Clinico-epidemiological Profile of Pediatric Rheumatology Disorders in Eastern India

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Abstract

Background: Data on rheumatological disorders in children from developing countries like India are scarce. Hence, this study aimed to understand the clinical and epidemiological profile of rheumatological disorders in children as this can help organize comprehensive evidence-based health care services. Methodology: A retrospective hospital-based study was designed in pediatric rheumatology clinic of the All India Institute of Medical Sciences, Patna, India, from January 2015 to December 2016. Clinical and laboratory findings and response to therapy of all children with rheumatological disorders were evaluated. Results: A total of 60 children with various rheumatological disorders were included in the study. Juvenile idiopathic arthritis (JIA) was the most common pediatric rheumatology disorder observed, and polyarticular JIA was the predominant subtype observed in our patients. The mean age of disease onset was 9.1 ± 3.6 years (age range: 1–16 years). Methotrexate was the most common disease modifying antirheumatic drug used. None of the children received newer biological agents due to financial constraints. The use of alternative medicine was observed in 10% of cases. Conclusion: The clinical and epidemiological profile of children with rheumatological disorders in our patient group was different from the European countries and Western world. There is a need for introspecting the lack of using biological agents and its potential impact in managing JIA in our patient group.

Keywords: Epidemiological profile, pediatrics, rheumatology disorders

INTRODUCTION

Pediatric rheumatological disorders are considered rare in the community more so in a developing economy like India where infectious diseases rule the roost in terms of morbidity and mortality. The data on pediatric rheumatology are sparse from India.[1] However, global data suggest that 2%–5% of the population suffers from rheumatological disorders,[2] which when extrapolated to the Indian population, throws up worrying numbers. It is also important to consider that about 10%–20% of the total burden of rheumatological disorders have their initial presentation in pediatric age groups, which on delayed intervention leads to significant disability, loss of economic productivity, and disability-adjusted life years.[3] Profile of many of the pediatric rheumatological disorders is different in children from Western countries.[4] Hence, clinico-epidemiological statistics are warranted to increase awareness in the medical community along with strengthening of ancillary services to cater to these patients.[5]

METHODOLOGY

A hospital-based descriptive study was designed in the All India Institute of Medical Sciences, Patna, India, from January 2015 to December 2016. The study included all the children who were enrolled and being followed up in the pediatric rheumatology clinic. The clinic manned by one consultant and two senior residents provides comprehensive outpatient care for children diagnosed with various rheumatological disorders. It has a facility for the physiotherapy and medical rehabilitation. Data, including residence, parental education, age of onset of the underlying disease, age of presentation, use of Nonsteroidal anti-inflammatory drugs (NSAID), use of disease modifying antirheumatic drugs (DMARDs), use of steroids, duration of treatment, duration of follow-up and

Access this article online

Quick Response Code:

Website:
www.jnsbm.org

DOI:
10.4103/jnsbm.JNSBM_80_17

How to cite this article: Patra PK, Kumar M. Clinico-epidemiological profile of pediatric rheumatology disorders in Eastern India. J Nat Sc Biol Med 2018;9:19-22.
those lost to follow, remission on drugs or off drugs, and use of alternative medication were collected in a structured format. All the files had detailed record of history, musculoskeletal examination by pediatric arms, leg, spine, clinical diagnosis, and relevant investigations. Children with juvenile idiopathic arthritis (JIA) are classified as per the International League against Rheumatism. The laboratory parameters collected for the study purpose were total leucocyte counts, erythrocytes sedimentation rate, C-reactive protein, rheumatoid factors (RFs), antinuclear antibody (ANA), hepatitis B, hepatitis C, human immunodeficiency virus, perinuclear antineutrophilic cytoplasmic antibody, antinuclear cytoplasmic antibody, and HLA-B-27. The study was conducted in accordance with the Declaration of Helsinki and was approved by the Ethics Committee of the Institution.

**Statistical analysis**

Microsoft Excel 2007 and Epi-info version 7.2 (Centre of disease control, Atlanta, USA) used for data storage and analysis. Qualitative data are presented as proportions, whereas continuous data are presented as mean ± standard deviation.

**RESULTS**

A total of sixty patients registered in pediatric immunology clinic with complete records were analyzed. Out of 60 children, 37 were male and 23 were female with a male-to-female ratio of 1.6:1. The minimum age of presentation was 1 year, whereas the maximum age was 16 years. The majority of children (54) enrolled in the clinic hailed from the rural area. Review of the parental education status showed that 13% of the parents were not literate. The mean age of patients at the time of enrollment in pediatric rheumatology clinic was 11.2 ± 3.3 years while the mean age of onset of disease was 9.1 ± 3.6 years. There were two peaks of the age of presentation [Figure 1].

JIA was the most common rheumatological disorder with 48 out of 60 children suffering from JIA. The distribution of different clinical diagnosis is summarized in Figure 2. Out of 4 cases of vasculitis, 2 cases were Henoch–Schonlein purpura and 1 case of polyarteritis nodosa was observed while 1 case of vasculitis remained undifferentiated. One-third of cases of JIA (16/48) were a polyarticular (rheumatoid factor negative) subtype, making it the most common JIA subtype in our cohort followed by oligoarticular persistent subtype (present in 25% of cases) [Figure 3].

Distribution of joint involvement showed that the knee joint was the most affected joint followed by the ankle joint [Figure 4]. Only 2 cases had evidence of uveitis on ophthalmological screening with none among them suffering from debilitating eye symptoms. The serological profile of patients in this cohort is summarized in Figure 5.

After enrollment, 86% of children received NSAIDs. A total of 34 children (56%) received steroids. Among 48 cases of JIA, systemic steroid was used in 27 cases, of which one patient received IV steroids while 26 children received oral steroids. Five children received steroid for prolonged duration with a diagnosis of systemic onset JIA while bridging steroids were used in 22 cases. Intra-articular steroid was given to three children. Among DMARDs, methotrexate was the most commonly used drug. The combination of DMARDs was used in five children. Biologics could not be used in any case due to financial constraints. All patients attending pediatric rheumatology clinic were on regular ophthalmology and physiotherapy care and rehabilitation follow-up. At the time of analysis, remission was not achieved in 9 cases (15% of cases). Out of 34 cases that were in remission, 3 cases were off medications. Seventeen cases (28%) were failed to follow up.

**DISCUSSION**

The prevalence of various rheumatological disorders has conventionally been difficult to estimate. Some population-based estimates from developed nations exist, however, data regarding rheumatological disorders in childhood from developing nation like India are scarce. This study attempts to review data from children registered in pediatric rheumatology clinic of a tertiary institute of the Eastern part of India to provide a sneak peek into the clinico-epidemiological profile of children suffering from rheumatological disorders.
other parts of the world. A recent study from Singapore documented JIA as the most common rheumatologic entity presenting to immunology clinic. The proportion of pediatric vasculitis cases in this study is similar to the studies done earlier from Canada and South Africa. Polyarticular JIA was the most common subtype noted in our cohort which is similar to earlier studies done from Northern and Eastern India. Interestingly, the spectrum of JIA in this study was different from that documented in the United States of America and Europe where oligoarticular subtype was reported to be more prevalent. This finding is in concurrence with the data from the studies, which documented a lower proportion of oligoarticular JIA in children from non-European ancestry. A comparison of JIA subtypes noted in this study with that from other cohorts from India and other developing nations is summarized in Table 1.

Uveitis was only observed in 0.03% of cases of JIA in our cohorts. The relative rarity of uveitis in Asian population is well documented, and this is similar to a large population-based study from Taiwan. ANA was positive only in 10% of cases of JIA in our cohort which is comparable to the relative rarity of ANA positivity seen in other Indian cohorts of JIA.

Our treatment patterns were comparable to the Childhood Arthritis Prospective Study (CAPS) in terms of use of steroid and DMARD. In our 1-year-old cohort, steroid was used in 56% of cases while 86% of children in CAPS cohort received steroids in first 3 years after the presentation to rheumatology clinic. Methotrexate was most commonly prescribed DMARD in this study similar to other cohorts. The major difference noted was in use of biological agents. Unlike Western countries where biologicals are used in about 20%–28.2% of cases of JIA, none of the cases from our cohort received biologicals despite medical indications due to financial constraints. This handicap translated to a greater number of cases, in which remission was not achieved. Prohibitory costs of biologicals agents limiting their use, and therefore, scant experience in Indian settings is a known limitation.

On an average, 5.3 years elapsed from onset of symptoms to enrollment at our clinic. This significant delay in seeking specialized immunology opinion may be reflective of lack of recognition of childhood immunological and rheumatological conditions in general medical community. This delay may also be attributable to community’s reliance on alternative medicine for rheumatological conditions in our state. Before enrollment at our center, every tenth child was being treated with an alternative medicine such as Homeopathy, Ayurveda, or Yunani. Another issue observed in this study was significant attrition rate, with 28% of cases failing to follow up. While nonachievement of remission may be a significant reason, inability to understand the chronicity of these conditions by parents and caregivers was also contributory factor. The fact that 13% of cases had illiterate parents made counseling challenging. With 90% of cases in our cohort residing in villages, poor connectivity in terms of transportation was a hindrance in ensuring regular follow-up.

The limitation of our study being a retrospective hospital-based study and extrapolation of its findings to the general community may be open to questions. However, it does provide a sneak peek into inadequacies of our current health infrastructure in dealing with childhood rheumatological conditions. There is an urgent need for sensitizing medical community in general and pediatricians in particular regarding pediatric immunological and rheumatological diseases. A system of back and forth referrals between tertiary care hospitals and primary health care setups to ensure comprehensive and multimodality care of rheumatological conditions is needed. Endeavors to make treatment with biological affordable through government subsidies and development of more cost-effective protocols for usage of biologicals in Indian settings are also warranted.
**Table 1**: Subtypes of juvenile idiopathic drugs in different studies

| Authors          | Year | Country     | OJIA (%) | POJIA (%) | SoJIA (%) | ERA (%) | Others (%) |
|------------------|------|-------------|----------|-----------|-----------|---------|------------|
| Seth et al[1]    | 1996 | India       | 30       | 46        | 27        | -       | -          |
| Nandi et al[11]  | 2009 | India       | 33       | 34        | 19        | 3       | 11         |
| Singh et al[13]  | 199  | India       | 47       | 37        | 14        | -       | -          |
| Kunjir et al[14] | 2010 | India       | 21       | 29        | 8         | 36      | -          |
| Weakley et al[15] | 2012 | South Africa | 27      | 40        | 8         | 23      | -          |
| Chiepta et al[16] | 2013 | Zambia      | 32       | 46        | 14        | 6       | 1          |
| Our study        | 2016 | India       | 25       | 33        | 10        | 8       | -          |

**CONCLUSION**

We conclude that the clinical and epidemiological profile of children with rheumatological disorders from our region is different from European countries and Western world.

There is a need for further research, especially, pertinent to use of biologicals and their response in our children. The data described here will be useful for both researchers and government agencies in understanding the unique burden of immunological and rheumatological diseases of childhood in Bihar and Eastern India.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

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