RESEARCH PAPER

Economic Burden of Juvenile Idiopathic Arthritis in India

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Background: Published Indian studies on the economic burden of juvenile idiopathic arthritis (JIA) are lacking. **Methods**: A prospective observational study recruited pediatric patients aged from 1 to 12 years with JIA in the pediatric rheumatology clinic of a public sector tertiary care hospital. Direct healthcare costs and indirect costs for transportation, accommodation of the caregivers, and productivity loss for work absenteeism were assessed. **Results**: The proportions of direct annualized cost assessed in 60 patients (mean (SD) age 8.46 (2.24) year) spent on outpatient visits, blood tests, imaging investigations, other tests, medications and hospitalization were 0.85%, 12.8%, 9.0%, 2.9%, 41.7% and 32.7%, respectively. Direct healthcare costs for blood tests and medicine were lowest in oligoarticular JIA and highest in systemic onset JIA and (P=0.043 and 0.001 respectively). The direct and indirect costs were higher with the use of biologic agents (*n*=9) than in those without (*n*=51). **Conclusions**: JIA imposes considerable economic burden with the largest share attributable to medicines, and maximum in those with systemic onset JIA.

Keywords: *Cost analysis, Biologic agents, Pharmacoeconomic study, Financial burden, Medication cost.*

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Juvenile idiopathic arthritis (JIA), the commonest rheumatic disorder in children, imposes a formidable burden on patients and caregivers in terms of reduced quality of life and economic hardship [1,2]. The financial burden is attributable to the cost of multiple clinic visits, laboratory tests, imaging investigations, expensive medications (biologics), occasional hospitalization, and work-absenteeism. The burden is particularly severe for patients with systemic onset JIA (sJIA) [1]. Literature on the economic burden of JIA are available from other countries [2,3] but are lacking from India. We, therefore, conducted this pharmaco-economic study of JIA patients attending a public sector hospital.

METHODS

This observational study was conducted between April, 2017 and March, 2018, in the pediatric rheumatology clinic of a tertiary-care teaching hospital in Eastern India. Patients aged 1-12 years and diagnosed with JIA were included after taking written informed consent from either parent. The study protocol was approved by the institutional ethics committee. The sampling strategy was purposive as children not accompanied by caregivers within the family and those with limited follow-up visits for at least 12 months were excluded. JIA subgroups were categorized as oligoarticular, polyarticular, systemic onset (sJIA), enthesitis related and undifferentiated.

Data were collected over a period of 12 months for direct healthcare costs and indirect costs. Direct healthcare costs covered six elements; out-patient clinic (OPD) visits, blood tests, imaging investigations, other tests, medications, and hospitalization. Most of the medications, investigations and hospitalization did not incur any costs as per the hospital's policy. The medication cost was calculated from the centralized procurement cost of the generic medicines supplied. The market prices of the brands supplied were considered at 20% discount for medicines supplied through local purchase. The cost of laboratory and imaging investigations were imputed from the government approved rates when offered by authorized public-private partnership units. The actual cost of investigations was recorded if done from outside. OPD visit costs were calculated at actuals which included the OPD registration charges and the expenditure on food and drink during the waiting time. Cost of hospitalization was applicable for only a proportion of patients and was inferred from the

bed charges (general pediatric ward) of three nearby not-for-profit hospitals in the non-governmental organization (NGO) sector. The costing was estimated to derive annualized costs and compared between different subgroups of JIA.

Indirect healthcare costs included transportation expenses for initial and follow-up visits for patients and their caregivers, and accommodation costs of the caregivers if from out-station. The productivity loss was calculated from the number of days of work-absenteeism and the loss of daily wages for daily wage earner caregivers or deduction from salary for salaried caregivers as declared.

Statistical analyses: Numerical variables were compared between biologic and no-biologic subgroups by Student unpaired *t* test or Mann-Whitney *U* test, depending upon whether normal distribution assumption was satisfied or not. Normality assumption was tested by Kolmogorov-Smirnov goodness-of-fit test. One-way analysis of variance (ANOVA) or Kruskal-Wallis ANOVA, as applicable, was applied for comparison between the multiple JIA subgroups. Statistical significance was inferred if *P* value was less than 0.05.

RESULTS

A total of seventy children were approached which excluded three who didn't consent, four with unreliable records and three who were lost to follow-up, to finally include 60 (53.5% males) children in the study with mean (SD) age 8.46 (2.24) years. The mean (SD) of number of OPD visits were 12.5 (2.56) (range 7-20) and inpatient admissions ranged from 0-6. The mean (SD) number of OPD visits in sJIA subgroup was 13.2 (2) which was the maximum among all subgroups with most hospital admissions [median (IQR) 3 (0-6). The proportions of the total annualized cost incurred on OPD visits, blood tests, imaging investigations, other tests, medication and hospitalization were 0.8%, 12.8%, 9%, 2.9%, 41.7% and 32.7%, respectively.

The direct and indirect costs in the whole JIA cohort have been presented in **Table I**. The mean (SD) medication costs and hospitalization costs were Rs. 9011.5 (4217.53) and Rs. 8489.3 (4789.88) in patients with sJIA, which were highest than all other subgroups; P =0.001 and 0.090 respectively. The mean (SD) medication cost was lowest at Rs. 3225.1 (1046.96) in the oligoarticular subgroup as compared to sJIA subgroup; *P* <0.001. Children with oligoarticular did not require hospitalization. The medication and hospitalization costs were comparable between polyarticular, enthesitis related and undifferentiated types; *P* >0.05 (data not shown).

The median (IQR) transportation cost was highest for patients with sJIA and lowest in oligoarticular JIA [Rs. 2737 (2230-6040) and 1280 (1120-2000); *P* = 0.05]. The median (IQR) accommodation cost was Rs. 1585 (0-2876) in sJIA which was the highest among all subgroups; *P*=0.001. Self-declared income loss was comparable between subgroups. **Table II** compares the costs in JIA patients who received treatment with biologics (*n*=9) and those who did not.

DISCUSSION

The provisional cost analysis of JIA in a public-sector hospital revealed considerable economic burden, majorly for the cost of medicines. The burden was highest in patients with sJIA and was higher with uses of biologics.

The type of JIA and active joint count are predictors of direct costs, with higher costs for patients with

Table I Healthcare Related Costs (INR) for Children With Juvenile Idiopathic Arthritis (N=60)

Category	*Cost*
Direct costs	
OPD visits	120 (110-140)
Blood tests	1555 (1350-2340.5)
Imaging	772.5 (460-1453)
Other tests	312.5 (145-630)
Medicines[a]	6185.2 (2984.34)
Hospitalization	3168 (1522-5216)
Indirect costs	
Transport	2155.5 (1275-3000)
Accommodation	0 (0-1705)
Income loss	5195 (2890-7229)

Cost in median (IQR) or [a]mean (SD); OPD-Outpatient department.

Table II Healthcare Costs (INR) for Children With JIA According to Usage of Biologic Agents

Cost (INR)	*Treatment with biologics (n=9)*	*Treated without biologics (n=51)*
OPD visits	120 (120-120)	120 (110-140)
Blood tests	1350 (1247-1845)	1624 (1365-2465)
Imaging	550 (260-780)	825 (460-1467)
Medicines[a,b]	11737.14 (4518.74)	5844.47 (2107.93)
Hospitalization[b]	11956 (11210-12040)	2390 (1325-3268)
Transport[b]	2970 (2230-5132)	1722 (1270-2970)
Accommodation[b]	2400 (1600-3428)	0 (0-1257)

Data shown as median (IQR) and [a]mean (SD); [b]P value <0.05.

WHAT THIS STUDY ADDS?

- A provisional costs analysis of juvenile idiopathic arthritis in a public-sector teaching hospital revealed considerable economic burden, which was higher in systemic onset JIA and with the use of biologics.

polyarthritis (rheumatoid factor positive or negative) or sJIA [4]. The cost increases with disease activity, disease duration, and time period from symptom onset to first pediatric rheumatologist visit [5].

The major share of the total direct healthcare cost burden was accounted for by medicines in the present study, similar to other studies [4-6]. The investigation costs tend to be relatively higher in the developed countries. With the increasing availability and use of biologics for more severe disease, medication costs can be expected to further increase.

A study [7], from New England reported that out-patient cost in JIA was 3.3 times higher than the inpatient costs. The mean in-patient cost comprising hospitalization and medicine cost was higher than the outpatient cost in the present study when all the cases were pooled together. The direct healthcare costs accounted for 46% of total costs, direct non-healthcare costs for 26.4% and productivity losses for 27.6% in another study in the United Kingdom [6]. A German study [5] also indicated considerable proportion of indirect cost due to time lost from work.

In India, studies on economic burden of childhood diseases are scarce. The growth velocity is significantly reduced in seropositive JIA subjects [8]; treatment for which is costly. A recent systematic review [10], also concluded considerable economic impact of JIA with data largely reflecting European and North American costs.

The present study was conducted in a government hospital where the salary of professional personnel and other logistical costs were not accounted for. The estimated costs would be invariably less than the costing in private set-ups. The expenditure on evaluation and management before coming to the index hospital were not accounted. The sample size was limited and children only up to twelve years were included which may also affect generalizability of the study findings.

More studies on the economic impact of juvenile idiopathic arthritis in the Indian scenario are needed from different regions and health-settings before a plea for social support to these children can be made to the public health authorities.

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