


## SPECIAL POPULATIONS SECTION

# Understanding the Pain Management Landscape Within the US Bleeding Disorder Community: A Multi-Center Survey

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## Abstract

**Objectives.** Pain is a known complication in persons with hemophilia (PWH) as a result of muscle and joint bleeding. Little is known regarding national Hemophilia Treatment Center (HTC) practice patterns related to pain management. The aim of this study was to: 1) Describe pain management practice patterns of HTC providers, 2) Identify gaps and areas of alignment with the CDC pain guidelines, and 3) Address educational opportunities for pain management. This survey is the first extensive description of multidisciplinary practice patterns of pain management for PWH. **Methods.** This descriptive study involved physicians, nurse practitioners, nurses, physical therapists, and social workers from federally funded Hemophilia Treatment Centers (HTC) eligible to complete an online survey exploring pain management practice patterns within the CDC pain guidelines. **Results.** Results of this survey shed light on areas of strength and cohesiveness between HTC providers, including the following: dedication to effective pain management, utilization of non-pharmacological pain options, trial of non-opioid medications first before opioids, maintaining follow-up with patients after opioid prescription initiation, recognizing and utilizing clinically important findings before prescribing opioids, and counseling their patients regarding potential risk factors. **Conclusions.** There remain opportunities to incorporate into clinical practice consistent use of tools such as formal screening questionnaires, opioid use agreements, written measurable goals, ongoing prescription monitoring, and written plans for discontinuation of opioid therapy. These results provide opportunities for improvement in education of HTC team members thus optimizing pain management in persons with bleeding disorders.

**Key Words:** Hemophilia; Bleeding Disorders; Pain; CDC Pain Guidelines; Chronic Pain; Hemophilia Treatment Centers

## Introduction

### Background

Hemophilia is a rare, X-linked genetic bleeding disorder caused by the body's inability to produce normal levels of blood clotting proteins, resulting in delayed clot formation. People with hemophilia (PWH) have a tendency to bleed in muscles and joints, often with little or no trauma. PWH have low levels of either factor VIII (hemophilia A) or factor IX (hemophilia B) [1]. The severity of hemophilia is determined by the amount of factor in the blood where low levels (< 1%) result in greater risk for bleeding (normal factor levels 50%–150%). It is estimated that there are approximately 29,000 persons in the United States with hemophilia, where hemophilia A is 3 times more common than hemophilia B [2].

For many patients with bleeding disorders (PWBD), pain is a part of daily life. PWBD, including PWH, typically experience pain resulting from two interrelated etiologies: 1) bleeds into joints and muscles (i.e., acute pain) and 2) repeated bleeds into the same joints and connective tissues resulting in synovitis, arthritis, and other joint diseases (i.e., chronic pain) [3–6]. Joint bleeds, which account for 70%–80% of all bleeding episodes in persons with severe hemophilia (<1% factor level) can be extremely painful and predispose to a pathologic cycle of repeated bleeding [4, 5, 7]. Repeated hemarthrosis triggers irreversible progressive damage to the joint cartilage and other structures, resulting in hemophilic arthropathy [8]. This joint damage affects bone health, resulting in chronic pain, limitations in mobility, and reduced quality of life (QoL) [9]. The frequency and severity of pain for PWBD varies according to disease, severity, and the effectiveness of management approaches. Ongoing, daily pain in one or more joints affects as many as two-thirds of persons with severe hemophilia [9]. Chronic pain is often associated with depression and anxiety, also observed in PWBD [9, 10]. Approximately 1/3 of PWH report having difficulty coping with pain associated with their disease [11]. Therefore, addressing pain is a critical aspect of hemophilia management and requires management strategies involving multiple disciplines.

### Factor Replacement

On-demand therapy (factor replacement treatment in response to a bleed), if infused promptly, ensures rapid bleed resolution resulting in pain relief [12]. Prophylaxis (scheduled factor replacement to prevent bleeding episodes) reduces the likelihood of bleeds and thus, bleed-related pain, and slows down the progression of joint disease lessening the development of chronic pain [13]. Newer treatment options are now becoming available including gene therapy, which may reduce the bleed frequency and pain issues experienced by PWH.

## Current Strategies in Managing Pain in PWH

Research from the 2012 National Pain Study (NPS) provided a foundation for what is known about pain management among PWH. The average daily pain reported by 764 PWH was 4.22/10 on a 0–10 pain rating scale [14].

### Non-Pharmacological Treatment Options

Outside of an infusion of clotting factor, the standard approach for treating acute pain associated with bleeds is rest, ice, compression, and elevation (RICE). Although the RICE method is a mainstay in the overall medical community for treating injuries, data from the NPS showed that it is underused by PWH [14]. Additionally, the NPS reported limited utilization of a variety of non-pharmacological treatment options for pain management [14]. Research in this area with respect to PWBD is lacking [15]. With the growing concern over opioid misuse and abuse as well as the general stigma tied to prescription pain management, [15] alternative pharmacologic and non-pharmacologic modalities for coping and managing chronic pain are crucial in PWBD including physical therapy, cognitive-behavioral therapies and meditation to name a few. These therapies have grown in popularity in recent years and offer a comprehensive approach as a complement/alternative to traditional pain management techniques [15].

### Oral Pain Medication Options

According to the NPS, the most commonly used drugs by PWH were acetaminophen and non-steroidal anti-inflammatory drugs (NSAIDs) [14]. Other drugs commonly prescribed for short-term pain relief included opioids such as oxycodone, morphine, methadone, and codeine [14, 16].

### Cannabinoids

Cannabinoids are therapeutic options that have received increasing attention, especially as more states have allowed medical or recreational use. There is no research specific to the use of cannabinoids in PWBD. However, in studies done prior to cannabinoid widespread legalization, patients reported some use when asked about illicit drugs [9, 14] to manage pain. In other disease states, such as arthritis, cannabis extracts and synthetic cannabinoids have been shown to be effective in treating acute or chronic pain in clinical and preclinical trials [17].

### Pain Guidelines in the United State

Prevailing approaches for addressing pain, including in PWBD, reflect consensus trends in pain management. In the United States, these trends have been shaped by attitudes toward pain, emerging evidence, and available treatment modalities. The use of opioids for management of acute and chronic pain rose sharply in the United States during the last 30 years.

Between 1999 and 2016, >630,000 persons in the United States died from drug overdoses; most of these drug-related deaths were due to opioids prescribed for pain [18]. The numbers are not known specifically for the bleeding disorders community although anecdotally, there have been a number of overdoses due to opioids and suicides said to be related to uncontrolled pain. These alarming trends in opioid-related fatalities have led to several efforts to decrease use. As part of the urgent response to opioid-related deaths, the Centers for Disease Control and Prevention (CDC) issued the *CDC Guideline for Prescribing Opioids for Chronic Pain* in 2016 [19] with the goal of helping providers select the safest and most effective treatment for their patients. The CDC document proposed solutions and guidance for pain management, including guidelines, policies, monitoring, and the promotion of alternative medicine. Specifically, the guideline addresses determination of initiating and/or continuing opioid pain management; opioid selection, dosage, duration, follow-up, and discontinuation; and assessing risk and addressing harms of opioid use.

### Hemophilia Care and Pain Management

PWBD require coordinated expert hematologic and multidisciplinary care and other ancillary services to achieve optimal outcomes, including those pertaining to pain management. This specialized care is delivered by a core team that includes hematologist, nurse coordinators, physical therapists, and social workers through approximately 140 federally funded Hemophilia Treatment Centers (HTCs) nationwide, which are divided into 12 regions by the CDC [20]. Beyond this core group, other care providers (e.g., dental professionals, genetic counselors, clinical psychologists, orthopedists, etc.) are either available on-site or via established partnerships. HTCs are experts in the diagnosis and management of bleeding and therefore, frequently tasked with the management of pain associated with these conditions [20].

### Rationale for Current Study

A knowledge and attitudes study of HTC providers regarding pain management in 2015 identified substantial knowledge deficits [21]. Additionally, a subsequent study demonstrated significant differences with respect to pain perceptions between providers, young adult and adolescent PWH, and their caregivers [22]. In 2015, the National Hemophilia Foundation's (NHF) Medical and Scientific Advisory Committee (MASAC) recognized the need for a multidisciplinary focused approach to pain management. A pain initiative taskforce (the taskforce) was developed with the charge to identify key issues within the community and avenues to address them. With the advent of the *2016 CDC Guidelines for Prescribing Opioids for Chronic Pain* [19] and emerging therapeutic modalities including medical cannabis, the

taskforce felt a comprehensive assessment of the HTC pain management practices integrating the opinions and viewpoints of the multi-disciplinary HTC team was necessary. Once this assessment was completed, recommendations could be submitted to MASAC that would guide the bleeding disorders community in a more focused approach to managing pain that encompassed the CDC Guidelines. Educational programs could be developed targeted to the areas noted to be weakest in the assessment.

To those ends, the taskforce developed a survey to better understand the HTC's multidisciplinary providers pain management practices. Specific study aims included:

- a. Describe pain management practice patterns of HTC providers
- b. Identify gaps and areas of alignment with the CDC pain guidelines, and
- c. Address educational opportunities for pain management

### Methods

This study was approved by the Institutional Review Board at Munson Medical Center in Traverse City, Michigan. The Pain Initiative Multidisciplinary Subcommittee consisted of physicians (3); nurse practitioners (2); doctor of nursing practice (1); registered nurse (1); social workers (2); psychologists (2); physical therapists (3); patients (2); and a statistician. This sub-committee developed the survey questions based upon the CDC's *2016 Guidelines for Prescribing Opioids for Chronic Pain* and included: a) HTC demographics; location, patient population served, percentage of PWBD with pain, and percentage on opioids, b) practice patterns of providers including use of pain assessment tools, frequency of assessment, use of non-pharmacological pain options, oral pain medication options, comfort level with pain management, access to additional services, and c) tools to monitor opioid management and access to pain specialists. Additional survey questions were contributed by discipline-specific subcommittee members who provide care for PWBD. The survey was not formally validated or previously tested. Qualtrics™ was utilized as the survey platform.

All providers working in federally funded HTCs identified through the CDC's HTC Staff Directory [23] were included; those who provide hemophilia care outside of the federally funded HTCs were excluded. Participants were divided into three groups: physicians/advance practice providers/nurses (P-APP-N), physical therapists (PT), and social workers (SW). It was noted that not all physical therapists were listed in the CDC HTC directory; therefore, physical therapists listed with the National Hemophilia Organization PT group were contacted directly to request participation, and then sent the link via email.

Surveys were sent weekly with an introductory email to every provider for 9 weeks beginning on May 7, 2018

until one of the following happened a) the participant took the survey, b) the participant opted-out, or c) the 9-week survey time frame ended. A “click to consent” page prior to the survey provided informed consent before the confidential and non-anonymous survey was made available to the participant. Data were stored on Qualtrics™, a secure server. Raw data with identifiers were sent electronically directly from Qualtrics™ to the statistician at Munson Medical Center, and then moved to a secure server at Munson Medical Center. Data from individual participants were used only for the purpose of the study. Published data were summarized, de-identified, and reported in aggregate form. Participants could stop the survey and exit at any time. There were no identifiable risks to participants. Data were analyzed using Stata 15 [24]. T-tests and ANOVA with Bonferroni correction for multiple comparisons were used for interval-level data, and the Fisher exact test was used for frequency counts and categorical data.

## Results

A total of 1038 providers received invitations to participate in the survey. Thirty-seven emails bounced back. Of the 330 who started the survey, 236 providers answered all survey questions (response completion rate = 22.7%). Not all providers were required to answer every question due to the response logic used and there were several questions with a small number of missing data for a few providers. All 12 US regions were represented, although 3 participants did not identify their region. There were no statistically significant differences in the ratio of provider types between regions. On average, the survey took 16 minutes to complete.

The P-APP-N group included 77 physicians who identified as hematologists (54.6%), 42 registered nurses (29.8%), 18 nurse practitioners (12.8%), and 4 physician assistants (2.8%) for a total of 141 in the group. Although the highest percentage of all provider groups saw patients across the lifespan, the P-APP-N group were more likely to provide care for either pediatric (51/36.2%) or adult patients only (33/23.4%), while PTs (36/61%) and SWs (27/75%) were more likely to see the lifespan (both children and adults) compared to (57/40.3%) in the P-APP-N group ( $P = .002$ ).

Sixteen survey questions were common between all three provider groups, three were common between two groups, and 42 were specific to a single provider group (P-APP-N, SW, or PT groups, respectively). [Supplementary Data](#) list the survey questions with a crosswalk indicating the questions that were common between provider groups and those that were specific to individual groups.

## Effectiveness of Pain Management

Most respondents in each group believe that their HTC managed pain either moderately or very well. SWs were

more likely than the other two groups to indicate that their HTC managed pain either not well at all or only slightly well (12/33.3%) compared to the P-APP-N group (14/9.9%) and the PT group (4/6.8%).

## Professional Role in Evaluation and Treatment

Over 90% of respondents in the P-APP-N (133/94.3%) and PT (56/94.9%) groups personally evaluated pain in PWBD, compared to 47.2% (17) of those in the SW group ( $P < .0001$ ). The most common reason given for not personally evaluating pain was that it was assessed by another discipline, and one PT and two SWs specifically indicated it was not part of their evaluation. In the comments section of the survey, one SW reported being actively discouraged from evaluating or providing any interventions for pain by other providers in the HTC.

Of those who evaluate pain, the P-APP-N (65/48.9%) and PT groups (33/58.9%) were more likely to use a formal pain measurement tool than those in the SW group (1/5.9%) ( $P < .001$ ), with the majority (> 60%) using the numeric rating scale (see [Table 1](#)).

Survey questions specific to the P-APP-N group focused on medications, comfort level of pain management, tools utilized and follow up assessment plans. Survey questions unique to the PT group focused on how PTs function within the HTC multidisciplinary team, and survey questions specific to the SW group focused on types of pain management counseling provided. Additional detail on the results from these unique survey items will be available in future publications.

## Non-Pharmacologic Treatment

The PT and SW groups, but not the P-APP-N group, were asked about specific recommendations for non-pharmacologic management. The PT group felt moderately to very comfortable with non-pharmacologic pain management (52/88.1%). Most SW respondents (21/58.3%) were at least somewhat comfortable providing pain management counseling to PWBD. The majority of SWs provided pain management counseling at the annual comprehensive clinic only (15/83.3%) followed by telephone consultation (14/77.8%), sessions based on individual need (7/38.9%), home visit (3/16.7%), and teleconsult (2/11.1%). SW respondents who do not provide pain management counseling cited they were not expected to do so (11/61.1%) or that they did not have the training or skills to do so (10/55.6%). Less than half (16/44.4%) of SW respondents indicated that they assessed and provided intervention for needle phobia/aversion as clotting factor is given intravenously. SWs and PTs were asked to list their top five recommended non-pharmacological pain treatment options (see [Table 2](#)). PT respondents were more likely to recommend physical interventions such as orthotics ( $P < .001$ ), splints or braces ( $P < .001$ ), aquatic therapy ( $P < .001$ ), surgery ( $P < .001$ ), massage ( $P = .004$ ), and yoga ( $P = .015$ ),



**Table 1.** Use of formal pain measurement tools

|  | P-APP-N<br>(n = 133) | Physical<br>Therapists<br>(n = 56) | Social Worker<br>(n = 17) | Main<br>effect | P-APP-N<br>vs PT | P-APP-N<br>vs SW | PT vs SW |
|--|----------------------|------------------------------------|---------------------------|----------------|------------------|------------------|----------|
| If you evaluate pain in your bleeding disorder patients, do you consistently use a formal pain measurement tool? | n = Yes (%)          | n = Yes (%)                        | n = Yes (%)               |                |                  |                  |          |
| Yes  | 65 (48.87%)          | 33 (58.93%)                        | 1 (5.88%)                 | <0.001         | 0.135            | <0.001           | <0.001   |
| No   | 68 (51.13%)          | 23 (41.07%)                        | 16 (94.12%)               |                |                  |                  |          |
|  | P-APP-N<br>(n = 65)  | Physical<br>Therapist<br>(n = 33)  | Social worker<br>(n = 1)  | main<br>effect | P-APP-N<br>vs PT |                  |          |
| If yes to the above, which pain measurement tool do you most frequently use?                                     | n = Yes (%)          | n = Yes (%)                        | n = Yes (%)               | 0.561          | 0.349            |                  |          |
| Numeric Rating Scale (0 to 10 scale)   | 40 (61.54%)          | 21 (63.64%)                        | 1 (100.00%)               |                | 1.000            |                  |          |
| Verbal Ratings Scale (word descriptors)  | 4 (6.15%)            | 0 (0.00%)                          |                           |                | 0.297            |                  |          |
| Brief Pain Inventory (BPI)   | 8 (12.31%)           | 6 (18.18%)                         |                           |                | 0.552            |                  |          |
| Face type rating scale (ex: FACES, Oucher, Wong-Baker)   | 12 (18.46%)          | 4 (12.12%)                         |                           |                | 0.567            |                  |          |
| Scale based on function  | 1 (1.54%)            | 2 (6.06%)                          |                           |                | 0.262            |                  |          |

Gray Shading=statistically significant.

while SW respondents were more likely to recommend cognitive interventions such as mindfulness ( $P < .001$ ), cognitive behavioral therapy ( $P < .001$ ), distraction ( $P < .001$ ), and meditation ( $P = .009$ ).

Respondents in the P-APP-N (134/95%), and PT groups (59/100%) were more likely than those in the SW group (31/86%) to educate their PWBD on non-pharmacologic or complementary treatment modalities for pain management ( $P < .001$ ). The average percentage of patients educated on non-pharmacologic or complementary treatment by each group was 58% (P-APP-N), 65% (PT), and 28% (SW), respectively. The most common barriers to recommending non-pharmacologic/complementary treatment modalities were reported to be lack of knowledge regarding risks or costs, distance traveled by PWBD, no time, and treatment not offered in their HTC (see Table 3). SWs were more likely than the other two groups to identify “no time” as one of the top three barriers ( $P = .016$ ).

## Pharmacologic Treatment

### Non-Opioid Management

The P-APP-N group was the only group asked about pharmacologic therapy. Acetaminophen was most frequently recommended for pain management (128/90.8%), followed by Cox II NSAIDs (116/82.3%), and factor replacement (88/62.4%), followed by neuroleptics (54/38.3%), antidepressants (29/20.6%), and Cox I NSAIDs (27/19.2%). Eighty-eight providers (62.4%) said that at least three of the six above therapies were trialed before initiation of opioids. Only two respondents

(1.4%) did not recommend other pharmacologic treatment prior to initiating opioid therapy, although 16 (11.4%) indicated that opioids should be a first line therapy for severe chronic pain.

### Opioid Therapy

P-APP-Ns were asked three general questions about opioid therapy for acute and chronic pain even if they did not actually prescribe opioids. When asked to estimate the percentage of patients who use opioids for acute pain, 23 of 141 respondents (16.3%) stated none of their patients used opioids for acute pain. When asked to estimate the percentage of patients who use opioids for chronic pain, 45 of 139 respondents (32.4%) stated none of their patients used opioids for chronic pain. When asked to estimate the percentage of patients who use opioids for both acute and chronic pain, 73 of 141 respondents (51.8%) stated none of their patients used opioids for both acute and chronic pain.

Of the 141 providers in the P-APP-N group, 97 prescribed opioids. Of the 44 respondents who did not prescribe opioids, 42 were RNs who indicated it was outside of their scope of practice and two with prescriptive authority indicated they chose not to prescribe opioids due to concerns of patients potentially misusing the medications or difficulty monitoring opioid use.

Of 97 prescribers, 14 (14.4%) said that none of their patients received opioids for chronic pain. Of those who did prescribe opioids, 41/96 (42.7%) indicated that none of their patients were treated with both short and long opioids for chronic pain, 67/97 (69.1%) indicated that

**Table 2.** Non-pharmacologic/complementary treatments recommended to PWBD who have pain

|                      | Physical<br>Therapist<br>(n = 59)<br>n = yes (%) | Social<br>Worker<br>(n = 36)<br>n = yes (%) | P     |
|----------------------|--|---|-------|
| Acupuncture          | 3 (5.08%)  | 6 (16.67%)                                  | .078  |
| Aromatherapy         | 0 (0.00%)  | 0 (0.00%)                                   | N/A   |
| Acupressure          | 1 (1.69%)  | 0 (0.00%)                                   | 1.000 |
| Acceptance therapy   | 0 (0.00%)  | 6 (16.67%)                                  | .002  |
| Aquatic therapy      | 44 (74.58%)                                      | 9 (25.00%)                                  | <.001 |
| Biofeedback          | 5 (8.47%)  | 4 (11.11%)                                  | .726  |
| BuzzyBee™            | 0 (0.00%)  | 3 (8.33%)                                   | .052  |
| Chiropractic         | 0 (0.00%)  | 0 (0.00%)                                   | N/A   |
| CBT                  | 3 (5.08%)  | 17 (47.22%)                                 | <.001 |
| Distraction          | 6 (10.17%)                                       | 17 (47.22%)                                 | <.001 |
| Essential oils       | 0 (0.00%)  | 0 (0.00%)                                   | N/A   |
| Imagery              | 2 (3.39%)  | 13 (36.11%)                                 | <.001 |
| Herbal-Vitamins      | 6 (10.17%)                                       | 0 (0.00%)                                   | .080  |
| Therapeutic touch    | 2 (3.39%)  | 1 (2.78%)                                   | 1.000 |
| Humor                | 3 (5.08%)  | 3 (8.33%)                                   | .670  |
| Hypnosis             | 0 (0.00%)  | 1 (2.78%)                                   | .379  |
| Joint injections     | 12 (20.34%)                                      | 1 (2.78%)                                   | .015  |
| Magnet therapy       | 0 (0.00%)  | 0 (0.00%)                                   | N/A   |
| Magic Glove™         | 0 (0.00%)  | 2 (5.56%)                                   | .141  |
| Massage              | 15 (25.42%)                                      | 1 (2.78%)                                   | .004  |
| Meditation           | 3 (5.08%)  | 9 (25.00%)                                  | .009  |
| Mindfulness          | 10 (16.95%)                                      | 23 (63.89%)                                 | <.001 |
| Music or art therapy | 7 (11.86%)                                       | 3 (8.33%)                                   | .737  |
| Orthotics            | 42 (71.19%)                                      | 3 (8.33%)                                   | <.001 |
| Pet therapy          | 0 (0.00%)  | 1 (2.78%)                                   | .379  |
| Psychology           | 4 (6.78%)  | 10 (27.78%)                                 | .007  |
| Reiki                | 0 (0.00%)  | 1 (2.78%)                                   | .379  |
| Splints or braces    | 50 (84.75%)                                      | 7 (19.44%)                                  | <.001 |
| Surgery              | 28 (47.46%)                                      | 2 (5.56%)                                   | <.001 |
| TENS                 | 8 (13.56%)                                       | 1 (2.78%)                                   | .146  |
| Virtual reality      | 0 (0.00%)  | 0 (0.00%)                                   | N/A   |
| Yoga                 | 19 (32.20%)                                      | 4 (11.11%)                                  | .015  |
| None                 | 0 (0.00%)  | 3 (8.33%)                                   | .052  |

CBT=cognitive behavioral therapy; TENS=Transcutaneous electrical nerve stimulation.

Gray Shading=statistically significant.

none of their patients were treated with long acting opioids for chronic pain without use of short acting opioids, and 34/97 (35.1%) indicated that none of their patients were treated with short-acting opioids without also prescribing long-acting opioids.

When initiating or reviewing opioid therapy, 93 (95.9%) of the 97 opioid prescribers consistently reviewed risks and benefits of opioid therapy with their patients, but fewer than half used opioid agreements (43/44.3%), written measurable goals (24/24.7%), a written plan for discontinuation of opioid therapy (24/24.7%), or formal screening tools (19/19.6%). Of the 24 providers who established written measurable goals, the goals focused on a functional or pain interference scale (21/87.5%), adverse effects of medication (13/54.2%), self-management techniques (13/54.2%), or the numeric pain scale (10/41.7%). The majority (69/71.1%) of those who prescribed opioids contacted patients in <9 days to evaluate the balance between benefit and harm.

Only 19/97 (19.6%) used a morphine milligram daily equivalent (MMDE) dose limit which ranged from 10 mg to >200 mg. The majority (89/91.8%) utilized the state prescription drug monitoring program (PDMP) either monthly (31/32.0%), every 3 months (11/11.3%), yearly (3/3.1%) or every 6 months (2/2.1%), only if a problem suspected (26/26.8%), or randomly (16/16.5%). Notably, eight (8.2%) respondents never checked the PDMP. Urine drug screening was utilized by 57(58.8%) opioid prescribers either when there was a suspected problem (27/27.8%), randomly (21/21.7%), monthly (4/4.1%), yearly (3/3.1%), or every 3 months (2/2.1%).

Although eight (8.2%) of the 97 prescribers did not answer the question on the number of days that opioids were prescribed for an acute bleed, the majority of those who answered (69/71.1%) this question limited it to less than 10 days. When the prescriber was aware that a patient used cannabis, 19 (19.6%) would not prescribe an opioid. However, the majority (78/80.4%) would prescribe an opioid on either a case-by-case basis (59/60.8%) or when indicated (19/19.6%) in the presence of cannabis. Prescribers were asked to rank on a 1–5 scale (with 5 being the most important) the importance of 22 different clinical findings when considering the decision to prescribe opioids. Responses with an average rating of 4 or above included: prior nonfatal overdose, current regimen including opioid or benzodiazepine use, history of positive drug screen for illicit substances, personal or family history of substance use disorder, patient description of pain symptoms, intensity of pain, and functional ability (see Figure 1).

### Cannabis Use

Questions about patient's cannabis use were asked of all respondents. PTs were less likely than the other two groups to ask their patients about cannabis use for pain management either with or without a prescription (n=30,  $P = .002$ ), or able to estimate the percentage of their patients who used cannabis for pain management ( $P = .003$ ). Among those able to make an estimate, those in the P-APP-N group had a lower estimate of cannabis use for pain management in their PWBD (15.2%) when compared to either the PT (26.9%,  $P = .029$ ) or SW groups (24.1%,  $P = <.001$ ). The accuracy of respondents' knowledge regarding the legality of cannabis in their state was queried. Three (1.3%) respondents did not provide identification on location, and nine (3.8%) were uncertain if cannabis was legal or not legal in their state. Of the remaining 224 respondents within all groups, 188 (83.9%) correctly classified cannabis as being either not legal or legal in some form. Respondents were more likely to incorrectly classify cannabis as illegal (31/13.8%). Although the frequency of incorrect responses was not significantly different between provider groups overall, more SW incorrectly classified cannabis as legal (3/8.6%) than P-APP-N providers (2/1.4%) or PT (1/2.0%), ( $P = .01$ ).

**Table 3.** Barriers that prevent the recommendation of non-pharmacological treatment options

|   | P-APP-N (n = 141)<br>N = yes (%) | Physical<br>Therapist<br>(n = 59)<br>n = yes (%) | Social Worker<br>(n = 36)<br>N = yes (%) | Main effect | P-APP-N vs PT | P-APP-N vs SW | PT vs SW |
|---|----------------------------------|--|--|-------------|---------------|---------------|----------|
| Lack proven benefit                           | 16 (11.34%)                      | 7 (11.86%)                                       | 1 (2.78%)                                | 0.287       | 1.000         | 0.202         | 0.252    |
| Lack of knowledge re-<br>garding risk or cost | 47 (33.33%)                      | 13 (22.03%)                                      | 14 (38.89%)                              | 0.159       | 0.129         | 0.559         | 0.102    |
| Not offered by our center                     | 38 (26.95%)                      | 17 (28.81%)                                      | 13 (36.11%)                              | 0.535       | 0.862         | 0.306         | 0.500    |
| Pt too far away                               | 44 (31.21%)                      | 27 (45.76%)                                      | 10 (27.78%)                              | 0.100       | 0.054         | 0.840         | 0.089    |
| No time                                       | 40 (28.37%)                      | 10 (16.95%)                                      | 16 (44.44%)                              | 0.016       | 0.108         | 0.073         | 0.005    |
| No insurance                                  | 22 (15.60%)                      | 11 (18.64%)                                      | 4 (11.11%)                               | 0.628       | 0.677         | 0.606         | 0.396    |
| Don't believe insurance<br>covers             | 24 (17.027%)                     | 9 (15.25%)                                       | 3 (8.33%)                                | 0.556       | 0.837         | 0.298         | 0.526    |
| No barrier                                    | 30 (21.28%)                      | 13 (22.03%)                                      | 9 (25.00%)                               | 0.880       | 1.000         | 0.655         | 0.804    |
| Outside scope of practice                     | 15 (10.64%)                      | 2 (3.39%)  | 4 (11.11%)                               | 0.226       | 0.161         | 1.000         | 0.196    |

Gray Shading=statistically significant.

### Resources Used for Managing Chronic Pain

All provider groups were asked about resources used or accessed during the past 12 months when managing chronic pain in PWBD (see Table 4). Significant differences were noted between the groups with respect to resources utilized including: use of opioid risk tools, state opioid monitoring programs, urine drug screens and referrals to a behavioral health provider, pain specialist, and psychologists ( $P < .001$ ), and MASAC guidelines ( $P = .032$ ). While the most common resource utilized was a pain specialist or physical medicine and rehabilitation provider, all groups identified that their patients had at least some difficulty in accessing pain specialists (P-APP-N; 87/61.7%, PT; 21/35.6%, SW; 24/66.7%). The most common barriers to accessing pain specialists in the combined group of providers who reported difficulty with access (132/55.9%) were lack of providers (100/75.8%), insurance or cost (88/66.7%), and pain specialist unwilling to manage opioids (71/53.8%).

### Substance Abuse Treatment

Forty-seven respondents (33.3%) in the P-APP-N group did not know if their PWBD had access to medication-assisted treatment (MAT) with buprenorphine or methadone for opioid use disorder and an additional 17.7% (25) were not familiar with MAT. Of those who were certain that their patients had access to MAT (55/39.0%), over half had not referred any PWBD for MAT during the past 12 months (31/56.4%). Social workers were significantly more confident in their ability to estimate the number of PWBD referred (or would have referred if services were available) for substance abuse treatment ( $P = .016$ ) when compared to the P-APP-N group. In the P-APP-N group, 56 (49%) indicated that they referred (or would have referred if services were available) at least one PWBD for substance abuse services compared to 19 (86.4%) SWs ( $P = .004$ ). Of those who could estimate the number of their PWBD who were currently in a substance use disorder program (P-APP-Ns 78/55.3%; SWs 19/52.8%), SWs (17/89.5%) were more likely

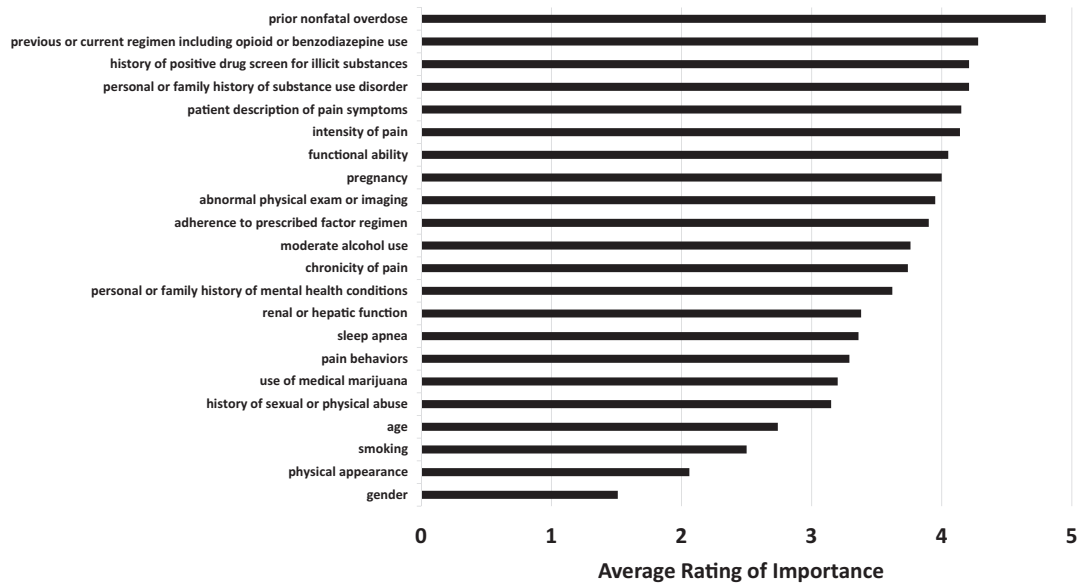
to estimate that at least one of their PWBD were currently in a program compared to P-APP-Ns (25/32.1%) ( $P < .001$ ).

### Discussion

This survey is the first extensive description of multidisciplinary practice patterns of pain management in HTCs across the United States. Conversely, practice patterns with respect to pain management has been more extensively studied among providers at European HTCs, noting suboptimal assessment and resultant treatment, as well as a lack of evidence-based guidelines [25, 26]. Similarly, in the US-based NPS, 39% of patients who sought pain management through their HTC did not feel their pain was well controlled while in this study all disciplines believed their HTC managed pain moderately to extremely well [14]. There are several years between this and the current study, and pain management approaches have changed significantly in the interim. Furthermore, patients and providers may often have differing opinions on pain management. While this dissonance may not mean that pain is being addressed inappropriately, it does offer opportunities for improvement. Further inclusion of the community in pain research would help understand the reasons behind this dissonance. It may be issues such as a difference between goals in pain management.

Comprehensive care of PWBD includes pain management specific to their disease state [6]. As such, it is the responsibility of HTC providers to address pain issues as an important component of care. Pain management should always be multidisciplinary, with each team member having a clear understanding of their responsibilities ensuring that all aspects of pain are addressed in an integrated manner.

Although all of the provider groups reported personal involvement in assessing pain, only 47.2% of the SW group indicated that they personally assess pain.



**Figure 1.** Ratings for importance of specific clinical findings when considering opioid prescribing.

**Table 4.** Resources accessed/used in the past 12 months for managing chronic pain

|                          | P-APP-N<br>(n = 141) | Physical Therapist<br>(n = 59) | Social Worker<br>(n = 36) | main<br>effect | P-APP-N<br>vs PT | P-APP-N<br>vs SW | PT vs SW |
|--------------------------|----------------------|--------------------------------|---------------------------|----------------|------------------|------------------|----------|
|                          | n = Yes (%)          | yes (%)                        | yes (%)                   |                |                  |                  |          |
| Behavior health provider | 38 (26.95%)          | 6 (10.17%)                     | 18 (50.00%)               | <0.001         | 0.009            | 0.015            | <0.001   |
| CAM resources            | 32 (22.7%)           | 16 (27.12%)                    | 6 (16.67%)                | 0.521          | 0.586            | 0.502            | 0.319    |
| CDC 2016                 | 31 (22.00%)          | 9 (15.25%)                     | 4 (11.11%)                | 0.260          | 0.335            | 0.167            | 0.760    |
| MASAC guidelines         | 1 (0.71%)            | 4 (6.78%)                      | 0 (0.00%)                 | 0.032          | 0.027            | 1.000            | 0.294    |
| Opioid risk tools        | 32 (22.7%)           | 0 (0.00%)                      | 8 (22.22%)                | <0.001         | <0.001           | 1.000            | <0.001   |
| PMR provider             | 64 (45.39%)          | 35 (59.32%)                    | 13 (36.11%)               | 0.064          | 0.050            | 0.351            | 0.035    |
| Pain specialist          | 114 (80.85%)         | 31 (52.54%)                    | 26 (72.22%)               | <0.001         | <0.001           | 0.258            | 0.084    |
| Psychology               | 67 (47.52%)          | 8 (13.56%)                     | 13 (36.11%)               | <0.001         | <0.001           | 0.262            | 0.011    |
| State monitoring program | 88 (62.41%)          | 1 (1.69%)                      | 9 (25.00%)                | <0.001         | N/A*             | N/A*             | N/A*     |
| Urine drug screen        | 38 (26.95%)          | 0 (0.00%)                      | 5 (13.89%)                | <0.001         | <0.001           | 0.129            | 0.007    |
| WFH guidelines           | 1 (0.71%)            | 3 (5.08%)                      | 0 (0.00%)                 | 0.070          | 0.078            | 1.000            | 0.286    |
| Other                    | 9 (6.38%)            | 7 (11.86%)                     | 5 (13.89%)                | 0.191          | 0.252            | 0.164            | 0.761    |

CAM=complementary alternative medicine; CDC=Centers for Disease Control and Prevention; MASAC=Medical and Scientific Advisory Committee; PMR=Physical Medicine Rehabilitation; N/A=not applicable; WFH=World Federation of Hemophilia.

\*PT and SW have variable access to state monitoring programs and are therefore not comparable to P-APP-N in this capacity.

Gray Shading=statistically significant.

While SWs were not always directly involved in pain assessment, they were frequently involved in its management providing and/or recommending specific non-pharmacologic treatments and managing many of the psychosocial complications of pain in PWBD. The results suggest inconsistency in the SW role responsibilities in HTC pain management and present an opportunity to recognize the importance of ensuring active collaboration of the multidisciplinary team in assessing and managing pain. Anecdotally, social workers have significant variability in their role and responsibilities within the HTC -some provide ongoing individual non-pharmacologic treatment for pain and others focus primarily on assisting the patient to access resources

within the community. Understanding and defining the role of the social worker in the HTC is an opportunity for future research.

The use of formalized pain assessments tools has been well established in the field of pain management and previous research has validated the use of specific ones in hemophilia yet this study demonstrated a lack of consistent use of any tool, let alone those validated in the hemophilia population [27]. Results identify opportunities to routinely include standardization of assessments during comprehensive clinic visits by the multidisciplinary team including functional evaluations in every initial assessment, with ongoing re-assessment to improve pain evaluation and management in this population.



Despite including opioids in their pain management plan and the 2016 CDC Guidelines recommending their usage, there remain opportunities for consistent use of opioid assessment tools such as formal screening tools, opioid use agreements, written measurable goals, or a written plan for discontinuation of opioid therapy as part of their practice. According to the Guidelines, consistent usage of these tools is an important step toward standardizing pain management practices [19]. The importance of pain management agreements cannot be overemphasized, setting the stage for commitment and collaboration between the HTC provider and their patient, delineating a personalized plan of care, and provide clear expectations for both parties -all resulting in optimal pain management. Areas for opportunity with P-APP-Ns to improve conforming to the CDC Guidelines include ongoing use of state PMDP programs, a MMDE dose limits, urine drug screening, and follow-up.

While illegal federally, the legal status of medical cannabis varies from state to state. Regardless of their location, there were gaps in exploring patients' use of cannabis for pain management as well as in providers' actual knowledge of their state's legal status. While a provider may personally disagree with cannabis use, it is important to understand patient's habits to recognize the implications of their actions in their treatment plan. These identified gaps suggest a need for increased training and/or education for HTC providers in this area incorporating an understanding of the physiologic, psychological, and legal issues surrounding cannabis use as well as state cannabis regulations. In addition, similar to the general population landscape, research in cannabis use for pain management in the bleeding disorders community is lacking, thus providing future research opportunities.

### CDC Guidelines Alignment

All providers recognized the importance of pain management in PWH. Prior to initiating opioid therapy, a vast majority of prescribers trialed non-opioid medications first. The P-APP-N group was consistent in contacting patients post opioid prescription initiation although the time frame varied from between 1 and 3 days to as long as >31 days. Prescribers recognized and utilized clinically important findings before prescribing opioids and 96% counseled their patients regarding potential risk factors (see Figure 1).

Both the PT and SW groups reported utilizing non-pharmacologic options most often within their scope of practice when treating pain in PWBD. A pain specialist was the most frequently accessed resource by the SW and P-APP-N groups and PMR providers were the most frequently accessed by the PT group but there remain challenges to referrals as well as access to these specialists. Insurance companies as well as HTC's should be challenged to explore provisions to provide specialist

consultations and evidence-based non-pharmacologic options to decrease costs and, most importantly, improve pain management in PWBD. In this era of opioid misuse and/or abuse, non-pharmacological options remain viable multimodal pain management opportunities. Results of this study identify opportunities for greater use of these modalities in PWBD suffering pain. It is important for all HTC providers to become educated within this domain, thus gaining more confidence and comfort with their recommendation and usage.

The survey results strengthen the need for evidence-based standards of pain management within the context of the HTC and the multidisciplinary team. A step-wise approach toward achieving this goal within the HTC umbrella would include the development of a road-map that i) delineates the responsibilities of each HTC team member optimizing care and minimizing the duplication of efforts; ii) includes regular educational sessions to improve comfort levels with pain management strategies including non-pharmacologic interventions; iii) offers a stepwise approach to opioid usage with clear recommendations for use of opioid screening tools, and iv) utilizes standard assessment tools including functional based instruments for optimal assessment and re-assessment of pain.

To begin that journey, the taskforce submitted recommendations to the larger MASAC Committee for review and recommendation in February of 2020. After deliberation and discussion, MASAC, and subsequently the NHF Board, approved and published these recommendations (<https://www.hemophilia.org/Researchers-Healthcare-Providers/Medical-and-Scientific-Advisory-Council-MASAC/MASAC-Recommendations/Management-of-Chronic-Pain-in-Persons-with-Bleeding-Disorders-Guidance-for-Practical-Application-of-The-Centers-for-Disease-Control-s-Opioid-Prescribing>). The bleeding disorders community can now look to these recommendations for guidance in managing pain in the inherited bleeding disorders population.

### Limitations

While every attempt was made to include all providers within the bleeding disorders community in this survey, utilizing the CDC's HTC directory, although comprehensive, it may not include every provider working in all HTCs; therefore, some may have not received a survey. Additionally, some surveys may have been blocked by institutional or individual firewalls and some providers chose not to participate. The data collected are specific to PWBD and would be difficult to generalize to other disease states. A greater response rate would have improved statistical power and achieved a more representative sample of providers. It is possible the low response rate may have resulted due to the time needed to complete the online study questions. In an attempt to be conscientious of the length of the survey for subjects, the questions

pertaining to non-pharmacological treatment options recommended to PWBD were not asked of the A-APP-N group, although they were queried regarding barriers to recommending non-pharmacologic/complementary modalities. This additional information could have added depth to this pain management area and provides a future research opportunity to further compare and contrast results with all HTC team members. The regional response rate of respondents may not have accurately reflected the true number of providers who manage PWBD pain. Specific regional practice patterns within this paper were not addressed, but can provide a future opportunity to explore. This study was designed to survey providers only, and the community member's voice will need to be assessed, described, compared, and reported elsewhere.

## Conclusion

While this survey sheds light on areas of strength and cohesiveness between HTC providers (all team members are dedicated to effective pain management), many opportunities for improvement were also noted. Potential solutions to these gaps include the following:

- a. HTC team members involved in pain assessment and management would benefit from continuing education opportunities, due the increasing complexity of pain assessment and management. Given the regionalization of the HTCs, this continuing education may be most effective if it is first standardized at a national level and subsequently disseminated at regional multidisciplinary meetings. Opportunities exist at yearly national meetings of HTC providers, as well as on-line webinar formats. In addition, a separate pain conference specific to HTC providers should be considered.
- b. Standardization of role responsibilities of all team members to maximize effective deployment of their specific training and skills to ensure a comprehensive assessment and ongoing review and management of pain.
- c. Incorporating the CDC opioid guidelines into a specific one for PWBD providing a unique perspective to this disease state which would include:
- d. Routine implementation of opioid agreements to facilitate ongoing discussions between prescribers and patients regarding development of specific and achievable goals, personalizing treatment plans, and establishing initial guidelines for care.
- e. Use of standardized measurement of pain using patient reported outcome tools to provide a reference point for those communicating about patient pain both within and across HTCs,
- f. Establish routine measures to include initial screening of patient opioid use,

as well as ongoing assessment of patients while maintained on opioids as currently recommended within the CDC pain guidelines.

(c) Encourage HTCs to participate in quality improvement programs at both the local and regional level with the use of specific guidelines for benchmarking.

This field is ripe for further research on the impact that changes in pain management has on practice

patterns within HTCs and the effects that these changes have on the health, safety, and well-being of PWBD. This survey, when compared to the NPS, demonstrates a dissonance between providers and patients as to the quality of pain management care provided by HTCs. Continued ongoing research will be required to monitor and assess response to the suggested changes.

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## Supplementary Data

Supplementary data are available at *Pain Medicine* online.

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