

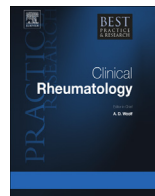


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Models of Care for addressing chronic musculoskeletal pain and health in children and adolescents



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Jennifer Stinson, Senior Scientist, Associate Professor ^{a, *},
Mark Connelly, Professor of Pediatrics ^{b, 1},
Steven J. Kamper, Senior Research Fellow ^{c, 2},
Troels Herlin, Professor of Pediatric Rheumatology ^{d, 3},
Karine Toupin April, Associate Scientist and Assistant
Professor ^{e, f, 4}

^a The Hospital for Sick Children, Lawrence S. Bloomberg, Faculty of Nursing, University of Toronto, Peter Gilgan Centre for Research and Learning, 686 Bay Street, Room 069715, Toronto, ON, M5G 0A4, Canada

^b Division of Developmental and Behavioral Sciences, The Children's Mercy Hospital, 2401 Gillham Road, Kansas City, MO 64108, USA

^c The George Institute, University of Sydney, PO Box M201 Missenden Rd, Camperdown, NSW 2050 Australia

^d Department of Pediatrics, Aarhus University Hospital, Palle Juul-Jensens Boulevard 99, DK-8200 Aarhus N, Denmark

^e Children's Hospital of Eastern Ontario Research Institute, 401 Smyth Road, Ottawa, Ontario, K1H 8L1, Canada

^f Department of Pediatrics, Faculty of Medicine, University of Ottawa, 401 Smyth Road Ottawa, Ontario, K1H 8L1, Canada

A B S T R A C T

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Chronic musculoskeletal pain among children and adolescents is common and can negatively affect quality of life. It also represents a high burden on the health system. Effective models of care for addressing the prevention and management of pediatric musculoskeletal pain are imperative. This chapter will address the following key questions:

* Corresponding author. Fax: +1 416 813 8501.

E-mail addresses: jennifer.stinson@sickkids.ca (J. Stinson), mconnelly1@cmh.edu (M. Connelly), Skamper@georgeinstitute.org.au (S.J. Kamper), troeherl@rm.dk (T. Herlin), ktoupin@cheo.on.ca (K. Toupin April).

¹ Fax: +816 460 1080.

² Fax: +61 2 8052 4301.

³ Fax: +45 7845 1669.

⁴ Fax: +613 738 4210.

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- (1) Why are pediatric-specific models of pain care needed?
- (2) What is the burden of chronic musculoskeletal pain among children and adolescents?
- (3) What are the best practice approaches for early identification and prevention of chronic musculoskeletal pain in children and adolescents?
- (4) What are the recommended strategies for clinical management of chronic pain, including pharmacological, physical, psychological and complementary, and alternative approaches?
- (5) What are the most effective strategies for implementing models of pain care across different care settings?
- (6) What are the research priorities to improve models of care for children and adolescents with chronic musculoskeletal pain?

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Why are pediatric-specific models of pain care needed?

A model of care (MoC) is defined as “an evidence-informed policy or framework that outlines the optimal manner in which condition-specific care should be made available and delivered to consumers.” [1] MoCs bridge the gap between what we know works or does not work in care delivery and practice by describing the “what” and “how” to do it within a health system [1]. A model of service delivery (MoSD), however, converts the principles of a MoC into operational activity and operational recommendations, relevant to the local context. The MoSD acts as a blueprint for the modes of service delivery and evaluation, considering resources, infrastructure, and workforce capacity requirements [1].

Young people (children and adolescents) with chronic musculoskeletal pain (CMP) should not be viewed as “little adults.” The developmental differences among children, adolescents, and adults mean that adult MoCs for CMP cannot simply be applied to pediatrics. CMP is best conceptualized within a biopsychosocial model where pain is influenced by biological/physical factors (e.g., sex, genetic factors, anatomical pathology, and physiological process), psychological factors (e.g., mood, cognitions, and beliefs), environmental factors (e.g., history of parental pain, parent coping and adjustment, school, and socioeconomic factors), and social factors (e.g., peer relationships; see Fig. 1). In each of these domains there are important differences between children and adults. For example, childhood and adolescence is a time of growth of the musculoskeletal (MSK) system with changes to structural properties, biomechanics, and sensorimotor control, before the system stabilizes in adulthood. Similarly, substantial cognitive and emotional developments occur during this time, and hormonal changes regulate mood and emotions differently. Services for young people with CMP need to be sensitive to the issues of growing self-determination and independence, and academic and vocational success, along with physical, psychological, and sexual development. Healthcare providers should have special training to assess and address these developmental needs. Furthermore, the management of young people requires consideration of the complexities commonly at play at this time of life, such as substance use, family dynamics, and mental health issues. Finally, the social relationships, expectations, and environments experienced by children are distinct from those of adults [2]. These differences provide good reasons why specific MoCs are needed for pediatric CMP.

What is the burden of CMP among children and adolescents?

CMP in children and adolescents is defined as any prolonged pain of a MSK origin that lasts at least 3 months or pain that occurs at least three times in a period of 3 months [3]. CMP in children is the result of a dynamic integration of biological processes, psychological factors, and sociocultural context, considered within a developmental trajectory [3]. The pain may fluctuate in severity, quality, regularity, and predictability. MSK pain can be nociceptive (pain that arises from activation of nociceptors due to actual or threatened damage to nonneural tissue), neuropathic (pain caused by a lesion or disease of

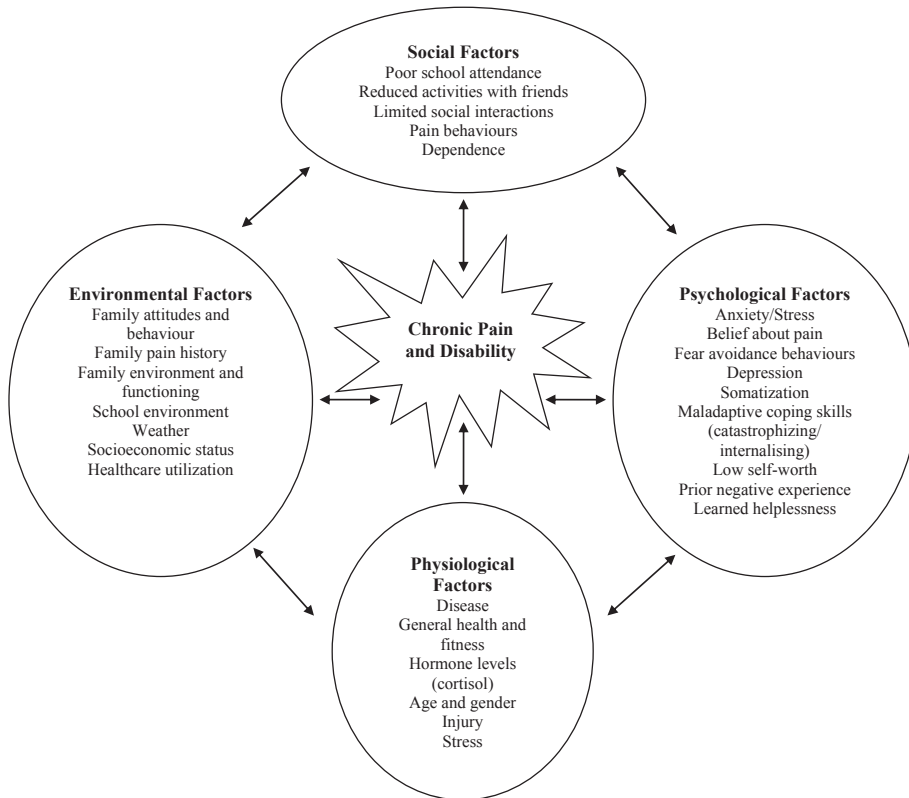


Fig. 1. Factors associated with Children's and Adolescent's Chronic Pain.

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the somatosensory nervous system), or mixed (both nociceptive and neuropathic) [4]. The focus of this chapter will be on CMP.

Prevalence

CMP in children and adolescents is one of the most common reasons to seek medical attention, is costly, and yet remains underrecognized and undertreated. In a systematic review of recurrent and chronic pain in children, the prevalence rates of MSK pain ranged from 4% to 40% increasing with age and being more common in women [5]. The most common pediatric CMP conditions are nonspecific or idiopathic (occur without an identifiable pathological–anatomical basis) and include widespread MSK pain (e.g., juvenile primary fibromyalgia), benign hypermobility syndrome, complex regional pain syndrome (CRPS) Type 1, low back pain, and benign nocturnal limb pain. CMP also occurs in a subset of youth with specific medical conditions such as juvenile idiopathic arthritis (JIA), chronic noninfectious osteomyelitis, hemophilia, scoliosis, etc., and can persist even if disease activity is controlled [6]. Table 1 shows the differential diagnoses. Evidence is also available that CMP in childhood can persist throughout adolescence and increases the risk of experiencing chronic pain in adulthood [7].

Effect

CMP can result in significant disability in a subset of children and adolescents. For example, in the most recent WHO global burden of disease study, low back and neck pain ranked third among

Table 1

Considerations for differential diagnosis and service delivery, as a component of models of care, for the management of musculoskeletal pain in children.

Differential diagnosis	Things to consider in history and physical examination	Suggested laboratory studies, radiographs, and initial treatment
Traumatic <ul style="list-style-type: none"> ■ Overuse injuries 	<ul style="list-style-type: none"> ■ Pattern of repetitive physical activity or repetitive relatively minor trauma 	<ul style="list-style-type: none"> ■ Plain films as an initial study or a bone scan to detect subtle fractures ■ Repeat imaging if pain persists for >2 weeks ■ Treat pain with NSAIDs and ice for comfort. Consider resting the affected area.
Orthopedic/Mechanical <ul style="list-style-type: none"> ■ Legg–Calvé–Perthes ■ Slipped capital femoral epiphysis (SCFE) ■ Osgood–Schlatter (OS) ■ Hypermobility 	<ul style="list-style-type: none"> ■ Pain is likely to have insidious onset, involve point tenderness, and be worse at the end of the day or with increased activity ■ Often presents with a limp in younger children ■ OS presents with point tenderness and occasional swelling at tibial tuberosity ■ Hypermobility syndrome will typically be evident from the physical examination 	<ul style="list-style-type: none"> ■ If Legg–Calvé–Perthes disease is suspected and plain films are negative, obtain MRI or consider bone scan to assess for avascular necrosis ■ For SCFE, obtain plain films, including frog-leg lateral view; consult with or refer to an orthopedist as soon as possible and have the child avoid extended weight-bearing until orthopedic consultation is obtained ■ For OS, X-ray maybe helpful; treat with rest and NSAIDs ■ For hypermobility, use physical therapy with joint protection and NSAIDs for comfort
Infectious <ul style="list-style-type: none"> ■ Lyme Disease 	<ul style="list-style-type: none"> ■ Presents with significant joint (usually knee) swelling, or possible classic rash. Only check Lyme (Western blot of the serum) if traveled to an endemic region (e.g., Eastern Canada and Great Lakes regions of the US). 	<ul style="list-style-type: none"> ■ Obtain CBC with differential, ESR, and CRP for suspected infectious cause. Obtain serum Lyme ELISA and confirmatory Western blot if indicated by travel history.
Inflammatory <ul style="list-style-type: none"> ■ JIA (includes Spondyloarthritis and Psoriatic Arthritis) ■ Postinfectious arthritis (PIA; more acute and very painful) ■ Acute rheumatic fever (ARF) ■ Juvenile dermatomyositis (JDM) ■ Systemic lupus erythematosus ■ Mixed connective tissue disease (MCTD) ■ Transient synovitis of the hip (more acute and very painful) ■ Chronic noninfectious osteomyelitis (CNO)/chronic recurrent multifocal osteomyelitis (CRMO) 	<ul style="list-style-type: none"> ■ In JIA, SLE, MCTD, and JDM – stiffness is worse in the mornings or after naps or sitting for extended periods, and is better with movement/activity ■ Physical examination suggestive of inflammation (e.g., warmth, effusion, or synovial thickening; erythema more with septic joint; tenderness or pain with motion; and decreased or loss of range of motion) ■ ARF presents with painful migratory arthritis ■ In transient hip synovitis – refusal to walk in the absence of fever, decreased range of motion, a flexed, abducted, and externally rotated hip, and a history of an upper respiratory infection is often elicited in cases of transient hip synovitis – acute and very painful ■ Constitutional symptoms (fever, weight loss, and fatigue) may be present in PIA and ARF 	<ul style="list-style-type: none"> ■ Obtain CBC and ESR (decreased hemoglobin, elevated ESR), and increased acute-phase proteins and cytokines correlate with clinical symptoms of disease activity in JIA – most JIA patients have normal laboratory values. May have elevated ESR or CRP, but not typically; 20–30% of children have no pain. ■ Ultrasound can demonstrate fluid in the affected joint if there is arthritis (acute or chronic) ■ Refer to the rheumatology clinic for disease management. Typically treated with NSAIDs (e.g., naproxen) and disease-modifying agents (e.g., methotrexate) or biologic agents (e.g., etanercept) ■ Refer to a pediatric rheumatologist and consider obtaining a bone marrow examination before starting steroid treatment in children with fever, rash, and anemia
Neoplastic <ul style="list-style-type: none"> ■ Leukemia ■ Lymphoma ■ Neuroblastoma ■ Ewing's sarcoma ■ Osteoid osteoma ■ Osteogenic sarcoma ■ Chondrosarcoma ■ Histiocytosis X 	<ul style="list-style-type: none"> ■ Concerning features for neoplasm include severe back pain, night sweats, and non-articular bone pain ■ Unremitting fever and weight loss are additional red flags ■ Acute leukemia in particular can present as a limping child ■ Osteoid osteomas are associated with night pain, often relieved markedly by small doses of NSAIDs 	<ul style="list-style-type: none"> ■ CBC with peripheral smear, ESR, uric acid, and LDH are appropriate initial laboratory screening tests. ■ Imaging studies are dictated by the type of complaint (i.e., plain films may be appropriate for localized bony symptoms, whereas MRI could be considered for soft tissue findings, and a bone scan for more generalized pain).

(continued on next page)

Table 1 (continued)

Differential diagnosis	Things to consider in history and physical examination	Suggested laboratory studies, radiographs, and initial treatment
	<ul style="list-style-type: none"> ■ Bony tenderness (which extends beyond the joint capsule) is suggestive of leukemia ■ Relative thrombocytopenia: markedly elevated ESR and normal platelet count may suggest early finding. Expect platelets to increase with ESR as acute-phase reactant. 	<ul style="list-style-type: none"> ■ Referral to a pediatric hematologist/oncologist is necessary in cases of suspected malignancy. Obtaining bone marrow aspirations and extensive imaging studies before referral is costly and may delay diagnosis and treatment.
Idiopathic Pain Syndromes	<ul style="list-style-type: none"> ■ Growing pains present with “classic” complaints of symmetrical lower limb pain of short duration that awakens the child from sleep and resolves by morning; pain can be quite severe. ■ Idiopathic pain syndromes typically present with longstanding symptoms. Onset is often preceded by trauma (injury, illness, significant psychological stressors) or there is a history of mood, family, and/or behavior problems. ■ Physical examination may reveal tender points and/or pain with gentle touch in the absence of arthritis; for CRPS, affected area may also have swelling, skin color changes, warmth or coolness, sweating, and abnormal hair or nail growth ■ Children may show few pain behaviors and move around with no apparent difficulty during examination, although will often use unique descriptors for pain such as “miserable,” “intense,” “unbearable,” and report high-pain-intensity levels. 	<ul style="list-style-type: none"> ■ Growing pains can be treated without referral. Reassurance, massage, and analgesic therapy (typically NSAIDs) are generally sufficient ■ Laboratory tests including CBC, ESR, CRP, and TFTs are recommended to exclude other etiologies of persistent pain ■ Multimodal treatment (e.g., medication, aerobic exercise, pain coping skills training, stress management, and family therapy) should be initiated in cases with functional disability. Referral to a pediatric pain clinic is recommended when available. If not available, this can be initiated by a primary care physician using local resources.
<ul style="list-style-type: none"> ■ Growing pains ■ Juvenile primary fibromyalgia syndrome (JPFS) ■ Complex regional pain syndrome (CRPS) ■ Low back pain 		

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individuals living with disability within the age range of 15–19 years [2]. In addition to the physical limitations, CMP is associated with impairments in children's academic performance and school attendance, emotional functioning (anxiety, depression, and fear of pain), sleep, and social functioning [6,8,9]. Research on social influence has revealed that families of children with CMP have poorer family functioning (e.g., more conflict and less cohesion) than families of healthy children [10], and siblings are also negatively affected [11]. A subgroup of children and adolescents (5–8%) reported severe disability and distress associated with chronic pain [12]. Miró et al. found that children's self-concept as being disabled, hesitance to perform exercise because of fear of potential injury, and catastrophizing were associated with high disability [12]. Other studies also have found associations between catastrophizing and increased pain intensity and disability [13–16].

Cost

The economic burden associated with CMP in young people can be substantial for families and society. Childhood CMP brings significant direct and indirect costs from healthcare utilization and lost wages due to taking time off work [17]. A recent cost-of-illness study comprehensively estimated the economic costs of chronic pain in a cohort of treatment-seeking adolescents. The estimated mean cost per participant was US\$11,787. The primary driver of costs was direct medical costs, followed by productivity losses. Total costs to society for adolescents with moderate to severe chronic pain were extrapolated to \$19.5 billion annually in the United States [17]. Another study conservatively estimated

the annual financial burden of adolescent chronic pain on the UK economy at £3840 million [18]. Furthermore, 11.6% of children <18 years used complementary and alternative medicines (CAMs) in the United States, and its use was most common among youth with back and neck pain as well as with other MSK conditions [19]. Studies on the actual cost of CAMs used by children and adolescents are scarce.

The current state of contemporary MoCs for children and adolescents

Typically, children and adolescents with MSK pain first present to primary care providers. The majority of these patients can be adequately managed in primary care with reassurance (e.g., pain is not a sign of ongoing damage) and building confidence to return to normal activities of daily life. However, effective risk stratification to determine which patients need subspecialty referral is difficult [20]. Many primary care clinicians report a lack of confidence in their pediatric MSK clinical skills and thus may unnecessarily refer these children who may be difficult to access to subspecialists (pediatric rheumatologists or chronic pain programs) [21–23]; moreover, this access may vary within and between countries. Furthermore, there is a lack of awareness regarding early identification and prevention of risk factors contributing to the development of CMP (e.g., heavy school backpacks and obesity) [24]. Thus, care delivery (ranging from prevention efforts to simple management in primary care to interdisciplinary specialty team management) is not currently being tailored to different risk profiles among children with MSK pain. Consequently, subspecialists are overburdened, resources are managed inefficiently, and unnecessary costs incurred (e.g., children undergoing unnecessary, costly, and sometimes painful investigations) [25]. Children and young people from rural areas are particularly disadvantaged because of the long distances required to travel to access expert care [26–28].

An MoSD for pediatric CMP care is needed that provides an evidence-informed, stepped care, family-centered approach that matches the intensity of services with the presenting issues of the child (phenotype of CMP) and family, is coordinated, and one that is situated within a biopsychosocial framework. The “cradle to the grave” approach used by adult physicians in the past has failed to recognize issues of growth and development, while pediatric physicians continued to care for children into adulthood have been criticized for not recognizing the changing needs of the older adolescents and young adults. It is crucial that MoSD, as a component of MoCs, includes tools to identify those at risk of developing CMP to prevent its development as well as tools to identify barriers to resolution of pain-related disability (See Fig. 1). Furthermore, failure to provide a coordinated transition from pediatric to adult services results in poor adherence to therapy, reduced attendance at follow-up appointments, and suboptimal long-term outcomes [25–28]. Thus, MoCs that facilitate smooth transition and incorporate support for self-management are essential. Currently, there is no standardization or quality assurance in the care of young people with CMP, and there is a lack of research on optimal MoCs for CMP in children, especially concerning the best targets for prevention. Pediatric MoCs therefore require a more standardized approach and rigorous evaluation.

What are the best practice approaches for the early identification and prevention of CMP in children and adolescents?

Importance of identification and prevention as components of an MoC

It is well recognized that pain assessment is the first step in the early identification and effective management of pain. A comprehensive assessment of the severity and impact of pain and the patient's response to treatment is essential for its management. Pain is a multidimensional experience comprising sensory, affective, and cognitive–evaluative components [29]. There are numerous validated unidimensional tools for the self-reporting of pain intensity as well as multidimensional tools that include intensity, location, and word descriptors. The Bath Adolescent Pain Questionnaire (BAPQ) is a well-validated multidimensional instrument that can aid in the comprehensive assessment of CMP in adolescents [30]. Recently, Stinson and colleagues have validated a number of web-based and smartphone-based multidimensional electronic pain assessment tools (e-Ouch, Standardized Universal Pain Evaluation for Rheumatology providers (SUPER-KIDZ) and Pain-QuILT™) [31–33]. These tools can be used in primary care and subspecialty clinical settings to monitor CMP in children in real time [34].

The ability to identify individuals at risk of developing CMP is crucial because it will facilitate timely and appropriate management of those individuals who need care, and ensure that those who do not need it are reassured and not subjected to unnecessary tests and treatments. It is important that transient aches and pains associated with growth, development, and activity are not “medicalized.” Understanding factors surrounding the initial onset is fundamental to any effort at primary prevention. Early intervention in particular is the key to prevent more refractory and disabling pediatric MSK pain disorders [25], because untreated acute MSK pain can change to chronic pain with central sensitization (increased responsiveness of nociceptive neurons in the central nervous system to their normal or subthreshold afferent input) [4,35].

For those who do develop CMP, understanding the natural and clinical course of the conditions and factors associated with poor prognosis is important. There is a growing volume of work in adult populations with regional CPM conditions that aims to stratify risk of chronic pain according to clinical characteristics at presentation. Some of this work has been influential in providing care pathways in the adult primary care setting [36]. Although much of this adult work is recent and preliminary [37], and has yet to have a clinical influence, it holds promise as a fruitful direction for the pediatric field.

What are the best practice components of care for the management of chronic pain in children and adolescents including pharmacological, physical, psychological and complementary, and alternative approaches?

Contemporary MoCs highlight the need to adopt a biopsychosocial approach to manage CMP in children and adolescents using evidence-informed interventions to provide the right care, at the right time, by the right team, in the right place with the right resources [24,38,39]. The components of care that are important at a service delivery level include using a multimodal approach (physical, psychological, and CAM) that is tailored to the individual's needs and pain profile, and typically should include self-management strategies, psychoeducation (understanding the neurophysiology of pain), cognitive and behavioral approaches to pain (e.g., addressing catastrophizing, distress, fear avoidance behaviors, and pacing), and active movement-based therapies (see Table 2 for overview of the 3 P's approach using Pharmacological, Physical, and Psychological strategies). These strategies may need to be combined with pharmacological strategies as appropriate. Most young people with CMP can be adequately cared for in primary care settings by physicians, nurse practitioners, and physical therapists. If CMP is severe and refractory to these initial treatment approaches provided in primary care, children should be referred to specialized interdisciplinary rheumatology or chronic pain teams that are now the standard of care for children with complex chronic pain conditions [40].

Practice point

The ideal MoC is one that provides coordinated and integrated care for young people with CMP across primary and tertiary care settings and adopts a biopsychosocial approach to service delivery using evidence-informed interventions (3 P's approach).

Pharmacological approaches. Pharmacological treatments are used to create a therapeutic window during which self-management strategies can be implemented and reinforced as part of efforts to promote behavior change. According to the WHO guidelines on medical treatment of persistent pain in children, a tiered approach is recommended [41] starting with simple analgesics such as acetaminophen/paracetamol and nonsteroidal anti-inflammatory drugs (NSAIDs) either singly or in combination. In acute flares of pain, the use of rapidly acting NSAIDs (e.g., ibuprofen or naproxen), if necessary in combination with acetaminophen, can be helpful [42]. For more persistent pain, longer-acting preparations should be considered (e.g., celecoxib, diclofenac SR, meloxicam, and piroxicam).

Simple analgesics may be insufficient for controlling CMP in conditions that have symptoms of central sensitization (e.g., allodynia, hyperalgesia, and paresthesias), such as juvenile fibromyalgia or CRPS. In these cases, adjuvant analgesics such as antidepressants or anticonvulsants with activity against neuropathic pain may be indicated. Low-dose antidepressants such as amitriptyline have been shown to reduce pain in juvenile fibromyalgia [43], though one study found that amitriptyline was

Table 2

An overview of 3 P's model of service delivery, as a component of models of care, for managing chronic musculoskeletal pain in children.

Pharmacological approaches	Physical approaches	Psychological approaches
<ul style="list-style-type: none"> • Simple analgesics • Opioid analgesics • Anticonvulsants • Antidepressants • Antiarrhythmics • (alpha-adrenergic blockers) • Anxiolytics • Nerve blocks (i.e., CRPS) 	<ul style="list-style-type: none"> • Exercise • Thermal stimulation (heat, cold, and desensitization) • Physiotherapy • Occupational therapy • Massage • TENS • Acupuncture 	<ul style="list-style-type: none"> • Education (about pain experience and pain problem) • Sleep hygiene • Relaxation • Biofeedback • Behavioral therapies • Cognitive behavioral therapies (CBTs) • Acceptance and commitment therapy (ACT) • Mindfulness therapy • Family therapies

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unlikely to reduce pain by a clinically significant amount in children with polyarticular JIA [44]. Although not completely known, the mechanism of action may be by inhibition of the serotonin–norepinephrine neurotransmitter reuptake leading to reduced perception of pain, including an apparently anti-inflammatory effect. Another selective serotonin–norepinephrine reuptake inhibitor, duloxetine, is approved for the treatment of fibromyalgia in adults [45] and has occasionally been used in children, but pediatric efficacy data are not available. Similarly, calcium channel-blocking anticonvulsants (such as gabapentin and pregabalin) that limit the neuronal release of excitatory neurotransmitters are approved to treat fibromyalgia in adults, but efficacy data are not available for the treatment of pediatric CMP.

Tramadol (a weak mu-opioid receptor agonist and an inhibitor of serotonin and noradrenaline reuptake) has also been recommended for use when NSAIDs or acetaminophen fails [46,47]; however, long-term use and safety of this drug for children with CMP has not been reported. Other opioids are rarely indicated for CMP because of their side effects: drug tolerance, dependence, potential induction of hyperalgesia, and unknown effects on the developing brain [48]. Moreover, the use of opioids for the treatment of CMP has been widely debated. Kimura et al. distributed a questionnaire to pediatric rheumatologists who were members of the Children's Arthritis and Rheumatology Research Alliance (CARRA) network regarding current treatment practices for chronic pain in JIA [49]. No consensus was found regarding the use of opioids (in fact 60% disagreed) for the treatment of residual pain in JIA, and many expressed the concern of adverse effects and dependency when using the drugs. Referral to a pediatric chronic pain clinic may be indicated for cases where use of opioids is clearly indicated (such as avascular necrosis of the hip due to prolonged steroid use in JIA).

To the extent that medication is necessary, it is likely that primary care providers can care for the majority of patients with idiopathic CMP in their practices using simple analgesics. For those with refractory CMP or a condition that requires a pediatric rheumatologist (e.g., JIA) or chronic pain teams (e.g., CRPS), these teams should manage any necessary pharmacotherapy. Once started on the adjuvant pain therapies, most patients can be monitored by their primary care providers.

Practice point

Primary care providers can care for most patients with idiopathic CMP and medications may have a role when they are used as part of a multimodal framework (3 P's approach), although there is little evidence on their effectiveness.

Psychological approaches. Given the role of certain psychological variables in the maintenance, expression, progression, and treatment responsiveness of MSK pain in children, psychological therapies often are an important component of pain care [50]. Relevant to efforts at preventing CMP, there are data suggesting that children with acute or episodic pain who show high levels of worrisome

thoughts about the pain (i.e., catastrophize) are more likely to fall into a vicious cycle of ongoing pain, fear, and prolonged disability [51]. This being the case, screening for fear of pain in youth experiencing acute or episodic MSK pain could help determine the need for early psychological/psychoeducational approaches to reduce the risk of developing disabling CMP [14,15,52]. In this context, education regarding the understanding of chronic pain and its treatment from a biopsychosocial perspective is recommended in MoCs, and should be initiated as early as possible in primary care settings but requires appropriately skilled health-care providers [38].

With regard to formal psychological treatments, the type of psychological therapy studied the most for treating pediatric chronic pain is cognitive-behavioral therapy (CBT) [53]. This type of therapy seeks to engage the child as an agent of change in his/her care and is typically delivered weekly by specially trained psychologists (pain psychologists) over the course of several months [53]. Variables targeted in CBT are those shown to reduce pain perception and disability and include passive or avoidant-coping strategies, fear of pain or movement, catastrophic interpretations of pain, high levels of depression and anxiety, poor sleep habits, autonomic arousal, low acceptance of pain, and (if parents are involved) high parental protectiveness [54–56]. Psychologists with specialized training in pain assessment and pain-focused CBT and acceptance and commitment therapy are relatively rare, however, and are usually connected to academic medical centers and tertiary or quaternary care [57]. Conversely, general psychologists and therapists may be more available in the community but often experience discomfort in addressing pain concerns due to lack of training in pain and may actually do more harm than good by giving improper recommendations [58]. CBT-based treatment delivered remotely (i.e., eHealth interventions supported by computer or phone) to treat chronic pain in youth (including CMP) have been developed in part to address this limited supply of pain psychology resources and could be initiated at the primary care level. Efficacy studies suggest that electronic platforms for providing CBT for pediatric chronic pain are comparable in outcomes to face-to-face therapies [59,60]. For severely disabled youth with CMP, more intensive psychological therapy often is integrated as part of inpatient or day-treatment pain rehabilitation programs. Although the incremental or unique benefit of psychological therapy in this context cannot be determined from existing studies, large and sustained improvements in functional disability have been observed in studies of intensive interdisciplinary pain treatment in mixed samples of youth with chronic pain (including CMP) [61]. A challenge for a successful MoSD, as a component of MoCs for pediatric CMP, however, is to determine when, through what medium, and at what intensity to most efficiently incorporate psychological services.

Practice point

Psychological approaches for preventing and managing CMP range from basic psychoeducation to intensive cognitive-behavioral treatment in a specialty pain rehabilitation setting. Remote electronic delivery of psychological approaches to CMP management may be one means to efficiently address access barriers to psychologists having specialty training to prevent or manage CMP.

Physical approaches. Pediatric patients with pain may receive physical or movement-based treatment from a variety of practitioners. These include medical practitioners, physiotherapists, chiropractors, osteopaths, and exercise physiologists. This therapy may be delivered in primary care from first-contact practitioners or after referral from medical practitioners or insurers. Robust evaluations of interventions for children with MSK pain are few. As an example, a recent systematic review of conservative treatments for back pain in children and adolescents identified in only four randomized controlled trials (RCTs); this can be compared with the thousands of studies conducted in adult populations [62]. Recommendations for an active approach to treating pain (especially CMP) are primarily based on guidelines for treatment of these conditions in adults and the few RCTs that tested exercise-based interventions [63,64]. Paucity of direct evidence aside, active approaches including exercises delivered within a cognitive-behavioral framework that includes techniques such as pacing and goal setting form the standard in pediatric populations and align with current recommendations from pediatric MoCs [26]. Evidence does not support the use of passive therapies such as manual and electrotherapies.

Complementary and alternative medicines

CAMs have historically encompassed the following therapies: (1) natural health products (vitamins, minerals, and herbal therapies); (2) mind and body therapies (meditation and relaxation therapy); (3) physical and movement therapies (massage therapy and yoga); (4) energy therapies (acupuncture and reiki); and (5) whole systems (Chinese medicine, naturopathic medicine and Aboriginal healing traditions). A recent trend within health care is the movement toward integrative medicine, in which evidence-informed CAM therapies are used alongside conventional medical treatments and healthcare is provided by a diverse, interprofessional team of health professionals that includes both conventional and CAM health-care providers [61–63]. Despite the high use of CAM in this population, there is little evidence of their efficacy and safety in pediatric CMP [65,66]. RCTs conducted in youth with JIA for massage, pilates, and yoga have shown encouraging results in terms of pain reduction without any adverse events [67–69].

How are these MoCs implemented?

Integration of services

Coordinated and integrated health services across sectors are an essential ingredient to the successful implementation of CMP MoCs [1]. Access to pain management services is inconsistent both within and across nations, and available health services differ markedly in the types of care offered and how they are delivered and paid for. The result is fragmented care for children and adolescents with CMP, and variable outcomes and significant avoidable healthcare costs [18]. However, there are also many enablers to promote the adoption of best practices in MoSD as a component of MoCs, in pediatric CPM. This requires whole-of-sector change at the system level, at the service delivery level, and at the clinician/consumer level. For example, some of the care disparities may be amenable to policy shifts (system level) that promote team management (service delivery level), support consumers (consumer level) with the knowledge and skills to take an active role in their care, and harness the potential of digital health technologies (system level) to overcome access barriers [21]. Developing innovative MoCs based on building the capacity for comanagement by youth and families as well as a skilled workforce with expertise in CMP care are essential to the sustainable delivery of best-practice MSK MoCs [21].

Harnessing technology to improve access

There is a movement toward building consumer capacity to reduce care disparities related to geographical, financial, or health literacy barriers through digital health technologies such as telehealth, online interventions, and mobile apps [70]. An advantage of these therapies is that they can enable nonspecialists and peers to be trained to provide pain management support, thereby improving access particularly to children and adolescents living in remote and rural areas. Stinson et al. [71] developed and evaluated an online self-management program for youth with JIA that has demonstrated efficacy for increasing disease knowledge and reducing pain intensity using a phased approach. We conducted a needs assessment with youth, their parents, and healthcare providers, and conducted a review of currently available Internet sites for JIA that confirmed that there were no websites providing information on the unique needs of this population. We then used an interdisciplinary team of healthcare providers from the pediatric rheumatology centers in Canada to develop the content for the program. The program will be made freely available on the aboutkidshealth website in the fall of 2016. This research group has also recently developed the iPeer2Peer program, which is a tailored peer-mentorship program that provides modeling and reinforcement by peers (trained young adults with chronic pain and JIA aged 18–25 years who have learned to successfully manage their pain) using 10 Skype video calls over the course of 8 weeks [70]. Again a phased approach was used starting with a needs assessment of youth with JIA to determine the need. We then did a systematic review of peer support programs for youth. An interdisciplinary team of clinicians, researchers, and patients was established to develop the training content. When compared with controls, adolescents who completed the iPeer2Peer program had significant improvement in self-management skills and their

coping efforts were more successful [87]. These novel interventions may be a first step in a tiered approach to provide education and psychological therapies to help children and their families learn to better manage pain and ultimately improve health-related quality of life.

Promoting shared decision making

Although families of youth with CMP wish to obtain information on various treatment strategies to manage pain [67–69,71], it is challenging for healthcare providers to offer evidence-based information about a wide variety of treatment options (e.g., medication, exercises, psychological strategies, and CAM). It is also important to engage youth in decision making and self-management and consider their values and preferences, consistent with contemporary MSK MoC recommendations for implementation. However, clinicians have been shown to deliver care that matches their own preferences [72], which may lead to clinician-centered decisions, possibly hindering their patients' engagement in following their treatment plan. Shared decision making is an approach to care that allows patients and healthcare providers to make a joint decision that takes into account the best available evidence of treatment options and the patient's values and preferences [73]. This process is effective in improving knowledge, helping patients to make decisions, which are consistent with their values, and facilitating patient engagement in decision making, especially when using decision support interventions such as decision aids and decision coaching [74,75]. Efforts should be made to improve shared decision making for CMP within the model of service delivery.

Training of healthcare workforce

One of the core components of a CMP MoC requires having highly trained healthcare providers across primary and tertiary care settings to facilitate the early diagnosis, triage, and management of CMP [40]. Currently, there is a lack of training of healthcare providers in pediatric MSK pain in generalist (primary care and public health) and subspecialty/tertiary care. Training is most commonly delivered in profession-specific curriculum models, thereby failing to capture the necessity for a cross-disciplinary approach, given the multidimensional nature of pain that frequently includes comorbidities. This is highlighted well by the findings of a recent survey of Canadian prelicensure pain curricula documents for dentistry, medicine, nursing, occupational therapy, pharmacy, and physiotherapy students showing that respondents representing the majority (67.5%) of health science programs could not specify designated hours for pain course content or clinical conferences. Veterinary medicine curricula documents were also surveyed for comparison and had five times more pain content than medicine [76]. Furthermore, pain competencies are minimal or not evident in regulatory requirements for licensure of health professionals at the beginning of their careers [77]. Addressing pain from a biopsychosocial perspective requires a healthcare practitioner workforce that understands this broader theoretical model and is willing to integrate the various components of multidimensional care. Ideally, this skill training should be provided at the level of preprofessional training, although the development of postgraduate units, perhaps administered by the professional organizations (e.g., Pain Australia [78]) alongside universities may also be required [79]. There are current efforts to build the primary care workforce in CMP. The University of Toronto Centre for Study of Pain has successfully implemented a Pain Curriculum for prelicensure health science students from six faculties since 2002 [79]. In addition, Campbell et al. have developed an online pediatric pain curriculum that is practically oriented to support and encourage adoption by front-line clinicians [80].

Continuous quality improvement

An important attribute of a successful MoC for pediatric MSK pain is that it is adaptive rather than static. Specifically, model implementation should be continuously evaluated and optimized by prospective large-scale data collection that is accessible and meaningful to relevant stakeholders. A necessary first step toward this end is to standardize (or have some consensus around) the metrics that are most critical for providing care of pediatric CMP. The Pediatric Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (PedIMMPACT) has established core outcomes to measure in

clinical trials of treatments for pediatric chronic pain that also have been applied when standardizing data collection procedures for pediatric MSK pain [32]. Clinical registries with significant bioinformatics support are needed to facilitate tracking of, and regular reporting on, the established key metrics over time, so that MoCs can be evaluated and adapt with new data. Existing examples include the CARRA registry [81] and the European Pharmachild registry, which report multifaceted outcomes in JIA using the Juvenile Arthritis Multidimensional Assessment Report (JAMAR) [82,83]. A comparable large-scale registry specifically for pediatric CMP has recently been developed in Australia [84] and will be important for answering critical MoC questions such as “which treatment types administered by what means and at what intensity produce the maximum benefit and lowest risk/cost for which clinical phenotypes of youth with chronic MSK pain?”

Cost-effectiveness of MoCs

Finally, we have limited data on the cost-effectiveness of multidisciplinary services for children with painful rheumatic conditions and/or CMP related to metrics such as healthcare utilization and clinical outcomes such as reduction in pain, improved school attendance, quality of life, disability, etc. Recently, Campbell et al. evaluated the impact of attendance at an Ontario tertiary-referral pediatric interprofessional chronic pain clinic on health-care utilization [85]. Following the initial appointment and subsequent ongoing care at the chronic pain clinic, there was a significant decrease in cost per patient pre- and postprogram enrolment, the number of emergency room admittances, physician consultations and follow-up appointments, and overall physician services, which sustained for more than five subsequent years [85].

What are the research priorities to improve MoCs for children and adolescents with CMP?

Despite concerning data regarding the prevalence, effect, and long-term consequences of CMP in children and adolescents, the field has not been subject to a concerted and systematic research effort [77]. Consequently, there are major gaps in our understanding of these conditions, which leaves clinicians charged with treating young people with little empirical evidence to help guide their management decisions. Key areas for future research include the following:

- Identifying risk factors for the development and maintenance of CMP in children and adolescents and the predictors of increased use of health services and costs.
- Evaluating ways to train healthcare providers in the interprofessional assessment and management of CMP in children and adolescents.
- Comparative effectiveness research based on registries and/or pragmatic trials to determine the minimum intensity/level of services required for maximum effect for various CMP phenotypes. This information could be used for iterations of pediatric MSK MoCs.
- Exploring how implementation science frameworks such as Consolidated Framework for Research Implementation (CFRI) and A Framework to Evaluate Musculoskeletal Models of Care could be useful in understanding the facilitators and barriers to wide-scale adoption of pediatric MoCs for CMP [86].
- Identifying patient and family research priorities to advocate for funding agencies and policymakers to invest resources into innovative MoCs and service delivery.

Summary

The ideal MoC for children and youth with CMP should provide care that is coordinated and integrated across primary and tertiary care settings by using a workforce that is skilled and trained to work in an interprofessional environment and is underpinned by a biopsychosocial framework using evidence-informed interventions (using 3 P's approach) that are consistent with patients' characteristics, values, and preferences. Unanswered questions that should inform the research agenda include:

- What are the risk factors associated with the development and maintenance of CMP in children and adolescents and the predictors of increased use of health services and costs?
- How can appropriate pain treatments be tailored to individual patients based on their unique pain profiles and preferences?
- What are the minimum intensity/level of services required for maximum effect for various CMP phenotypes?

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