients. The socioeconomic status was assessed using the socioeconomic status scale for health research in Egypt, which is scoring system, based on seven domains (education and cultural, occupation, family, family possessions, economic, home sanitation and health care) with a total score of 84. Higher score indicating better SES. Socioeconomic level to be classified into very low, low, middle and high levels depending on the quartiles of the score calculated. This socioeconomic status scale has adequate factor reliability and validity **[6]**

Disease activity

By using the Juvenile Arthritis Disease Activity Score based on 27 joints (JADAS-27) that included the following four measures: physician global assessment of disease activity, measured on a 10-cm visual analog scale (VAS) where 0 = no activity and 10 = maximum activity; parent/ patient global assessment of well-being, measured on a 10-cm VAS where 0 = very well and 10 = very poor; count of joints with active disease; and erythrocyte sedimentation rate. The JADAS-27 was calculated as the simple linear sum of the scores of its 4 components, which yields a global score of 0–57: 0 = no disease activity and 57 = maximum disease activity **[7].**

Functional ability

Childhood Health Assessment Questionnaire (CHAQ) describes the child's usual activities in eight domains over the past week. It includes dressing, getting up, eating, walking, with or without aids or assistive devices, hygiene, reaching overhead objects, grip and activities. Each question is scored from 0 to 3 (0 = no difficulty, 1 = some difficulty, 2 = much difficulty and 3 = unable to do). The score for each of the eight functional areas was averaged to calculate the disability index. The Arabic form of CHAQ was used in this study **[8].**

Depressive symptoms

Participants completed the Children's Depression Inventory (CDI). The CDI is a 27-item self-report inventory of childhood depression that taps a variety of depressive symptoms. Items assess negative mood, interpersonal difficulties, negative self-esteem, ineffectiveness and anhedonia in children aged 7–17 years. Each item offers respondents three alternatives scored 0, 1 or 2, and accordingly, raw scores range from 0 to 54 and then convert raw scores to T-scores. Higher scores indicate higher levels of depressive symptoms **[9]**. The CDI was translated and normalized for Arab

children by Gharib **[10]**. Reliability and validity data for the Arabic version are comparable to those provided for the original instrument. For this study, we considered T-Score ≥70 clinically significant score as in previous study **[11]**.

Statistical Methods:

All statistical analyses were performed using IBM SPSS Statistics, version 24 **(SPSS Inc., 2016).** Continuous variables were presented as the mean (SD) if normally distributed or median (range) if not normally distributed. Chi-square tests for categorical variables and unpaired t tests for continuous variables were used. All variables were dichotomized to make interpretation more explicit. A multiple logistic regression model was built up to identify factors associated with functional disability.

T-scores are commonly used for neuropsychological normative data A T-score is a z-score that has been transformed to eliminate negative values. The mean and standard deviation of z-scores is zero and one, respectively; the mean and standard deviation of T-scores is 50 and 10, respectively. The results are presented as odds ratios (ORs) with 95 % confidence intervals (CIs). P-value<.05 indicates a statistically significant difference.

RESULTS

The mean age was 12.3(2.4) years, the median disease duration was 4(1–10) years, and the majority were females (71%), live in rural areas (67 %) with a low socioeconomic status (67%). Approximately 27% of patients stopped schooling.

Table 1 Demographic, socioeconomic and clinical characteristics

Variables	**n=48**
Age (years)	
Mean (SD)	12.3 (2.4)
Sex, n (%)	
Female	34 (71)
Male	14 (29)
Residency, n (%)	
Urban	16 (33)
Rural	32 (67)
School status, n (%)	
Continue	35 (73)
Stop	13 (27)
Educational level, n (%)	
<6 academic years	15 (31)
≥ 6 academic years	33 (67)
Socioeconomic status, n (%)	
Very low	2 (4)
Low	32 (67)
Moderate	14 (29)
High	0 (0)

JIA subtype, n (%)	
Oligoarthritis	8 (17)
Polyarthritis	28 (58)
Systemic	12 (25)
Psoriatic	0 (0)
Enthesitis related arthritis	0 (0)
Disease duration (years)	
Mean (SD)	5 (2.5)
Median (range)	4 (1-10)
Rheumatoid factor, n (%)	
Positive	20 (42)
Negative	28 (58)
JADAS-27	
Mean (SD)	22.1 (13.4)
Median (range)	15.5 (5-46)
CDI total raw scores	
Mean (SD)	9.7 (4.7)
Median (range)	9.5 (0-19)
CHAQ score	
Mean (SD)	0.77 (0.76)
Median (range)	0.5 (0-2.7)
Medication, n(%)	
Methotrexate	38 (79)
Steroids	45 (94)
Hydroxychlorquine	19 (40)
Leflunomide	4 (8)
Biologicals	10 (21)
Sulfasalazine	5 (10)

Table 2 Univariate analyses of factors associated with functional disability

Variables	**Patients with functional disability, n=39**	**Unadjusted odds ratio (95% CI)**	**P-value**
	N (%)		
Age		1.67 (0.346 - 8.03)	0.53
Child (<11 years), n=12	9 (75)		
Adolescent (≥ 11 years), n=36	30 (83)		
Sex, n		1.56 (0.28 to 8.62)	0.61
Female, n=34	27 (79)		
Male, n=14	12 (86)		
Residency, n		-----	0.99
Rural, n=32	23 (81)		
Urban, n=16	16 (100)		
School status		---	0.99
Stop, n=13	13 (100)		
Continue, n=35	26 (74)		

Educational level		1.13 (.240 to 5.27)	0.88
<6 academic years, n=15	12 (80)		
≥ 6 academic years, n=33	27 (82)		
Socioeconomic level		---	0.99
Low and moderate**,** n=48	39 (100)		
High and very high, **,** n=0	0 (0)		
JIA subtype, n		7 (1.31 to 37.3)	**0.023**
Oligoarthritis, n=8	4 (50)		
Other subtypes, n=40	35 (88)		
Disease duration		2.23 (0.514 to 9.69)	0.028
(<4 years), n=19	14 (73)		
(≥ 4years), n=29	25 (86)		
Rheumatoid factor, n, (%)		---	0.99
Positive, n=20	20 (100)		
Negative, n=28	19 (68)		
JADAS-27		13.6 (2.55 to 72.53)	**0.002**
(<13), n=11	5 (46)		
(≥13), n=37	34 (92)		
CDI T scores (median of T scores)		---	0.99
(<70), n=45			
(≥70), n=3	36 (80)		
	3 (100)		

Table 3 Factors associated with functional disability by multiple logistic regressions

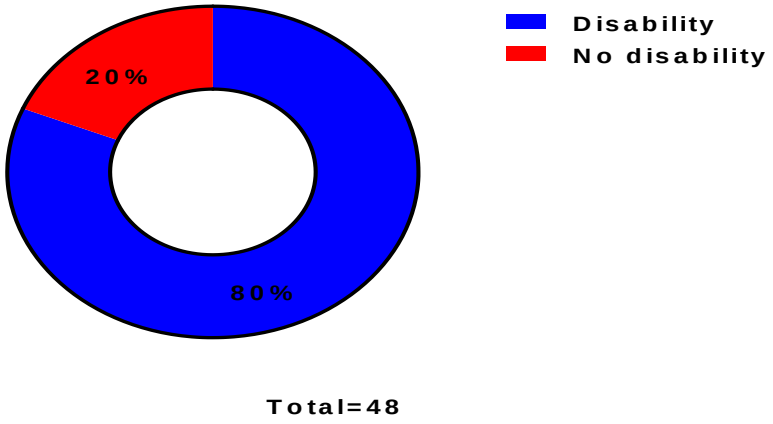
Variables	**CHAQ/functional disability,** n (%)				**Test of significance**	**P-value**
	No	**Mild**	**Moderate**	**Severe**		
	n=9	n=21	n=10	n=8		
Socioeconomic status, n (%) Very low	0 (0)	2 (10)	0 (0)	0 (0)	Fisher's Exact Test=16.2	**0.002**
Low	9 (100)	16 (76)	5 (50)	2 (25)		
Moderate	0 (0)	3 (14)	5 (50)	6 (75)		
High	0 (0)	0 (0)	0 (0)	0 (0)		
JIA subtypes, n (%)					Fisher's Exact Test=23.8	**0.001**
Oligoarthritis	4 (44.4)	4 (19)	0 (0)	0 (0)		
Polyarthritis (positive RF)	0 (0)	4 (19)	8 (80)	4 (50)		
Polyarthritis (negatives RF)	4 (44.4)	7 (33.3)	1 (10)	0 (0)		
Systemic	1 (11.1)	6 (28.6)	1 (10)	4 (50)		
Psoriatic	0 (0)	0 (0)	0 (0)	0 (0)		
Enthesitis-related arthritis	0 (0)	0 (0)	0 (0)	0 (0)		

JADAS 27, n(%) Low	6(67)	5(24)	0(0)	0(0)		Fisher's Exact Test=27.4	**<0.001**
Intermediate	3(33)	16(76)	4(40)	4(50)			
High	0(0)	0(0)	6(60)	4(50)			

Table 4 Multivariable logistic regression analysis of various predictors with functional disability in juvenile idiopathic arthritis (JIA) patients

Variables	Adjusted odds ratio (95% CI)	P-value
CDI (≥9.5)	20.78(1.64 to 262.91)	**0.019**
JADAS-27 (≥ 15.5)	17.49(2.16 to 141.62)	**0.007**
Socioeconomic status (low)	10.43(1.24 to 87.57)	**0.031**

Figure 1 Donut chart representing percentage of functional disability in patients with JIA



DISCUSSION

Juvenile idiopathic arthritis (JIA) is a heterogeneous group of diseases characterized by arthritis of unknown origin with the onset before the age of 16. The etiology is not completely understood but it is considered as multi-factorial with an essential role of both genetic and environmental factors [12]**.** It is one of the most common chronic diseases of childhood and the most prevalent of pediatric rheumatic illness [13]**.**

The extent of impact on the various aspects of the patients', families' and society's functioning is clear. It imposes a societal burden of significant health care costs and utilization. Possible continuing disease activity, medication-associated morbidity, life-long disability and risk for emotional and social dysfunction are the main problems that may face JIA children who have reach adulthood [14]**.**

In this cross-sectional study, we assessed the frequency of functional disability, as well as we examined the association between functional disability and a comprehensive set of variables in a representative sample of Egyptian children and adolescents with JIA. Our study shows that 80% of patients have functional disability while 20% of patients have no disability at median disease duration of 4 years.

In our study, age, sex, duration of disease and treatment were not associated with functional impairment. Similarly, **Adib et al.** [15] found that duration did not correlate with C-HAQ score at presentation. This was probably because they depended on data collected during first presentation. In contrast, previous studies [16] showed that the strongest predictors of CHAQ were; disease duration, female gender.

Rheumatoid factor positive polyarticular onset is the most frequently observed type found in our patients. In contrast to some countries as France and Spain described the oligoarticular onset was the commonest [17, 18]. This would be explained by the differences in environmental and genetic factors with possible exposure to different types of infections.

Our study also showed that there was no statistically significant association between the type of medication and the functional disability.

Regarding the influence of socioeconomic status (SES) on functional ability in the present study, we found that patients with low to moderate socioeconomic status have higher risk of moderate to severe impaired functional ability. This could be explained by the fact that the risk factors associated with low socioeconomic may affect not only family but also the children [19].

Similarly**,** previous study found that, despite similar disease activity scores obtained in the clinic, patients from a low-SES background rated their general disease activity, pain, and functional disability worse than patients from a high-SES background [20].

In our study the functional status of children was assessed using childhood health assessment questionnaire (CHAQ). It was seen that 18.7% children did not have disability, 43.7% had mild disability, 20.8% had moderate disability and 16.6% cases had severe disability. In contrast to our findings, **Packham and Hall** [21] in their cross-sectional study found that functional disability contributed 18% of total variance explained in pain.

In the current study, we found that JIA subtype was a determinant for functional disability. Patients with systemic onset and rheumatoid factor positive polyarticular disease have a higher risk of moderate to severe functional limitation than other types. Similarly, another study [22] found that systemic JIA had more severe disability compared to patients with oligoarticular JIA who exhibited mild disability. This may be attributable to the more number of joints affected and multiple system organs affection.

Results also showed that physical function measured by CHAQ significantly associated with the degree of disease activity measured by JADAS-27, patients with higher JADAS-27 scores (≥13) were 13.6 times more likely to have functional disability than those with lower JADAS-27 scores (<13). This is consistent with the previous study [22] that found that C-HAQ significantly correlated with the majority of disease activity variables.

CONCLUSION

Unfortunately even with improved treatment modalities, children and adolescents with JIA are at significant increase in possibility of having functional disability.

Poor psychological outcome and severe disease activity are powerful determinants of functional disability. Thus, the incorporation of disease outcome measures of juvenile idiopathic arthritis in daily care requires the use of simple and feasible tools.

Declaration of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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