Healthcare Use Patterns and Economic Burden of Chronic Musculoskeletal Pain in Children before Diagnosis

Frances Tian, MD[1], Patsy Guittar, RN[2], Melissa Moore-Clingenpeel, MA, MAS[2,3], Gloria Higgins, MD[2,4], Stacy P. Ardoin, MD, MSc[2,4], Charles H. Spencer, MD[2,4], Karla Jones, RN, MS, CPNP[2], Bethanne Thomas, RN, MS, CPNP[2], Shoghik Akoghlanian, MD[2,4], and Sharon Bout-Tabaku, MD, MSCE[5]

Objectives To evaluate the healthcare use and costs of amplified musculoskeletal pain syndrome (AMPS) in children before diagnosis.

Study design We performed a retrospective study in children with AMPS at a pediatric rheumatology clinic between 2010 and 2014. Data were abstracted on 80 patients after primary rheumatic diseases were excluded. Healthcare visits, medications and diagnostic testing that occurred in the years before diagnosis were collected. The Medical Expenditure Panel Survey was used to estimate visit costs.

Results Patients were adolescent females (89%) and white (86%). The median time to diagnosis was 10.2 months. The median pain score was 6.5 and the median Childhood Health Assessment Questionnaire score was 1.1. In this cohort, 29% had at least 1 ED visit and 5% were hospitalized. All patients saw a rheumatologist and 41% had visited another specialist, typically orthopedics and sports medicine. More than one-half had at least 1 radiographic study and 21% had at least 1 magnetic resonance imaging. The total cost for office, emergency department, and hospital visits for AMPS in all 80 patients was $152 853. The mean cost per patient over the entire study period (2008-2014) was $1911 ± $3808, and 43% of costs were outpatient visits.

Conclusions Children with AMPS have high levels of disability and take a long time to be diagnosed. As a result, even before diagnosis, they have high levels of healthcare use, diagnostic testing, and medical costs. Early recognition of disability and quicker referral to trained subspecialists may improve the prognosis, reduce unnecessary testing, and reduce the overall costs of healthcare. *(J Pediatr 2018;■■:■■-■■).*

Musculoskeletal pain affects approximately 1 in 4 children.[1-3] A subset of children experience chronic musculoskeletal pain syndromes, which can include including complex regional pain syndrome (I and II, the former previously known as reflex sympathetic dystrophy), localized and diffuse idiopathic pain, and juvenile-onset fibromyalgia.[4] These diagnoses have overlapping features, so we use the descriptive term amplified musculoskeletal pain syndrome (AMPS).[5] The etiology of AMPS is understood through a biopsychosocial model that includes triggers (physical or other trauma), and modifiable social, environmental, and biological factors.[6] Disordered sleep, joint hypermobility, and heightened peripheral sensory reception are some co-occurring conditions.[1-3,7,8] Patients with localized or widespread musculoskeletal pain can have associated symptoms, including chronic fatigue, cognitive and mood difficulties, headaches, irritable bowel syndrome, and subjective soft tissue swelling, which make the diagnosis challenging.[1] The most effective treatment is a multidisciplinary approach that includes psychological and physical therapy.[7,9-11]

Children with AMPS have poor social, emotional, and physical functioning relative to those without chronic musculoskeletal pain.[12,13] The monetary costs of AMPS are significant because children with chronic musculoskeletal pain seek increased health and social services, including outpatient appointments, occupational therapy, complementary therapy, mental health services, and pain medications.[13,14] In 1 study, approximately 66% of adult family members of children with AMPS reported lost employment time related to the child's pain.[13] Across tertiary care pain centers in the US, the mean annual cost of caring for a child with persistent pain is estimated to be $11 787 and the median cost to be $6770.[11] This study found that the annual societal cost was on par with the high costs of attention deficiency hyperactivity disorder, which is prevalent in children and adolescents.[11] Another study estimated the mean annual cost per patient

AMPS	Amplified musculoskeletal pain syndrome
CHAQ	Childhood Health Assessment Questionnaire
ED	Emergency department
EMR	Electronic medical record
MEPS	Medical Expenditure Panel Survey
NCH	Nationwide Children's Hospital

From the [1]University of Illinois COM-Chicago, Chicago, IL; [2]Nationwide Children's Hospital; [3]Research Institute at Nationwide Children's Hospital; [4]The Ohio State University College of Medicine, Columbus, OH; and [5]Sidra Medicine, Doha, Qatar

Supported by Columbus Medical Research Foundation (to S.B.T.]) and funded by the Rheumatology Research Foundation's (Investigator Award, #82120615). The authors declare no conflicts of interest.

Portions of this study were presented at the American College of Rheumatology annual meeting, November 11-16, 2016, Washington, DC.

https://doi.org10.1016/j.jpeds.2018.01.076

with chronic pain in rheumatology and tertiary pain clinics in the UK to be $16 400.[13]

Studies have documented the cost of chronic pain associated with treatment, yet no studies have assessed the economic burden or healthcare use of patients with AMPS before diagnosis. It is likely that there are great additional costs before diagnosis because diagnosis of pain syndromes is frequently delayed, children with chronic pain and their families often feel minimized and their pain ignored in the medical system, so they seek multiple doctors and multiple tests, and seek unconventional providers and treatments, all leading to greater medical testing and out-of-pocket costs. Our objectives were to describe children presenting to a general pediatric rheumatology clinic and to evaluate their healthcare use patterns and cost estimates before being diagnosed with AMPS. We hypothesized that the duration of symptoms before diagnosis would be associated with higher use, cost estimates, and pain intensity, and that psychological comorbidity would be prevalent.

Methods

A retrospective chart review was approved by the Institutional Review Board at Nationwide Children's Hospital (NCH). The study was conducted using the rheumatology clinic electronic medical record (EMR). Children with a diagnosis from a pediatric rheumatologist of primary chronic musculoskeletal pain, including juvenile-onset fibromyalgia, reflex sympathetic dystrophy, and complex regional pain syndrome between January 1, 2010, and December 31, 2014, were identified. We excluded patients who had an established chronic musculoskeletal pain diagnosis before their initial rheumatology clinic visit, patients with prior treatment at a specialized pain treatment center, and patients with a primary inflammatory diagnosis (such as juvenile idiopathic arthritis or systemic lupus erythematosus). We excluded children with joint hypermobility who often have AMPS as well, so that we could best capture children with primary AMPS. Data abstracted from the NCH EMR by the investigators included approximate month and year of symptom onset, date of diagnosis, number of NCH emergency department (ED) and urgent care visits for pain, number of NCH hospital admissions for pain, duration of hospital admissions, number and type of NCH outpatient visits for pain, medications used before diagnoses for pain, and imaging obtained at NCH before diagnoses for pain not related to injures. Time to diagnosis was calculated based on final diagnosis date minus symptom onset. We recorded visual analog pain scores (scale 0-10) and Childhood Health Assessment Questionnaire (CHAQ) scores that are routinely obtained in our clinic. We assessed psychological comorbidities from the EMR based on recorded diagnoses and confirmed visits with any psychologist and/or psychiatrist. Finally, the data for each subject were reviewed for accuracy by 2 pediatric rheumatology providers.

We used the direct healthcare expenditures captured in the Medical Expenditure Panel Survey (MEPS) database to calculate estimated costs associated with office-based visits, ED visits, and hospitalizations conducted for presenting complaints of pain. MEPS is a survey of noninstitutionalized US families and individuals, and their healthcare providers, that provides nationally representative estimates of healthcare use, costs, payment sources, and insurance coverage. To calculate the costs for a MEPS participant receiving a medical service, we used the total expenditure variable, including costs paid by insurance, out of pocket, or by other sources. We only considered costs for services that involved face-to-face contact with a service provider. Phone calls or visits that were not billed for were not counted. Unit costs for hospital admissions, ED, and ambulatory healthcare visits costs were taken from individual years 2008-2013. We used data from 2008 because many of the costs were incurred up to 2 years before eventual diagnosis. We used 2013 estimates for costs incurred in 2014 because there were no data available for 2014. All mean cost values were calculated using MEPS survey weights. The costs were tabulated for the entire year per subject and averaged to obtain mean estimates. Only visit data from NCH was captured because we did not have access to data from other institutions where the patients may have obtained care. Costs of prescription medications, imaging, and health insurance premiums are not captured by MEPS and, therefore, were not included in cost estimates. Institutional costs for medication and imaging were not tabulated because true out-of-pocket costs vary widely and would not be generalizable.

All analyses were conducted using SAS 9.3 (SAS Institute, Cary, North Carolina) with 2-sided P values considered statistically significant. Group comparisons were assessed using Wilcoxon rank-sum tests for continuous variables and the χ^2 or Fisher exact tests for categorical variables. A signed rank test was used to determine whether the typical time to diagnosis exceeds 3 months. Correlations with time to diagnosis were assessed with Spearman correlation coefficients.

Results

Initially, 342 patients were identified; however, after applying exclusion criteria, 80 patients were included in the study. These were mostly female (89%), white (86%), and had a median age at diagnosis of 15 years (**Table I**). The median time to diagnosis was 10.2 months (IQR, 6.3-12.7) and more than one-half of patients were diagnosed at the first rheumatology visit. At the first visit, the median pain score was 6.5 and the median CHAQ score was 1.1.

As shown in **Figure 1**, over the entire study period and before a diagnosis of AMPS, more than one-quarter of the patients (29%) had at least 1 ED visit but only 4 patients (5%) were hospitalized. All had seen a rheumatologist to receive a diagnosis of AMPS; 41% of patients had visited another specialist before diagnosis. After rheumatology, orthopedic visits were most common, with 11 patients having at least 1 visit, followed by sports medicine with 9 patients having at least 1 visit. Diagnostic laboratory testing was most commonly ordered by the rheumatologist, primary care physician, and other specialists. More than one-half of the patients had at least 1 radiographic study, and 6% of patients had 5 or more

FLA 5.5.0 DTD ■ YMPD9782_proof ■ April 3, 2018