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volume 120 · issue 1 · april 2021

The Sudden Savant

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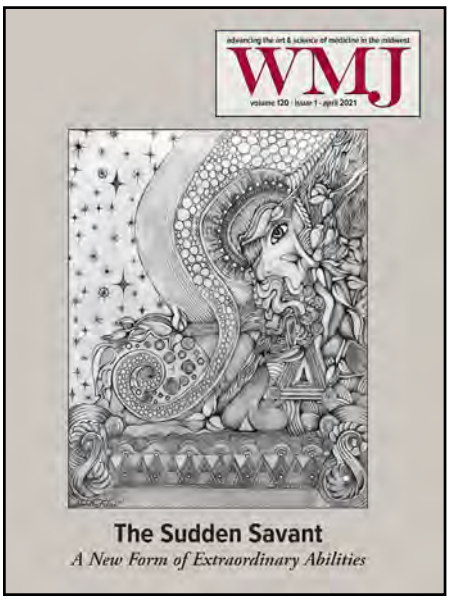

COVER THEME

The Sudden Savant

Savant syndrome previously has been characterized as either congenital or acquired. A report in this issue of WMJ*, however, describes sudden savant syndrome, in which neurotypical persons have the sudden emergence of savant skills without underlying disability or brain injury and without prior interest or ability in the newly emerged skill areas. These skills include music, mathematics, language, and art—which is the case for artist Michelle Felan, whose artwork is featured on the cover.*

Cover illustration: "The Mayan" by Michelle Felan. *Reproduced with permission.*

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EDITORIAL/FEATURE

ORIGINAL RESEARCH

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COVID-19 Outbreak Associated With Ice Fishing

Ice fishing is a popular pastime in northern Wisconsin and Minnesota. Makeshift villages show up on area lakes every winter and remain there most of the winter. Wisconsin mandates that ice houses must be removed from lakes by March 15. This may be the first documented outbreak of COVID-19 associated with ice fishing.

During my regular clinic on Friday, January 29, I was assigned to do COVID testing. Our small clinic at a critical access hospital allows asymptomatic people to be tested by laboratory personnel, but symptomatic people are supposed to be seen by a provider. The first 2 patients were a couple in their 30s. They requested testing after finding out the previous day that someone with whom they had been ice fishing on a lake in northern Polk County, Wisconsin, during a family outing on January 23 had tested positive. Both patients had started to develop mild upper respiratory symptoms on January 27. Rapid antigen testing (Quidel) was positive for both patients. The family then brought multiple other members to our clinic over the course of the day and 9/9 tests were positive.

The patients ranged in age from 4 to 67 years. No one reported sharing beverages, but several meals were consumed and they used 3 ice houses. In total, 11 people were affected, including the presumed index case who was tested early in the week with a polymerase chain reaction (PCR) test at another clinic. Three patients were pediatric (2 of whom were asymptomatic). Subsequently, all 9 positive rapid antigen tests were confirmed positive by a send-out PCR test. Further, genetic analyses indicated that 8 samples shared very similar sequences (0-2 mutations apart), suggesting a common source and/or direct transmission (4 had identical sequences).

When I saw the 67-year-old family member, I alerted her to the availability of administering monoclonal antibodies to mitigate the risk for severe disease. Her 69-year-old husband had tested positive at a drive-through site in Polk County, Wisconsin on January 28 but was not notified about the availability of treatment. He then came into the office and was evaluated as well. He did have a symptomatic hypertrophic cardiomyopathy but was otherwise healthy. After receiving an exam, being provided with the appropriate educational material, and a discussion of risks and benefits, the 67-year-old patient and her husband received infusions of bamlanivamab on the same day.

The 69-year-old husband reported some low-grade fevers and chest pain overnight, such that he presented to our emergency department the following day. Because of an abnormal electrocardiogram and a mildly elevated troponin, he was transferred to the Twin Cities (Minneapolis/St Paul, Minnesota) for evaluation but was discharged after an overnight period of observation without any intervention. The 67-year-old female patient had complete resolution of her symptoms within 2 days.

While preparing this report, our clinic subsequently found a second outbreak associated with ice fishing on a Burnett County, Wisconsin lake.

These may be the first reported outbreaks of COVID-19 associated with ice fishing. As demonstrated by these cases, while ice fishing is usually thought of as a safe outdoor activity, fishing within the more comfortable ice houses available do represent a potential unrecognized hazard.

—Blaise Vitale, MD, FAAFP

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Adapting Medical School Curriculum to Millennial and Generation Z Learners

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mentoring program at The Medical College of Wisconsin,[5] where medical students were paired with undergraduate students at Marquette University and the University of Wisconsin-Oshkosh. This small study evaluated the experience of both groups and suggested positive outcomes. It is a model that deserves further exploration.

Among the variety of other papers in this issue, we are proud to publish a paper by Darold Treffert, MD, and Hunter Ries on the "sudden savant."[6] Dr Treffert, who was a former member of the *WMJ* Editorial Board and died this past December, was an internationally respected researcher in autism, hyperlexia, and savant syndrome who has published widely, including other papers and commentaries in the *WMJ*. (See accompanying essay by John J. Frey, III, MD.[7]) He leaves behind a remarkable legacy and will be missed.

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Adapting Medical School Curriculum to Millennial and Generation Z Learners

Sarina Schrager, MD, MS, *WMJ* Editor-in-Chief

The millennial generation—people born in the early 1980s to 1996—makes up a majority of today's medical students. These young people have grown up with the internet and an almost continuous source of information. They are more diverse, socially responsible, and virtually connected than any generation before them. Millennials also have different learning styles than students in previous generations. Many have shorter attention spans, are used to instant gratification, and want learning to be connected to technology. As such, teaching millennial learners has to adapt to their strengths. Optimal education for millennial learners packages information into bite-sized pieces and provides interactive, experiential, and collaborative learning. The 5 R's of optimal education for millennial learners provides a framework to adapt existing curriculum (See Box).[1]

Box. The 5 R's of Curriculum for Millennial Learners[1]

Research-based methods	Need a variety of different learning approaches; new focus on how each individual learns rather than just standard didactic transfer of information.
Relevance	How does the information relate to their lives and what they will be doing in the future?
Rationale	Need to understand the "why" of content that they are learning.
Relaxed	Low-pressure environment. Millennials do not like hierarchy and enjoy learning in a more casual situation.
Rapport	Focus on personal relationships between the teachers and the learners. Learning in small groups or working in teams.

Generation Z learners (born 1997-2012) are beginning to matriculate in medical school, and they, too, have a penchant for interactive learning, heavy on technology.

Traditional medical school curriculum is full of didactic lectures designed to transfer massive amounts of material from the instructors to the students. The instructor usually creates a Powerpoint presentation with large numbers of slides covering a topic in the basic or clinical sciences. Previous generations of medical students have learned this way, studied the material, and taken tests. And while this learning method worked well for Baby Boomers and Generation X students, it does not match the learning styles of either Millennial or Generation Z learners. Learners from these younger groups want alternative methods of information delivery, in addition to traditional lectures. So, the instructor can add videos or supplemental online content to their slides. Use of cases and clear examples of how the material connects with the outside world is effective. Young learners want the material to be relevant to their lives and the world. So, a lecture on the Krebs cycle should have some clinical application or an example of how the knowledge can be used in a practical way. A focus on milestones and competency-based assessment methods may be an optimal adaptation for medical school curricula.[2] Millennials have grown up receiving almost continuous feedback; they want direct, frequent feedback from their teachers, and assessment of competencies is a good way of providing feedback.

Mentors for Millennial and Generation Z residents also must adapt to their needs.[3] Mentorship will be most successful if it is done in a flexible way. Instead of scheduling appointments, many millennial residents and fellows desire access to their mentors in a much more relaxed and fluid manner. They also look to peers and other team members for mentoring.

A paper in this issue of the *WMJ*[4] describes a study where the researchers surveyed both medical students and faculty to assess how each group perceived how much of the curriculum was lecture only and how much was delivered via alternative methods (like online content or virtual lecture). Interestingly, the faculty felt that they were using alternative methods of information delivery much more than did the students. The students were frequently looking for supplemental information online, which the faculty did not know about. This mismatch in experience underscores the difficulty in teaching transgenerationally.

Another paper in this issue looked at a peer-

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A Tribute to Darold Treffert, MD

John J. Frey, III, MD

Darold Treffert was a remarkable person and physician in so many ways it is difficult to list them all. He was a terrific public-minded clinician, and his tremendous warmth and intelligence no doubt let him explore areas of autism and families into which ordinary clinicians would never have been allowed. He was genuinely interested in the lives of these remarkable people, and he was trusted at a time when medicine was beginning to lose its place of trust in the public eye.

Darold Treffert, MD, 1933-2020

He was an enthusiastic and observant clinician who understood and studied savant syndrome, a variant of the Autism Spectrum Disorder and, through that work, established and led a worldwide network of families and clinicians who helped each other and helped him become a widely known expert in savant syndrome and its manifestations. He wrote about autism and savant syndrome for clinicians and communities and families. He was an engaged and committed member of the psychiatric profession and consulted anywhere he felt that the medical community could benefit from a deeper understanding of an increasingly public behavioral health issue.

He served his profession as a leader of institutions – mental health institutes and psychiatric societies – and of medicine in the state of Wisconsin – he was Wisconsin Medical Society president and board chair and for decades was involved with the State Medical Examining Board, helping physicians in difficulty by holding all of us accountable to our profession's highest ideals. He served the Appleton and Fox Valley region through his work with community organizations and professional service. He was very much in the historical mode of clinicians willing to step up for their profession and civic-minded leaders willing to serve their communities.

When I began with the editorial board of the *WMJ*, Darold was already on the board. He was a warm and compassionate person with a clear idea of the qualities of a caring physician, which he demonstrated through example. All of us benefitted from his tremendous experience and the long view of mental health and of physician behavior he brought to the table and from his willingness to reflect with us on some of the thorny problems of reviewing, editing, and publishing the work of physicians in the state and region. As editor, I would frequently seek his advice on how to deal with people, decisions, and issues, such as making rejections less painful for authors. He was a continued presence in the *WMJ* until his death. And he continued to write and publish new and interesting data and observations about savant syndrome gleaned from the enormous network of families and physicians he created, and he continued to publish his work in the *WMJ*. It was humbling to see his submissions, knowing that he could have published them in a "bigger" and more prestigious journal. Instead, he supported the *WMJ* and undoubtedly helped its visibility and added to its prestige.

So many of us benefitted from Darold's thoughtful counsel: patients and families, colleagues, students and residents, and neighbors. It was a privilege to know him and experience the joy with which he approached medicine and life.

• • •

Dr Frey served as *WMJ* Editor-in-Chief from 2006 to 2019.

The Adaptive Learner: How Faculty and Medical Students' Perceptions of Learning Needs and Desires Differ

Crystal J. Graff, MD; Kristina Kaljo, PhD; Robert W. Treat, PhD; Kate Dielentheis, MD

ABSTRACT

Problem Considered: Medical schools historically have utilized instructor-centered lectures to teach medical students the basic sciences. Several commercial electronic-based resources are now available to enhance lecture-based content. This study examines perceptions between students and faculty regarding the efficacy of lecture-based teaching and learning strategies used by students overall.

Research Methods: The authors distributed surveys to medical students and basic science teaching faculty at the Medical College of Wisconsin. Survey items used categorical and 10-point scales and open-ended text response. Mean scores were compared with independent *t* tests and Cohen *d* effect sizes. Pearson (*r*) and Spearman rho (ρ) correlations were used for relational analysis. IBM SPSS 24.0 was used for statistical analysis, NVivo 11 was used for qualitative analysis.

Results: Faculty's perception of meeting students' learning needs was rated significantly higher (mean [SD] = 7.3 [1.3]) than students (5.9 [2.0]) (Cohen d=1.0/P < .001). There was a significant negative correlation between lectures meeting students' learning needs and time students spent outside of lecture seeking supplemental learning resources (ρ = -0.4/P < .001). Students highlighted their use of personal learning strategies, desire for equitable access to resources, and preparation for national board examinations. Faculty emphasized their perceptions of learning resources, recognition of learning styles, time restrictions, and desire to utilize diverse teaching methods.

Conclusions: Student and faculty perceptions regarding student learning needs were significantly different. Students use lectures extensively, but additionally add to the financial burden of medical school by personally funding supplemental resources. This study helps bridge the gap between medical students and faculty regarding what educational tools are best suited to support a student population with increasingly diverse learning needs.

• • •

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INTRODUCTION

A significant challenge facing the advancement of medical education is the ability to provide an optimal learning environment that addresses the individualized needs of medical students. Since the advent of the Flexner report in 1910, the first 2 years of medical school have classically utilized instructor-centered didactic lectures as the primary modality for presenting basic science content.[1] According to the Association of American Medical Colleges (AAMC) Curriculum Inventory, in the 2018-2019 academic year, the majority of medical schools continued to utilize didactic lectures as their primary source of disseminating material.[2] Nonetheless, fewer than half of medical students report attending lecture "most of the time" or "often."[3] Reasons for forgoing lecture attendance include inability to concentrate for long periods of time and low quality of lectures, as well as time saved and flexibility afforded from watching recorded lectures despite no drop in—or even improvement—in performance on exams.[4-6]

Present-day medical students expect to be heavily engaged by learning material as a result of having technology integrated into nearly every aspect of their lives since birth.[7,8] Students' enjoyment and comprehension of material is enhanced by interactive, multimedia education that incorporates multiple learning modalities.[9-11] The integration of faculty-made technological resources, to be used outside the traditional lecture setting, has been described as highly favored by med-

ical students.[12-14] The prevailing reasons that students prefer these supplemental tools include access to up-to-date information, ease of usability, increased flexibility, improved gratification, and personalization of their learning experience.[15-17]

Despite the availability of faculty-made multimedia resources available at some institutions, many students turn to and purchase higher-quality commercial resources that are often not formally provided by medical education institutions.[5,15] Why students seek out these resources has not been extensively studied, although many hypothesize that it is students' familiarity with technology, in addition to utilization of commercial resources to study for the United States Medical Licensing Examination (USMLE) Step examinations.[18] Another consideration includes the marketing strategy that centralizes on the students' perception regarding a "shortcut to success" and unfounded claims that licensing scores can be purchased.[19] Across the nation, many students begin studying for the Step 1 exam with these commercial resources alongside their preclinical curriculum, thus creating a sort of "self-directed parallel curriculum." The most common reported resources include the USMLE First Aid review book, UWorld question bank, and Pathoma review series.[20] Determining how and which technological resources to use posts a challenge to both educators and learners alike, as there are new and improved modalities developed every day.[8]

Several reported studies have surveyed medical students regarding the relevant medical educational technologies used to study for the USMLE Step 1 examination. To the authors' knowledge, no previous studies have assessed faculty's awareness of these resources or how students utilize them to supplement formal medical school curriculum. The purpose of this study was to analyze perceptions between medical students and faculty regarding the overall efficacy of lecture-based teaching and the corresponding learning strategies that students employ to solidify knowledge. Overall, we sought to evaluate the current educational model in the basic sciences curriculum to determine if it is adequate to meet the needs of present-day medical students.

Table 1. Medical Student Survey

Question	Answer Options
What is your gender?	Male, female, choose not to answer
What is your age?	18-22, 23-27, 28 and above, choose not to answer
During the 2016-2017 academic year, what year in school were you at MCW?	M1, M2, M3, choose not to answer
How do you describe yourself (select all that apply):	White or Caucasian; Black or African American; Asian or Asian American; American Indian or Alaska Native; Hawaiian or Other Pacific Islander; Hispanic or Latino; choose not to answer
I prefer to use lectures as my main source of learning material	Strongly disagree, disagree, neither agree or disagree, agree, strongly agree, not applicable
I attend lecture mainly to absorb as much of the material as possible	
I prefer when lecturers incorporate multiple teaching modalities into their presentations	
I re-watch lectures to review material	
I find that educational resources (other than lecture) often explain material better than lecturers	
I believe that lecturers often relate basic science principles to clinical practice	
In the past academic year, lectures met my learning needs. (Likert scale: 1-10)	(1 = not at all, 10 = very much)
How often do you spend time seeking out additional educational resources to supplement lectures?	Every day, every other day, once per week, rarely
What, if any of the following resources do you regularly use outside of lectures?	Textbooks, individual tutoring, academic enhancement, public websites, evidence-based journals, Sketchy, First Aid, Pathoma, YouTube, Anki, Draw-it-to-Know It, Firecracker, study groups, premade study charts
What, if any educational resources would you like the school to provide for you?	(free response)
Rank the following learning styles in the order that describes you best. (1 = most describes me, 4 = least describes me)	Visual, auditory, read/write, kinesthetic
How often do you attend class/lecture in person?	Rarely, 25%-50% of the time, 50%-75% of the time, nearly 100% of the time
If you don't attend class in person, select the choice below that best fits how you listen to lecture the majority of the time	Live-stream lecture, watch lecture recording at normal speed, watch lecture recording at a faster speed
Tell me more about your personal learning and study habits	(free response)

Abbreviations: MCW, Medical College of Wisconsin; M1, first-year medical student; M2, second-year medical student; M3, third-year medical student.

METHODS

In August 2017, medical students and faculty from the Medical College of Wisconsin (MCW) were invited to participate in this study. MCW consists of 1 central campus in Milwaukee, Wisconsin and 2 regional campuses in Green Bay and Wausau, Wisconsin. Course material in the basic science years (first and second years of medical school) are primarily presented by way of didactic lectures, but also includes a limited number of small-group problem-based sessions. Separate and anonymous web-based surveys were developed using Qualtrics for both student and faculty groups. Survey questions were developed by a team

Table 2. Faculty Survey

Question	Answer Options
What is your gender?	Male, female, choose not to answer
What is your age? choose not to answer	29 and below, 30-39, 40-49, 50 and above,
How many years have you taught M1/M2 classes?	1-5, 6-10, 10 and above, choose not to answer
What is your faculty status at MCW? lecturer, choose not to answer	Assistant professor, associate professor, professor,
How do you describe yourself (select all that apply) Asian or Asian American; American Indian or Alaska Native; Hawaiian or Other Pacific Islander; Hispanic or Latino; choose not to answer	White or Caucasian; Black or African American;
I think students use lectures as their primary source of learning material	Strongly disagree, disagree, neither agree or disagree, agree, strongly agree, not applicable
I think that students attend lecture to absorb as much material as possible	
I often incorporate multiple teaching modalities into my lectures	
I feel that my lectures are organized effectively to best support learning	
I often relate basic science principles to clinical practice in my lectures	
I often wish I had more time to present my lecture material to students	
I think students often seek outside resources because lectures are confusing	
I regularly recommend outside resources to supplement lecture material (other than textbooks	
I am aware of and/or think that students regularly use the following outside resources to supplement lectures journals, Sketchy, First Aid, Pathoma, YouTube, Anki, Draw-it-to-Know It, Firecracker, study groups, premade study charts	Textbooks, individual tutoring, academic enhancement, public websites, evidence-based
In the past academic year, I feel that my teaching methods met the learning needs of the students.	Likert scale: 1-10 (1 = not at all, 10 = very much)
Rank the following learning styles in the order that describes you best. (1 = most describes me, 4 = least describes me)	Visual, auditory, read/write, kinesthetic
What learning style do you believe is most common among M1/M2 medical students?	Visual, auditory, read/write, kinesthetic
Tell me more about your teaching style	(free response)

Abbreviations: MCW, Medical College of Wisconsin; M1, first-year medical student; M2, second-year medical student.

of researchers with extensive experience in medical education research. Questions were analyzed individually by each team member and vetted for statistical quality. Utilizing a listserv provided by the school, a 13-item survey (Table 1) was sent via email to current first-year (M1), second-year (M2), and third-year (M3) students at MCW-Milwaukee, Green Bay, and Central Wisconsin. M4 students were not included given their remoteness to the basic science curriculum and concern that recall of their study habits from 2 years prior may not be accurate. An 11-item survey (Table 2) was sent to basic science teaching faculty who teach 1 or more lectures to the M1 and/or M2 students at MCW. Participants were given 2 weeks to complete the survey and received a reminder email 1 week following the initial email.

Survey items used categorical, 10-point scales (10 = high), and open-ended text-response formats. Mean scores were analyzed with independent *t* tests and Cohen *d* effect sizes. Median scores were compared with Mann-Whitney *U* tests. Frequencies and percentages were analysed with Pearson chi-square tests. Spearman rho (ρ) correlations and stepwise multivariate linear regressions were used for relational analysis. IBM SPSS 24.0 was used for statistical analysis.

Two members of the research team (CG and KK) qualitatively analyzed the open-ended text responses. Beginning with open coding, responses were read independently by each reviewer line-by-line, word-by-word and systematically categorized into recurring concepts. Using the constant comparison method, incidents identified by faculty and students were further distilled into cross-cutting themes to address the research question. The MCW Institutional Review Board approved this study.

RESULTS

Quantitative Results

Twenty-two percent (155/711) of students and 22% (81 of 376) of faculty responded to their respective surveys. Table 3 displays the demographics collected on student and faculty participants. Tables 4 and 5 provide descriptive statistical results corresponding to research questions in the student and faculty surveys that are otherwise unmentioned elsewhere due to low effect size. Faculty's perception of meeting students' learning needs via lectures was rated significantly higher (mean [SD] = 7.3 [1.3]) than students (5.9 [2.0]) (Cohen $d = 1.0$, $P < 0.001$). No significant difference in learning needs being met by lectures was reported between medical student years ($d = 0.4$, $P < 0.069$). Students prefer when lecturers incorporate multiple teaching modalities into their presentations (61%; 96/155). Faculty declared that they often incorporate multiple teaching modalities into their lectures (55%; 45/81). Several students reported attending lectures in person less than 50% of the time (43%; 66/155). Of those students who do not attend class in person, the majority reported watching the lecture recording at a faster speed than normal (63%; 97/155).

Students reported utilizing a significantly higher number of supplemental educational resources (5.9 [2.0]) than faculty (4.7 [2.1]) perceived (Cohen $d = 0.6$, $P < .001$). The top 5 resources used by students included SketchyMedical (114/155, 74%), First Aid (99/155, 64%), YouTube (98/155, 63%), Pathoma (84/155, 54%), and Academic Enhancement (MCW's student-led group tutoring program) (80/155, 52%). Faculty perceived that students predominantly used textbooks (63/81, 78%), public websites (55/81, 68%), study groups (55/81, 68%), First Aid (34/81, 42%), and Pre-Made Study Charts (30/81, 37%) to supplement lectures. The Figure portrays the extent to which students reported utilizing these resources versus those that faculty were aware of and/or thought students were using. On the other hand, 70% (78/112) of M1/M2 medical students preferred to use lectures as their main source of learning materials compared to 39% (14/36) of M3 students ($P < 0.001$). There was a significant negative correlation between meeting learning needs and time spent outside of lecture seeking supplemental learning resources ($\rho = -0.4$, $P < 0.001$). Supplemental educational resources often explain material better than lectures, according to students (56%; 87/155). Students seek out additional educational resources to supplement lectures every day or every other day (68%; 106/155). Many faculty agree that students often seek outside resources because lectures are confusing (35%; 28/81).

Of note, there were no statistically significant differences in responses based on student or faculty demographics such as sex, age, or ethnicity. Furthermore, there were no differences in faculty responses based on faculty status or amount of years teaching M1 and M2 medical students.

Table 3. Student and Faculty Demographics (in % total)

	MCW Students (n = 155)	US Students[a] (n = 92,758)	MCW Faculty (n = 81)	US Faculty[b] (n = 179,238)
Sex				
Male	52.9	49.4	45.7	57.6
Female	45.8	50.6	51.9	42.3
Choose not to answer	0.6	NA	2.5	NA
Age				
18-22	1.3			
23-27	78.1			
28 and above (students)	18.1			
29 and below (faculty)			0	(unknown)
30-39			28.4	
40-49			19.8	
50 and above (faculty)			48.1	
Choose not to answer	1.3		2.5	
Ethnicity				
White/Caucasian	71.6	49.8	79.0	63.5
Black/African American	3.2	7.3	0	3.6
Asian/Asian American	14.2	22.5	13.6	19.9
Hispanic/Latino	3.2	6.5	1.2	3.3
American Indian/Alaska Native	0	0.2	0	0.1
Hawaiian/Other Pacific Islander	0	0	0	0
Other	NA	12.6	NA	5.1
Choose not to answer	6.5	0.9	4.9	4.2
Year in school				
M1	42.6			
M2	29.7			
M3	23.2			
Choose not to answer	0.6			
Years taught M1/M2 classes				
1-5			46.9	(unknown)
6-10			17.3	
10 and above			34.6	
Choose not to answer			0	
Faculty status				
Assistant professor			30.9	46.4
Associate professor			29.6	20.5
Professor			30.9	21.5
Lecturer			2.5	8.9
Other			4.9	2.8
Choose not to answer			0	NA

Abbreviations: M1, first-year medical student; M2, second-year medical student; M3, third-year medical student.

[a]Association of American Medical Colleges. FACTS: Applicants, Matriculants, Enrollment, Graduates, MD-PhD, and Residency Applicants Data. Accessed May 20, 2020. https://www.aamc.org/data-reports/students-residents/report/facts

[b]Association of American Medical Colleges. Faculty Roster: U.S. Medical School Faculty. Accessed May 20, 2020. https://www.aamc.org/data-reports/faculty-institutions/report/faculty-roster-us-medical-school-faculty

Qualitative Student Responses

Upon coding the student responses, 3 themes emerged: (1) recognition of personal learning strategies as an adaptive learner, (2) desire for equitable access to supplemental academic resources, and (3) methods to prepare for national exams.

Recognition of Personal Learning Strategies as an Adaptive Learner – The majority of students who participated in this survey were acutely aware of the individualized learning strategies needed for success. Some students delineated elaborate study plans with multiple steps, while others admitted to listening only to online prerecorded lectures. One student offered this strategy:

> Spend the first few days of a rotation studying from high yield resources (First Aid, Sketchy, Osmosis, Pathoma), study new flashcards, reviewing old flashcards. Only after I feel like I have a good bird's eye view do I start to watch recorded lectures on 2x speed, pausing to take notes when needed to supplement my learning (male, age 28 and above, M2).

These learning strategies and preferences speak to the ability medical students have in serving as "master adaptive learners" or the ability to rely on previous lived experiences to inform the acquisition of new content to meet and exceed the expectations of the curriculum.[21] Another example includes harnessing learning preference in an attempt to make better use of time, when faced with the inability to maintain focus during long periods of lecture time:

> I am not able to pay attention for the entire 4-hour lecture periods. Watching the lectures later allows me to speed up sections I am more familiar with, pause and replay sec-

Table 4. Student Likert Question Answers

Questions		Answers (valid % [raw number])				
Please rank how much you agree or disagree with each of the following statements	**Median (IQR)**	**SD**	**D**	**NAD**	**A**	**SA**
I prefer to use lectures as my main source of learning material	4.0 (2)	8.5 (12)	13.5 (19)	12.8 (18)	6.9 (52)	28.4 (40)
I attend lecture mainly to absorb as much of the material as possible	3.0 (2)	23.2 (32)	21.0 (29)	10.9 (15)	30.4 (42)	14.5 (20)
I prefer when lecturers incorporate multiple teaching modalities into their presentations	4.0 (2)	2.9 (4)	4.4 (6)	22.1 (30)	40.4) (55)	30.1 (41)
I rewatch lectures to review material	2.0 (3)	28.3 (39)	28.3 (39)	11.6 (16)	15.2 (21)	16.7 (23)
I find that educational resources (other than lecture) often explain material better than lecturers	4.0 (2)	0.7 (1)	9.7 (13)	24.6 (33)	34.3 (46)	30.6 (41)
I believe that lecturers often relate basic science principles to clinical practice	4.0 (1)	2.2 (3)	18.0 (25)	24.5 (34)	50.4 (70)	5.0 (7)

Abbreviations: SD, strongly disagree; D, disagree; NAD, neither agree or disagree; A, agree; SA, strongly agree; IQR, interquartile range.

Table 5. Faculty Likert Question Answers

Questions		Answers (valid % [raw number])				
Please rank how much you agree or disagree with each of the following statements	**Median (IQR)**	**SD**	**D**	**NAD**	**A**	**SA**
I think students use lectures as their primary source of learning material	4.0 (1)	1.3 (1)	19.5 (15)	22.1 (17)	37.7 (29)	19.5 (15)
I think that students attend lecture to absorb as much material as possible	4.0 (1)	1.3 (1)	19.5 (15)	24.7 (19)	46.8 (36)	7.8 (6)
I often incorporate multiple teaching modalities into my lectures	4.0 (1)	0 (0)	17.6 (13)	21.6 (16)	43.2 (32)	17.6 (13)
I feel that my lectures are organized effectively to best support learning	4.0 (1)	0 (0)	1.3 (1)	11.8 (9)	57.9 (44)	28.9 (22)
I often relate basic science principles to clinical practice in my lectures	4.0 (1)	0 (0)	4.2 (3)	8.3 (6)	45.8 (33)	41.7 (30)
I often wish I had more time to present my lecture material to students	3.0 (2)	2.7 (2)	31.1 (23)	27.0 (20)	24.3 (18)	14.9 (11)
I think students often seek outside resources because lectures are confusing	3.0 (2)	5.3 (4)	26.3 (20)	31.6 (24)	25.0 (19)	11.8 (9)
I regularly recommend outside resources to supplement lecture material (other than textbooks)	3.0 (2)	2.7 (2)	32.4 (24)	24.3 (18)	28.4 (21)	12.2 (9)

Abbreviations: SD, strongly disagree; D, disagree; NAD, neither agree or disagree; A, agree; SA, strongly agree; IQR, interquartile range.

> tions that are unclear, and pause to take breaks to increase my efficiency overall…I'm not a morning person either, so watching the lectures later in the day allows me to take full use of my most productive times of the day (female, age 23-27, M1).

Students also acknowledged that not much content was lost if lecture was not physically attended:

> I never attended lecture. Prior to watching the lectures, I would make outlines or charts of the lecture PowerPoints…Before a test, I then would review my outlines/charts a few times. This worked extremely well for me, and there were several other students who I knew that had very similar study habits as mine. This allowed me to make my own schedule and gave way more flexibility (female, age 23-27, M3).

Desire for Equitable Access to Supplemental Academic Resources – Excessive cost of ancillary materials and, in turn, a competitive disadvantage emerged as an unanticipated theme. The high cost of medical school tuition coupled with the perceived need to purchase additional study tools becomes financially challenging for students. One student acknowledged:

> There are some students who choose not to buy the UWorld USMLE 1 and USMLE 2 question banks to save money but ultimately, in the long run, these question banks are what lead to success on the boards and NBMEs. Both of these question banks are essential to success and should be included in our tuition (female, age 23-27, M3).

Another student admitted, "I could not afford UWorld myself…I felt this could potentially have put me at a disadvantage compared to my peers at this institution and beyond" (female, age 28 and above, M3). Another student explained, "Review books and materials are pricey, and I found nowhere in our financial aid plan for this, especially third year" (female, age 23-27, M2). Finally, students recognized that they would prefer to make their own choices when purchasing supplemental materials based on their learning style, "Give my money back for the resource the school purchased and let me put it towards review materials or a question bank of my choice" (male, age 23-27, M2).

These comments indicate that students are conflicted by the cost of ancillary resources and are frustrated that access to supplemental resources encourages an unfair advantage for their peers with no financial restrictions. Students did recommend that the school should provide the popular educational resources upon matriculation to ensure equity for students from all backgrounds.

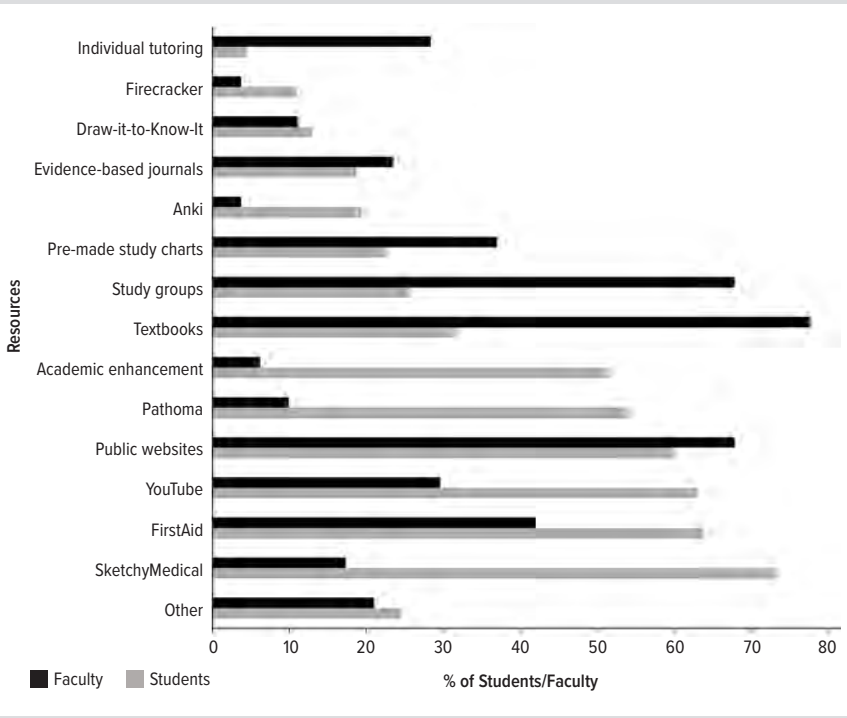
Methods to Prepare for National Exams – Successful completion of the USMLE Step 1 and various national subject exams weigh

heavily on students as they recognize the importance of these test scores on future career choices. This guides their intended methods of study and engagement in the medical school course work. One student explained, "In the first half of the year, I attended lecture all the time, but as Step 1 came, I stopped attending to better study" (male, age 23-27, M2).

The pressures of external, national exams are insurmountable. A reasonable consideration includes access to supplemental resources that parallel course work with national exams to alleviate the associated stress. One student said, "I found outside resources to be much more helpful than a majority of the first- and second-year lectures, as lecture material was not important for Step 1" (female, age 28 and above, M3), while another said, "I think my [NBME subject exam] scores would've been even better if I wasn't required to go to lectures all the time" (female, age 23-27, M3). One student expressed a perception that future residency programs will value Step 1 results more than attained course grades and went so far as to provide the number of hours saved when not attending a class in person:

> In my mind, doing well on Step 1 was so much more important than getting Honors in pre-clinical classes, because Step 1 is what residency programs value the most. Consequently, I gave up getting the extra credit points and didn't go to class in person so that I could have an extra 10 hours per week to study for Step 1 (watching 20 hours of lectures double speed leaves 10 hours saved, plus transportation time to and from school). I think this gave me a good foundation in medical knowledge, and it helped me to do pretty well on the Step 1 exam (male, age 23-27, M2).

Figure. Student vs Faculty Use and/or Awareness of Ancillary Resources



Qualitative Faculty Responses

Faculty open-text survey results highlighted four themes: faculty perceptions of utilized learning resources, recognition of multiple learning styles, restrictions on time in the learning environment and desires for utilization of other teaching methods.

Faculty Perceptions of Utilized Learning Resources – A large percentage of the faculty respondents believed that students primarily and regularly used textbooks to supplement lecture. However, 1 faculty member emphasized that only "a FEW students use the required textbooks," further suggesting that some teaching faculty are cognizant that textbooks may not be as popular or as widely used as others believe. Faculty also perceived students using public websites such as YouTube, WebMD, and Wikipedia in addition to individual tutoring. One faculty (female, associate professor, 6-10 years teaching M1/M2 classes) did recognize that she had no knowledge of the resources students might be supplementing in addition to the traditional lectures and whether they are included in their tuition. Another faculty member indicated their perception of how students want to learn and the implications that has on personal teaching style:

> My experience is that they [students] want information delivered as efficiently as possible, which is a well-designed lecture. Sure, there is a lot of room for other modalities, but lecture bashing is very disappointing to those of us who do an excellent job of it (male, associate professor, >10 years teaching M1/M2 classes).

Recognition of Multiple Learning Styles and How to Address Them – A majority of faculty did recognize that medical students have multiple learning styles. From the survey responses, it appears that faculty are trying to meet the needs of their learners: "…Recognizing that there are multiple learning styles makes it important to present this information in multiple ways – which can be challenging" (female, professor, >10 years teaching M1/M2 classes). To address multiple learning styles, faculty self-identified a diverse set of pedagogical skills when facilitating the required didactic sessions for medical students. In the survey itself, many faculty wrote out an extensive outline on how they prefer to conduct the scheduled didactic time. Some faculty

mentioned gathering student background knowledge, teaching in smaller groups, and utilizing audience response systems:

> I like to survey the students first to see where they are in their learning…This is done in small groups. I do this to ensure that students know what they know. If students truly know the material, I encourage them to teach those that are not 100% clear on the material. I step away but keep my ears open. If the student teacher makes a vital mistake, I step in. Once the students feel they have mastered the material, I give them a clinical situation to test them. Based on their understanding, we adjust from there (unknown sex, assistant professor, 1-5 years teaching M1/M2 classes).

This level of thoughtfulness indicates that faculty are knowledgeable of teaching methodologies, student learning styles, and how to attempt to integrate those in the basic science education: "I aim to use both auditory and visual cues at a minimum and aim to have at least 1 application/kinesthetic activity in each didactic" (female, assistant professor, 1-5 years teaching M1/M2 classes). Another faculty member said, "When in large group teaching sessions, I like to mix methods. I usually do mini-lectures broken up by student small-group work. I often use audience response. I like to use humor as well" (male, associate p[rofessor, >10 years teaching M1/M2 classes).

Restrictions on Time in the Learning Environment – One of the most prevalent themes that emerged from the survey data was time constraints when teaching. A majority of the faculty responded that due to severe time restrictions and a large student body, lecture was the most concise and reliable method to get content to students. This was noted by both junior and senior faculty: "Time is THE constraint and lectures remain the best way to communicate large amounts of information" (male, associate professor, 10 or more years teaching). There is also mention of the negative connotation when referencing "lectures:" "Lectures are often considered a 'dirty word' as noted by one of our participants; however, when time is limited and content needs to be disseminated to a large body of students, faculty doubt that there are many tangible solutions" (male, associate professor, >10 years teaching M1/M2 classes).

Desires for Utilization of Other Teaching Methods – Conversely, a few faculty responded that lecture was not the favored method and that they would prefer to incorporate different strategies:

> Would prefer to move away from lecture toward small group discussions/focus groups in which students apply material that they have been given prior to class time. I do not like passive learning environment of lecture formats and would prefer a more active/dynamic format afforded by small group discussion requiring the learner to self-learn from previously deposited resources (male, assistant professor, 1-5 years teaching M1/M2 classes).

DISCUSSION

This study further informs the argument that traditional medical education does not adequately meet the needs and goals of medical students. As hypothesized, student and faculty perceptions regarding student learning needs were significantly different. The large effect size, coupled with a difference of over 1 expensive resource requiring the added burden of cost, time to locate, and review, indicates the educational relevance of the finding. Our study echoed the results of other studies in regard to lecture attendance and how students are utilizing lecture recordings. It was also clear that students prefer acquiring knowledge from interactive learning modalities and that faculty, in fact, desire – and many times attempt – to provide this to students through their instruction. Most medical students in this study reported using lectures as their main source of material but are spending time and money on additional educational resources available outside of the formal curriculum to enhance their learning and success on national exams. Thus, utilization of these outside educational resources is supplementing, or even replacing, material learned in the didactic lecture setting.

The USMLE Step 1 examination historically has had an inescapable effect on residency selection and career choices.[22,23] As a result, this exam has an unfortunate but very significant impact on medical student study patterns. However, in the spring of 2020, the USMLE announced that scoring for the USMLE Step 1 examination would switch to pass/fail in the near future.[24] In this announcement, stakeholders highlight the current "overemphasis" on the numeric score of the Step 1 examination, as well as its detrimental effect on medical student "well-being." While many perceive this as a step in the right direction for medical education as a whole, there are several implications that must be considered. These include a possible shift in emphasis to the numeric score of the USMLE Step 2 CK examination, as well as relying more heavily on medical school prestige in selection of residency applicants.[25] Supplemental commercial resource companies have already picked up on this trend, including the brand SketchyMedical, which released videos to assist in studying for the Step 2 CK examination shortly after this announcement.

The large volume of available products poses a challenge to learners as they must evaluate these resources for their usefulness and credibility prior to usage. For instance, the list of resources included in our survey is nowhere near comprehensive and was simply created via informal survey of MCW students. Therefore, learners allocate time and money seeking out and investing in these educational tools, which are used to supplement lecture-learning and prepare for institutional and/or national board examinations. Unexpectedly, the results of our study highlighted the large financial strain that individually purchasing ancillary resources places on students. Medical school graduates often complete their training with loans exceeding

$200,000.[26] Greater amounts of medical school debt have been shown to influence career decisions such as specialty selection, as well as personal choices regarding when to get married and have children.[27] Students often discover that they must allocate loan money to purchase perceived essential ancillary educational resources. Students who cannot afford these resources are placed at a competitive disadvantage, which may reduce their potential success in medical school and beyond. In prior studies, a financial need was significantly correlated with lower Step 1 scores.[22] Unfortunately, medical students learn early on in their medical education that any disadvantage that prevailed prior to matriculation may continue to plague their future goals – whether that is attainment of core competencies necessary for physicians, graduation requirements, or even specialty selection.

Results generated from self-reported surveys have some limitations. The overall response rate was relatively low, which decreases construct validity and interpretation of analytical results. Data from a single institution limits the generalizability of results. The survey was developed specifically for this study, thus limiting some elements of validity. Content validity is reduced by the survey's finite list of educational software. Concurrent validity of survey outcomes with student performance was precluded as survey results were anonymous.

The strength of our study included the fact that we garnered views from both students and the faculty who teach them, thus allowing for intriguing comparisons. Of the participants who responded, there was a reasonable spread between year in school of students and time that faculty had spent teaching M1 and M2 students.

Our study yielded compelling results on the use of supplemental resources by medical students, but additional research must be generated. Well-developed didactic lectures can remain a vital part of curricula but must be balanced with active-learning strategies to maximize student learning. Ultimately, 1 common theme remains certain: learner-centered medical education should be a principal focus of all medical curricula, in order to support adaptive learners who are prepared for lifelong learning.[28-31]

CONCLUSION

Student and faculty perceptions regarding student learning needs were significantly different. Students use lectures extensively but additionally add to the financial burden of medical school by personally funding supplemental resources. This study helps bridge the gap between medical students and faculty regarding what educational tools are best-suited to support a student population with increasingly diverse learning needs.

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Establishment and Retrospective Analysis of a Pilot Peer Mentorship Program

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ABSTRACT

Background: Studies suggest widespread advantages to peer mentoring programs; however, there is minimal data pertaining to medical students mentoring undergraduate students.

Objectives: To determine the feasibility and perceived effectiveness of a medical student-undergraduate student peer mentorship program.

Methods: A needs assessment guided the development of Pre-Med Pair Up, a program connecting medical student mentors from the Medical College of Wisconsin and other US medical schools to undergraduates at Marquette University and the University of Wisconsin-Oshkosh to provide peer mentorship, premedical resources, and global health information. After 6 months, surveys were distributed to 43 premedical and 26 medical students to evaluate the program. Descriptive statistics and Pearson correlations (*r*) were used to assess the relational strength between program components and student confidence and knowledge.

Results: Eleven undergraduate and 26 medical students completed surveys. Most undergraduates expressed increased confidence in abilities as premedical students associated with program involvement (18.2% great, 27.3% moderate, 45.5% minimal, 9.1% no improvement). Increased confidence was strongly correlated with knowledge of volunteer opportunities ($r = 0.887$, $P < 0.001$) and feelings of preparedness for the medical school application process ($r = 0.854$, $P = 0.001$) and curriculum ($r = 0.871$, $P < 0.001$).

Conclusion: While self-reported confidence improved and overall positive program outcomes were statistically significant, the number of participants was low and the number who completed mid-year surveys was even lower. Therefore, no conclusions about program effectiveness were made. Instead, a lessons-learned approach was used to discuss the pilot development, implementation, and suggestions for future program installment.

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INTRODUCTION

Data released by the Association of American Medical Colleges (AAMC) in 2015 reported that the number of students enrolling in US medical school programs increased 25% between 2002 and 2015. The number of first-time applicants increased by 4.8%, while total number of applicants increased 6.2% from the previous year.[1] This rise in application rates indicates an increasing interest in medicine and suggests that there are many individuals who could benefit from a mentorship program that facilitates partnerships with medical students.

Literature suggests widespread advantages of peer mentoring for both mentors and mentees,[2-5] including skill building, community engagement, knowledge acquisition, cultural competency, and feeling valued and supported.[6] The Medical Student Mentorship Program at John A. Burns School of Medicine in Hawaii has focused on fostering relationships between undergraduate students, medical students, and faculty to guide undergraduate premedical students through the medical school application process since 2002.[7]

More recently, the University of California Irvine School of Medicine created and publicized an innovative summer enrichment program to incorporate multiple levels of mentorship that, over its first 3 summers (2010-2012), involved 253 high school students, 48 undergraduate students, 12 medical students, and several faculty and additional staff, such as registered nurses.[8] All undergraduate and medical school students self-reported enhancement of teaching and leadership skills, self-confidence, and motivation toward careers in academic medicine.[8]

Figure 1. Program Logo

In the past decade, virtual programs have become more popular. Virtual programs involving telementoring offer the possibility for cost-effective, large-scale programs that are more widely available and accessible.[9] The Society for Academic Emergency Medicine sponsored a virtual advisor program to provide meaningful career guidance to national and international students either without access to emergency medicine providers at their home institution or who desired counsel in a specific area within the specialty.[10] In its pilot academic year 2001-2002, it facilitated mentorship pairs between 264 medical students and 121 emergency medicine faculty mentors.[10] Feedback about the program from participants was generally positive, and the virtual nature of the program allowed mentorship to be provided at a distance.[10]

Although there are many examples of mentorship programs in the medical field, very few published programs focus on pairing premedical students with medical students who attended the same undergraduate university. The purpose of this manuscript is to describe the development and evaluation of "Pre-Med Pair Up," a unique medical student-undergraduate student distance peer mentorship program that offers guidance for medical school preparation, enhances global health awareness, and identifies local health care outreach opportunities.

METHODS

Program Development

Three medical students at the Medical College of Wisconsin (MCW) developed Pre-Med Pair Up: A Medical Mentorship and Global Awareness Program (PMPU) in 2015 to facilitate mentorship between currently enrolled medical students and undergraduate premedical students. The program incorporated global health education, as this was of growing interest to many students, and it exposed students to opportunities in medicine and volunteerism. The 1-year pilot program was implemented in 2016, with intentions of renewing the program for additional years depending on outcomes.

Marquette University and the University of Wisconsin-Oshkosh (UW-Oshkosh) were the 2 undergraduate institutions selected to participate in the pilot year of PMPU based on their affiliation with the medical students who founded the project. One student from each undergraduate institution was appointed as a campus representative. Communication between MCW and the 2 undergraduate institutions was maintained through these campus representatives during initial program development. The campus representatives informally gauged interest in development of a mentorship program by word-of-mouth at institutional premedical society and AAMC chapter meetings. Mentor interest was obtained by emailing enrolled medical students at MCW and other medical schools across the United States who graduated from Marquette University and UW-Oshkosh.

Once interest in mentorship was identified, formal program development began. A PMPU logo was created (Figure 1) and a program-specific email account was set up. An online application and needs assessment were both created using Google Forms. The application included a brief program summary, a section to provide name and contact information, and 2 additional sections—one specific to current medical students and one specific to undergraduate students. The section for current medical students requested undergraduate institution, medical college, anticipated MD or DO graduation year, and any additional program interests (eg, PhD, MS, JD, military, rural program). The section for undergraduates requested undergraduate institution, major and minor, anticipated undergraduate graduation date, interest in MD/DO/both, interest in additional programs (eg, PhD, MS, JD, military, rural program), and the following short-answer questions: "Why do you want to be a part of this program?", "What questions do you have for your future medical student mentor?" and "What resources would be of interest to you?" The 11-item needs assessment was created to identify resources that would be beneficial for undergraduate participants. A link to the application and corresponding needs assessment was emailed to 59 undergraduate students at Marquette University and UW-Oshkosh and 34 current medical students at 9 medical schools across the US who indicated interest in the program. The application and needs assessment were available to both groups for 21 days. Descriptive statistics were used to compile and analyze the needs assessment data, which were then utilized to create program content.

Program Implementation

Program founders read all needs assessments and used them to match students. Forty-three undergraduate mentees and 26 medical student mentors were joined in pairs or triplets to form mentorship groups based on similarities in application responses. Highest priority for matching criteria was undergraduate institution attended, followed by additional program interests, such as dual degree. Participants were encouraged to communicate regularly through face-to-face meetings, email, or phone. Resources, guided by the needs assessment, were provided to participants. These resources are detailed in the Results section.

Program Assessment

Six months following program implementation, an evaluation was conducted to investigate the effectiveness of PMPU. A 12-item medical student survey and 17-item undergraduate student survey were created and distributed. Both surveys assessed several components:

- Report of frequency and method of communication with mentee/mentor
- Perceived value of monthly e-newsletter
- Self-reported knowledge of global health issues
- Perceived strengths and weaknesses of program

Additionally, the undergraduate survey assessed perceived benefit of resources, confidence, and understanding of and preparedness for medical school and the application process, while the medical student survey assessed student perceived confidence as a mentor. The retrospective survey was approved by the MCW Institutional Review Board.

Surveys were available through SurveyGizmo (now Alchemer, www.alchemer.com) to participants for 14 days. A link to the survey was emailed to all enrolled students: 26 medical students and 43 undergraduate students. Participants were informed that the survey was optional and results would be used to evaluate and improve the program. Surveys were anonymous and no incentive was offered for completion. A statistician calculated descriptive statistics and Pearson correlations (r) to assess the relational strength between program components and student confidence and knowledge. Analysis was generated by IBM SPSS 24.0.

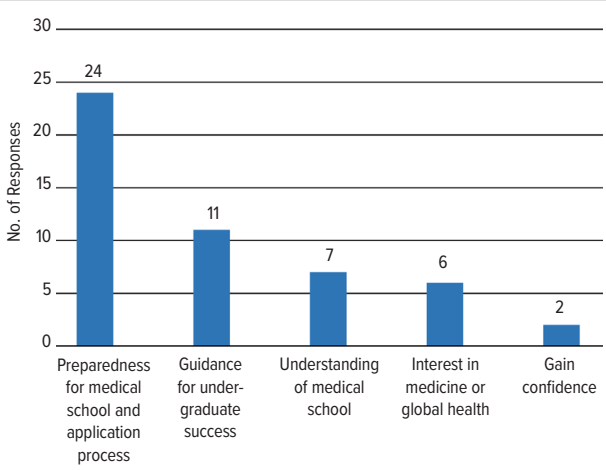
RESULTS

Program Development

Forty-three undergraduate students and 26 medical students completed the enrollment application and accompanying needs assessment. Undergraduate students cited several reasons for joining PMPU, including wanting to understand and feel prepared for medical school, seeking guidance for success in undergraduate courses, and having a general interest in medicine or global health (Figure 2). They identified the following resources as having the most potential to be helpful: a month by month checklist (n = 35), volunteer resource guide (n = 34), and advice pertaining to the Medical College Admission Test (MCAT), personal statement writing, and interviewing (n = 35). Data collected from the needs assessment guided PMPU content, which included:

- Month-by-month checklist of activities recommended for premedical undergraduate students specific to year in school, eg, when to take the MCAT, ask for letters of recommendation, etc.
- Volunteer resource guides specific to location of undergraduate institution.
- Monthly e-newsletter: global health article, "Words of Wisdom" section, and "Pre-Med Prep" section with tips for the premedical process.

Figure 2. Premedical Student Reasons for Joining Pre-Med Pair Up (N = 43, multiple responses allowed)



- Dedicated webpage, including featured program information and resources readily available (http://www.mcw.edu/MedicalSchool/PMPU-Mentorship-Program.htm).

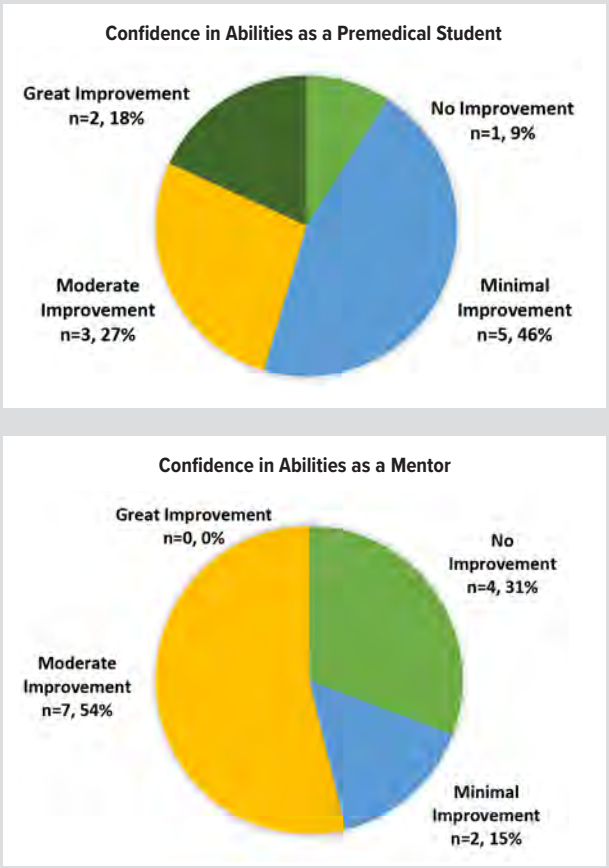
Program Implementation

Peer mentorship pairs and triplets were assigned as described above. The time commitment for program facilitators varied throughout the school year. Monitoring participant interest, establishing program curriculum, and pairing mentors with mentees required the most time—about 15 to 30 hours weekly divided among program leaders. Undergraduate campus representatives assisted with curriculum specific to their location. After the initial program startup phase, the time requirement was limited to creating monthly communication—approximately 1 to 10 hours per week divided among program leaders.

Program Assessment

The 6-month program evaluation survey had 11 undergraduate respondents (25.6%) and 13 medical student respondents (50.0%). Most undergraduate students reported communication with their mentors 3 to 4 times (36.4%) or greater than 6 times (36.4%) over 6 months. Most medical student respondents reported communication with their mentees 2 to 3 times (92.3%) over 6 months. The majority of both undergraduate students (n = 8, 72.7%) and medical students (n = 12, 92.3%) reported email as a method of communication with their mentor. Students were able to choose all methods of communication that applied. Less than half of the respondents reported reading the e-newsletter each month (45.5% undergraduate students and 23.1% medical students, respectively). However, more than half of the undergraduates self-reported improvement in knowledge of global health issues after reading the monthly e-newsletters (9.1% great improvement, 36.4% moderate improvement, 9.1% minimal improvement). Fewer than half (46.2%) of medical students

Figure 3. Mid-year survey responses



Reported confidence in abilities as a premedical student (n = 11) and as a mentor (n = 13) after completion of the program.

acknowledged improvement in global health knowledge after receiving monthly e-newsletters.

Undergraduate student responses indicated that most students felt that their confidence in abilities as a premedical student improved with program involvement (18.2% great improvement, 27.3% moderate improvement, 45.5% minimal improvement, 9.1% no improvement) (Figure 3). This confidence was strongly correlated between students' knowledge of volunteer opportunities ($r = 0.887$, $P < 0.001$) and feelings of preparedness for the medical school application process ($r = 0.854$, $P = 0.001$) and medical school curriculum ($r = 0.871$, $P < 0.001$). In terms of the resources and advice offered, undergraduate students indicated varying degrees of improvement in their knowledge of global health issues, volunteer opportunities, and understanding and preparedness for medical school and the application process (Table).

More than half of medical student respondents reported their confidence in their abilities as a mentor improved following program involvement (54% moderate improvement, 15% minimal improvement, 31% no improvement) (Figure 3). Medical students' confidence in abilities as a mentor was correlated with their feelings of success as a mentor ($r = 1.0$, $P < 0.001$).

DISCUSSION

PMPU provided undergraduate students the opportunity to seek advice regarding specific undergraduate coursework and schedule, MCAT preparation, medical school application process, and medical school curriculum and structure. Most undergraduate students reported at least minimal improvement in confidence in their abilities as a premedical student after program involvement. This confidence correlated with their knowledge of volunteer opportunities and feeling of preparedness for the application process and medical school curriculum.

The program aimed to facilitate the medical student role as mentor, providing an opportunity for professional development. More than half of the medical students self-reported moderate improvement in confidence in their abilities as mentors. Learning and honing mentoring skills will be beneficial throughout their training and future careers, potentially affecting many future medical trainees.

The strengths of this program include the number of resources it provides to premedical students and its facilitation of mentorship relationships between premedical students seeking advice and medical students who have recently and successfully completed both prerequisites at the same undergraduate institution and the medical school application process. As interest in medicine continues to grow and the number of medical school applicants increases, this type of program may be in high demand and particularly helpful for undergraduate students.

Lessons Learned

There were many lessons learned from this pilot study which, if applied, would help enhance the program and allow for more rigorous study of its success. Participants were very excited about the idea of a program that matched premedical students with current medical students from the same undergraduate institutions. The program goal and incorporation of global health was appreciated from a subjective perspective; however, after analyzing surveys, it was apparent that there may be better ways to accomplish program goals. The program was implemented primarily as a distance mentoring program that relied heavily on communicating information via the monthly e-newsletter and posting it on the website. Unfortunately, most participants reported that they did not read the e-newsletters. It is possible that different methods of presenting information, such as virtual meetings or recorded webinars, would be more appealing to participants.

The PMPU website was a central location for housing information, however, it was not promoted as much as it could have been. The website consisted of a program introduction and drop-down menus with carefully organized information. It could be organized so it is more reader-friendly and conveys answers to frequently asked questions in a similar fashion to the Medical Student Mentorship Program organized by students at the University of Hawaii.[7] This website is very inclusive of topics of interest for

medical students and is easy to navigate. However, it should be noted that a question-and-answer format is at risk for misinformation and requires meticulous efforts to ensure the information is accurate and up-to-date.

Due to the low reported use of the e-newsletter, challenges of maintaining a website, and overall program goals of personal mentorship, the main focus going forward should be interactions between program participants. This may include direct guidance for undergraduate students to explore and prepare for medical school. Activities to incorporate could include meet-and-greet sessions, shadowing days, and mock interview workshops. Some of these experiences could be provided virtually due to the distance mentoring nature of the program.

Table. Premedical Student Mid-Year Survey Responses, N=11

Program Goal	No Improvement n (%)	Minimal Improvement n (%)	Moderate Improvement n (%)	Great Improvement n (%)
Knowledge of volunteer opportunities	3 (27.3%)	2 (18.2%)	4 (36.4%)	2 (18.2%)
Understanding of and preparation for medical school	2 (18.2%)	4 (36.4%)	1 (9.1%)	4 (36.4%)
Medical school application process	3 (27.3%)	3 (27.3%)	2 (18.2%)	3 (27.3%)
Knowledge of current national and global health issues	5 (45.5%)	1 (9.1%)	4 (36.4%)	1 (9.1%)
MCAT preparation	3 (27.3 (%)	4 (36.4%)	1 (9.1%)	3 (27.3%)
Resources	**Not Useful n (%)**	**Minimally Useful n (%)**	**Moderately Useful n (%)**	**Very Useful n (%)**
List of volunteer opportunities	4 (36.4%)	2 (18.2%)	3 (27.3%)	2 (18.2%)
Month-by-month checklists	5 (45.5%)	1 (9.1%)	1 (9.1%)	4 (36.4%)

Abbreviation: MCAT, Medical College Admission Test.

To more rigorously evaluate the program, carefully created pre/post surveys could be implemented for new participants. These surveys could include items such as confidence scales for mentor and mentee, as well as objective, knowledge-based questions pertaining to the medical school application process for undergraduate students and questions pertaining to global health for both undergraduates and medical students. Surveys could be identified using a numerical system to maintain anonymity but ensure that pre/post participation answers could be compared and reasons for poor response rates could be investigated. The timeline for program participation also could be tracked through the survey. Pre/post survey answer comparisons could be further correlated with mentor/mentee characteristics, mode of mentor/mentee communication, number of interactions/mentor involvement, and duration of participation. Long-term studies could measure behavior change and impact on high stakes outcomes. For example, behavior change could be measured by medical student self-report of later involvement in mentorship programs or receipt of mentor awards. Impact could be measured by comparing undergraduate student PMPU program participant acceptance into medical school versus nonparticipants.

Limitations

Given this was a pilot study, there are multiple limitations that provide ideas for future research. First, this study was restricted to a single mentorship program. Thus, data collected cannot yet be generalized to peer mentorship programs involving other institutions. Small sample size limits statistical power and the interpretation of analytical results. Due to the nature of the pilot program, the sample size was small at the program's initiation (n = 43 undergraduates, n = 26 medical students). Even fewer participants completed mid-year surveys (n = 11 undergraduates, n = 13 medical students). It is unknown whether the remaining students dropped out of the program or continued but declined to complete the mid-year survey. That information would be helpful to determine the usefulness and success of the pilot program. The study did not collect follow-up data to learn if undergraduates were accepted into medical school or medical students became successful mentors in residency. Finally, this study did not account for all potential confounding variables; thus, it cannot be concluded that the program alone influenced premedical student understanding and preparedness for medical school or medical student feeling of success as a mentor.

Program Update

PMPU was continued at MCW during the 2017-2018 academic year. Already-established peer mentorship pairs were encouraged to remain in contact. The medical student founders and the undergraduate campus representatives passed their responsibilities to new student leaders. The program expanded to include additional undergraduate campuses, based on the new leaders' pre-existing relationships at undergraduate institutions. All prior leaders were available by email, phone, or in person to facilitate a smooth transition.

CONCLUSION

The many lessons learned through this pilot study could be used to improve the resources of the distance mentoring program PMPU, promote its long-term sustainability, and lead to future more rigorous studies to better determine its objective effect. While this study showed some promising self-reported improvement in confidence of undergraduate premedical student abilities and medical student mentor abilities, it has the potential to make an impact

on undergraduate students as they navigate the process of medical school admission and medical students as they develop mentorship skills that can be used throughout their careers.

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Leadership Views on the Barriers and Incentives to Clinical Preceptorship

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ABSTRACT

Background: Clinical education often relies on a one-to-one student-preceptor model. Recruiting and retaining quality preceptors to sustain this model has become increasingly difficult at academic institutions across the nation. While ample literature describes preceptor barriers and incentives as viewed by physician educators, few studies explore the issue from institutional leadership perspectives.

Objectives: This study aimed to describe leadership perceptions across an academic institution to better understand knowledge gaps, system barriers, and proposed solutions to help institutions take action and address preceptor shortages.

Methods: Between February and July 2019, the researchers conducted one-on-one semi-structured interviews with sampled representation of Medical College of Wisconsin leadership. The researchers reviewed transcriptions of each interview verbatim and used a qualitative grounded theory approach to generate content codes and themes. Researchers iteratively refined codes using the constant comparison method until all interviews were analyzed and final themes and subthemes were defined.

Results: Twelve institutional leaders participated, of whom 5 were clinical executives, 1 was an academic executive, 4 were academic deans, and 2 were educational directors. Analysis yielded 4 major themes: student impact, recognition, physician well-being, and leadership.

Conclusion: Each content theme highlighted areas to consider when addressing preceptor issues within an institution: (1) leadership knowledge gaps regarding the scope of preceptor challenges, particularly time commitments and the number of preceptors required; (2) improving career advancement or promotion criteria to recognize teaching efforts; (3) enhanced physician well-being from teaching, while important, may no longer be sufficient for participation, especially without financial compensation; (4) distributed leadership may be needed to address issues at the course, clinic, and system levels.

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INTRODUCTION

Many medical schools use one-to-one student-preceptor models to teach in the clinical environment. This model provides a rich personalized learning opportunity for students, but its success typically depends on creating a large sustainable pool of qualified preceptors. Building and maintaining such a preceptor pool is a major logistical challenge. Unfortunately, this issue is widespread. In the 2013 Clerkship Survey of all MD- and DO-granting medical schools, 80% of respondents were concerned about the number of clinical sites available for clinical education.[1,2] Understanding and addressing the challenges of building robust preceptor pools is essential for preserving and augmenting high quality clinical education.

The difficulty in preceptor recruitment results from numerous factors, including preceptors experiencing lower clinical productivity, longer work hours, and limited recognition.[3,4] Physician barriers and incentives to preceptorship have been long reported in the literature; however, preceptor shortages remain. Few studies explore the issue from academic and clinical leadership perspectives to understand why institutions have seemingly not lowered barriers or offered incentives to address preceptor concerns. In this study, we used qualitative analysis of semistructured interviews with institutional leadership to better understand knowledge gaps, existing incentives, potential solutions, and barriers to solution implementation.

Box 1. Leadership Semi-Structured Interview Questions

- Please name barriers to physician participation as a clinical preceptor.
- In what ways would you minimize the above barriers?
- How much time is added to a physician's half day of clinic to teach a medical student?
- What incentives are currently in place to entice physicians to serve as preceptors to medical students?
- What are other incentives you suggest (may not be in place currently or have been tried in the past)?
- Do you think physicians should be monetarily compensated for participating as a preceptor? If so, what would be a reasonable amount?
- Is there anything else we have not discussed that you would like to comment on?

Box 2. Themes and Subthemes Identified Through Qualitative Analysis

Student Impact: How students affect the clinical environment
- Time
- Clinical productivity
- Infrastructure
- Competing clinical and administrative demands

Recognition: Benefit to preceptors for educating students
- Financial compensation
- Career advancement
- Tokens of appreciation

Physician Well-being: Factors that influence a physician's well-being
- Work-life balance
- Flexibility
- Professional development

Leadership: Roles of clinical and educational leaders
- Establish expectations for preceptors
- Clear communication of institutional teaching requirements
- Need to enable champions and local leaders

METHODS

Between February and July 2019, we conducted semistructured interviews with a purposeful sample of Medical College of Wisconsin (MCW) leadership. Researchers chose individuals with a stake in medical education who, collectively, represented leadership across all MCW-affiliated institutions. Most of the MCW-affiliated institutions are located within the greater Milwaukee area, including Froedtert & MCW, Children's Hospital of Wisconsin, and Clement J. Zablocki VA Medical Center. MCW also has 2 regional campuses located in Green Bay and Central Wisconsin. Study participants did not receive compensation. Ethical approval was granted by the MCW Institutional Review Board on November 29, 2018.

Researchers employed a grounded theory approach[5,6] to best understand participants' views on preceptor issues. We developed a semistructured interview protocol based on a review of literature regarding physician barriers and incentives for preceptorship.[2-4] Institutional medical education experts reviewed interview questions for clarity. One individual of the study team (PH) facilitated all of the interviews using the interview protocol (Box 1). Each interview lasted approximately 30 minutes. PH recorded each session using a digital handheld recorder and transcribed interviews verbatim, excluding any identifiers.

Three members of the study team (JB, PH, TM) analyzed transcripts though an iterative coding process. Initially, the study team independently reviewed 2 interviews to establish preliminary codes of recurring ideas and experiences. The researchers then met to jointly review, refine, and finalize the coding structure. Employing a constant comparative method, the researchers iteratively analyzed the remainder of interviews and then compared interviews to synthesize codes into overarching themes.

JB and TM are physicians who also serve as course directors and clinical preceptors. PH is an instructional designer. While we are MCW employees, none work with the study participants as part of our daily responsibilities.

RESULTS

Of the 12 participants, 5 were clinical executives, 1 was an academic executive, 4 were academic deans, and 2 were educational directors. Participants collectively represented all of the MCW-affiliated institutions: 5 from Froedtert & MCW, 2 from Children's Hospital of Wisconsin, 2 from Clement J. Zablocki VA Medical Center, 2 from the Green Bay campus, and 1 from the Central Wisconsin campus. All invited participants agreed to be interviewed. The qualitative analysis resulted in 4 major themes: student impact, recognition, physician well-being, and leadership. Box 2 provides all major themes and subthemes. We identified no additional conceptual codes in reviewing the final transcript, suggesting theoretical saturation.

Student Impact

All participants discussed how students affect physicians and their clinical environment. The participants observed mainly the students' impact on time, productivity, and infrastructure. The most prevalent of these subthemes was time. Participants estimated physicians spend an additional zero to 60 minutes per 4-hour block of clinic when precepting. One participant pointed out that the increase in time affected all clinic staff:

> My medical assistant who is working with me is now staying half an hour, 45 minutes late. The person at the front desk who checks out the last patient can't leave until they're gone. The lab person who waits until I'm done to know if they're going to get a lab can't leave until that last person goes. So if somebody runs late, there's a whole cascade of other people who support them who have to hang around longer.

Multiple participants attributed the barrier of time to the increasing number of physician demands within a clinic session. One participant perceived this as "[the] collapsing of time in the clinical arena." Examples of competing tasks included charting requirements, quality metrics, and high patient volumes resulting from the "incredible competition in town now." Simplified

workflows and innovative learning models were mentioned as possible solutions:

> Somehow, we have to get to a place where this doesn't become just 1 more thing and find ...build some simplicity. Leverage the skill set that the students bring to the table, and build, figure out where that partnership can happen.

Participants' ideas on how to accomplish this simplicity included improving student productivity, for example through student-led learning or billable student documentation.

Some participants felt an increasing pressure to take more than 1 student into their clinical space:

> …our docs and our APPs (advanced practice providers) are being contacted by every educational institution under the sun in southeast Wisconsin that has an APP program, or post-doc program, or just go down the list—and they're being inundated with these requests to precept.

Regarding the magnitude of student impact, some participants estimated the academic faculty workforce was sufficient to fulfill preceptor needs: "…we have about 1500 faculty, we have a lot of other doctors who are on staff…I guess what we need is 200 or 250 or something [as preceptors]."

Meanwhile the number of academic faculty available for teaching may be decreasing as career paths specialize:

> It's just all over the country that this [trend towards specialization] is what's happening. They'll be teams of great researchers, they'll be some clinician educators… and they'll be an army of clinicians generating the resources to support everything.

Recognition

A majority of participants discussed the importance of recognition, particularly in the form of financial compensation and career advancement. Participants' beliefs conflicted regarding the appropriateness and amount of financial reward.

In general, participants felt academic physicians should not receive financial compensation for teaching, rather it was an expectation of their self-chosen career: "… for primary care doctors or most specialty doctors, there's no reason to be at an academic medical center unless you are interested in teaching." No participant mentioned a specific amount of time expected of faculty to be spent teaching. Two participants mentioned "Teaching Value Units," analogous to "Relative Value Units." as a method for quantifying teaching efforts.

Conversely, most participants were in favor of compensating community physicians. Regarding the amount, one stated that the community physician should be "made whole" to balance out their loss in clinical productivity. Similarly, another participant stated, "…we just need to have a level playing field, so people don't think they'll be penalized for doing this mission." Participants said the importance of compensation was growing as the sense of moral obligation to teach may be declining:

> Schools often assume that [physicians] want to work for free, and that probably has less of an altruistic appeal for people who trained in the 80s and 90s [than for those] who trained before then.

Another participant acknowledged:

> I think that personal satisfaction and giving back is a big part of that incentive, but it's not enough to rely on to get the kind of engagement that I think we want to have or need to have moving forward.

Others pondered the impact of compensation. One participant expressed concern that monetary reward was a "perverse incentive" and would yield low quality preceptors. Conversely, another participant envisioned the potential for improving education quality through compensation:

> If I'm compensating a preceptor and expect changes, I expect that they're going to be very engaged in learning about student education, about proving their teaching skills, [and] they will be available.

Many participants questioned the feasibility of financial compensation, especially those who assumed this responsibility fell on the academic institution. Two participants, however, identified the health care system as the responsible financial party, noting the perks of a talent pipeline and recognition as an academic affiliate within the community.

Participants also viewed career advancement as an important form of recognition. Career advancement was viewed universally as being of higher value to academic physicians versus community physicians. Multiple participants mentioned the need for explicit expectations for teaching and a clear pathway for teaching to contribute to career advancement. Participants were unsure if current promotion criteria accounted for teaching efforts; as stated by 1 participant, "I think, hopefully, we're giving strong credit for this [precepting] in our promotion pathways."

Physician Well-being

Participants universally stated that an intrinsic interest in teaching is key for participation as a preceptor. Multiple participants described the personal fulfillment gained from teaching, such as the "feeling of giving back" and "self-satisfaction of contributing to a person's education." Others felt inspired as a physician model, noting, "[precepting] forces you to be the best doctor you can be. It's one of the great things about having learners around."

While students may elevate well-being, 1 participant expressed concern that they disrupt physician work-life balance and noted that the importance of this balance may be on an upswing:

> My presumption is that this is a generation of Millenials/GenXers [who] will have a much better sense of work-life

> balance and much better sense of family at the end of the day because they are making choices saying both [family and career] are important.

Participants said flexibility may help physicians maintain work-life balance and could be achieved by allowing physicians to easily opt in and out of teaching over time, dynamic clinic scheduling, and partnerships with other physicians.

Lastly, participants noted professional development in clinical education was important to one's well-being. Specifically, participants mentioned that activities such as sharing best practices and networking build confidence, provide a sense of connectedness, and promote physician satisfaction.

Leadership

All participants described the need for strong leadership across multiple organizational levels. At the course level, participants voiced that leadership was needed to ensure clear preceptor expectations, noting "…the biggest barrier is maybe a misconception… by our faculty or those who could have the role of how much effort is required in order to be a preceptor."

To achieve effective communication of expectations, multiple participants envisioned the role of local champions:

> The best thing we could do, if possible, is to create some champions…within every department or…every division that could say 'look, you know I've done this for the last 4 to 5 years and it's really not that big [of a time commitment]'…rather than a mass email.

Participants suggested that local champions could support physicians by explaining the role of a preceptor, helping faculty prepare for students, and engaging faculty in quality improvement.

Participants also discussed institutional leadership in the context of developing community partnerships. One participant observed that health care systems have become key stakeholders as more physicians shift away from private practice models:

> In the old days, I would call in favors. I'd pick up the phone, call somebody that was in private practice, and say 'oh, could you take a student for a certain period of time for this purpose?' And they would say yes or no. Now, because of the employment status, it's frequently not the physician that is the decision-maker. Maybe it's the clinic manager, maybe somebody higher up in their organization structure. It may be somebody completely removed from their practice. So I think that one of the things we have to work on in the community is… the docs are willing, but the health systems aren't for a variety of reasons.

DISCUSSION

Collectively, institutional leadership-perceived incentives and barriers align well with the literature.[7-12] One major barrier, which participants and the literature alike describe, is time.[13-15] Studies have shown that physicians report increased clinical and nonclinical workloads, which may explain why the time required to precept is an ongoing barrier.[7,10] Further compounding the issue of time is that the demand for preceptors is increasing as the number of health professional trainees grows.[10] While participants in this study recognized the issues of time and preceptor demand, a few participants underestimated their magnitude. The literature shows a student adds at least 30 minutes to a clinic half-day,[13,14] yet, notably, some of study participants thought a student adds no additional time. Further, a few participants thought the preceptor demand could be fulfilled by academic faculty alone. At MCW, the first 2 years of medical school require the participation of over 500 preceptors, not including preceptors needed for third- and fourth-year clerkships, medical residencies, and other health sciences programs. As 1 participant pointed out, there simply is not enough academic faculty to meet the institution's teaching needs: "I have 103 primary care pediatricians that work for Children's [Hospital of Wisconsin] who are nonacademic. We have 3 academic general pediatricians." These findings may present an opportunity for improved communication to institutional leadership regarding the scope of the preceptor issue.

Study participants and the literature recognize that the additional time spent teaching by preceptors has a financial cost,[16] which comes in the form of decreased clinical productivity, increased administrative needs, and infrastructure.[14,16] To estimate the clinical productivity cost, one may equate the extra 30 minutes spent with a student per clinic half-day to 1 level 4 ambulatory visit, for which the Centers for Medicare and Medicaid Services (CMS) estimated payment is $90.[17] During 1 of the early MCW clinical courses, approximately 215 students participate in 20 half-day clinic sessions over the course of a year. Thus, it may be reasonable to estimate the annual clinical productivity cost incurred by this course alone to be $387,000 (215 x 20 x $90). The debate comes when determining who should absorb this cost – physicians, academic institutions, or health care systems.

Traditionally, physicians have been willing to bear the brunt of this cost by volunteering their time. This altruism stemmed from the intrinsic joy of teaching.[11,12,15] Whether this altruism remains sufficient for participation is questionable. Similar to finding in the literature, participants of this study were split on whether or not it is time to financially compensate preceptors. Many studies show that preceptors are still primarily incentivized by the intrinsic rewards of teaching; however, others have found nonpreceptor physicians placed an increased importance on financial reward.[11,12,18-21] Additionally, while studies show many in academic leadership feel monetary compensation would help recruit and retain preceptors, like our study participants, they doubted its feasibility.[1,18,22] Some study participants suggested the financial responsibility should be shared with participating health care systems given the benefits of a talent pipeline and recognition as an academic partner. Complicating the issue is the unclear impact of

financial compensation, such as its effect on education quality or the spurring of institutional competition, which may place publicly funded institutions at a disadvantage.[21,23,24] Thus, the decision to compensate preceptors is complex and may entail understanding local physicians' expectations, negotiating with health care systems, and coordinating actions with other institutions within a region.

To counter financial costs imposed on physicians, academic institutions have turned to other extrinsic rewards. Participants in this study highlighted career advancement as a key reward for academic faculty. However, participants were unclear whether or not teaching was currently a factor for promotion. They also pointed out that a method for quantifying teaching effort was important but absent. Overall, clear quantifiable expectations for career advancement may be a key area of focus for growing and maintaining preceptor pools.

As for intrinsic reward, participants in this study correctly recognized that the personal fulfillment of teaching remains a main motivating factor for precepting.[8,11,12] The sense of well-being accompanied by teaching is of critical importance, particularly in an era of increasing physician burnout and menial administrative workload.[7] However, all good things must come in moderation. Excessive expectations for preceptor teaching may hasten burnout. Study participants offered solutions to help preserve personal reward from clinical teaching, such as allowing for preceptor flexibility in clinic scheduling or the ability to easily opt-in or out of teaching at any time.

In regard to minimizing barriers, study participants pointed out the need to support preceptors administratively and develop innovative teaching models that foster both learning and clinical productivity. These aims mirror the literature[25] and specifically align with efforts made by organizations such as the Society of Teachers of Family Medicine (STFM).[4,26,27] STFM has published tools, such as a student passport, to minimize administrative burdens and effectively led CMS policy change to allow for billable aspects of student documentation.[26-28]

While solutions offered by organizations like STFM are helpful, much work is still needed for their successful implementation. Institutions need to tailor solutions to meet the individual needs of physicians and practices. To accomplish this, study participants expressed the need for strong leadership across multiple organizational levels. Currently, at many institutions, course directors take on much of this work. Unfortunately, they may be poorly positioned to implement systems change, influence promotion criteria or budget proposals, partner with community organizations, negotiate with health care systems, or represent the institution on policy change. Further, the efforts of individual course directors are often fragmented, which produces inefficiencies. For example, communication to physicians may be redundant and inconsistent, thus fatiguing its recipients to preceptor requests. As a result, some medical schools have created a centralized office for managing clinical educators.[29] A recent director role advertised for such an office within the University of California San Francisco School of Dentistry entailed accounting for clinical education budgets; a central preceptor database; continuing education; quarterly site visits; and quality metrics to assess sites, preceptors, and program successes.[30] Allocating centralized resources to support leaders distributed at the level of courses, clinics, and institutions may allow for both comprehensive and streamlined solutions.

Limitations

Limitations of this study include participant representation from a single institution, which may restrict generalization of its results. Additionally, researchers deliberately, rather than randomly, selected participants, which may have introduced result bias.

CONCLUSION

Preceptor shortages challenge academic institutions across the nation. Much is known about preceptor issues based on the perspectives of preceptor physicians. Yet shortages remain. This study is the first to explore perspectives of leaders across an academic institution to better understand knowledge gaps, system barriers, and proposed solutions to address preceptor shortages. Each content theme highlighted areas to consider when addressing preceptor issues within an institution: (1) leadership knowledge gaps regarding the scope of preceptor challenges, particularly time commitments and the number of preceptors required; (2) improving career advancement or promotion criteria to recognize teaching efforts; (3) enhanced physician well-being from teaching, while important, may no longer be sufficient for participation, especially without financial compensation; (4) distributed leadership may be needed to address issues at the course, clinic, and system levels.

Prompt attention and investment to addressing preceptor issues are critical as the stakes to clinical education are high. The return on investment to academic institutions will be the success of its trainees. After all, as 1 participant stated, "We rely on our students and our residents and our fellows to really be the spokespeople [for our institution]."

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Health Policy Advocacy Engagement: A Physician Survey

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ABSTRACT

Purpose: Physicians can play an important role in shaping health policy. The purpose of this study was to determine characteristics of physicians participating in health policy and barriers and facilitators to their advocacy.

Methods: A modified previously validated survey instrument was mailed to physicians affiliated with the University of Wisconsin on October 12, 2018. Three follow-up emails were sent, and the response period closed January 30, 2019. Twenty-eight items were included in the survey tool. Respondents were considered highly engaged if they: (a) reported involvement in predetermined high impact areas, (b) had self-reported weekly or monthly advocacy involvement, or (c) had more than 10% dedicated work time for advocacy.

Results: Eight hundred eighty-six of 1,432 physicians responded (61.9%), of which 133 (15.0%) were highly engaged. Highly engaged respondents were more commonly male (57.1%), White (90.2%), of nonsurgical specialties (80.5%), and Democrat (55.6%) or Independent (27.1%). Those not highly engaged were more likely to report "I don't know how to get involved." Less than half of all respondents received any advocacy education, with professional organizations providing the majority of education through conferences and distribution of materials. Only 2.5% of respondents had more than 10% of work time dedicated to health policy.

Conclusions: Engagement in health policy exists on a spectrum, but only a small percent of physicians are highly engaged, and very few have dedicated work time for advocacy. Certain demographics predominate the advocacy voice, and health policy training opportunities are lacking.

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INTRODUCTION

In 2018, health care spending accounted for $3.6 trillion, amounting to 17.7% of the United States Gross Domestic Product (GDP). Over the next decade, health expenditures are expected to outpace overall GDP growth, accounting for a greater proportion of the domestic economy each year.[1] These expenses outpace every other nation but do not translate to improved health or life expectancy.[2]

As such, physicians are at the front line of health care spending, utilization, and quality, and poised and equipped to influence legislative and regulatory national health policy reform and expenditures with the primary goal of improved patient care and care delivery as the end point. While doctors and other health professionals visibly contribute to policy discussions on specific topics, such as firearm safety,[3,4] health care reform,[5] and more recently in the COVID-19 pandemic, at other times physicians seem absent from critical policy discussions. National organizations provide some structure to advocacy and encourage physician involvement,[6] but a variety of barriers exist.[7,8] As a result, physicians may not have a voice in critical decisions directly impacting their patients or profession.

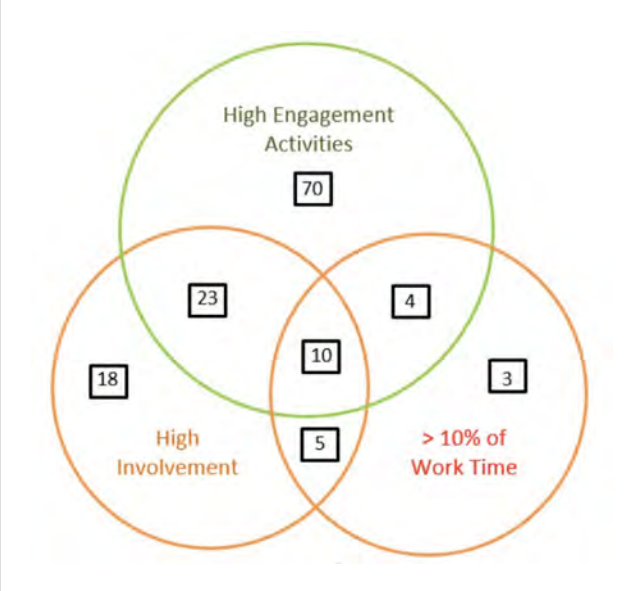
Despite its importance, little is known about physician engagement in health policy advocacy, including what defines engagement, how many physicians are engaged, which types of physicians are involved, what barriers exist to policy engagement, and how physicians learn how to effectively advocate for

Table 1. Baseline Characteristics

	All n=886		High Engagement n=133 (15.0%)		Not High Engagement n=753 (85.0%)	
Age						
0-44	428	48.3%	43	32.3%	384	51.0%
45-64	390	44.0%	77	57.9%	313	41.6%
65 and older	68	7.7%	13	9.8%	55	7.3%
Gender						
Male	491	55.4%	76	57.1%	415	55.1%
Female	366	41.3%	54	40.6%	312	41.4%
Prefer not to answer or omitted	29	3.3%	2	1.5%	22	2.9%
Race[a]						
White	736	83.1%	120	90.2%	616	81.8%
Asian/Pacific Islander	82	9.3%	7	5.3%	75	10.0%
Other[b]	39	4.4%	2	1.5%	37	4.9%
Prefer not to answer	41	4.6%	6	4.5%	35	4.6%
Specialty						
Surgical	185	20.9%	26	19.5%	159	21.1%
Nonsurgical	667	75.3%	107	80.5%	560	74.4%
Did not answer	34	3.8%	0	0.0%	34	4.5%
Primary political affiliation						
Democrat	480	54.2%	74	55.6%	406	53.9%
Republican	65	7.3%	6	4.5%	59	7.8%
Independent	208	23.5%	36	27.1%	172	22.8%
Other	29	3.3%	7	5.3%	22	2.9%
Prefer not to respond	101	11.4%	10	7.5%	91	12.1%

If "did not answer" exceeded 1%, it is specified in the Table.
[a]Respondents were asked to choose all that applied.
[b]Other includes those that identified as Black, Latinx, Native American, or Other. These were grouped soley due to small sample size and desire to protect participant identity.

Figure. High Physician Engagement by Activities, Involvement, or Work Time



133 unique respondents were considered highly engaged, with 91 meeting 1 criteria, 32 meeting 2 criteria, and 10 meeting all 3 criteria.

health policy priorities. The purpose of this study was to determine specifically physician views of, and level of engagement in, health policy advocacy, and to identify barriers and facilitators of that involvement.

METHODS

General Methods

The University of Wisconsin (UW) Institutional Review Board approved this study. We conducted a survey of all active UW Health physicians from October 12, 2018 to January 30, 2019. UW Health is the "integrated health system of the University of Wisconsin-Madison caring for more than 600,000 patients each year with 1,785 employed physicians and 21,000 employees at 7 hospitals and 87 outpatient clinics."[9] The academic medical center, located in Madison, Wisconsin, consists of a 505-bed adult University Hospital and 87-bed pediatric hospital. The system also includes a 50-bed inpatient rehabilitation facility and 4 community hospitals ranging from 34 to 448 beds.[9]

Providers were identified on a list provided by the University of Wisconsin School of Medicine and Public Health. A total of 1,542 surveys were sent, which was reduced by 110 after the list revealed accidental inclusion of nurse anesthetists (CRNA) and PhD-only faculty. Therefore, 1,432 surveys were included as the denominator.

Survey Tool

The 28-question survey tool was developed and conducted with support of the University of Wisconsin Survey Center. The questionnaire was derived from a previous validated survey to evaluate the involvement of health educators in health policy advocacy and included questions on demographics such as age, career level, specialty, gender, board certifications, race, and personal political affiliation.[10] The instrument also included questions on personal and professional advocacy during the past 2 years, work time or personal time dedicated to advocacy, perceived barriers and benefits to advocacy participation, and training in policy. In this study, public policy was defined as "a system of laws, courses of action, and priorities directing a government action." Health policy was defined as "decisions, plans, and actions that are undertaken to achieve specific health care goals within a society." Advocacy was defined as "attempting to influence public policy through education, lobbying, or political pressure." We define health policy advocacy as the activities used to influence specific health goals of society via action taken by physicians to inform and educate public officials or policies.

Defining Health Policy Advocacy Engagement

A priori, respondents were defined to have high overall health policy advocacy engagement (highly engaged) if they met 1 or more of the following criteria:

1. Participation in high effort activity: Participation in any high

effort and/or time intensive advocacy activity done outside of other professional responsibilities that would typically involve additional training, experience and collaboration, and/or coordination with others. (Example: testifying at a formal legislative hearing).

2. High involvement: Self-reported level of activity defined as monthly or weekly activity involvement.
3. High work time: Ten percent or more of work time dedicated to health policy.

Survey Process and Response

An initial mailing was sent on October 12, 2018 to the physician list. This mailing included a link to the survey to be completed online, in addition to a $5 incentive. Follow up emails were sent October 17, October 23, and October 31, 2018.

Of the 1,432 surveys sent, 29 mailed letters were unable to be delivered, and 22 emails were undeliverable. A total of 476 responses came from the letter prompt, and 410 came from a directed email link, for a total of 886 respondents. The survey response period closed January 30, 2019. The authors were blinded to respondents and were not included in the distribution list.

Statistical Analysis

Survey data was analyzed with the assistance of biostatisticians of the Department of Surgery. Descriptive statistics were used to summarize the data results. Skipped or omitted questions are reported in the results if they exceed 1% of the sample. Other was used to include those who identified as Black, Hispanic, Native American, or Other. These were grouped solely due to small sample size and desire to protect participant identity.

RESULTS

A total of 886 of 1,432 (61.9%) survey responses were obtained. Of respondents, the majority were male (n = 491, 55.4%), White (n = 736, 83.1%), and under age 65 (n = 818, 92.3%). Nonsurgical respondents (n = 667, 75.3%) were more common than surgical (n = 185, 20.9%). Most identified as Democrats (n = 480, 54.2%), followed by Independents (n = 208, 23.5%) and Republicans (n = 65, 7.3%) (Table 1).

A total of 107 (12.1%) unique respondents participated in 1 or more high effort activity, 56 (6.3%) met high involvement criteria, and 22 (2.5%) reported high work time (10% or more of work time for health policy advocacy) (Table 2). Of these 3 definitions, a total of 133 (15.0%) unique respondents were identified as highly engaged, with 91 meeting 1 criteria, 32 meeting 2 criteria, and 10 meeting all 3 (Table 2, Figure 1).

Overall, highly engaged respondents were more likely to be male (57.1%), White (90.2%), identify as a nonsurgical specialty (80.5%) and self-report as Democrat (55.6%) or Independent (27.1%) (Table 1). Highly engaged respondents cited more benefits to participating in health policy advocacy. Those not highly engaged were more likely to report "I don't know how to get involved" (Table 3), which may be due to a significant difference in having health policy advocacy training (highly engaged, 48.9%; not engaged, 18.3%). Very few overall respondents (n = 43, 4.9%) reported health policy advocacy training in either college or medical school. Engaged respondents received education most often at conference sessions (n = 44, 33.1%), from materials provided by professional organizations (n = 39, 29.3%) and on-the-job experience (n = 39, 29.3%) (Table 3).

Table 2. Phyisican Self Reported Health Policy Engagement: Activities, Involvement, and Work Time (n=886)

Activity Participation[a]	N	%
None		
• Not engaged in health policy or advocacy	189	21.3%
Low: Advocacy activity that would be expected to be performed as basic portion of physician professionalism		
• Member of organized medicine or professional society	512	57.8%
• Contacting (calling or writing) legislators	315	35.6%
Moderate: Physician seeks out additional activity related to health policy advocacy, but this activity can be relatively easily incorporated amongst other patient care, administrative and/or academic responsibilities		
• Provided health policy-related information to patients, professionals	169	19.1%
• Used mass media or public events to address health policy issues	80	9.0%
• Actively involved in organized medicine or professional society	215	24.3%
• Attended a medical advocacy summit or event	86	9.7%
• Contribute to a medical political action committee (PAC)	137	15.5%
High: Requires effort and/or time by the physician of significance above and outside of other professional responsibilities[b]		
• Provided written reports, research, recommendations, or other medical related expertise/assistance to a public official	83	9.4%
• Drafted legislation or developed a resolution	13	1.5%
• Testified at a formal legislative hearing	20	2.3%
• Testified or did research for a legal action (lawsuit)	20	2.3%
Physician self-described health policy involvement		
• No activity	329	37.1%
• Slightly involved (1-2 times per year)	361	40.7%
• Moderately involved (more than 1-2 times per year, but less than monthly)	134	15.1%
• Very involved (monthly) or extremely involved (weekly)	56	6.3%
Dedicated percent of work time for health policy		
• None	435	49.1%
• Less than 10%	427	48.2%
• More than 10%	22	2.5%

[a]Respondents were asked to select all that were applicable

[b]Of the 136 respondents reporting a high engagement activity, there were 107 unique respondents, meaning 29 respondents participated in more than 1 of the 4 listed categories.

DISCUSSION

In this study of physician health policy advocacy engagement, we determined that 15% of physician respondents in our health system are highly engaged, although very few had dedicated work time for health policy advocacy. Given that lack of time and com-

Table 3. Benefits of, Barriers to, and Training in Health Policy Participation[a]

	High Engagement n=133	%	Not High Engagement n=753	%
Benefits				
Improving a situation or issue	110	82.7%	564	74.9%
Improving the health of the public	118	88.7%	608	80.7%
Affecting many patients at once	103	77.4%	463	61.5%
Making a difference in others' lives	103	77.4%	447	59.4%
Being able to get involved and participate	68	51.1%	211	28.0%
Potential to get resources such as funding or staffing	39	29.3%	119	15.8%
Personal gratification	80	60.2%	259	34.4%
Barriers				
Lack of time	109	82.0%	563	74.8%
Not important	1	0.8%	15	2.0%
I don't know how to get involved	7	5.3%	260	34.5%
Other priorities	58	43.6%	354	47.0%
Lack of support from others	27	20.3%	87	11.6%
Takes too long to see a difference	9	6.8%	98	13.0%
Frustration with the process	39	29.3%	239	31.7%
Uncertain outcome	17	12.8%	116	15.4%
Probably won't make a difference	13	9.8%	136	18.1%
Using money or resources in other ways	11	8.3%	89	11.8%
Uncomfortable confronting others with opposing views/large funds/influence	21	15.8%	105	13.9%
Policymakers attitude/viewpoints	34	25.6%	133	17.7%
Can't be involved due to employment	14	10.5%	59	7.8%
Training (Any)	65	48.9%	138	18.3%
Type of Training				
College coursework	3	2.3%	2	0.3%
Medical school coursework	10	7.5%	28	3.7%
Other advanced degree coursework	9	6.8%	18	2.4%
Workshops	37	27.8%	33	4.4%
Professional journals	12	9.0%	23	3.1%
Professional colleagues	37	27.8%	51	6.8%
Sessions at conferences	44	33.1%	68	9.0%
Materials from professional organizations	39	29.3%	45	6.0%
Mass media	12	9.0%	11	1.5%
On-the-job experience	39	29.3%	26	3.5%

[a]Respondents were asked to select all that applied.

peting priorities were the most common reasons cited for lack of involvement, health care organizations will need to invest in dedicated professional time and resources if physician advocates are to impact health policy.

Importantly, our results show that less than half of all highly engaged respondents reported any advocacy training, and many fewer (1 in 5) for those not actively engaged. Not highly engaged physicians were also more likely to report they did not know how to get involved in health policy. For those reporting health policy training, surprisingly few physicians reported training in formal degree programs or university course work, with far more citing training obtained at conferences and from professional organizations. While this may demonstrate a need for more degree programs and courses dedicated to advocacy, it may alternatively show that continuing medical education and other more accessible opportunities are preferred, making advocacy instruction more easily accessible to physicians at all career levels. Professional organizations should consider whether health policy advocacy could be a larger part of professional meeting agendas and materials. Undergraduate and graduate medical education also may consider this addition to their curriculums if not already available.

We found several important differences between the highly engaged physicians and, thus, those more likely to be affecting policy, and those not highly engaged. In particular, highly engaged physicians tend to be White, male, Democratic or Independent, and practicing in nonsurgical specialties. Although these data also generally reflected the population surveyed, these demographic and training backgrounds could impact or bias policy positions and priorities, and academic medical centers and their physicians should be aware of this potential. While the population surveyed, and that which responded, was quite congruous, we believe the voice of more diverse physicians in advocacy is critical and needs to be the highest priority for the future of medicine.

Interestingly, benefits and barriers of engagement tracked similarly between those who reported being engaged and those not engaged. Both groups reported "improving a situation or issue" and "improving the health of the public" as the primary benefits of health policy engagement. This highlights physician motivation as the patient and patient care being the center of their work focus. Likewise, with regards to barriers, almost none reported that policy was "not important," but "lack of time" and "other priorities" were commonly cited barriers by both groups. This strongly suggests physician interest in helping patients through policy but that physicians need more dedicated time to engage in this work.

Finally, we used this survey as an opportunity to propose definitions of physician engagement that may be used in future studies of physician advocacy. In addition, these definitions may be used to encourage stepwise involvement in health policy by outlining low barrier activities that could serve as entry points for new physician advocates. For example, the majority of respondents were members of a professional society, and about a third had recently contacted their representative. This shows that engagement in health policy exists on a spectrum and, hopefully, this may encourage interested physicians to engage in higher levels of advocacy and impact. However, future work and professional discourse will be required to determine optimal physician engagement targets at individual institutions and within different specialties. Although beyond the scope of this survey, study of nonphysician medical provider engagement, motivations driving highly engaged physicians, and preferred training for advocacy engagement should be next steps in this line of investigation.

Our findings and conclusions are limited by being from a single Midwestern academic medical center. Although there is no reason to believe our academic medical center is significantly different than other large public institutions, we cannot generalize to nonacademic medical centers or physicians employed by health systems that may differ in other important ways. We also cannot draw conclusions from our findings about those of other health care professionals. Finally, we cannot say with certainty that our survey respondents are representative of our entire physician population. However, we believe our 61.9% response rate to be a study strength that mitigates some of that concern.

CONCLUSION

By engaging in health policy advocacy by responding to legislation, government regulations, and administrative actions, physicians can impact the care of patients and the practice of medicine. This engagement exists on a spectrum; future work could address barriers and needs identified in this introductory survey.

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Obstructive Sleep Apnea in Pregnancy: Early Lessons From Our Sleep Pregnancy Clinic

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ABSTRACT

Problem Considered: Obstructive sleep apnea (OSA) is underdiagnosed during pregnancy, but there is strong theoretical and some empiric evidence that treatment may improve obstetric outcomes. Barriers to screening, testing, and treatment are common during pregnancy. The goal of this described intervention was to reduce these barriers and improve detection of OSA in pregnancy.

Methods: Representatives from sleep medicine and perinatology established a cross-disciplinary, collaborative Sleep Pregnancy Clinic offering a streamlined referral process for multimodal screening, testing, and treatment of OSA during pregnancy. This is a retrospective analysis of data from the clinic's first 19 months.

Results: Between June 2017 and December 2018, 134 pregnant women were referred for OSA testing. Sixty-three (47.0%) completed objective sleep testing, and 38 (60.3%) of the women who completed testing met diagnostic criteria for OSA. This intervention resulted in a statistically significant increase in the number of diagnostic sleep apnea tests performed (average 22.4 tests per year pre-intervention, 77 per year post-intervention [$P = 0.0012$]).

Discussion and Conclusions: Despite a streamlined referral pipeline, completion rates of OSA testing in pregnant women remained below 50%. However, the overall number of women referred and who completed testing increased significantly during this time period. Of those who completed testing, the majority were diagnosed with OSA. Since starting this clinic, we have created resources to familiarize patients with the equipment and worked to reduce other barriers. Assessment of these interventions and the impact of treatment on obstetric outcomes is ongoing, as is assessment of reasons women do not complete diagnostic testing.

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INTRODUCTION

Obesity is the most common comorbid condition of pregnancy, and obstructive sleep apnea (OSA) is a common comorbidity of obesity.[1] When OSA occurs in pregnancy, it is independently associated with increased risk of gestational hypertension, preeclampsia, gestational diabetes and, possibly, fetal growth restriction and other adverse neonatal outcomes.[2-13]

The prevalence of OSA during pregnancy is unclear, but evidence suggests it complicates 8% to 32% of pregnancies, depending on comorbidities such as obesity and gestational age at time of testing.[14-16] While the literature indicates OSA during pregnancy may be relatively frequent, it remains underdiagnosed.[3-15] There is also evidence, albeit limited, to suggest that continuous positive airway pressure (CPAP)—the first-line treatment for OSA—may have therapeutic benefit for blood pressure control in preeclampsia.[17-18]

Given that treatment may reduce the risk of adverse pregnancy outcomes—specifically preeclampsia, which has limited proven prevention options[19]—identifying OSA during pregnancy is timely and critical. Therefore, we sought to identify and reduce barriers to diagnosing OSA in pregnant patients via a multimodal approach. We aimed to increase screening using the best screening tool available for pregnant patients and to reduce barriers to referral and referral completion. Thereafter, we aspired to increase treatment initiation for those who required treatment, with an overarching goal of reducing adverse obstetric outcomes related to OSA.

Here, we describe the interventions we undertook to increase the number of women completing indicated testing for OSA during pregnancy. We also report our findings on the characteristics of pregnant women who completed versus did not complete referrals and display the general trend in completed referrals both before and after our interventions.

METHODS

Study Design/ Intervention

This project was a joint venture between the Division of Maternal-Fetal Medicine at the University of Wisconsin-Madison/UnityPoint Health-Meriter and the Wisconsin Sleep Clinic at the University of Wisconsin-Madison (UW-Madison). The analysis described was determined consistent with quality improvement and did not meet criteria for human subject research; it was, therefore, deemed exempt from oversight by the Institutional Review Board for both UnityPoint Health-Meriter and the UW-Madison (May 2018).

Together, representatives from sleep medicine (MHB) at Wisconsin Sleep/UW-Madison and perinatology (KMA) at UW-Madison/UnityPoint Health-Meriter met with clinic managers from each site to establish a cross-disciplinary, collaborative Sleep Pregnancy Clinic. This clinic offers a streamlined referral process for multimodal screening, diagnostic testing, and treatment of OSA during pregnancy.

Simultaneously, the Department of Obstetrics and Gynecology at UW-Madison and UnityPoint Health-Meriter created task forces aimed at optimizing care of pregnant women with obesity and jointly generated clinical care guidelines based on best available evidence and national and society recommendations. Overviews of these guidelines were presented to both academic and private obstetricians and gynecologists, certified nurse midwives, and family medicine physicians at faculty meetings and perinatal summit events. Electronic and printed copies of the guidelines were disseminated at these events, and the guidelines remain with other clinical guidelines both on UnityPoint Health-Meriter's website and within the shared files utilized for direct clinical care. These guidelines also were used to generate streamlined order sets and note templates in the electronic health record (UW Healthlink Epic, Hyperspace 2018, Epic Systems Corporation, Verona, Wisconsin) to facilitate order placement. One part of these updated guidelines prompted the obstetric care provider to complete OSA screening using at least 1 published and validated tool, thus ensuring that pregnant women with at least 1 risk factor (pre-pregnancy body mass index [BMI] $\geq$30 kg/m^2) were being screened for OSA.[20-22] Screening tools utilized included the 4-variable tool by Facco and STOP-BANG for all women.[20-22] The Facco tool was published in 2012. In this model, age and BMI are summed and 15 points each are added if the woman has chronic hypertension or snores > 3 nights per week.[20] STOP-BANG, which is commonly used by anesthesia, was also included per the Sleep Clinic's intake protocol.[23] STOP-BANG is an 8-item questionnaire that assigns 1 point for each positive answer to questions about snoring, tiredness, observed apneas, hypertension, BMI >35 kg/m^2, age > 50, neck size >16 inches, and male sex.[21,22] Scores of 0 to 2 are low risk for sleep apnea, with higher scores consistent with intermediate to high risk.[21,22]

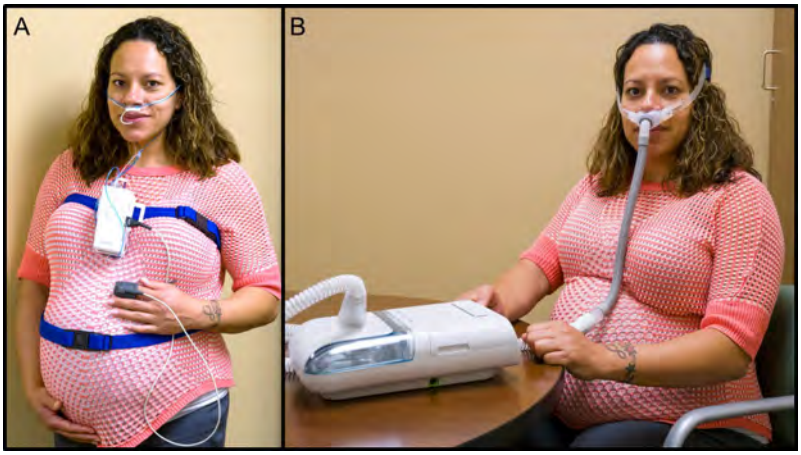
On the sleep clinic end, to expedite testing, the decision was made to utilize portable 4-channel home sleep apnea testing (HSAT) equipment (Respironics Alice PDx) currently used at Wisconsin Sleep for home diagnostics in nonpregnant patients as well, rather than in-lab polysomnography (PSG). Prolonged wait-times up to 2 to 3 months are common for PSG testing, but HSAT results are typically available the same day or within a few weeks, if insurance prior authorization is required. We also hypothesized that home testing might reduce barriers to test completion for some pregnant women who might be hesitant to spend a night away from children and family members and in the potentially intimidating and cumbersome environment of the sleep lab. Finally, there is precedent for using portable OSA diagnostic devices in pregnancy, including within the largest prospective study of OSA during pregnancy to date, the nuMoM2b sleep disordered breathing substudy.[4]

While establishing this collaborative Sleep Pregnancy Clinic, patient-facing materials were developed. Some materials addressed barriers to completing sleep testing reported by pregnant women. For example, many pregnant women cited concerns about bulky or cumbersome testing equipment and noisy and uncomfortable treatment choices. To address these concerns, photographs were taken of a pregnant consenting volunteer wearing both the home sleep apnea testing device and a continuous positive airway pressure (CPAP) nasal pillows interface or mask, with the CPAP machine on a table next to her for scale (Figure 1A and B). We also created a brochure listing the symptoms of OSA, its significance during pregnancy, and potential treatment benefits (Appendix).

After the aforementioned clinical guideline and patient-facing materials were generated, the typical workflow was comprised of (1) screening pregnant women with a pre-pregnancy BMI $\geq$ 30 kg/m^2 for sleep apnea with screening questionnaires,[20-22] (2) displaying images of a pregnant woman wearing the sleep testing device and CPAP mask, (3) distributing brochures, and (4) placing referrals to the Wisconsin Sleep Clinic, wherein patients receive expedited triage per an established "pregnancy protocol" to HSAT with the Alice PDx 4-channel system (Koninklijke Philips NV, Amsterdam, Netherlands).

To maximally expedite testing and clinical evaluation in the time-sensitive period of pregnancy, significant changes were made to the typical triage for OSA referrals at Wisconsin Sleep. Specifically, whereas triage normally requires evidence of documented symptoms (eg, snoring, insomnia, and/or excessive day-

Figure 1. Images of a Pregnant Woman Wearing (A) Sleep Device (Alice PDx 4-channel System) and (B) Nasal Pillows Interface/Mask



Images are shown to patients during their obstetrical visit if they screen positive for possible sleep apnea, prompting a referral for testing.

time sleepiness) AND documented airway, cardiovascular, and pulmonary examination by the referring provider, pregnancy referrals were triaged to home OSA testing directly. Medical comorbidities of chronic hypertension and obesity were used as criteria for testing, akin to their use in the STOP-BANG OSA questionnaire[21,22] and Berlin Questionnaire[24] in nonpregnant populations. Both the STOP-BANG and Berlin Questionnaire are short questionnaires (8 and 10 questions, respectively) that emphasize obesity and hypertension as risk factors for sleep apnea, in addition to subjective measures of sleepiness and snoring. Frequent (>3 days per week on average), not just loud snoring was admissible as a symptom. Triage questions where in-lab PSG or clinic visit as the initial step were considered (eg, with documented morbid obesity, previous CPAP noncompliance, and presence of additional sleep disorders such as restless leg syndrome) were forwarded to 1 sleep physician (MHB) for ultimate decision. Finally, in circumstances where face-to-face clinic visits were mandated (for patients with Medicare insurance), such visits were expedited by MHB.

Referrals for the analysis period were tracked via entry into a clinical database created to track sleep testing referrals and results. Data were collected and managed using Research Electronic Data Capture (REDCap) tools hosted at the UW-Madison School of Medicine and Public Health.[25] REDCap is a secure, web-based application designed to support data capture for research studies, providing (1) an intuitive interface for validated data entry, (2) audit trails for tracking data manipulation and export procedures, (3) automated export procedures for seamless data downloads to common statistical packages, and (4) procedures for importing data from external sources.

For the purpose of this analysis, OSA was defined as a respiratory effort index (REI) with portable HSAT (the vast majority of cases) or, in the case of in-lab PSG testing, apnea-hypopnea index (AHI) or respiratory disturbance index (RDI) >5 events/hour.[26] Both 3% and 4% desaturation criteria were used, as Wisconsin Sleep uses 4% desaturation criteria as default, but sleep physicians can interpret studies using the American Academy of Sleep Medicine's (AASM) 3% desaturation criteria per their discretion. REI, AHI, or RDI of 5 to 15 per hour were classified as mild, >15 to 30 per hour as moderate, and >30 events per hour as severe, as is standard practice.[27] These severity designations remain widely used in research and clinical practice, with predetermined symptoms or medical comorbidities required for insurance coverage for CPAP therapy for OSA in the mild category. We offered CPAP therapy to all pregnant women with studies demonstrating AHI/RDI or REI >5 events per hour. In the majority of cases, there were both symptoms and comorbidities that justified CPAP therapy in the "mild" OSA category. In the 3 instances where insurance denied coverage, MHB initiated appeals to the home care company/insurance.

Relevant interventions include development of the clinic and rigorous referral tracking starting in June 2017, the use of photographs to show HSAT and treatment CPAP equipment being worn by a pregnant woman starting in September 2017, and implementation of the standardized obesity order sets, prompting the use of sleep apnea screening questionnaires in September 2018.

Descriptions of barriers encountered were as reported to sleep clinic scheduling personnel and were ascertained pragmatically and for clinical purposes.

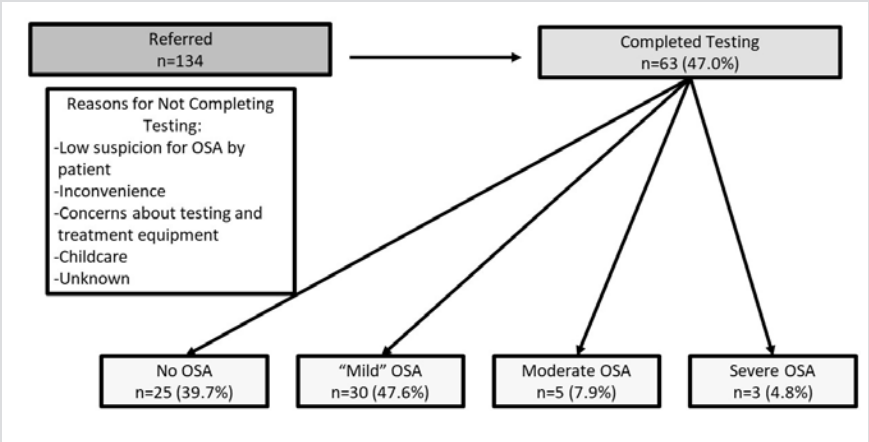
We conducted 2 analyses. First, we analyzed demographic variables associated with sleep apnea study completion for women referred between June 2017 and December 2018. Demographic variables of women who did and who did not complete sleep studies were analyzed using Pearson's chi-square test and Student *t* test as appropriate. Second, we analyzed whether our rate of referral completion (determined by sleep test completion) or the number of sleep apnea tests in pregnant women increased prior to or after our interventions. This was accomplished via query of the electronic health record (UW Healthlink Epic, Hyperspace 2018, Epic Systems Corporation, Verona, Wisconsin) for the number of referrals and completed clinic visits. Data for this analysis were analyzed by 12-month interval from January 2012 through October 2019. All statistical analyses were performed utilizing Excel (Microsoft Excel, 2013, Redmond, Washington) and

STATA 16.0 (StataCorp, 2017, College Station, Texas).

RESULTS

Between June 2017 and December 2018, 134 pregnant women were referred for OSA testing. Sixty-three (47.0%) completed objective sleep testing (Figure 2). Of those who completed testing, 38 (60.3%) met diagnostic criteria for OSA. Thirty (78.9%) had "mild," 5 (13.2%) had "moderate," and 3 (7.9%) had "severe" OSA. Women who did not complete objective sleep testing cited low suspicion for OSA, inconvenience, and concerns about the testing and treatment equipment.

Figure 2. Obstructive Sleep Apnea Testing Referrals and Results



Of 134 Women Referred for Sleep Testing, 63 (47.0%) Completed Testing. Of those who completed testing, 60.3% met criteria for obstructive sleep apnea.

The Table shows the demographic and maternal characteristics of pregnant women who did or did not complete sleep testing. Those who did not complete sleep testing were less likely to be married and more likely to have a diagnosis of hypothyroidism. There was a nonstatistically significant trend toward lower testing completion for referrals that occurred later in pregnancy.

When assessing whether the total number of sleep clinic visits by pregnant women changed over time, the average number of annual referrals pre-intervention was 44.4 per year (SD 3.8), rising to an average of 139.7 referrals per year (SD 34.9) post-intervention, a statistically significant increase ($P < 0.001$). This intervention also resulted in a statistically significant increase in sleep tests performed (average 22.4 tests per year [SD 5.3] pre-intervention and 67 [SD 16.6] post-intervention [$P = 0.0012$]). However, the sleep study completion rate (as a percent of completed tests per referral) did not improve, as referral completion was 50.3% (SD 10.2%) pre-intervention and 48.0% (SD 2.4%) post-intervention ($P = 0.718$). (See Figure 3.)

Table. Maternal Characteristics by Sleep Test Completion

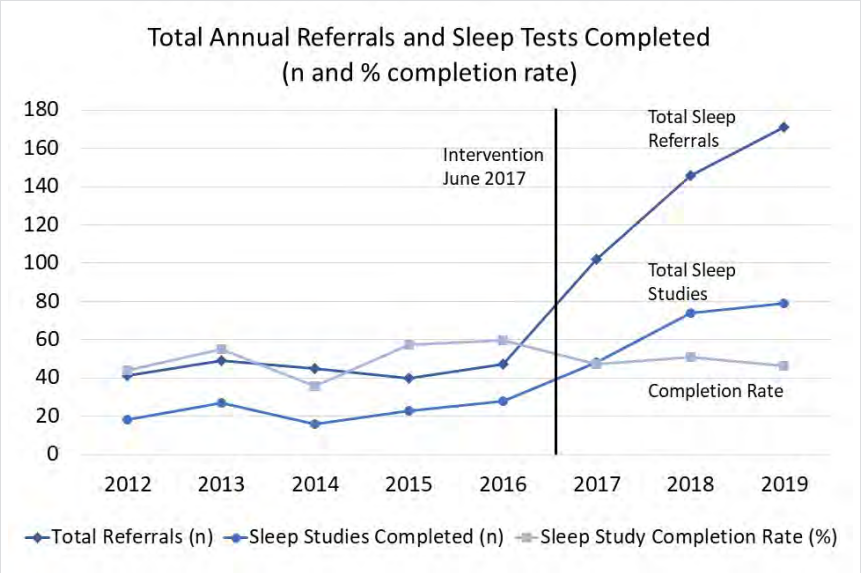
	Sleep Test Not Completed N = 71*	Sleep Test Completed N = 63 [a]	*P* value
Age, years, mean (SD)	32.8 (5.0)	34.3 (5.6)	0.106
Age, years by group, n (%)			
18-34	48 (67.6)	35 (55.6)	
≥35	23 (32.4)	28 (44.4)	0.152
Race/ethnicity, n (%)			
White	50 (74.6)	50 (79.4)	
Black or African American	10 (14.9)	5 (7.9)	
Hispanic	2 (3.0)	3 (4.8)	
Asian	2 (3.0)	2 (3.17)	
Other or not reported	3 (4.5)	3 (4.8)	0.782
Marital status, n (%)			
Single	8 (14.3)	3 (5.17)	
Married or committed	15 (26.8)	38 (65.5)	
Divorced or separated	2 (3.6)	0 (0.0)	
Unknown	31 (55.4)	17 (29.3)	< 0.001
Parity	1.2 (1.5)	0.9 (1.2)	0.176
Trimester at time of referral			
First trimester	27 (40.3)	33 (54.1)	
Second trimester	26 (38.8)	24 (39.3)	
Third trimester	14 (20.9)	4 (6.6)	0.051
BMI, kg/m^2, mean (SD)	42.2 (8.7)	40.4 (8.7)	0.256
BMI, kg/m^2, by group, n (%)			
< 30	5 (7.9)	5 (8.0)	
30-39.99	22 (34.9)	23 (37.1)	
≥ 40	36 (57.1)	34 (54.8)	0.965
Medical comorbidities (n,%)			
Hypertension	27 (39.1)	20 (31.8)	0.376
Pregestational diabetes	13 (18.3)	8 (12.7)	0.372
Hypothyroidism	4 (6.0)	19 (31.2)	< 0.001
Smoking	28 (41.8)	17 (27.4)	0.087
Excess gestational weight gain (n,%)	18 (26.9)	26 (41.3)	0.083

[a] Some electronic health records lacked data

DISCUSSION AND CONCLUSIONS

We describe in replicate detail the steps our clinics undertook to increase indicated testing for OSA during pregnancy. While completion rates of referrals—ie, referrals resulting in completion of sleep apnea testing—did not change, the overall number of pregnant women referred and tested for OSA increased significantly, both statistically and clinically. This increase occurred following the creation of a standardized protocol and order set for managing pregnant women with obesity inclusive of sleep apnea screening (as described above) and a streamlined referral process for objective sleep apnea testing. Of women who completed testing, the majority met diagnostic criteria for OSA, with flexible use of both 4% and 3% desaturation criteria. This underlines the high prevalence of sleep-disordered breathing in pregnancies complicated by obesity, and the importance of considering and pursuing evalu-

Figure 3. Referrals and Sleep Studies Completed by Pregnant Women at the Wisconsin Sleep Clinic From January 2012 Through October 2019.



There was a statistically significant increase in the number of referrals and completed studies performed on pregnant women after the intervention (average 22.4 tests per year before [SD 5.3] and 67 [SD 16.6] after the intervention [P=0.0012]).

ation of OSA in pregnancy. However, the percentage of women who tested positive cannot be extrapolated to the nontested population, because there is likely ascertainment bias.

While the majority of pregnant women with OSA had "mild" sleep apnea, over 20% had "moderate or severe" sleep apnea. The classification of sleep apnea severity based on the number of respiratory events per hour (mild for AHI/RDI or REI 5 to 15/hr and moderate to severe for >15/hr) was formally adopted in 1999 and is based exclusively on PSG data, with scoring of respiratory events incorporating arousals, in addition to desaturations, to arrive at the AHI/RDI. Thus, designations of "mild" versus "moderate to severe" apnea severity remain empiric, have not been adjusted to reflect revised scoring criteria and 4-channel portable testing, and are unlikely to accurately define pathophysiological and clinical consequences of the unique physiology of OSA in pregnancy. In addition, use of 3% versus 4% desaturation criteria can easily change the diagnosis from snoring to OSA or move a diagnostic test from the mild to the moderate to severe category.[28] These limitations remain a challenge in the field of sleep medicine in general and are beyond the scope of this publication, outside of advocating for the use of less restrictive and likely more physiologically relevant 3% diagnostic criteria in the evaluation of OSA in pregnancy.

Women who did and did not complete testing had demographic differences that we will use to inform our ongoing efforts to increase diagnostic testing. Women who did not complete recommended sleep testing were less likely to be married and may be concerned that a sleep test would require them to be away from home, which would pose challenges if there are additional children in the family that require care. Thus, assuaging these concerns will be a priority for future interventions; the sleep test used in our clinic is a home test, and we will emphasize that. We are also working to determine best ways of distributing the PSAT devices to women at their obstetric clinic rather than requiring a trip to the sleep clinic to pick up the equipment, as the sleep clinic may not be near their home or work. Recommended sleep testing completion was also higher among women with hypothyroid disease, which may reflect worsened symptoms of fatigue. We also noted that testing completion was (nonstatistically) lowest in advanced pregnancy and, therefore, suggest screening in early to mid-pregnancy. This would also allow any indicated treatment to commence—and potentially have an effect—earlier in the pregnancy.

When OSA is diagnosed, systematic reviews and meta-analyses have demonstrated increased risk of adverse pregnancy outcomes.[2-4,6-12] Assessment of the impact of OSA on obstetric outcomes in our population is ongoing, as is assessment of whether treatment is beneficial. Here we demonstrate that while the overall referral completion rate was low, of the women who completed testing, over half were diagnosed with OSA, and treatment was recommended. CPAP remains first-line therapy for OSA.[3] While large trials of CPAP tolerance and efficacy in pregnancy are lacking, the findings of small studies suggest that treatment may improve obstetric outcomes with regards to preeclampsia.[17,18]

Strengths of this study include the detailed review of the sleep testing results and treatment plan to ensure that all women with sleep apnea were accurately diagnosed and evaluated in the sleep clinic and treatment was initiated expeditiously.

Limitations include the use of the electronic health record (EHR) to extract referrals retrospectively. The EHR occasionally lacks diagnostic codes for pregnancy, particularly for women whose prenatal care is not within the same health care system or EHR as the system where the sleep test occurs. This would be expected to reduce the capture of pregnant women seen at the sleep clinic. To account for this source of bias, our analyses of referral completion and sleep clinic visits utilized data exclusively from the electronic health record query and did not include data from our clinical database, as this would introduce understandable ascertainment bias. Retrospective analysis occasionally also lacks relevant clinical information. Some referrals lacked demographic characteristics as seen in the Table. Another important limitation

is the performance of sleep apnea screening questionnaires during pregnancy. Most screening tools used for the nonpregnant population perform poorly during pregnancy, including the Berlin and STOP-BANG questionnaires and the Epworth Sleepiness Scale, although each have some useful components.[14,23,29,30] We opted for Facco's 4-variable tool, which had the highest accuracy for predicting sleep apnea in pregnancy at the time our intervention was designed, although more recently, it has been demonstrated to have poor specificity among women with BMI ≥ 40 kg/m^2.[20,30] Our assessment of reported barriers was obtained by the sleep clinic schedulers and was limited by ascertainment bias due to the retrospective approach.

Future analyses of our clinical database will focus on assessing obstetric outcomes associated with sleep apnea diagnosis in this population, evaluating the currently utilized sleep screening tools, and measuring the impact of treatment on established obstetric outcomes. We will also plan a patient-focused exploration of the barriers to OSA testing via systematically performed interviews of recently delivered postpartum women and will study the percentage of tests performed that yielded positive results before and after the intervention. Our intention with this manuscript is to demonstrate 1 feasible workflow to allow other clinics to emulate this model, improve the number of women referred and screened for OSA during pregnancy and, hopefully, reduce adverse obstetric events associated with OSA.

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Evaluating the Impact of Provider Type and Patient Diagnosis on Patient No-Shows to Vascular Clinic

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ABSTRACT

Background: No-shows are a source of burden that lead to wasted resources. While prior research has established that many patient-level factors affect no-show rates, the impact of referring provider-level factors, in particular the type of referring provider and specific diagnosis, are still largely unknown.

Materials and Methods: Retrospective chart review examining new patient consults scheduled for outpatient vascular surgery clinic from August 1, 2014 through February 28, 2015 was conducted. The specialty types of the referring physicians and the reason for referral (patient diagnosis) were recorded.

Results: Of 227 new patient consults scheduled, 30% were no-shows to their appointment. No-show rates were significantly higher when the patient was referred by a primary care physician versus a specialist and differed significantly based on patient diagnosis.

Conclusions: Given that referring provider type and patient diagnosis significantly affect no-show rates, interventions that integrate the community of providers are needed to reduce no-shows.

INTRODUCTION

Health care expenditure in the United States has grown steadily over the last decade, reaching nearly 18% of our national gross domestic product (GDP) in 2017.[1] This figure amounts to roughly $10,739 spent per person, with a total of $3.5 trillion spent per year.[1] As costs continue to rise and resources become more limited, it is vital to evaluate sources of unnecessary health care expenditure and usage. Patient no-shows, or clinic cancellations, are one such problem that permeates all aspects of modern medical care and present a significant area of opportunity for improvement in vascular clinics. No-shows to clinics range from 4% to 80% of scheduled appointments, averaging approximately 23% of all clinic appointments and costing the overall health care system over $150 billion per year.[2,3] The United States Veterans Affairs (VA) system is the United States' largest integrated health care system, and a study in 2008 found that the average cost of no-show per patient in the VA system was $196.[4] In addition to monetary losses, no-shows lead to decreased provider productivity and wasted clinic time.[5,6] Given that providers are reserving clinic time for patients who ultimately do not show up, no-shows often lead to clinics being unnecessarily booked for long periods of time, leading to longer wait times for patients to schedule appointments and overall patient dissatisfaction.[5,6] Moreover, they can interfere with patient access to care, worsening patient outcomes.[7]

Prior research has established that patient-level factors, such as age and insurance, affect no-show rates.[2] However, the impact of referring provider specialty and patient vascular diagnosis on no-shows is still largely unknown. This study aims to provide an initial look into the impact of the referring provider's specialty and patient diagnosis on new patient no-shows to a large VA vascular surgery clinic. Referrals to a VA vascular surgery clinic were used as the subject of this investigation because the clinic is high-volume, the clinic does not typically see large variability in diagnosis among patients (allowing for more focused analysis of the impact of diagnosis type), and patient records in the VA system are well maintained.

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Table 1. No-shows by Referring Provider Specialty

Referring Provider Specialty	No. of Consult Appointments (n=227)	No. and Percentage of No-shows[a] (n=68)
Primary care provider	154	54 (35%)
Nephrology	26	5 (19%)
Inpatient medicine team	19	3 (16%)
Other specialties	28	6 (21%)

[a] $P = 0.047$. Analysis groups patients referred by nephrology with those referred by other specialities.

Table 2. No-shows by Reason for Referral

Diagnosis Listed as Reason for Referral	No. of Consult Appointments (n=227)	No. and Percentage of No-shows[a] (n=68)
Carotid stenosis	30	12 (40%)
Peripheral vascular disease	99	34 (34%)
Abdominal aortic aneurysm	47	9 (19%)
End-stage renal disease	40	9 (23%)
Other	11	4 (36%)

[a] $P = 0.044$. Analysis excludes patients with "Other" diagnoses.

METHODS

This retrospective study examined new patient consults scheduled for outpatient vascular surgery clinic from August 1, 2014 through February 28, 2015 using patient electronic medical records. Appointments were classified as either no-show or completed appointment, and the specialty types of the referring physicians were recorded. We also examined the reason for referral (patient diagnosis). While demographic and other patient characteristics were not collected, all patients were treated within the VA system. All statistical analyses comparing patients referred by different specialities and patients with varying diagnoses were performed using Fischer exact tests and chi-square tests with the Freeman-Halton extension.[8] Analyses were done using GraphPad PRISM 8 (La Jolla, California), and significance was set as $P < 0.05$.

This study was granted Institutional Review Board exemption, as this was deemed a quality improvement study with fully deidentified data. No patient information or images are disclosed in the report.

RESULTS

There were 227 new patient consults scheduled for the vascular surgery clinic over the 7-month study period. This number of newly referred patients is similar to that of other no-show analyses in the literature.[9-13] A total of 30% of patients were no-shows to their appointment (n = 68). No-show rates were highest among patients referred by their primary care provider versus patients referred by specialists (including nephrologists and other specialists) or by inpatient medicine teams (35% vs 19% /21% vs 16%, respectively; $P = 0.047$) (Table 1).

There were also significant differences in rates of no-shows among patients with different referral diagnoses ($P = 0.044$). When stratified by reason for referral, no-show rates were highest among patients referred for carotid stenosis (40%). The no-show rates of patients with peripheral vascular disease, abdominal aortic aneurysm, and end-stage renal disease were 34%, 19%, and 23%, respectively (Table 2). Patients with diagnoses other than those listed above comprised only 11 total referrals, of which 4 were no-shows (36%).

DISCUSSION

Studies have demonstrated that patient-level factors, namely socioeconomic factors such as age, race, income, insurance status, and history of previously missed appointments, affect no-show rates.[2,14] Additionally, other reasons, such as lack of transportation or time, forgetting appointments, distrust in staffing, unclear scheduling protocols, or just overall fear of a new diagnosis, all have been shown to play a role in no-shows.[15,16] These and many other variables have been shown to impact patient no-shows in clinic, but few studies have looked at referring providers. Although our study does not collect patient demographic or socioeconomic data of patients, all patients were treated under the VA health care system, providing a degree of uniformity to insurance status (although this is not exactly the same across patients, as the level of copay for procedures and subspecialty consults can vary). Here, we demonstrate that the referring provider's specialty and reason for referral may both affect no-show rates, drawing attention to some of the possible complex reasons outside of socioeconomic status that may lead to increased patient hesitancy to show up for their appointments. Though this investigation was a small pilot study and conducted at a VA clinic, the variables studied apply across virtually all specialty clinic referrals, as patients visiting these clinics often have a referring provider and reason for referral.

The challenges surrounding no-shows for new patients to a specialty clinic are unique. Specialty physicians generally focus on just one aspect of a patient's care plan, and these providers may see patients less frequently than a primary care provider. This places burden on the specialty care provider to quickly build rapport with their new patients and avoid diminishing any symptoms that they cannot appropriately address as a specialist. Additionally, new specialty referrals are often made for diagnoses that are new to the patient or involve new symptoms not adequately addressed by their referring physician alone. This unfamiliarity with their diagnosis and how it has changed over time, coupled with unfamiliarity with a new specialist, may increase a patient's fear, a known factor that leads to an increase in no-show events.[15] These examples help to highlight the importance of identifying individual barriers that each patient faces with a new

referral and working to alleviate these barriers. This burden also may fall partly on physicians receiving referrals. For example, by acknowledging that certain diagnoses are more likely to result in a no-show event, physicians receiving referrals could produce targeted education materials with information on at-risk diagnoses that could be given to referring providers for distribution to patients prior to referral.

Our study found that the no-show rate for patients referred by inpatient medical teams was less than half that of patients referred by a primary care provider. A possible explanation may be that subjectively, the inpatient setting connotates a higher degree of "seriousness" for patients than a scheduled visit to their primary care physician. Inpatient care requires a significant change to a patient's daily schedule in order to stay in the hospital as compared to routine primary care visits. In this sense, the inpatient setting may better emphasize the severity of a diagnosis, especially if the new diagnosis contributed to their inpatient stay. Further research is warranted in exploring why patients referred from inpatient medicine care chose to attend their scheduled appointments.

The reason for referral (the patient's diagnosis) also correlated with differing no-show rates. The highest no-show rates were among patients referred for a diagnosis of carotid stenosis, followed by peripheral vascular disease, abdominal aortic aneyurism and, finally, end-stage renal disease. Similar findings of diagnosis being a significant factor in patient no-shows have been reported in various fields, including cardiology, endocrinology, neurosurgery, infectious disease, and psychiatry.[2,17-22] Similar to our study, these studies do not collect granular data elucidating specific reasons for why patient diagnosis correlated with no-shows, but rather, in retrospective review, found a relationship between diagnosis and no-show rates.

Many factors may contribute to differing no-show rates among diagnoses. While detailed data of patient clinical status were not collected in this study, one reason may be disease severity. For example, it is possible that a higher proportion of patients with carotid stenosis and peripheral vascular disease were, in general, asymptomatic and in stable health, leading to a higher no-show rate. This is described by Zailinawati et al, who found that patients with coronary artery disease who were asymptomatic had high no-show rates.[23] In addition, similar results were found for neurosurgery clinic by Mark et al, who found that patients with chronic subdural hematomas had higher no-show rates compared to patients with symptomatic tumors and subarachnoid hemorrhages, citing that the difference may have been because most of the patients with chronic subdural hematomas had fewer complaints.[21] Furthermore, we found that patients referred by inpatient medicine teams had lower no-show rates than those referred by primary care providers. Patients receiving inpatient hospital care may have been sicker than those referred by primary care providers, further suggesting that severity of disease may contribute to no-show rates. Symptomatic patients or those with more severe manifestations of disease may have lower no-show rates because their referring provider may convey a greater sense of urgency, or the desire to alleviate symptoms may motivate patients to attend their appointments. Further studies more closely examining disease severity with no-shows are warranted. Given that diagnosis affects patient no-shows to vascular surgery clinic, it may be beneficial for physicians in this setting to have a more in-depth discussion of diagnosis with their patients as well as the importance of the future scheduled appointment in relation to their diagnosis.

Limitations

This pilot study has limitations. The study only examined patient referrals to 1 vascular surgery clinic in the VA system, limiting the generalizability of the findings. This is especially notable because there is no financial penalty for no-shows in the VA system, in contrast with most private health care systems. Similarly, the age, sex, and comorbidity status may be different between the VA population and the general public patient population, again limiting the generalizability of the findings. Despite these differences, we found that no-show rates overall were similar to what has been previously published for private clinics as well as within the VA system.[2] In addition, the study sample size was relatively small. The lack of measuring multiple demographic and patient-specific variables precluded the ability to perform a multivariate analysis of the data. This limited the conclusions that could be drawn when examining the impact of referring provider and patient diagnosis, leaving discussion to be primarily based on correlations observed.

CONCLUSIONS

Our results support the idea that patient no-shows and clinic cancellations are affected by multiple factors, including provider-level factors, such as specialty and reason for referral. It is likely these factors play into already studied reasons for no-shows, such as fear of the diagnosis, socioeconomic factors, or misunderstanding of a new scheduling system. However, the data shown in this study represent referring specialties and diagnoses that are particularly susceptible to no-shows, alerting physicians to at-risk patient populations. This suggests referring physicians should take time to identify and mitigate factors regarding referring provider and diagnosis that may contribute to no-show rates prior to the no-show event, if appropriate. Further study correcting for other factors and reasons that patients may have to not keep appointments could illuminate further how significant the impact diagnosis and physician specialty have on no-show rates. Understanding no-show patterns can improve future decisions and practices to decrease no-show rates and alleviate the associated economic burdens. Based on our findings, interventions to decrease no-shows at specialty clinics should take into consideration the whole com-

munity of providers involved in the patient's care, including the referring providers. Such an intervention may be accomplished through strategies such as changing the outpatient consult process to query if the reason for referral has been discussed with the patient and if the patient agrees to the consult.

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Barriers to Self-Disclosing Level of Maternal Care: What Are Wisconsin Hospitals Worried About?

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ABSTRACT

Objective: The American College of Obstetrics and Gynecology (ACOG) has recommended every hospital disclose their level of maternal care (LOMC) to categorize the capabilities of their birthing center and regionalize perinatal care. Of the 98 birthing centers in Wisconsin, 44% have self-disclosed their LOMC. In many states, disclosing LOMC is mandated but, despite evidence and professional association recommendations, Wisconsin relies on voluntary self-reporting. We surveyed all birthing centers in Wisconsin to better understand the barriers to disclosing their LOMC.

Study Design: An anonymous survey was sent to all 98 birthing centers in Wisconsin. Survey recipients were hospital administrators, nursing supervisors, or physician directors of obstetric units. The survey sought information on perceived barriers to completing self-assessments and disclosing their hospital's LOMC. Quantitative descriptive statistics were used for data analysis.

Results: Of 98 birth centers in Wisconsin, 40 (40.8%) responded. Fifteen of the 40 responses were from birthing centers that have not yet disclosed their LOMC. Of these, 93% were unsure how to disclose, 73% found the paperwork confusing, and 80% did not have the time or staff to complete the paperwork. Respondents did not report lack of departmental support, concerns about losing business or reputation, or future physician recruitment as barriers. Of all respondents, 77.5% were aware of ACOG's LOMC recommendations, but only 35% thought disclosing their LOMC would be beneficial to maternal care.

Conclusions: Birthing centers in Wisconsin need further guidance on how to complete a self-assessment of their LOMC. In order to increase self-disclosure of LOMC, statewide perinatal organizations will need to continue to emphasize the benefits of releasing this information. Organizations should also provide additional support to level 1 and 2 birthing centers and improve maternal and neonatal care overall.

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INTRODUCTION

In 1976, *Toward Improving the Outcome of Pregnancy* was published, which recommended the development of regional centers for perinatal health services.[1] Studies showed timely access to centers equipped to manage high-risk obstetrical patients improved perinatal outcomes and, thus, designating levels of perinatal services was suggested.[1] Over the ensuing 3 decades, the American College of Obstetricians and Gynecologists, American Academy of Pediatrics, and the March of Dimes have pushed for more consistent regionalization of perinatal care.[2-4]

Other specialty areas, such as trauma surgery and emergency departments, have provided excellent examples of regionalization of care, establishing networks for effective patient care and transports, and verification of disclosed levels.[5,6] Levels of neonatal and pediatric care also have been implemented. However, despite statements provided by The American Academy of Pediatrics in 2004 and 2012[7-9] that defined neonatal intensive care unit (NICU) levels of care and recommended universal disclosure, a recent state-by-state review found only 22 states with relevant policy as well as significant variation in definitions, criteria, and enforcement of reporting.[10]

The American College of Obstetricians and Gynecologists (ACOG) and the Society for Maternal-Fetal Medicine (SMFM) released a consensus document supporting disclosure of levels of maternal care (LOMC) in 2015 and revised it in 2019.[11,12]

Table 1. Brief Overview of Required Criteria for Each Proposed Level of Maternal Care

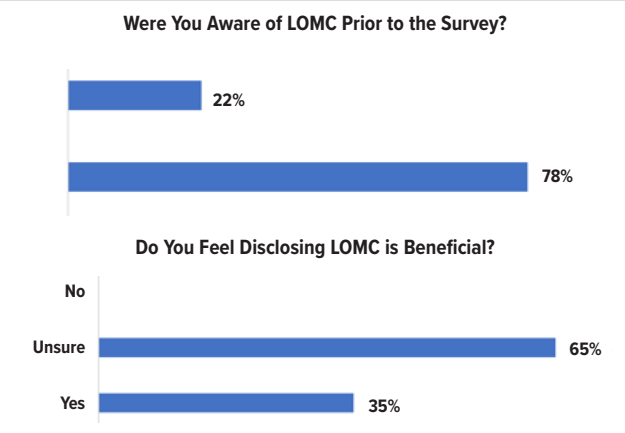
Level of Maternal Care	Criteria
Level 1[a] (Basic Care)	• Limited obstetric ultrasound available at all times • Support services available at all times (blood bank, laboratory testing) • Ability to start emergency Cesarean in a timely fashion • Ability at all times to initiate a massive transfusion protocol • Established procedures for patient stabilization and transport
Level 2[a] (Specialty Care)	Level 1 care plus: • OBGYN readily available at all times • Maternal-Fetal Medicine (MFM) specialist available at all times (telemedicine, telephone, in-person) • Anesthesiologist readily available at all times • Access to radiology and other imaging available daily • Obstetric ultrasound with interpretation available at all times
Level 3[a] (Subspecialty Care)	Level 2 care plus: • Board-certified obestetrician-gynecologist present at all times • Board-certified MFM specialist readily available in-person at all times • Board certified anesthesiologist present at all times • On-site medical, surgical intensive care unit • Access to radiology and other imaging at all times, including interventional radiology • Established procedures to accept patient transports
Level 4[a] (Regional Perinatal Healthcare Center)	Level 3 care plus: • Adult subspecialty consultants who can provide complex antepartum, intrapartum, and postpartum care of mother and infant • At least one of the following adult subspecialties readily available at all times: neurosurgery, cardiac surgery, or transplant

[a]Please refer to ACOG Consensus Statement Number 9 for full criteria. Adapted from: The American College of Obstetricians and Gynecologists and Society for Maternal-Fetal Medicine. Levels of Maternal Care. Obstetric Care Consensus No. 9. *Obstet Gynecol.* 2019;134:e41-55.

Table 2. Disclosed Levels of Maternal Care for Representatives of 40 of the 98 Hospitals With Birthing Centers Who Responded to the Survey

Disclosed Levels of Maternal Care	No. of Respondents
Level 1	15 (37.5%)
Level 2	5 (12.5%)
Level 3	2 (5%)
Level 4	3 (7.5%)
Unknown	15 (37.5%)

Figure 1. Survey Responses



Abbreviation: LOMC, levels of maternal care

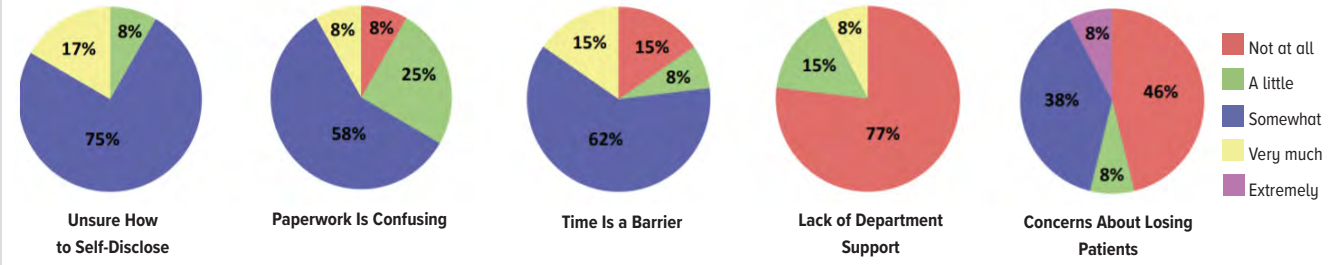
The 2019 consensus was endorsed or supported by the American Academy of Family Physicians, American Association of Birth Centers/Commission for the Accreditation for Birth Centers, American College of Nurse-Midwives, Association of Women's Health, Obstetric and Neonatal Nurses, and the Society for Obstetric Anesthesia and Perinatology.[12] The purpose of this statement was to standardize definitions of levels of maternal care, promote quality improvement, and develop risk-appropriate health care systems.[11]

In the last few years, attention has focused on birthing centers disclosing their LOMC in order to help improve maternal morbidity and mortality in the United States.[12] The Wisconsin Association of Perinatal Care (WAPC) is the organization that administers the voluntary application and approval process for LOMC in Wisconsin. The WAPC criteria are based on the ACOG/SMFM Levels of Maternal Care guidelines, the ACOG *Guidelines for Perinatal Care* 8th edition[11,13] (Appendix).

Knowledge of a hospital's LOMC conveys information about what resources are or are not available and also can elucidate hospitals that are under-resourced and may benefit from assistance. A brief overview of criteria required for each level of care is provided in Table 1. (See ACOG's *Obstetric Care Consensus on Levels of Maternal Care* for further details regarding criteria.[11,13]) A cross-sectional survey of California hospitals demonstrated that over half did not meet criteria for the lowest level of maternal care.[14] There is also evidence that high-acuity women who deliver at low-acuity hospitals have a higher risk of severe morbidity.[15]

Wisconsin is plagued by high rates of maternal and neonatal morbidity and mortality and some of the highest rates of maternal and infant racial health disparities in the country.[16,17] It is also important to note that women residing in rural areas face their own set of health disparities, specifically access to risk-appropriate care.[18] In September 2019, the Wisconsin Department of Health Services Maternal Mortality Review Team (MMRT) was awarded a 5-year federally funded Centers for Disease Control and Prevention ERASE grant (Enhancing Reviews and Surveillance to Eliminate Maternal Mortality). The MMRT enters deidentified confidential death review reports into a nationwide data system, the first step to understanding the causes of maternal mortality and eliminating preventable maternal deaths. Reports now include the LOMC. If the level of maternal care has not been assessed for the facility, that is noted

Figure 2. Perceived Barriers to Self-Disclosing Level of Maternal Care (N=15)[a]



[a]Data is from 15 birthing centers that have not disclosed levels of maternal care.

in the data system and shared with the review team. This data is necessary to make recommendations and improve care. Despite significant ongoing efforts by WAPC, when this project was designed, only 44% of Wisconsin birth centers—mostly from urban centers—had self-disclosed their LOMC.[19] There is an urgent need to understand the barriers to self-disclosing LOMC and to close gaps in reporting the state.

MATERIALS AND METHODS

Study Design

Between May 2019 and August 2019, online surveys were sent to representatives of all hospitals in Wisconsin that provide labor and delivery services. These representatives included chief executive officers, chief nursing officers, physician directors, and nursing supervisors of the 98 birthing centers. A total of 170 surveys were sent via email. Each representative was sent 3 emails with an opportunity to complete the survey at 2-week intervals. The survey was sent to up to 3 representatives from each institution. This study was approved by the University of Wisconsin-Madison Institutional Review Board (2019-0356).

Questionnaire

With the assistance of the University of Wisconsin-Madison Survey Center, an anonymous online survey was composed using REDCap (Research Electronic Data Capture, Vanderbilt University). Respondents were given an opportunity to state whether their institution had already disclosed their LOMC. Branching logic was then used to separate those that have self-disclosed and those that have not. The survey aimed to understand the birthing center's current resources, staffing, and consultants; their institution's barriers to self-disclosing their LOMC; and their perceptions of how LOMC would influence perinatal care. Each question posed a statement; respondents were asked to gauge the degree to which they agreed with the statement, from "not at all" to "extremely" or "immensely." There were also free text boxes provided for respondents to indicate whether additional barriers or concerns existed.

Survey responses were anonymous; however, investigators were privy to which birthing center representatives completed the study. Respondents provided consent to participate in the survey, and no incentive was offered for completion. Quantitative descriptive statistics were used for data analysis.

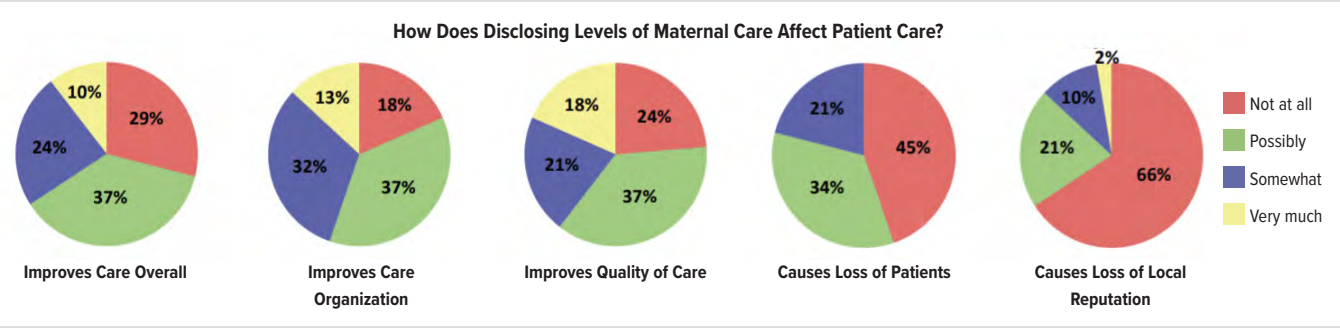
RESULTS

Of 98 hospitals with birthing centers, representatives from 40 birthing centers (40.8%) responded to the survey (Table 2). Fifteen of 40 responses were from birthing centers that stated they had not yet disclosed their LOMC. Respondents were asked if they were aware of ACOG's LOMC recommendations[11] prior to receiving the survey and 78% agreed. When asked if they felt disclosing LOMC was beneficial in general, 65% of respondents were unsure, while 35% agreed it was beneficial (Figure 1).

Of the 15 birthing centers that had not self-disclosed their LOMC, 92% of respondents were at least somewhat unsure how to disclose. The paperwork was at least somewhat confusing to 66% of respondents, and 77% indicated that time was at least somewhat a barrier. Respondents did not report lack of departmental support, concerns about losing business or reputation, or future physician recruitment as barriers. However, 46% reported they were somewhat (38%) or extremely (8%) concerned about losing patients (Figure 2). Two respondents omitted answering certain portions of the questions related to barriers to disclosing their LOMC.

The survey asked respondents if they felt disclosing their LOMC would be beneficial to perinatal care overall. All respondents indicated it would be at least somewhat beneficial, but no one thought it would be "immensely" beneficial. Of all respondents, 35% said disclosing LOMC would at least somewhat improve patient care, 45% said there would be somewhat of an improvement in care organization, and 39% said it would at least somewhat improve the quality of care. A majority indicated they did not think disclosing LOMC would affect patient numbers or local reputation, but 33% at least somewhat had concerns about effects on local reputation (Figure 3). No respondents said that self-disclosing LOMC would immensely benefit the organization or quality of care.

Figure 3. Data From All Respondents Regarding Their Thoughts About Disclosing Levels of Maternal Care (N=40)



DISCUSSION

Barriers to disclosing LOMC experienced by Wisconsin birthing centers included lack of knowledge about how to self-disclose, lack of administrative time, confusing paperwork, and some concern regarding loss of patients. No respondents said they felt that self-disclosing LOMC would immensely benefit patients, the organization, or quality of care. It is also important to note that 7 respondents did express concern regarding loss of patients or local reputation.

While 78% of survey respondents were aware of ACOG's LOMC guidelines, 65% of respondents were unsure whether disclosing their LOMC was beneficial to perinatal care. This is in contrast to other states, such as Texas and Illinois, that have mandated all birthing centers disclose their LOMC in an effort to regionalize perinatal care. It is important to note states like Texas and Illinois are similar to Wisconsin, specifically as they all have large rural areas with smaller hospitals and fewer resources. Disclosing LOMC will foster the establishment of connections between birth facilities to ensure that appropriate resources are available for women with high-acuity conditions. These connections can provide consultative support, facilitate appropriate patient transports and care organization, impart educational resources, and create collaborative efforts to improve the care of moms and babies throughout the state. Prior studies have shown that timely access to centers equipped to manage high-risk obstetrical patients improved perinatal outcomes.[1]

One significant obstacle we did not anticipate was the inability to procure an updated, verified list of contacts for birthing center administrators. The initial list we identified was comprised of hospital chief executive officers or chief nursing officers who were not always the appropriate contact personnel to complete the survey, nor are they likely to be the primary official responsible for disclosing LOMC. This prompted us to create a list of obstetric-care administrative contacts for Wisconsin hospitals. We were able to contact 72 birthing centers and confirmed contact information for 63 of the facilities. This list of contacts was shared with WAPC and the Wisconsin Perinatal Quality Collaborative to maintain and update.

A critical limitation to our study was a low response rate. We believe this can be attributed to the lack of appropriate contact information mentioned above and also lack of time on behalf of health care administrators. However, we received responses from a geographically diverse group of birthing centers that also differ in delivery volume; therefore, we believe the results are likely generalizable to institutions in Wisconsin. The questions within the survey were also mostly close-ended questions. Therefore, there may be other key barriers that were not captured within our survey. As of November 25, 2019—after our study period was complete—an additional 6 birthing centers disclosed their LOMC. Because the survey was anonymous, we cannot recalculate our data to fit the most updated information, nor can we identify if those were birthing centers that acknowledged our survey.

Based upon our data, we suggest that future statewide efforts focus on emphasizing downstream benefits of disclosing LOMC for the purpose of regionalizing perinatal care and improving perinatal outcomes. It is well documented that integrating systems of care across levels improves outcomes. Many states have used a level of care process to monitor access and quality of maternal and infant care. Understanding a hospital's level of maternal care is a critical step toward ensuring that women are giving birth at a hospital that is equipped to adequately meet the level of risk. However, it is critical to also implement monitoring of adherence to best practices aligned with each level.

There remains significant variation in state endorsement of LOMC disclosure. Currently, implementation of LOMC is at the state's discretion, and there is not a federal mandate in place for birthing centers to disclose their LOMC. However, LOMC has caught the attention of federal programs, specifically the Centers for Disease Control and Prevention (CDC). The CDC's Maternal and Child Health Epidemiology Program has worked to design a tool to help birthing centers assess their capabilities—the CDC LOCATe tool.[20] The tool is easily accessible and has been used by other states, such as Texas and Illinois, to assess a birthing center's capabilities and help regionalize perinatal care. Texas has led these efforts and, as of March 2018,

has required every birthing center disclose their LOMC and participate in an onsite verification process in order to receive Medicaid funds.[21] Birthing centers in Texas will be required to disclose their LOMC by August 2021 to receive Medicaid reimbursement for obstetrical care.[22] The state of Illinois also has been able to implement perinatal regionalization using a state-specific adaptation of the CDC LOCATe Tool.[23,24] Illinois also is currently part of a 3-state program implementing onsite verification of LOCATe results.[12]

Despite success in other states, Wisconsin has not prioritized policies regarding disclosing LOMC. These results likely align with data from California suggesting many birthing centers may not meet criteria for even the lowest level of maternity care and, therefore, may need additional support to reach these criteria.[14] It is important to note these efforts also may address the benefits of knowing the level of maternity care for one's own hospital, as well as regional hospitals, in order to effectively regionalize perinatal care. Knowing the level of maternity care can also aid hospitals and clinicians in stratifying risk-appropriate care for their patients, given that high-acuity patients who deliver at low-acuity hospitals have increased morbidity compared to high-acuity patients who deliver at high-acuity hospitals.[15,20] LOMC disclosure also can aid in facilitating a network for maternal transfers and ambulance direction. The level of maternal care cannot be assumed to be the same as the level of neonatal care, as these levels are often discordant.[14] The primary issues surrounding access to risk-appropriate care are difficult access to tertiary care centers for some women in rural areas, the patient's lack of awareness of the birthing center's capabilities, and the frank unpredictability of pregnant and postpartum women's medical status. We believe providers understand their hospital's limitations; however, patients may present to them in emergency situations they may be ill-equipped to manage.

Because the mandates in both Texas and Illinois are recent, without full implementation until the future, outcomes cannot yet be assessed. Regardless, implementing statewide LOMC is still recommended by all of the organizations listed above. We encourage all statewide organizations that provide care to pregnant women to also support efforts to encourage birthing centers to complete their self-assessment of LOMC.

Statewide organizations may also focus on establishing avenues for disseminating information, which may include regional forums where hands-on sessions could assist participants to access the website and complete their LOMC. Birthing centers should also be given the opportunity to evaluate the application or request feedback in order to improve the application process. Lastly, state organizations should keep a statewide repository of updated contact information for birthing center administrators to improve communication amongst institutions providing perinatal services. By providing clear communication about the resources available at each birthing center, statewide implementation of LOMC should make childbirth a safer experience for mothers in Wisconsin.

CONCLUSION

Birthing centers in Wisconsin need further guidance on how to complete a self-assessment of their LOMC. In order to increase self-disclosure of LOMC, statewide perinatal organizations will need to continue to emphasize the benefits of releasing this information. Organizations should also provide additional support to level 1 and 2 birthing centers and improve maternal and neonatal care overall.

Acknowledgements: We would like to thank Kathy Kostrivas, Wisconsin Association for Perinatal Care, the Wisconsin Perinatal Quality Collaborative, and the Wisconsin Section of the American College of Obstetricians and Gynecologists for their collaboration on this project. Survey design support was provided by the University of Wisconsin Survey Center. We also thank all hospitals with birthing centers in Wisconsin for their commitment to providing accessible and safe care to women in this state.

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Appendix: Available at www.wmjonline.org.

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Prevention of Neonatal Hypoglycemia With Oral Glucose Gel for High-Risk Newborns

Mark Deyo-Svendsen, MD; Sara Herrmann, MD; Christina Andrist, DO; Michael Phillips, MD; Matthew Cabrera Svendsen, MD; Rachel Oldfather

ABSTRACT

Background: Neonatal hypoglycemia (glucose <47) is the most common metabolic problem in newborns (incidence 5% - 15%) and can cause adverse outcomes, even in the absence of noticeable symptoms. Oral glucose gel (OGG) is safe and effective for treatment of neonatal hypoglycemia. In order to reduce interventions such as intravenous (IV) dextrose administration and neonatal intensive care unit (NICU) transfer, in October 2017, we implemented a protocol in our Level 1 rural community hospital to identify newborns with asymptomatic hypoglycemia based on risk factors and treat them with OGG. Risk factors include large or small size for gestational age, maternal gestational diabetes, preterm and late preterm birth, and newborns requiring resuscitation.

Methods: Chart review was performed for all infants born at our hospital from October 1, 2016 through September 30, 2018. Data for year 1—the period before protocol implementation (October 2016-September 2017)—was compared to post implementation data from year 2 (October 2017-September 2018).

Results: There was a significant risk reduction in newborns requiring interventions due to hypoglycemia after protocol implementation ($P = 0.029$, Student t test). In year one, 7 of 310 total newborns required IV dextrose or NICU transfer related to neonatal hypoglycemia. In year two, 108 out of 250 total newborns were tested for asymptomatic hypoglycemia based on risk factors identified in the protocol. Of those tested, 31 newborns demonstrated hypoglycemia and received OGG. None of the 250 newborns required further associated interventions.

Conclusion: Protocol-based hypoglycemia testing based on risk factors with subsequent OGG administration was effective in reducing the need for IV dextrose and NICU transfer from our Level 1 rural community hospital.

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INTRODUCTION

Neonatal hypoglycemia is the most common metabolic problem in newborns and is a preventable cause of neurological impairment.[1] The incidence of neonatal hypoglycemia ranges from 5% to 15% in the first few days after birth[2,3] and increases to more than 50% in newborns with certain risk factors. For example, milestone data indicate hypoglycemia rates of 19% to 52% in premature newborns, 42% to 54% in newborns small for gestational age, 10% to 47% in newborns large for gestational age, and 33% to 48% in newborns born to mothers with diabetes.[4,5]

Because of the critical role glucose plays in brain metabolism, short- and long-term neurological impairment can result from neonatal hypoglycemia—even if asymptomatic—depending on the severity and duration.[6,7] Transient hypoglycemia can be physiologic following birth,[6] but hypoglycemia also may be the initial presenting sign for complex metabolic disorders, such as hyperinsulinemia, glycogen storage disease, congenital disorders of glycosylation, galactosemia, fatty acid oxidation defects, growth hormone deficiency, and adrenal insufficiency.[8]

The definition of neonatal hypoglycemia and the glucose value at which adverse outcomes occur is controversial. The Pediatric Endocrine Society opined that neurological injury may occur at a glucose level less than 47 mg/dL,[9] and others support this risk threshold.[7] However, after evaluating newborns with asymptomatic hypoglycemia and poor long-term neurodevelop-

Table. Risk Factors Contributing to Neonatal Hypoglycemia Per Chart Review

Newborns Screened	Year 1[a]	Year 2[b]
Large for gestational age	34	29
Gestational diabetes mellitus	22	19
Small for gestational age	12	10
Maternal type 1 and type 2 diabetes mellitus	2	0

[a]October 1, 2017 to September 30, 2017, pre-implementation.
[b]October 1, 2017 to September 30, 2018, post-implementation.

mental outcomes, the American Academy of Pediatrics (AAP) indicated that injury may occur when glucose levels are below 30 mg/dL.[10]

Traditionally, oral glucose gel (OGG) has been used to reverse hypoglycemia in persons with diabetes. Sublingual and buccal administration of OGG is preferred over the oral route,[11] because it avoids first-pass metabolism and improves bioavailability for fast treatment results. The 2013 Sugar Babies Study[12] found that compared to formula feeding, OGG reduces treatment failure, as defined by hypoglycemia after 2 treatment attempts. It also found OGG to be both safe and effective for neonates.

The AAP generally endorses selective screening for hypoglycemia in high-risk newborns. Specifically, in 2011, the AAP Committee on Fetus and Newborn guidelines provided screening guidance for newborns who are late-preterm, small or large for gestational age, or born to mothers with diabetes.[10] Based on these guidelines, we implemented a neonatal hypoglycemia protocol to identify and treat newborns with asymptomatic hypoglycemia in our nursery.[10] The goal of this quality improvement initiative was to reduce interventions such as intravenous (IV) dextrose administration and neonatal intensive care unit (NICU) transfer.

Our Level 1 rural hospital is part of an integrated network of hospitals and clinics in the Midwest United States, serving approximately 45,000 people.[13] Eight family medicine physicians and 3 obstetrician-gynecologists provide prenatal care; 13 family medicine physicians and 2 pediatricians provide newborn care. The family birth center offers newborn care annually to 250 to 350 patients. In general, the rural population served by our nursery may benefit from actions taken to ensure adequate care as close to home as possible. Most families practice "rooming-in" (ie, the newborn is cared for in the postpartum room), and not all newborns designated as high risk are required to receive care in the nursery.[14] Medicaid is the most common payer for maternity care services (64%). Teenage pregnancy (16.7%), poverty (13.9%), and delayed or no prenatal care (4 or fewer prenatal visits 4.2%; 18.4% not in the first trimester) contribute to a high-risk population. The primary and repeat cesarean delivery rates are 15% and 9%, respectively, totaling 24% of deliveries. Most newborns (93.8%) are normal birth weight.[14]

METHODS

The hypoglycemia protocol in this study was designed to follow the AAP's 2011 clinical statement on the postnatal management of hypoglycemia in late-term and term newborns (Appendix).[10] Newborns identified as high-risk (Appendix) were provided early feeding and a glucose level was obtained. Those with a glucose level less than 45 mg/dL were provided OGG 0.2 grams/kg.[15] The newborn's mouth first was dried with gauze, then OGG was applied with a gloved finger and massaged into the buccal mucosa. Doses greater than 1 mL were divided and administered bilaterally to promote buccal absorption and to reduce swallowing. Following this procedure, nurses immediately offered feeding with human milk or at least 15 mL of formula. Providers were notified if newborns did not feed. Blood glucose levels were rechecked 30 minutes after feeding.

A labor and delivery registered nurse reviewed charts of all newborns born at our hospital from October 1, 2016 to September 30, 2018. Year 1 (October 1, 2016-September 30, 2017) data included newborns treated for hypoglycemia with formula, human milk, or IV dextrose infusion. These data were compared to post-implementation data from Year 2 (October 1, 2017-September 30, 2018).

The Institutional Review Board for Mayo Clinic Health System Northwest Wisconsin approved this research project in October 2017, prior to protocol implementation.

RESULTS

In year one, 122 neonates were screened for hypoglycemia. Seventy were identified with specific hypoglycemia risk factors, and 12 had hemodynamic instability or required resuscitation. Of the 310 total newborns, 7 with neonatal hypoglycemia required IV dextrose or NICU transfer due to hypoglycemia (2.25%, 95% CI, 1.80%-2.71%).

In year two, 108 neonates were screened for hypoglycemia. Fifty-eight had specific hypoglycemia risk factors, and 22 had hemodynamic instability or required resuscitation (Table). None of the 250 newborns required IV dextrose or NICU transfer due to hypoglycemia. This was a significant reduction in risk ($P = 0.029$, Student t test). Relative risk reduction is undefined as there were zero interventions in year 2.

It should be noted that 59 newborns with symptomatic hypoglycemia (37 in year 1; 22 in year 2) did not meet protocol criteria and were excluded from the study. Newborns transferred to a NICU for causes unrelated to hypoglycemia also were excluded; these newborns required resuscitation or were experiencing persistent tachycardia, respiratory distress, and hypothermia. All excluded patients were tested and treated as appropriate.

DISCUSSION

A hypoglycemia protocol adapted from the 2011 AAP guidelines[10] was implemented in our Level 1 rural community hospital nurs-

ery. Newborns identified with asymptomatic hypoglycemia based on risk factors were treated with OGG. In the year following protocol implementation, no newborns required treatment with IV dextrose or transfer to a higher level of care for treatment related to asymptomatic hypoglycemia.

Study limitations include the small sample size and the fact that the study design utilized retrospective chart review. Additionally, in year 1, infants were tested only if signs of hypoglycemia were present; if hypoglycemia was present, newborns were treated with early feeding or IV dextrose. It is not known whether at-risk infants were observed differently for signs of hypoglycemia—and potentially offered early feeding without testing—than those who were not at risk. Had this occurred, it may have resulted in lower year 1 interventions. Without this potential bias, the impact of testing and treating at-risk infants with OGG might have been even greater.

The findings from this quality improvement initiative support newborn screening for asymptomatic hypoglycemia based on select risk factors and subsequent treatment with OGG, and they are consistent with other recent reports of reduced interventions for hypoglycemia with the use of OGG in asymptomatic newborns. One retrospective analysis found that OGG effectively managed newborns deemed high risk for hypoglycemia and reported high-risk identifiers for newborns more likely to require a second dose of OGG or IV fluids with dextrose;[16] another reported a reduction in NICU transfer, improvement in breastfeeding rates, and an overall associated reduction in cost of care with OGG.[17]

Beyond reducing interventions for newborns, our findings have additional implications for practice. There is concern that intravenous infusion separates the mother and newborn and using formula to treat hypoglycemia may disrupt breastfeeding.[12,18] Particularly in a smaller newborn unit, starting and maintaining an IV might require transfer to a higher level of care facility.[12] Administration of OGG can occur quickly in the nursery while simultaneously encouraging maternal bonding. OGG has been shown to reduce hospital costs and supports breastfeeding.[12,17,19] OGG is a practical, cost-effective method to manage neonatal hypoglycemia in comparison to IV dextrose and formula.

CONCLUSION

Our findings indicate that protocol-based identification of newborns at risk for hypoglycemia with subsequent administration of OGG is an effective method to address asymptomatic neonatal hypoglycemia. It eliminated the need for more aggressive interventions, including peripheral IV line placement and IV dextrose infusion, and decreased the number of newborns transferred to a neonatal intensive care unit.

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Pharmacotherapy for Management of 'Kratom Use Disorder': A Systematic Literature Review With Survey of Experts

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ABSTRACT

Objectives: An increasing number of Americans are turning to kratom for self-management of various pain, anxiety, and mood states and as an opioid substitute. Addiction to this unique botanical develops and carries a high relapse risk and, to date, there are no guidelines on how to maintain long-term abstinence. The aim of this article is to compile all available information on management of "kratom use disorder" (KUD)—as coined here—from the literature, with evidence from the clinical practice of expert addictionologists in an attempt to develop a standard of care consensus.

Methods: A systematic literature search was conducted to capture all relevant cases pertaining to maintenance treatment for KUD. Results were supplemented with case reports and scientific posters gleaned from reliable online sources and conference proceedings. Additionally, a survey of members of the American Society of Addiction Medicine (ASAM) was administered to assess the practice patterns of experts who treat patients with KUD in isolation of a comorbid opioid use disorder (OUD).

Results: Based on a literature review, 14 reports exist of long-term management of KUD, half of which do not involve a comorbid OUD. Pharmacological modalities utilized include mostly buprenorphine but also a few cases of naltrexone and methadone, all with favorable outcomes. This is supported by the results of the expert survey, which demonstrated that those who have managed KUD in isolation of a comorbid OUD reported having utilized buprenorphine (89.5%), as well as the other medications for opioid use disorder (MOUD).

Conclusions: This is the first comprehensive review to examine the existing literature referring to management of KUD in combination with a survey of current experts' clinical consensus regarding pharmacological management. Based on this information, it seems reasonable that the indication for MOUD should be extended to cases of moderate to severe KUD.

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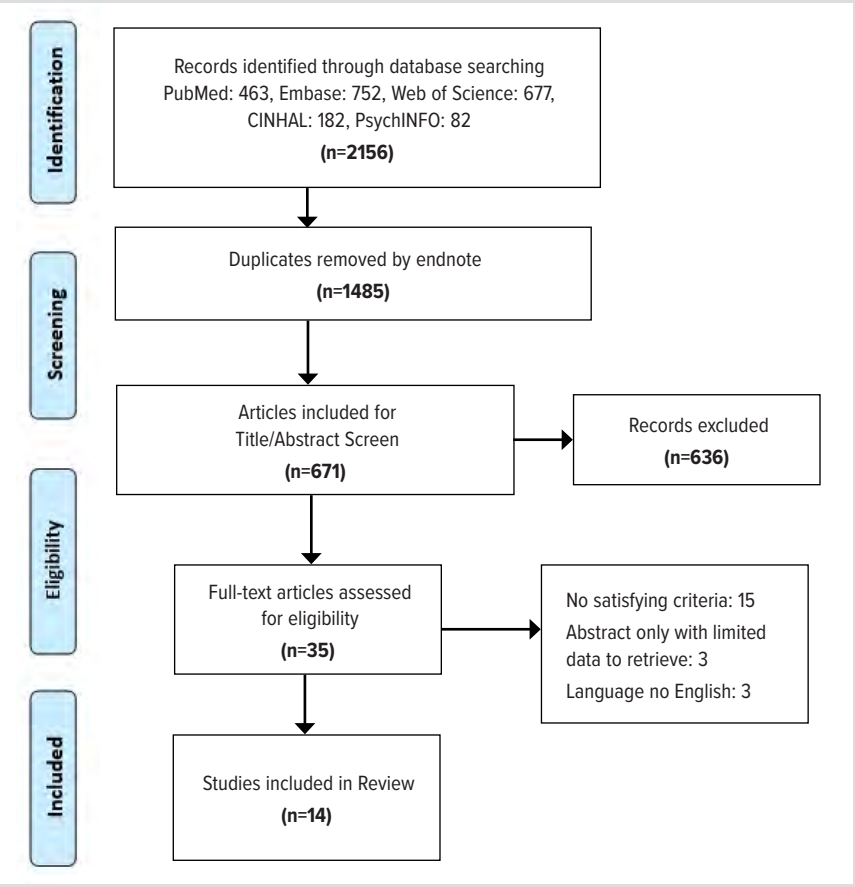
INTRODUCTION

The increasing consumption of kratom (*Mitragyna speciosa*) is emerging as a public health concern among Americans, and forecasting models indicate its use will continue to rise.[1] Aside from the Food and Drug Administration (FDA) reports of concern[2] and adverse effects exhibited through increased calls to poison control centers[3] and overdose deaths,[4] the notion of addiction is rapidly emerging. In Southeast Asia where this botanical is indigenous, 55% of regular users develop dependence and tolerance. Withdrawal and cravings also have been reported.[5-8] There is now substantial evidence showing it is possible for individual kratom users to meet all Diagnostic and Statistical Manual, Fifth Edition (DSM-5) criteria associated with a substance use disorder diagnosis.[9] A category for "kratom use disorder" (KUD)—as we coin in this paper—does not formally exist in the DSM-5, which was last revised in 2013. In the United States, a survey of 8,000 users conducted through American Kratom Association (AKA)[10] revealed that although some disclosed use with an underlying intent to self-manage opioid misuse including withdrawal, 68% reported using to self-manage chronic pain and 65% for anxiety or mood states, where opioids are not involved at all.

The effects of kratom to date are attributed primarily to the 2 active alkaloids—mitragynine (MG) and 7-hydroxymitragynine (7-HMG)—although more than 25 other alkaloids have been identified in the plant.[11] Both exert their primary action through agonism at the μ opiate receptor and weak antagonism at δ and κ receptors.[12,13] There is also evidence that MG is involved in sero-

tonergic (antagonist at serotonin 5-HT-2A receptors), dopaminergic (agonist at dopamine D1 receptors), and noradrenergic (agonist at postsynaptic alpha-2 receptors) pathways.[14-17] These translate to users experiencing stimulant-like and opioid-like intoxicating syndromes when either low or high doses are consumed. In traditional medicine, kratom leaves have been used for pain relief; to increase appetite, mood, energy, and sexual desires; to provide wound healing based on anti-inflammatory properties; as a local anesthetic; and to manage coughs, diarrhea, and intestinal infections, among other uses. It is apparent that MG, 7-HMG, and the rest of the plant's constituents are involved in a multitude of other pathways as well, which have yet to be determined. Although there have been efforts by the FDA to classify MG and 7-HMG as an opioid based on the Public Health Assessment via Structural Evaluation (PHASE) model,[18] this is a very complex botanical with much more unique pharmacodynamic and intracellular signaling actions, hence deserving its own category and classification.

Figure 1. PRISMA Flow Diagram of Literature Search



In a previous review of kratom withdrawal,[6] we outlined that symptoms respond akin to that of opioid withdrawal through symptomatic management of a hyperadrenergic state and/or use of opioid receptor agonists (methadone) or partial agonists (buprenorphine). We also alluded to the notion of cravings being present and that there is a high risk of relapse to use on cessation. To date, no guidelines exist regarding the long-term management of KUD. In medical terminology, the "standard of care" is established based on what the average physician in the appropriate specialty community would do when faced with a specific situation. When it comes to KUD management, there is a great need to establish such a standard of care. In this article we report on all the evidence currently available in the literature and combine it with survey information regarding pharmacological management by the addiction medicine specialty community. The aim here is to evaluate potentially beneficial pharmacotherapy only and not specifically any behavioral treatments.

METHODS

Literature Search

We searched PubMed/MEDLINE, PsycINFO, PsycARTICLES, CINAHL, EMBASE, Scopus, Cochrane, and Academic OneFile for English-language medical literature published between January 1, 1970, and January 1, 2020, using the search terms: "kratom," "mitragyna speciose," "mitragynine," and "7-hydroxymitragynine."

Regarding inclusion and exclusionary criteria, our interest revolved around clinical cases reporting the use of any pharmacotherapy in management of remission from kratom use in both humans and animals. Only English literature was considered.

The original search yielded a total of 2156 returns: PubMed (n = 463), Embase (n = 752), Web of Science (n = 677), CINHAL (n = 182), and PsychINFO (n = 82). After removing duplicates, 671 citations were left. Authors CS and BH examined each by title and abstract. After eliminating studies based on exclusionary criteria and applying the inclusion criteria, 14 papers met the original search criteria (Figure 1, Tables 1 and 2). Any disagreements would have been mediated for proper allocation by a third reviewer, but that was not required. Results were supplemented by references gleaned from recent reviews and citations of searched returns, as well as credible reports from academic conferences (Figure 1).

Survey

A survey was designed via Qualtrics (https://www.qualtrics.com) and distributed to the 40 state chapter presidents of the American Society of Addiction Medicine (ASAM), with a request to extend it to their specific membership group. At the time of the survey,

Table 1. Cases Reporting Maintenance Pharmacotherapy of Patients With Kratom Use Disorder and Opiod Use Disorders

Ref No.	Clinical Paradigm	Reason for Kratom Use	Extent of Kratom Used	Intervention	Maintenance Regimen	Outcome
16	43-year-old man with history of chronic pain from thoracic outlet syndrome treated with hydromorphone. Started subcutaneously injecting crushed 10 mg tablets of hydromorphone and using kratom to help ameliorate withdrawal when hydromorphone not available. Stopped hydromorphone 3.5 years before presenting and was strictly using kratom. Started taking modafinil 100 mg to help with alertness and presented to ED after experiencing a generalized tonic-clonic seizure. Following discharge, stopped kratom and reported a less intense but more protracted withdrawal compared to opioids persisting for 10 days.	Opioid substitution	Initially used unknown amount of kratom to manage episodic withdrawal from hydromorphone. Ultimately continued using unknown quantity of kratom as a tea 4 x/day; reported spending $15,000/year on kratom.	Started on BUP/NX following withdrawal from kratom to assist with cravings, 16-4 mg.	BUP/NX 16-4 mg/day	Ongoing abstinence confirmed by urine toxicology, maintained on BUP/NX 16-4 mg/day.
20	52-year-old woman with depression and chronic pain admitted to inpatient psychiatric unit for suicidal ideations. She was experiencing opioid-like withdrawal symptoms. Years prior had developed iatrogenic opioid addiction and switched to kratom 9 months prior to presentation.	Pain management	9 months of use. Gradually increased from 1 tbsp/day powdered plant matter to 1 tbsp 4-6 times/day.	As inpatient, BUP/NX induction occurred, requiring 16/4 mg on day 1 for withdrawal symptoms. Initial plan was for taper but, due to difficulty tapering, was discharged with 2-0.5 mg 4 times/day. BUP/NX increased to 8-2 mg 2x/day to manage cravings as outpatient.	BUP/NX 8-2mg 2x/day	Ongoing abstinence at 18 months, corroborated via negative urine toxicologies.
21	32-year-old man with history of PTSD, alcohol use disorder, and OUD in remission from heroin for 2 years. Presented to outpatient clinic for help with kratom dependence.	Energy	8 months of use. Started using 1 capsule kratom product/day; increased to 5-10 capsules/day.	As outpatient, started on BUP/NX 4-1 mg/day; increased to 16-4 mg/day due to withdrawal symptoms.	BUP/NX 16-4 mg/day	No cravings endorsed at follow-up visits; toxicology screens unremarkable.
22	28-year-old woman at 19 weeks of gestation with history of alcohol use disorder in remission, stimulant (methamphetamine) and OUD (heroin) complicated by a bipolar spectrum diagnosis; presented to ED for symptoms of withdrawal due to kratom use.	Opioid substitution	4 months of use prior to presentation via smoking; unknown amount, frequency.	Upon admission to inpatient unit, BUP/NX induction occurred. Discharged on 4-1 mg 4 times/day. At 36 weeks gestation, BUP/NX increased to 20-3 mg daily to address withdrawal symptoms.	BUP/NX 4-1 mg 4 x/day; increased to 20-3 mg/day at 36 weeks gestation	Upon induced delivery at 39 weeks, patient continued with BUP/NX 20-3 mg during hospitalization; discharged on it with ongoing abstinence at follow-up.
23	57-year-old man with chronic back pain, anxiety, depression; originally prescribed oxycodone but developed iatrogenic addiction. After oxycodone was discontinued, transitioned to using kratom 1 year prior to presenting. Noted withdrawal when without kratom and sought help.	Pain management	1 year of use; unknown dose, duration, frequency, route of administration. Purchased from online retailer; spent ~$2500/month.	Outpatient induction to BUP/NX was performed; patient transitioned to 24-6 mg/day for maintenance.	BUP/NX 24-6 mg daily	Abstinence maintained at 7-month follow-up; confirmed by urine toxicology.
24	54-year-old man with history of depression, anxiety, and 16-year history of iatrogenic opioid addiction. Used kratom to assist quitting opioids but experienced difficulty when trying to stop. Presented to outpatient addiction treatment clinic for help.	Opioid substitution	Unknown amount, formulation, duration.	Inducted on BUP/NX 8-2 mg on day 1; increased to 16-4 mg on day 2 to target withdrawal symptoms and cravings.	BUP/NX 8-2 mg 2x/day	Maintained abstinence at 2 months while on BUP/NX 8-2 mg 2x/day. Weeks 2-5 post induction, urine mitragynine levels were 52.7, 36.6, 1.2, and < 1 ng/mL (negative), respectively.
25	Report of 9 veterans using kratom in 2013 and 8 more between 2016 and 2017. Two-thirds used kratom daily. One used kratom solely for pain and had an alcohol use disorder. Remainder had history of severe OUD and other substance use disorders. Kratom listed as opioid of choice in 50%; 40% noted tolerance and withdrawal.	Opioid substitution, pain management	Two-thirds had reported daily use of kratom. Formulation included tea/drink, capsules, leaves added to food, or multiple means.		BUP/NX, methadone, naltrexone	All who were opioid dependent were treated with BUP/NX, referred to a methadone clinic, or treated with naltrexone.

Abbreviations: ED, emergency department; BUP/NX, buprenorphine/naloxone; tbsp, tablespoon; PTSD, posttraumatic stress disorder; OUD, opioid use disorder.

ASAM's membership was 6,365. By using formulas for the maximum error of the estimates, we determined that—for a 95% confidence interval and margin of error of 0.4—a sample size of 564 was required.[19] The survey was distributed initially on January 9, 2020 and was available for 10 days, with 1 brief communication reminder sent during this period to the ASAM chapter presidents. A total of 711 participation invites were sent. Participants were registered electronically through an individualized link, responses were anonymous, and no personal identifiers were collected.

The survey was intended to gauge whether specialists have encountered patients suffering from KUD and how they have managed abstinence in such cases. Our main interest was in pharmacological management of KUD in isolation of past or comorbid OUD histories. Specific questions and flow are detailed in Appendix A.

Eighty-two participants completed the survey, a response rate of 11.5%. Data generated were analyzed via Qualtrics. Some participants who had encountered KUD in isolation of OUD also entered comments regarding management and outcomes (see Appendix B).

RESULTS

Literature Search

The literature review yielded 14 reports involving patients for whom long-term maintenance of KUD was required, including 7 with concomitant OUD diagnoses. Of those 7 patients, all received buprenorphine for maintenance with doses of 16 mg daily; 1 patient required increase from 16 mg to 20 mg due to pregnancy, and another required 24 mg daily. All had switched to kratom use to replace their opioid addiction.

Of the 7 patients without concomitant OUD, 4 were using kratom for pain management, 1 for anxiety/insomnia, 1 for concentration and focus, and 1 patient's reason for use was unclear. For maintenance, 1 patient was started on naltrexone, and 5 were started on buprenorphine at the following doses: 8 mg eventually tapered to 2 mg prior to pregnancy, 16 mg, 6 mg (2 patients), and 4 mg daily. The other patient was on buprenorphine initially; however, due to chronic pain, he eventually was switched to methadone. See Tables 1 and 2 and Figure 1 for a summary.

Survey

Eighty-two ASAM members completed the survey, and 69 qualified for study inclusion based on their credentials (physicians only). A total of 57 (82.6%) endorsed having encountered patients with KUD, including 19 (27.5%) who had patients with KUD only—no past or comorbid OUD (Figure 2). In managing their abstinence, 17 used buprenorphine (17/19, 89.5%)—including 6 who combined it with talk therapy 1 used methadone, and 3 used naltrexone. Additionally, 1 respondent used buspirone in conjunction with therapy, and another used talk therapy only (Figure 3). (Some of the participant-reported outcomes are included in Appendix B.)

Statistical Analysis

A biostatistician analyzed 2 research questions: (1) Does the proportion of those with kratom addiction in isolation of comorbid OUD from the survey match that found through the literature review? and (2) Among those without comorbid OUD from the survey, does the profile of maintenance modalities match that from the literature review? To address these questions, the survey data was compared with the historical data via a 1-sample proportion test.

Out of the 69 qualifying participants who completed the survey, 57 encountered cases of KUD, including 19 (19/57, 33.3%) cases in isolation of comorbid OUD. This is contrasted to the 14 reports found in the literature, with 7 (7/14, 50%) in isolation of OUD comorbidity. In terms of the profile for maintenance modalities, 17 survey respondents (17/19, 89.5%) endorsed having used buprenorphine maintenance, compared to 6 (6/7, 85.7%) found in the literature. A 1-sample proportion test shows that the proportion in isolation of OUD from the survey is significantly different from the proportion of 0.50 found in the literature (95% CI, 0.22-0.47; $P = 0.02$). Given the small sample size of data and the fact that the upper limit of the confidence interval is close to 0.50, it is reasonable to believe that such a difference is not large. There is no significant difference between the profile of buprenorphine maintenance reported in the survey versus that found in the literatures (95% CI, 0.69-0.97; $P = 0.64$).

DISCUSSION

Kratom is a botanical with a known addiction liability and, in vulnerable individuals, dependence may develop rather quickly with tolerance noted at 3 months and 4- to 10-fold dose escalations required within the first few weeks.[31] Kratom addiction carries a relapse risk as high as 78% to 89% at 3 months post-cessation.[7,8,32] Although there are numerous pathways that kratom's constituents act upon, the opioid pathway has received the most interest with respect to mediation of withdrawal and addiction.[33,34] This is consistent with the notion that stimulant effects are noted at low doses—5 grams or less daily, while opioid effects at higher doses and the doses used by those addicted to it indeed seem to range from 14 grams to 42 grams daily.[31] Unfortunately, most of the cases included in our review do not reference doses. In the 3 that do (all without comorbid OUD), 1 describes an individual using 7 grams every 4 hours, and 2 involve doses of 30 grams daily. One of the experts surveyed also mentioned having managed patients with histories of 30 grams daily use.

There are 2 main pathways describing how individuals are introduced to kratom – opioid substitution by those with OUD[35,36] and self-management of various ailments (ie, anxiety and mood states, pain) by those without OUD. The cases included in this review corroborate this notion. For patients with OUD, relapse rates without MOUD are in the 90% range[37-39]–similar to relapse

Table 2. Cases Reporting Maintenance Pharmacotherapy of Patients With Kratom Use Disorder Without Co-occurring Opiod Use Disorder

Ref No.	Clinical Paradigm	Reason for Kratom Use	Extent of Kratom Used	Intervention	Maintenance Regimen	Outcome
22	32-year-old woman at 22 weeks gestation presented to specialty clinic for pregnant women with substance use disorders. Had previously undergone radiation for Hodgkin's lymphoma, resulting in chronic shoulder pain and anxiety. Managed on oxycodone until previous pregnancy, but had been self-managing with kratom for previous 7 months. Attempted to stop kratom at 16 weeks gestation but resumed due to withdrawal.	Pain management, anxiety	7 months of use; unknown dose, duration, frequency, and route of administration.	After kratom abstinence period, patient started on BUP as outpatient; reported good results with 8 mg/day. Given concern of neonatal abstinence syndrome, tapered off BUP over 2 weeks but experienced severe depression and was restarted and maintained on 2 mg for remainder of pregnancy.	BUP 2 mg during pregnancy	Upon planned C-section at 39 weeks gestation, patient maintained on BUP; abstinence maintained at follow-up visits.
23	60-year-old woman with chronic pain and history of alcohol dependence in sustained remission presented following unintentional overdose on illicit methadone. No history of OUD; endorsed kratom use and was on a long-term opioid regimen with tramadol and oxycodone with no evidence of misuse. Discharged following admission and stabilization, but presented several months later because of difficulty stopping kratom due to rebound pain and withdrawal symptoms.	Pain management	At time of evaluation, 0.25 ounces every 4 hours; purchased via online retailer.	Outpatient induction to BUP/NX performed; patient then transitioned to 4-1 mg 4 x/day maintenance.	BUP/NX 4-1 mg 4x/day	Abstinence maintained at 9-month follow-up; confirmed by urine toxicology.
26	37-year-old woman with history of postpartum depression and 2-year history of kratom use to self-manage pain stemming from fibromyalgia and after surgery for carpal tunnel syndrome. Experienced withdrawal symptoms when trying to cut back; attempted outpatient detox with low-dose clonidine without success. Contacted mental health and addiction service for inpatient kratom detox; ultimately admitted for inpatient detox.	Pain management	Started using unknown amount of kratom capsules; transitioned to using kratom extract purchased from online retailer over 2 years.	As inpatient, treated with symptom-triggered clonidine protocol and supportive medications for 3 days prior to discharge.	Naltrexone 50 mg/day	Patient discharged to partial hospitalization program and instructed to start oral naltrexone on day 7 post-discharge.
27	20-year-old man with history of ADHD (treated with stimulant) presented to office-based addiction treatment clinic for KUD management. Had used kratom past 2 years to manage anxiety and insomnia but developed tolerance. Cessation attempts led to opioid-like withdrawal.	Anxiety, insomnia	2 years of use; increased gradually to every 2 hours for 30 g total daily dose. Obtained from local gas station and mixed with water into tea.	Outpatient induction to BUP/NX performed, starting with 4-1 mg 12 hours after last kratom use and with moderate withdrawal. Attempt to taper to 2-0.5 mg over 4 days resulted in withdrawal symptoms and dose was brought back up.	BUP-NX 4-1 mg daily	Noted difficulty tapering off BUP/NX with supervision. After 3 months treatment, had 1 setback on kratom when out of BUP/NX. Has maintained sobriety after several months, working to taper off BUP/NX.
28	35-year-old male veteran presented to addiction treatment clinic reporting escalating kratom use over past 3 years. Started using kratom for concentration but use gradually increased and became singular focus over work, school, and personal activity. Was able to reduce from 30g daily to 5g/day following motivational interviewing, but experienced withdrawal.	Focus, concentration	Daily use increased from 10 g/day initially to 30 g/day. First obtained from gas station; consumed in smoothie or shake form.	Outpatient induction to BUP/NX performed, 4-1 mg 2x/day.	BUP/NX 8-2 mg/day for 16 months, then decreased to 6-1.5 mg/day	BUP/NX increased to 12-3 mg to target evening cravings; decreased back to 8-2 mg/day due to sedation. Maintained abstinence at 16 months, corroborated by urine toxicology screens for mitragynine. After 16 months, BUP/NX dose decreased to 6-1.5 mg/day, with goal of tapering off over 1 year.
29	24-year-old man with history of alcohol use disorder, Asperger's, and kratom use presented to ED after being found down, minimally responsive, hypothermic, and having a witnessed seizure by emergency medical personnel. Upon stabilization in ICU, was transferred to inpatient psychiatric unit.		Unclear duration, but was using 600 mg/day prior to presentation.	BUP 2 mg started on hospital day 13 on psychiatric ward to target kratom cravings. On day 25, BUP increased to 4 mg 2x/day due to persistent signs/symptoms of withdrawal. Discharged to a rehab center on day 28. BUP discontinued initially but restarted at 2-0.5 mg 3x/day due to withdrawal symptoms.	BUP/NX 2-0.5 mg 3x/day.	Tapered off BUP/NX after 45 days at rehab center and discharged home.

continued on next page

Table 2 continued. Cases Reporting Maintenance Pharmacotherapy of Patients With Kratom Use Disorder Without Co-occurring Opiod Use Disorder

Ref No.	Clinical Paradigm	Reason for Kratom Use	Extent of Kratom Used	Intervention	Maintenance Regimen	Outcome
30	44-year-old man with history of alcohol use disorder presented to detox unit for help stopping kratom. Began use after brief use of nonprescription oxycodone for chronic abdominal pain. Noted difficulty stopping after 1 year due to withdrawal.	Pain management	1 year of use. Initally used a "tincture" dosed by "dropper squeeze;" gradually increased to "6 dropper squeezes" every 4-6 hours.	Inpatient induction to BUP to help with withdrawal.		At 15 months post discharge revealed use of oral opiates, including methadone and oxycodone, for chronic pain syndrome.

Abbreviations: BUP/NX, buprenorphine/naloxone; OUD, opioid use disorder; detox, detoxification; ADHD, attention deficit hyperactivity disorder; ED, emergency department.

rates for KUD—versus less than 50% when MOUD are implemented.[7,8,32] Hence, for those with both OUD and KUD, it is logical to utilize MOUD. In all such cases reported above, buprenorphine was used with good results in terms of opioid and kratom abstinence.

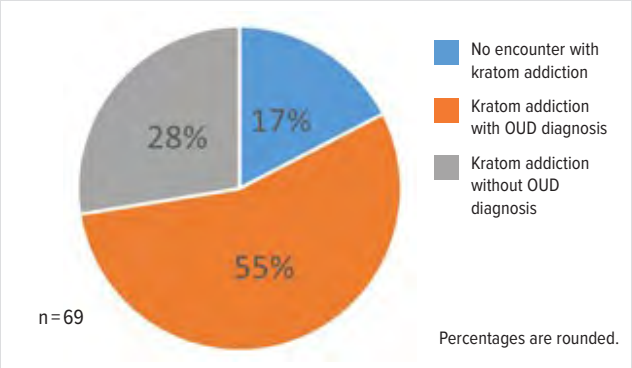
There is a clear need to establish a consensus on how to manage KUD independent of an OUD. As demonstrated in this review, there has been success with treating KUD using the same pharmacological agents as those approved for OUD. In the cases included here that did not involve a comorbid OUD diagnosis, clinicians have utilized naltrexone (n=1 case) and buprenorphine for maintenance. The use of MOUD to treat KUD has been hindered historically by the medicolegal aspects governing these agents, yet reports of treatment do exist and are corroborated by results of the survey conducted as part of this review.

There is pharmacodynamic evidence to suggest for those with OUD, ~70% mu receptor occupancy is required to achieve suppression of psychological aspects of opioid addiction.[40] Depending on the severity of one's OUD, for example high dose and intravenous use, upwards of 90% occupancy may be required.[41] Although the first may be achieved with 2-3 ng/mL plasma concentration of buprenorphine (corresponding with 8-16 mg oral dose), the latter would require 5-6 ng/mL (corresponding to 20-32 mg oral dose).[41] It is still uncertain what the opioid receptor dynamic with MG and 7-HMG is, however, it is believed that—at least for MG—it is very similar to buprenorphine.[12,13] From the cases included here, it appears that lower buprenorphine doses tend to be required for KUD in absence of OUD. Antagonist treatment has even been used in 1 case.

Limitations

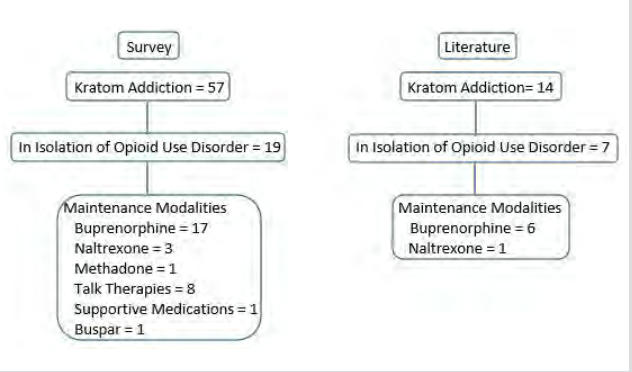
The cases resulting from the literature search and included in the analysis/comparison have a significant amount of heterogeneity in the descriptions, information provided (ie, kratom dose, route, etc), toxicology screens used for abstinence monitoring, reporting of maintenance follow-up duration, etc. Nonetheless, they all used buprenorphine or naltrexone for management of long-term abstinence as a general consensus.

Figure 2. Percentage of Survey Participants Who Have Encountered Any Kratom Addiction



Abbreviation: OUD, opioid use disorder

Figure 3. Pharmacological Modalities for Managing Kratom Use Disorder When Found in Isolation of Opioid Use Disorder



CONCLUSION

Through our survey, we assessed clinical practice patterns for management of KUD without the confounding OUD diagnosis, which would be a clear indication MOUD—the standard of care. A substantial number of respondents (82.6%) have encountered cases of KUD, of which the majority involved a comorbid OUD diagnosis. Those who endorsed treating cases of kratom addiction that did not involve a comorbid OUD reported having used primarily buprenorphine (89.5%) to manage abstinence, with the

rest using naltrexone and methadone. Based on some of the comments in Appendix B, the outcomes have been good and, like with OUD, counseling alone is not sufficient.

Together, the literature review and survey data suggest that a standard of care for maintenance of abstinence from kratom use in those with KUD hints towards the use of MOUD. This is especially true for individuals with histories of using in excess of 24 grams of kratom daily. The maintenance buprenorphine doses seem to be lower than those needed for OUD.

In light of the detrimental risks associated with growing reports of kratom use disorder and lack of any randomized controlled trials to explore treatment, this review provides sufficient evidence that the indication of MOUD should be extended to KUD as well. This is especially true if one's use of kratom involves high doses and meets DSM-5 diagnostic criteria for a moderate or severe substance use disorder.

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Current Trends in HPV Vaccine Uptake: Wisconsin and United States, 2016-2019

Alexandria Cull Weatherer, MPH; Stephanie L. Pritzl, MD; Sarah Kerch, MPH; Zhanhai Li, PhD; Noelle K. LoConte, MD

ABSTRACT

Background: Human papillomavirus (HPV) is a recognized cause of cancer in both males and females. HPV vaccination prevents development of HPV-associated diseases.

Methods: Wisconsin HPV vaccination rates (2016-2019) were obtained from the Wisconsin Immunization Registry. Data was stratified by age, sex, Medicaid status, race/ethnicity, and ZIP code. Wisconsin vaccination rates were compared with national trends using data from the 2016, 2018, and 2019 National Immunization Survey-Teen.

Results: Wisconsin HPV vaccination rates remain consistently below national averages. HPV vaccination rates are improving—especially among males; however, vaccine coverage at the recommended age of 11-12 remains low. Rates of vaccine uptake differ by race/ethnicity, rurality/urbanicity, and Medicaid status.

Conclusion: Further initiatives are needed to increase awareness and acceptance of HPV vaccination for cancer prevention throughout Wisconsin.

BACKGROUND

Persistent infection with high-risk human papillomavirus (HPV) genotypes is associated with cancer in men and women, including cervical, oropharyngeal, anal, vaginal, vulvar, and penile cancers.[1] Approximately 34,800 HPV-attributable cancers are diagnosed annually in the United States, with 600 reported each year in Wisconsin.[2] The HPV vaccine prevents new HPV infections and the development of HPV-associated diseases. The Advisory Committee on Immunization Practices (ACIP) recommends all individuals receive the HPV vaccine at 11 or 12 years of age.[3] Vaccination can be given at age 9, and catch-up vaccination is recommended for all individuals not adequately vaccinated through age 26.[3] In adults ages 27-45, shared clinical decision-making is recommended to determine which individuals may benefit.[3] There is limited research comparing national HPV vaccination trends to those in Wisconsin. Here, we have identified and summarized national and statewide immunization rates from 2016 through 2019 and demographic variables that may result in differences in HPV vaccine initiation and completion.

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METHODS

State and county-level HPV vaccination initiation and completion data was obtained from the Wisconsin Immunization Registry (WIR), a robust immunization information system. Data were stratified by sex at birth, age group (11-12 vs 13-17 years of age), Medicaid status (ever vs never), racial/ethnic group, and urbanicity. Urbanicity was determined by rural-urban commuting area (RUCA) codes by ZIP code, which classify census tracts using population density, urbanization, and daily commuting. RUCA codes 1 through 3 were defined as urban and 4 through 10 as rural.

Wisconsin vaccination rates were compared to data from the 2016, 2018, and 2019 National Immunization Survey-Teen (NIS-Teen), a survey that uses a representative sample to estimate adolescent vaccination coverage.[4] Data prior to 2016 was omitted given the HPV vaccine schedule change in 2015, making it difficult to ascertain if the change in vaccination coverage was due to HPV vaccine practice or the change in schedule.

RESULTS

HPV Vaccination Uptake: Wisconsin vs National Trends

Vaccine uptake at the recommended age of 11-12 from 2016 through 2019 has been slow. In 2016, 11% of Wisconsin adolescents ages 11-12 completed the HPV vaccine series. The completion rate increased to 15% as of 2019. All counties reported <30% vaccine completion rates for adolescents ages 11-12 in 2019 (Figure 1A). Despite the ACIP recommendation for HPV vaccine initiation and completion at ages 11-12, national HPV vaccination completion is reported for adolescents ages 13-17. As of 2019, Wisconsin falls short of its 80% coverage goal, and only 15 of 72 (21%) Wisconsin counties reported vaccine completion rates of at least 50% for adolescents ages 13-17 (Figure 1B).

When comparing Wisconsin and national HPV vaccination rates, the state consistently remained below national rates (Figure 1C). In 2016, 34.0% of Wisconsin adolescents ages 13-17 completed the HPV vaccine series compared to national estimates of 43.4%.[5] In 2019, 45.6% of Wisconsin adolescents were up-to-date with the HPV vaccine series compared to national estimates of 54.2%.[6] Vaccination rates for males remain slightly below vaccination completion rates for females. However, HPV vaccination rates for males have improved consistently from 2016 through 2019, and this gender gap is beginning to close (Figure 1C).[5-7]

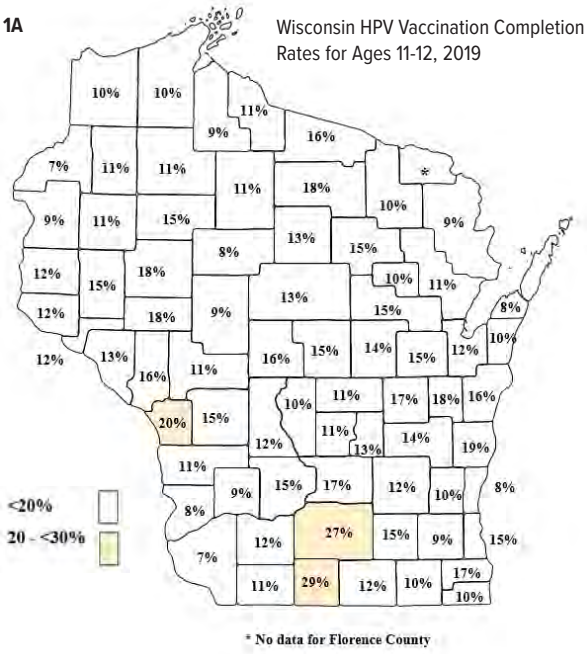
Differences in HPV Vaccination Rates: Rurality, Urbanicity, and Medicaid Status

HPV vaccination initiation and completion rates vary throughout Wisconsin. RUCA codes were used to evaluate differences in HPV vaccination by ZIP code based on rurality and urbanicity. Wisconsin adolescents in urban areas have statistically significant higher HPV vaccination initiation/completion rates compared to their rural peers ($P<0.0001$). In 2019, 46.8% of urban Wisconsin adolescents (ages 13-17) completed the HPV vaccine series compared to 42.7% of rural adolescents, and 60.4% of urban adolescents initiated the vaccine series versus 56.1% of rural adolescents (Figure 2A).

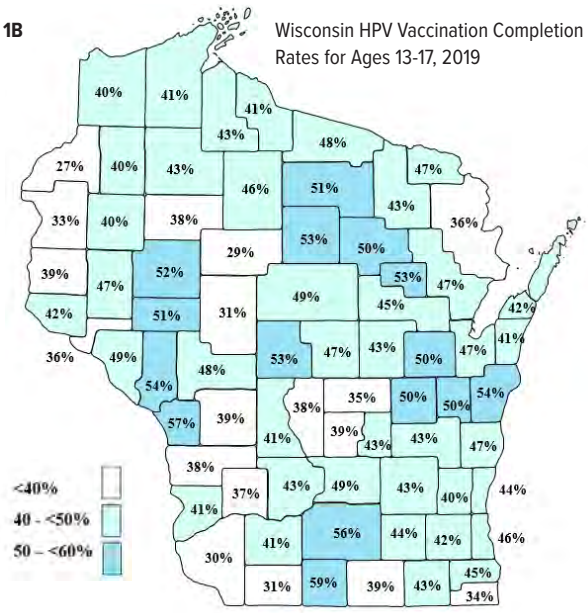
Similar trends are observed when reviewing national data. The NIS-Teen measures differences in vaccination status by rurality and urbanicity using metropolitan statistical areas (MSA). MSA status is defined by the US Census Bureau as having at least 1 urbanized area of at least 50,000 inhabitants. MSAs are grouped into 3 categories: MSA principal city (mostly urban areas), MSA non-principal city (mostly suburban areas), and non-MSA (mostly rural areas).[6] In 2016, HPV vaccine coverage was 14.8% lower for adolescents living in a non-MSA and 6.1% lower among those living in MSA non-principal cities compared to those living in MSA principal cities.[5] In 2019, HPV vaccine coverage was 9.8% lower for adolescents living in a non-MSA and 3.7% lower for adolescents living in MSA non-principal cities than those living in MSA principal cities (Figure 2B).[6] As in previous years, HPV up-to-date vaccination status continues to be lower among adolescents in a non-MSA; however, geographic disparities were statistically significant only for adolescents at or above the poverty level in 2019.[6]

Figure 1. HPV Vaccination Completion Rates

1A Wisconsin HPV Vaccination Completion Rates for Ages 11-12, 2019



1B Wisconsin HPV Vaccination Completion Rates for Ages 13-17, 2019



1C. HPV Vaccination Completion Rates, 2016-2019, Ages 13-17: United States vs Wisconsin

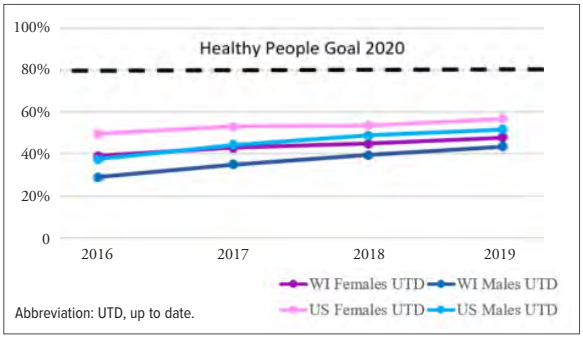
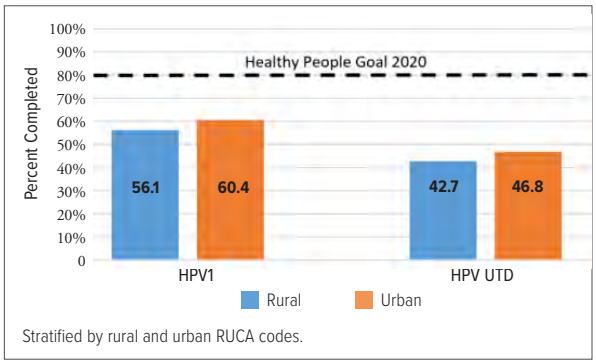


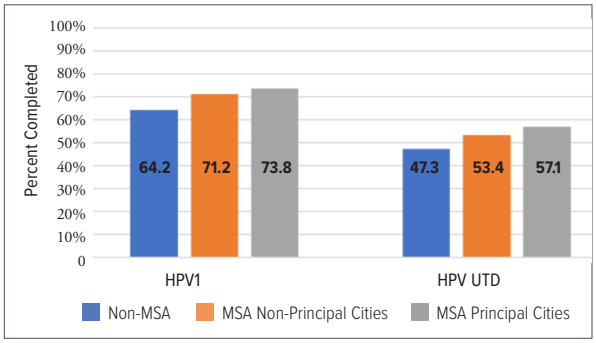
Figure 2. Vaccination Rates for Adolescents Ages 13-17

2A. 2019 HPV Vaccination Rates in Wisconsin: Rural vs Urban

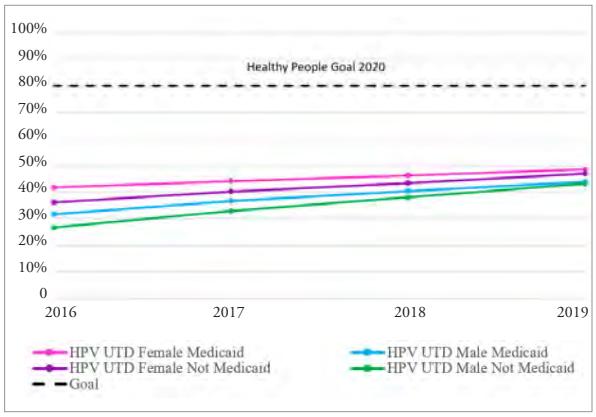


Stratified by rural and urban RUCA codes.

2B. 2019 HPV Vaccination Rates in the United States: Metropolitan Statistical Areas



2C. Wisconsin HPV Vaccination Completion Rates Stratified by Medicaid Status



Abbreviation: UTD, up to date.

HPV vaccination rates in Wisconsin vary by Medicaid status. Wisconsin adolescents (ages 13-17) who were ever on Medicaid have statistically significant higher rates of HPV vaccination initiation and completion compared with those who were never on Medicaid ($P < .0001$) (Figure 2C). While Medicaid status was not included in national estimates until 2017, the NIS-Teen in previous years demonstrated that those living below the federal poverty level had higher HPV vaccination coverage for both males and females compared with their peers living at or above the poverty level.[5] In 2019, US adolescents on Medicaid had statistically significant higher HPV vaccination coverage compared to those with private health insurance (4.4 and 6.0 percentage points higher for receipt of ≥1 dose and being up to date, respectively).[6]

Differences in HPV Vaccination Rates by Race and Ethnicity

HPV vaccination rates differ by race and ethnicity. In Wisconsin, Hispanics and non-Hispanic Blacks (ages 13-17) have higher HPV vaccine completion rates when compared to non-Hispanic Whites from 2016 through 2019 (Table). However, this gap is closing. Nationwide, non-Hispanic Whites have the lowest rates of HPV vaccine initiation and completion compared to all other racial and ethnicity groups.[5-7] In both 2016 and 2019, national HPV vaccine coverage was highest among Hispanic and Asian adolescents, both of which were statistically significant.[5-6]

DISCUSSION

Despite strong evidence supporting safety of the HPV vaccine and efficacy in cancer prevention, vaccination rates in Wisconsin and throughout the United States remain low. Wisconsin falls drastically short of the Healthy People goal of 80% vaccination by 2020 and the Wisconsin Cancer Plan 2020-2030 goal of 50% HPV vaccine completion.[8] Initiation and completion rates at the recommended age of 11-12 remain below targets. HPV vaccination at the recommended age is important for greater immunogenicity, administration before the onset of sexual activity and exposure to HPV, ability to bundle with other routine adolescent vaccines, and decreased dosing schedule (2 doses of HPV vaccine recommended if initiated before age 15 vs 3 doses if initiated at age 15 or later).[3,9] Barriers to HPV vaccination include limited understanding of HPV-associated diseases, safety concerns, discomfort discussing sexual behavior, missed clinical opportunities, and lack of a universal strong recommendation from health care providers.[9] While annual improvement in HPV vaccination initiation and completion have continued, the impact of COVID-19 will likely have an effect on routine vaccination uptake. Disease prevention measures, including routine vaccines and minimizing missed opportunities, are of utmost importance as the health care system continues to be burdened by the pandemic.

Barriers to HPV vaccination are multifactorial. A clear recommendation from health care providers is essential to increase HPV vaccine completion.[9] The prevalence of parents reporting receiving a recommendation for adolescent HPV vaccination varies, especially in rural areas.[7] A 2017 national survey found that the general public is now moderately aware of HPV, but awareness is higher among females and knowledge of noncervical HPV–related cancers remains low.[10] Further education is needed to raise awareness of other HPV-associated cancers, especially as oropharyngeal cancer in males continues to rise.[10]

Initiatives to improve access to HPV vaccination include addressing associated costs. Individuals ≤18 years of age who are Medicaid-eligible, uninsured, underinsured, or American Indian/

Alaska Native are eligible to receive vaccines through the Vaccines for Children (VFC) program.[7] The VFC program reduces barriers for low-income individuals. Both state- and national-level data demonstrate that adolescents on Medicaid have slightly higher HPV vaccination rates than those on private insurance. Coverage of the HPV vaccine under Medicaid and VFC programs improves access to vaccination services and should be continued and expanded when possible.[6,9] Socioeconomic status may be a moderating factor between HPV vaccination and rurality. Studies suggest that adolescents below the poverty level have higher HPV vaccination (possibly due to the VFC program), and those above the poverty level have lower HPV vaccination. More research is needed to explore this further. It is possible that lower HPV vaccine confidence in those above the poverty level may be a contributing factor, as these individuals have adequate insurance coverage to receive the vaccine.[6]

Table. 2016-2019 HPV Vaccination Completion Rates in Wisconsin for Adolescents Ages 13-17 Stratified by Racial and Ethnic Group

Racial/Ethnic Group	2016	*P* value	2017	*P* value	2018	*P* value	2019	*P* value
Females								
Hispanic	47.5%[a]	<.0001	50.2%[a]	<.0001	52.0%[a]	0.0002	54.9%	0.0524
Non-Hispanic Black	47%[a]	<.0001	49.2%[a]	<.0001	51.1%	0.2325	53.2%[a]	0.0113
Non-Hispanic White	44.0%		47.7%		50.7%		54.2%	
Other[b]	26.0%[a]	<.0001	28.0%[a]	<.0001	29.1%[a]	<.0001	31.0%[a]	<.0001
Unknown[c]	14.7%[a]	<.0001	16.0%[a]	<.0001	13.8%[a]	<.0001	14.9%[a]	<.0001
Males								
Hispanic	40.6%[a]	<.0001	45.4%[a]	<.0001	48.3%[a]	<.0001	51.7%[a]	<.0001
Non-Hispanic Black	39.4%[a]	<.0001	43.9%[a]	<.0001	47.1%[a]	<.0001	50.2%[a]	0.0024
Non-Hispanic White	31.8%		38.5%		43.7%		49.0%	
Other[b]	18.9%[a]	<.0001	21.9%[a]	<.0001	24.8%[a]	<.0001	27.4%[a]	<.0001
Unknown[c]	10.8%[a]	<.0001	13.0%[a]	<.0001	12.6%[a]	<.0001	14.1%[a]	<.0001

[a]Statistically significant difference (all $P < 0.05$) in estimated vaccination coverage by race/ethnicity; referent group was white, non-Hispanic adolescents.

[b]"Other" includes adolescents who identify as Asian, Native American and/or Multiracial.

[c]Unknown= racial data not captured in the WIR.

Limitations to our analysis include the use of 2 different data sources that are not directly comparable. National trends in HPV vaccination rates are measured for adolescents ages 13-17 rather than ages 11-12. Statewide data for the 11-12 age cohort help demonstrate the low rates of HPV vaccination completion. A small percentage of records are missing race/ethnicity data due to incomplete capture in the original electronic health record or incomplete file transfer.

CONCLUSION

While HPV vaccination rates continue to improve, further work is needed to promote HPV-attributable cancer prevention in males and females. A better understanding of how HPV vaccination coverage differs by age, race/ethnicity, rurality, and Medicaid status may help to further inform and identify strategies for improvement. Medical providers should universally and strongly recommend the HPV vaccine to every adolescent ages 11-12 for optimal protection. Additionally, vaccination can be given as young as age 9 and catch-up vaccination is recommended for all individuals not adequately vaccinated through age 26.[3] Vaccination status should be assessed at every medical encounter and offered when medically appropriate.

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Pediatrician Exposure to Neuromuscular Patients

Matthew Harmelink, MD; Erin Yale, MM

ABSTRACT

Background: Primary care providers (PCPs) provide general care to patients, including those who are followed by specialists. In the field of rare diseases, there is growing research that the primary care needs of these patients are unique to their individual disease state. The purpose of this study is to determine the prevalence of patients with pediatric neuromuscular diseases among a subset of pediatric practices in Southeastern Wisconsin.

Methods: A retrospective review of all patients with neuromuscular diseases seen at Children's Hospital of Wisconsin (CW) was conducted from January 1, 2016 through September 30, 2018. All patients who were seen by Children's Medical Group (CMG) providers were included, with a division of patients by provider.

Results: Eight hundred eleven (811) unique pediatric neuromuscular patients were identified; 188 patients were included in the study cohort. The median number of patients per provider was 2.5, mean number of patients was 2.68, and mode number of patients was 1.74; 51% of pediatricians within CMG did not care for a pediatric neuromuscular patient.

Discussion: The prevalence of patients with neuromuscular diseases followed by an individual CMG provider is low, with over half of the CMG providers not caring for any patients with neuromuscular diseases. Given the specific primary care knowledge needed to care for these patients, this suggests the need for a novel method of help support these providers.

BACKGROUND

The role of primary care providers (PCP) is to "provide definitive care to the undifferentiated patient at the point of first contact" and to refer to specialists for conditions that are beyond a PCP's scope of practice.[1] However, many PCPs continue to take "responsibility for providing the patient's comprehensive care."[1]

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While studies have shown that specialists contribute considerable time to primary care issues,[2] PCPs report delivering 88% of the primary care received by their patients.[3] For PCPs to provide general care alongside specialists, they need to have a basic understanding of the various conditions and diseases. The American Board of Pediatrics requires maintenance of certification, which includes continuing medical education credits for PCPs to stay up to date on current literature and practice guidelines. For patients with common conditions, PCPs can access and apply their knowledge and skills on a frequent basis so that they can maintain their competence in caring for those patients. However, this becomes more challenging for rare diseases, especially in fields that are rapidly changing.

One definition of a rare disease based on the Orphan Drug Act of 1993 is any disease or condition that affects less than 200,000 people in the United States.[4] Most pediatric neuromuscular diseases—diseases that affect the peripheral nerve, neuromuscular junction, muscle, or associated connective tissue—fall into this definition. While these diseases are rare, there are over 100 different types—all with unique pathology and treatment considerations. For example, the incidence of Duchenne muscular dystrophy is 1.51 to 2.05 per 10,000 boys and spinal muscular atrophy is 1 in 12,000 people.[5,6] Given this, our hypothesis is that pediatricians likely care for very few, if any, patients with neuromuscular diseases.

The purpose of this study is to evaluate the distribution of patients with neuromuscular diseases among a subset of pediatricians in Southeastern Wisconsin to assess the prevalence of patients with neuromuscular diseases per provider.

METHODS

Using the Insight Data Search Portal, a data query collected the name and primary care provider of all unique patients seen in the Pediatric Neuromuscular Clinic at Children's Wisconsin-Milwaukee campus from January 1, 2016 through September 30, 2018. The patients were selected if they were seen in the "CHW Neuromuscular" "Department" of the EPIC electronic health record (Epic Systems Corporation). This method of data inquiry captured all patients with neuromuscular diseases seen at this site while excluding those from the remote clinic in Neenah, Wisconsin. Only patients with completed visits were included; those who were scheduled but never seen in the clinic were excluded. All patient appointments in the symptom-specific "Hypotonia Clinic" within the Pediatric Neuromuscular Clinic were then excluded, as patients seen in that clinic are often diagnosed with non-neuromuscular diseases.

The number of patient visits and unique patients was collected in aggregate and by year or partial year for 2018. Each patient's PCP was identified, but patients were only included in the data analysis if their pediatricians were within the Children's Medical Group (CMG), where individual provider information was easily accessible. This data were analyzed using descriptive statistics.

RESULTS

A total of 1,790 patient encounters occurred within the time frame of this study for 811 unique patients. Of these, 188 patients had an identified pediatrician within the CMG. These patients were seen by 70 different pediatricians in 22 different practice locations (Table). The median number of patients per provider was 2.5, mean number of patients was 2.68, and mode number of patients was 1. Of the 144 pediatricians who comprised CMG, 74 (51.3%) did not have a pediatric neuromuscular patient in their patient panel. The highest number of patients seen by an individual pediatrician was 7.

Table. Prevalence of Pediatric Neuromuscular (NM) Patients in a Pediatrician's Practice

	n (%)
Number of neuromuscular patients	811
Number of pediatricians	144
Number of pediatricians with a NM patient in their practice	70 (49%)
Number of pediatricians without a NM patient in their practice	74 (51%)
Maximum number of NM patients in a pediatrician's practice	7
Median number of NM patients in a pediatrician's practice	2.5
Mean number of NM patients in a pediatrician's practice	2.68
Mode number of NM patients in a pediatrician's practice	1

DISCUSSION

This study demonstrates that the prevalence of patients with neuromuscular diseases followed by an individual pediatrician is low, with most pediatricians having no identified patients under their care. Those who did typically cared for only 1 patient. This suggests there is not a clustering of patients to certain providers.

These results confirm that for pediatric neuromuscular diseases, pediatricians do not have a high frequency of exposure and experience. And with a field rapidly changing with new diagnostic tests, new treatments, and ongoing clinical trials, a pediatrician cannot be expected to be maintain up-to-date knowledge of the changing recommendations. For example, with Duchenne's muscular dystrophy, the disease-specific standard growth curves published in 2013 are rapidly becoming outdated due to changing treatment algorithms and, thus, no longer accurately reflect the disease-specific percentiles of weights and lengths.[7]

There is literature indicating that better coordinated care can improve outcomes, such as emergency hospital admissions,[8] therefore, maintaining an up-to-date understanding of these diseases is important. Given limited specialists able to education pediatricians, and using results from this study, we addressed this need by developing a neuromuscular pediatrician who specializes in the general pediatric aspects of care for patients with neuromuscular diseases. The neuromuscular pediatrician role is similar to that of a complex care specialist: to act as a resource and answer questions from community providers, provide care for issues between the scopes of practice of PCPs and specialists, and to help manage general pediatric issues during routine multidisciplinary clinic visits.[9] Additionally, the neuromuscular pediatrician can assist with care during hospitalizations and help to coordinate transitions of care after discharge.

This study is limited due to the data collection method. Patients who are not seen at CW-Milwaukee campus were excluded from this study, although the excluded number is small. Analysis also was limited to pediatricians within CMG, which does not include all PCPs in Southeastern Wisconsin. Of note, a benefit of our methodology, compared to using ICD-10 or ICD-9 codes, is that the data was not affected by potential errors in diagnostic coding. As well, we were able to automatically exclude any patient who was not seen by the neuromuscular practice but was seen for other reasons in the institution.

CONCLUSION

In Southeastern Wisconsin, the prevalence of patients with neuromuscular diseases for individual pediatricians within the Children's Medical Group is low. Resources need to be developed to better support PCPs and general care for patients with neuromuscular diseases. Developing a neuromuscular pediatrician role was the model that our neuromuscular program implemented. Next steps are to evaluate the efficacy and utilization of this role by patients and their PCPs.

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The Sudden Savant: A New Form of Extraordinary Abilities

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ABSTRACT

Introduction: Savant Syndrome previously has been characterized as either congenital or acquired. This report describes sudden savant syndrome in which neurotypical persons have the sudden emergence of savant skills without underlying disability or brain injury and without prior interest or ability in the newly emerged skill areas.

Case Presentation: Eleven cases are described in which savant abilities suddenly and unexpectedly surfaced in neurotypical persons with no special prior interest or ability in the new skills, accompanied by an obsessive interest with and compulsive need to display the new abilities. All participants completed an online survey to record their demographics and skill characteristics.

Discussion: The acquired savant, and now the sudden savant, raise questions about the dormant potential for such buried skills in everyone. The challenge is to be able to tap such latent abilities without head injury or other precipitating events.

Conclusion: This paper documents 11 cases of sudden savant syndrome, which is a new and additional form of savant abilities surfacing in neurotypical persons without developmental disabilities (such as autism) or head or other brain injury (acquired savant syndrome). It opens new paths of inquiry for exploration of extraordinary abilities perhaps within everyone.

INTRODUCTION

Savant syndrome is a rare condition, with only 319 recorded cases, in which persons possess unexpected and sometimes prodigious abilities in stark juxtaposition to underlying neurodevelopmental disability or other central nervous system (CNS) disorder.[1-3] These abilities are most commonly in music, art, mathematics, calendar calculating, language, or visual-spatial/mechanical abilities.[3]

There are 2 previously reported classifications of savant syndrome: congenital and acquired.[1,2] In the congenital form, the savant skill surfaces in childhood. This skill is always accompanied by an underlying developmental disability, often—but not always—autistic spectrum disorder. Acquired savant syndrome occurs in previously neurotypical individuals who suffer head injury, stroke, dementia, or other central nervous system (CNS) event or disorder.[1,2] After this event, savant skills surface unexpectedly, sometimes at a prodigious level. Both congenital and acquired savant syndrome are rare, with their documented case totals being 287 and 32, respectively.[3]

In this report, we propose a new or alternate form of savant syndrome: sudden savant syndrome, in which savant-like abilities surface unexpectedly, sometimes at a prodigious level, in neurotypical persons with no prior interest or ability in the newfound skill, and with no apparent cause, injury, or underlying disability. Neurotypical persons with no particular art, music, or mathematical interests or abilities, for example, report an unanticipated, sudden, spontaneous burst of newfound abilities accompanied by an epiphany-like understanding of the "rules" and intricacies of the particular areas of specialization. Here, we report the demographics and skill characteristics of 11 cases of sudden savant syndrome.

METHODS

Initial Contact

The participants in this study sent emails or letters to the Treffert Center, located in Fond du Lac, Wisconsin, describing their experience with sudden new savant-like abilities and asking for more information about savant syndrome, with which they were somewhat familiar. These initial contacts always included a self-

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Figure 1. Samples of artwork by "MF"

Illustration 1: "The Mayan"

Illustration 2: "Self-Portrait"

report of the newfound abilities, which occurred spontaneously and without apparent cause. A follow-up email from the Treffert Center requested more information and, if the experience fit the description of sudden savant syndrome, the potential participant was asked if they wished to participate in a formal survey in order to receive more standardized and consistent information.

Sudden Savant Syndrome Survey

Secondary contact was made with the subjects to ensure the information within their initial email was accurate, as well as to create a route to send the Sudden Savant Syndrome survey. The survey was created to obtain current, comprehensive information regarding each subject's name, age, sex, location, race, ability, initial point of skill realization, follow-up course of abilities and other pertinent information. Thirteen consenting subjects were sent the survey; 8 of the 13 initial participants (61.5%) completed the entire survey.

SurveyMonkey (www.SurveyMonkey.com), an online survey tool, was used to develop, send, and receive the survey.[4,5] The survey consisted of 34 questions and was developed to be a comprehensive investigation with the purpose of elucidating the etiology of sudden savant syndrome. Following Institutional Review Board approval, informed consent was obtained electronically through the survey. Data obtained from the survey were analyzed using Microsoft Excel.

During the study period, DT published a blog post regarding the sudden savant titled "Brain Gain: a person can instantly blossom into a savant-and no one knows why" in the online magazine Scientific American.[6] This brought forth a large numbers of replies, some from suspected sudden savants. Subsequently, 3 sudden savants were added to the contact list and were prompted to complete the Sudden Savant survey, bringing to 11 the total number of sudden savants who completed the survey. MH, a case example described below, was not included as 1 of the 11 participants in this study due to the uniquely transient nature of their acquired abilities.

CASE EXAMPLES

MF

MF, a 43-year-old woman, woke up one night in December 2016 with what she called "the urgent need to draw a multitude of triangles, multiple geometric and triangular formations, which quickly evolved to a web of complex abstract designs." She recalled that she "stayed up into the morning with a compulsive need to draw, which continued over the next 3 days at an intense level." She had no prior interest or training in art. By the third day, she was working on a piece she named "The Mayan," which took her 2 weeks to complete (Illustration 1). Three months later she had created 15 pieces, with styles reminiscent of artists including Frida Khalo and Picasso. She currently spends about 8 hours per day on her artwork, in addition to working as a real estate agent. Incorporated into most of her pieces is a mandalic style of which she was totally unaware prior to her sudden art ability. "My art style is an ever-growing perpetual vision which leads me to create mandalic style art, sacred geometry, and mysterious creature beings all incorporated into art. Ideas are never-ending. It just flows so easily in my mind and on to paper." Illustration 2 is a self-portrait she created in 2017.

KA

KA, a 28-year-old man from Israel, provided a description of his epiphany moment. He was in a mall where there was a piano. Whereas he could play simple popular songs from rote memory before, "suddenly at age 28—after what I can best describe as a 'just getting it moment'—it all seemed so simple. I suddenly was playing like a well-educated pianist." His friends were astonished as he played and understood music in an entirely intricate way. "I suddenly realized what the major and minor scales were, what their chords were, and where to put my fingers in order to play certain parts of the scale. I was instantly able to recognize harmonies of the scales in songs I knew, as well as the ability to play melody by interval recognition." He began to search the internet for information on music theory and, to his amazement, "most of what they had to teach I already knew, which baffled me as to how I could know something I had never studied."

KA, an attorney, has a high IQ and has no history of any developmental disorder. He now makes part of his living doing musical performances. His epiphany is described in much more detail in the articles section of www.agnesian.com/page/savant-syndrome, as well as in *Islands of Genius: The Bountiful Mind of the Autistic, Acquired and Sudden Savant.*[2]

MH Temporary Sudden Savant

MH was a 31-year-old journalist stationed in West Germany in 1983 to 1985. While there, he attended some basic German language classes and picked up a rudimentary ability to speak or understand German using some basic phrases such as "please," "thank you," "how much," or "where is the train station?" When he tried to speak German, he said most people would ask him to use English since they were proficient in both languages. One evening, he met a middle-aged man in a bistro. MH tried speaking some German, such as giving his name, saying that he was in the Army and that he loved Germany. The acquaintance replied in German, but MH could not understand. At that point MH told the other man he had been in Germany 25 months. The man replied, "If you have been here 25 months, then you can speak German." At that point, MH said, "He had said that in an irrefutable declarative sentence and, much to my amazement, I discovered he was right, Suddenly, for the first time ever, I understood everything he said in German and I effortlessly replied in fluid German. For the next 15 to 20 minutes, we had a nonstop conversation—I understood and spoke German perfectly." But that ability was very short-lived. "Then came the moment when I became aware of the strangeness of the phenomenon and I tried to analyze the what and why of things. As soon as my thoughts changed from subjective to objective, poof, my newfound ability vanished and has not returned."

Then, at age 38, MH was a civilian doing some outreach work for the Mormon church. He visited a family one afternoon. There was a piano there and he sat down and played "little, very little." Growing up, MH said he played a piano in the basement that he "dinked around on," knowing only a few chords. The mother of the family he was visiting asked if he played the piano and he replied "no, not really." But then, "suddenly my mind opened up as it had in Germany. I began playing elaborately and spontaneously notes and chords—beautiful music, whatever it was. I had never heard it before, but it was beautiful, flawlessly performed. Not once did I hit an off or sour note. My sudden and inexplicable musical ability lasted 15 to 20 minutes." Just as with the German transient language ability, the musical ability vanished when he realized what he was doing and tried to analyze the "what and why of things." That musical genius disappeared as suddenly as it had appeared. MH said it "was gone forever and entirely. Yet my memory of playing the piano like a virtuoso is still quite clear and certain."

MH's experience is similar to that of foreign accent syndrome (FAS), which is described in *Islands of Genius*.[2] FAS is a rare but dramatic condition in which persons following head injury or other CNS incident begin to speak with the foreign accent or entire language of a country they may have never visited. Most pertinent here is the case of a Czech racecar driver who was knocked unconscious in a speedway accident. When he regained consciousness, although he knew only the most basic English phrases, he was conversing fluently in English with the paramedics attending him. After a full recovery, however, the fluency disappeared to where it was very difficult for him to make himself understood in English and he reverted to his native language. In most persons with FAS, however, the change in language or accent remains and is not transient.

RESULTS

The 11 participants who experienced sudden skill realization provided the following information via correspondence and the sudden savant survey.

Demographics

Most of the survey respondents currently reside in the United States (n = 7, 63.6%); others reside in Finland, Israel, Canada, or Japan (Table). Four of 11 respondents were male (36.4%), 7 were female (63.6%; Table). Participants had completed various levels of education before and after skill realization. Before skill realization, distribution was as follows: 5 high school degrees (45.5%), 1 associate degree (9.1%), 3 bachelor's degrees (27.3%), 1 master's degree (9.1%), and 1 doctoral degree (9.1%). Following skill realization, the education levels differed only with 1 high school degree participant earning an associate degree, changing the post-realization distribution to 4 high school degrees (36.4%) and 2 associate degrees (18.2%).

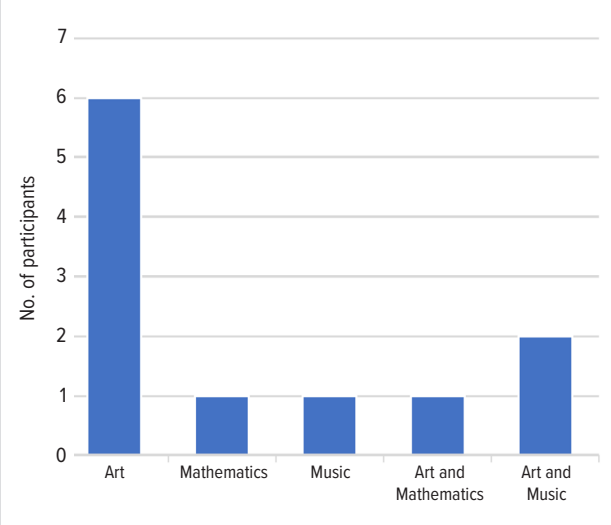
Abilities

The most prominent savant-like ability elucidated in our study was art (n = 9, 81.8%): 6 participants possessed savant-like abilities solely in art, 2 participants possessed abilities in art and music,

Table. Demographics and Skill Characteristics

Variables	N (%)
Sex	
Female	7 (63.6)
Male	4 (36.4)
Country of residence	
Finland	1 (9.1)
Israel	1 (9.1)
Japan	1 (9.1)
Canada	1 (9.1)
United States	7 (63.6)
Compulsion	
Yes	7 (63.6)
No	4 (36.4)
Familial history of acquired skill	
Yes	2 (18.2)
No	9 (81.8)
Change in ability over time	
Increase	10 (90.9)
Decrease	0 (0.0)
Neither	1 (9.1)
Synesthesia	
Yes	1 (9.1)
No	10 (90.9)

Figure 2. Quantity of Participants and Their Savant-like Skill Category/ Categories



and 1 participant possessed abilities in art and math. Within this subgroup, participants rapidly discovered abilities in drawing, painting, and sculpting. Two participants discovered mathematical abilities—1 specifically in calendar calculation and 1 with additional abilities in art. Three participants discovered musical abilities—1 in piano and guitar, while the other 2 possessed additional abilities in art. (See Figure 2.)

Modes of Skill Realization

The survey included a section for respondents to list any coincidental or collateral circumstances they believed may have accompanied the skill realization. Six of 11 respondents (54.5%) mentioned such a circumstance, which included spasmodic dysphonia, menopause, fever, sepsis, early onset dementia and pernicious anemia, traumatic stress, and prior head trauma. Two respondents had mentioned menopause as possibly playing a role in the development of their skill, as did 2 respondents regarding spasmodic dysphonia. The participant who reported prior head trauma indicated that the event occurred 14 years prior to skill realization.

Age at Skill Realization

The age of skill realization ranged from 14.9 to 79.8 years old. The mean was 44.6 years, with a standard deviation of 15.9 years. Outside the interquartile range, there were 3 outliers: 2 who had skill realizations at 19.0 and 26.8 years of age, respectively, and 1 who had skill realization at 79.8 years.

Skill Realization Timeframe

All participants experienced a sudden bout of skill realization and compulsive skill development. The timeframe for sudden compulsion toward their newfound ability was described as less than a week by 5 respondents (45.5%), 1 to 2 weeks by 2 respondents (18.2%), 1 to 3 months by 3 respondents (27.3%), and over a year by 1 respondent (9.1%). The participant with skill realization "over a year" stated that "the artistic skill acquisition immediately followed a musical skill acquisition" within this timeframe.

Time Spent on the New Skill and Compulsivity

Hours spent on the new skill varied from zero hours daily (n=3, 27%) to 1 hour (n=1, 9%); 1 to 3 hours (n=1, 9%); 3 to 8 hours (n=2, 18%); 8 hours per day (n=4, 37%). The time spent performing the ability after skill realization and control was rather polarized. Five respondents (45.5%) reported spending less than 3 hours per day performing compulsion(s) with much control; whereas 6 respondents (54.5%) reported spending more than 3 hours per day performing compulsion(s) with moderate control.

Familial History of Savant Skill

Two of 11 respondents (18.2%) reported that a known family member held some level of skill in the participant's particular savant skill (Table). One participant with a familial history of the skill wrote that both of their parents had "innate artistic abilities" and one of their brothers "was artistic from an early age" and "later went to commercial art school but quit drawing soon after and never picked it up again." The final participant who reported familial skill stated that one of their parents "paints, but not with the accuracy or detail that I seem to have."

Prior Interest in Savant Skill

Three of 11 respondents (27.3%) reported interest in the acquired skill prior to skill realization. One of the 3 participants with prior interest in their musical skill described themself as a "bonfire singer" with "crude piano skills" prior to skill realization. Another

participant described their savant skill as their main hobby prior to skill realization, where their abilities rapidly and compulsively advanced from drawing "basic line drawings, cartoons, and such" to "every fiber…every single bit of fur and change of hue" in a picture of a dog.

Change in Savant Skill Proficiency Over Time

Ten of 11 respondents (91%) reported an increase in newly acquired skill over time (Table). One participant reported neither an increase nor decrease in skill ability over time, experiencing mild lessening of compulsivity in art and music abilities.

Change in Memory Over Time

Six of 11 respondents (54.5%) reported some change in memory following skill realization. One of the 6 participants acquired synesthesia following skill realization and said that tasting or touching "anything that reminds [them] of [their] dreams" would trigger the recall of previous memories related to that stimuli; the participant would "feel, taste, and experience the moments like [they are] in them again." Of the remaining 5 respondents, 2 experienced an increase in long-term memory ability, 2 experienced a decrease, and 1 experienced no change. A participant who experienced an increase in long-term memory recall and decrease in short-term memory recall stated, "[My] past had always been a huge blur to me…since [skill realization], I've been able to recall many of my childhood memories in detail." In addition, 4 participants reported a decrease in short-term memory, with 1 participant denoting no change in short-term memory.

DISCUSSION

In congenital savant syndrome, the extraordinary abilities surface most often in childhood and are superimposed—or grafted on—some underlying developmental disability. In acquired savant syndrome, there is a specific brain injury or other CNS event that precipitates the emergence of the new extraordinary skill or ability. In sudden savant syndrome, the extraordinary abilities surface suddenly without an apparent underlying disability or brain injury.

Many ordinary persons develop a new interest or hobby at various points in their life, often later in life. So, what differentiates sudden savant syndrome from those more ordinary life changes? Several things:

- The ability has an abrupt onset with no prior interest in or talent for the newly acquired ability.
- There is no obvious precipitating event, CNS injury, or disease.
- The new skills are coupled with a detailed, epiphany-type knowledge of the underlying rules of music, art, or math, for example—none of which the person had previously studied in detail. Sudden savants appear to know concepts without having previously learned them or suddenly gain a deeper understanding they had not had before.
- The skill is initially accompanied with an obsessive-compulsive component; there is the overpowering need to play music, draw, or compute. It is as much a force as a gift, as is usually the case with both congenital and acquired savant syndrome.
- There is a fear the gift and compulsion are evidence of losing one's mind and a tendency to hide the new ability from others rather than display it.

While the term "sudden savant" is used here, by definition savant syndrome always includes some underlying disability from autistic spectrum disorder or, in the case acquired savant syndrome, from some head injury or other CNS disorder. But in sudden savant syndrome, there is no underlying disability, so technically a better term for the abrupt emergence of the sudden extraordinary new ability might be "sudden genius." Genius is a term generally used for the presence of extraordinary, prodigious abilities without underlying disability.

The underlying question for each form of savant syndrome is whether such capacity and ability savants demonstrate might reside dormant in everyone. The challenge is how to tap those hidden abilities without injury in the case of acquired savants, or more often and easily in the case of the sudden savant.

CONCLUSIONS

This study reports the first observations and analysis of sudden savant syndrome. Neurotypical participants experienced sudden, rapid, and unprompted development of savant skills in the absence of brain injury, autistic spectrum disorder, or other developmental disabilities. This preliminary investigation using case reports of 11 individuals provides a glimpse into a previously unreported and uninvestigated condition. Further work is necessary to document the incidence, prevalence, and mechanisms of sudden savant syndrome, along with its implications for better understanding both brain function and human potential.

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Varicella Zoster Meningitis in Immunocompetent Hosts: A Case Series and Review of the Literature

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ABSTRACT

Meningitis caused by varicella zoster virus (VZV) infection is uncommon in immunocompetent patients. We report 3 cases of VZV meningitis with rash in immunocompetent adults from a single academic institution over a 1-year period. The low prevalence of VZV meningitis in this population is attributed to lack of early recognition or underreporting. We highlight the importance of considering VZV as a possible cause of meningitis even in previously healthy young individuals.

INTRODUCTION

Meningitis is characterized by inflammation of the layers of tissue encasing the brain and spinal cord and is primarily caused by viral infections. Varicella zoster virus (VZV) is one of the common causes of viral meningitis and is rare in otherwise healthy individuals.[1] Following a primary VZV infection, which is often asymptomatic, viral particles may remain latent in cranial nerves, dorsal roots, and autonomic ganglia and can reactivate when cellular immunity is compromised. Reactivation of VZV, known as herpes zoster (HZ), typically presents as painful vesicles in a dermatomal distribution that can be associated with a variety of complications.[2] Risk factors for reactivation include any condition that weakens cell-mediated immunity such as trauma, advanced age, malignancy, chronic kidney or lung disease, and immunosuppression.[3] Acute central nervous system infection or reactivation of VZV in young immunocompetent adults is unusual, and only a few cases have been reported in literature to date. This case series describes 2 immunocompetent men and 1 immunocompetent woman who had VZV meningitis associated with rash.

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CASE 1

A 22-year-old man with a past medical history significant for primary varicella as an infant and mononucleosis in 8th grade presented with headache, fever, photophobia, and painful vesicular rash over the scalp. Vital signs were within normal limits except for a low-grade fever of 100.7° F. Physical exam was significant for generalized anterior cervical lymphadenopathy and a 1.5 cm x 3 cm left-sided retro-auricular lymph node. Nuchal rigidity with pain was noted; Brudzinski's and Kernig's signs were negative. The computed tomography (CT) scan of the brain did not show any temporal lobe leptomeningeal enhancement. Cerebrospinal fluid (CSF) analysis was suggestive of possible viral etiology with leukocyte count of 6 cells/μL, 94% lymphocytes, protein of 50 mg/dL, and normal glucose of 60 mg/dL. Subsequent polymerase chain reaction (PCR) analysis of the CSF was negative for herpes simplex virus 1 and 2, cytomegalovirus, enterovirus, and VZV. A skin biopsy of the lesions ultimately returned positive for VZV. Acyclovir and supportive treatment were started due to concern for viral meningitis. By the third day of admission, the patient's pain, nausea, vomiting, photophobia, neck stiffness, and fever improved, and he was subsequently discharged on intravenous (IV) acyclovir 10 mg/kg q8hr for total of 14 days. Upon follow-up, he had improved significantly and reported mild residual headache.

CASE 2

A 29-year-old man with no past medical history presented to the hospital with a rash over the left side of his scalp, eye swelling, photophobia, headaches, and neck stiffness. He had previously presented to the same hospital a day earlier with a 1-day-old rash

spreading from the left temporal scalp and periorbital skin across to his nasal bridge (Figure), for which the ophthalmologic service diagnosed him with herpes zoster ophthalmicus without corneal or intraocular involvement and discharged him on valacyclovir 1g orally 3 times per day. He was later discovered to be unimmunized for varicella by the Wisconsin Immunization Registry, although his mother stated otherwise. The patient had a history of 2 male partners in the past 5 years but was not currently sexually active and denied any history of sexually transmitted infections. Upon presentation, his vitals were within normal limits and physical exam was remarkable for a confluent vesicular rash with lesions in various stages of healing with crusting, as described above. His left eyelid was edematous (Figure) and the left conjunctiva injected; extraocular muscles and pupils were uninvolved. Brudzinki's and Kernig's signs demonstrated characteristic neck stiffness; the Hutchinson's sign was negative, however. CSF analysis was suggestive of aseptic meningitis with leukocytes of 101/uL, monocytes 61%, glucose and protein within normal limits. Multiplex PCR CSF analysis for common infectious pathogens was positive for VZV. Serum HIV screen was negative. The patient was treated with IV acyclovir 10 mg/kg 8 hourly with complete resolution of symptoms during a week of inpatient admission, and he was discharged with another 7 days of oral valacyclovir.

Figure. Herpes Zoster Rash on Periorbital Skin and Nasal Bridge of Left Eye With Eyelid Edema

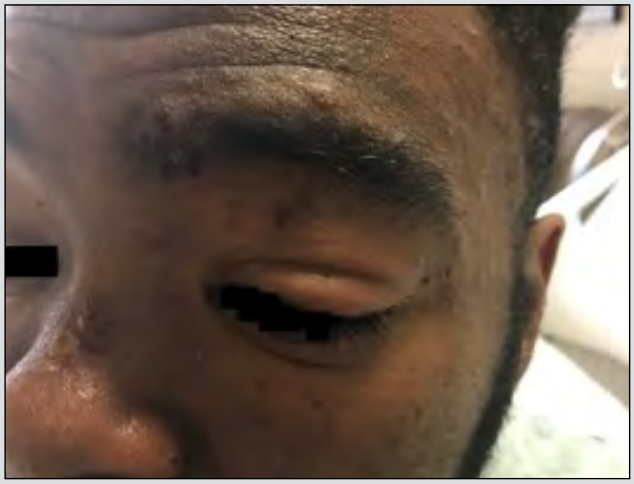

Photo consent obtained from patient. Parts of both eyes have been blackened to protect identity.

CASE 3

A 60-year-old woman with a past medical history significant for diabetes and schizoaffective disorder presented from a behavioral health facility with altered mental status and fever up to 102º F following a fall. Upon presentation, her vital signs were stable and initial lab workup was significant for a lactic acid of 2.8 mmol/L and a positive urinalysis. The CT scan of head was negative for acute changes. She initially was given ceftriaxone for a suspected urinary tract infection and was discharged the next day with a 5-day course of antibiotic. Two days following discharge, she presented to the emergency department with concerns from the facility staff for worsening lethargy and weakness. Physical exam was limited due to the patient's uncooperative state, but vesicles with an erythematous base and punctate healing lesions were noted over her right shoulder. CSF analysis revealed a white blood cell count of 106 cells/µL, 92% lymphocytes, 7% monocytes, protein of 71 mg dl, and glucose of 90 mg/dl. Positive VZV nucleic acid amplification test on CSF PCR analysis confirmed the diagnosis of VZV reactivation complicated by meningitis. She was started on 10 mg/kg IV acyclovir q8h. Once the lesions crusted, her weakness improved and she returned to her baseline mental state, her antiviral was transitioned to oral valacyclovir and she was discharged back to her mental health facility.

DISCUSSION

The incidence of HZ increases with age with an estimated 2.5 cases per 1,000 for the ages 21 to 50 years and 10.1 per 1,000 for those over 80 years.[4] While enteroviruses are the major causative agents, VZV accounts for 2.5% to 8% of cases of aseptic meningitis.[5,6] The exact pathophysiology of VZV meningitis is unknown. It is a rare disease that predominantly affects immunocompromised people, such as the elderly and solid organ transplant recipients. There are only a few reported cases of VZV meningitis in immunocompetent hosts (Table).[4,7-34] Both male sex and a craniocervical dermatomal distribution of the HZ rash are risk factors for the development of aseptic meningitis caused by VZV (30) as seen in 2 of our cases. However, in the majority of patients with VZV meningitis presenting with a dermatomal rash, the meningitis symptoms arose, on average, 6 days before the rash appeared, making rash an unreliable clinical finding to substantiate diagnosis as shown by a retrospective review of data over a 3-year period.[35]

Despite antivirals being the mainstay therapy for VZV meningitis, there is no published data supporting a change in the disease course with the use of antivirals such as acyclovir. One study found that patients receiving IV acyclovir remained hospitalized longer than those not receiving acyclovir; however, 3 of the 4 patients in this study were over 70 years old and other comorbidities likely contributed to their prolonged hospitalization.[35] Early initiation of antivirals may aid in preventing long term sequelae. Symptomatic management through hydration, antinausea medication, pain control, antipyretics, and anti-inflammatory medications remain the mainstay of treatment for uncomplicated cases of VZV meningitis in immunocompetent patients. The use of steroids in viral meningitis is controversial because of the resulting delayed immune response and clearance and is therefore reserved only for patients with evidence of increased intracranial pressure.[36] Further studies are needed to determine the efficacy and safety of concurrent steroids and antiviral therapy administration.

Table. Available Case Report/Series of Varicella Zoster Meningitis in Immunocompetent Patients, 1997-2018[a]

First Author, Year	Sex	Age (years)	Rash	Ref No.
Moriuchi, 1997	M	37	P	7
Hartzell, 2006	M	21	P	8
Pirounaki, 2007	M	27	P	9
Haargaard, 2008	F	64	P	10
Frantzidou, 2008	F, M	15, 72	NA	11
Mpaka, 2008	M	66	A	12
Leahy, 2008	M	14	A	13
Habib, 2009	F	26	A	14
Iyer, 2009	M	9	P	15
Pena, 2009	M	11	P	16
Spiegel, 2010	F	14	A	17
Klein, 2010	F	53	A	18
Han, 2011	M	7	P	19
Kangath, 2013	F	30	P	5
Mantero, 2013	F	17	A	20
Goyal, 2013	M	27	P	21
Esposito, 2013	M	14	P	22
El-Safadi, 2014	M	39	A	23
Pasedag, 2014	M	18	A	24
Sanguankeo, 2015	M	51	P	25
Abe, 2015	M	57	A	26
Ibrahim, 2015	F	15	A	27
Ganesan, 2016	F	70	P	28
Itoh, 2017	M, M	12, 12	P	29
Kim, 2017	16 M, 8 F	mean age 51.1	P	30
Gnoni, 2018	F	29	P	31
Suri, 2018	F	24	A	32
Spernovasilis, 2018	M	36	A	33
Khaliq, 2018	M	28	P	34

Abbreviations: Ref No., reference number; M, male; F, female; P, present; A, absent; NA, information not available.
[a]As found in the literature (PubMed-indexed).

This case series presents 2 young, immunocompetent men in their 20s and a 60-year-old immunocompetent woman who developed aseptic meningitis from the reactivation of HZ. Our first patient developed varicella in infancy, whereas the second patient is, to our knowledge, the first reported case of an unimmunized individual who developed VZV meningitis. The exact mechanism of illness in the second case is unknown; however, it illustrates an example where a patient was infected without the expected immunologic priming expected with either varicella vaccination or primary infection between the ages of 12 months and 6 years. Our third case demonstrates VZV meningitis in an immunocompetent patient with a HZ rash that started after meningitis symptoms began. This patient's immunization status and previous exposure could not be verified due to lack of records and cooperation by patient. All 3 cases recovered fully with resolution of symptoms and without focal neurological deficits.

In our first case, CSF VZV PCR was negative, but CSF analysis did support aseptic meningitis. Biopsy of rash is not performed routinely but, given the clinical presentation of meningitis and negative CSF PCR, we did obtain biopsy of scalp rash that supported our presumptive diagnosis of viral meningitis. Although PCR for VZV in the CSF is a useful confirmatory test with 95% specificity, it only has 30% sensitivity, which limits its reliability.[37,38] Based on our findings, we treated the patient appropriately with acyclovir and he had clinical improvement. This unusual presentation makes our case unique.

While these cases are not necessarily novel, the occurrence of 3 cases at our institution in immunocompetent patients suggests that the prevalence of VZV meningitis might be higher than what has been reported previously and highlights the variations in their clinical presentations. Whether VZV meningitis is simply underreported or misdiagnosed, there clearly exists a dearth of evidence in terms of management of VZV meningitis and further research in this critical area will fulfill this unmet need. While there is no concrete evidence that antivirals reduce mortality or changes disease outcomes in VZV meningitis, timely diagnosis is prudent to shorten length of hospital stay and prevent long-term sequelae associated with the disease.

CONCLUSION

Central nervous system involvement is a relatively uncommon complication following a VZV infection. Neurologic involvement is a life-threatening sequela and should be assessed early to facilitate prompt and appropriate treatment. VZV meningitis is a rare complication of varicella infection; the low prevalence reported likely suggests a lack of early recognition or reporting. The cases presented here highlight the need for more research to establish standard practice guidelines for the management of VZ meningitis.

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Platelet-Rich Plasma Injection for Quadriceps Tendinopathy: A Case Report

Ryan J. Wagner, DO; Karie N. Zach, MD

ABSTRACT

Introduction: Platelet-rich plasma (PRP) is a promising treatment for persistent symptoms caused by various musculoskeletal injuries, including tendinopathies.

Case Presentation: A 47-year-old woman presented with chronic left knee pain following a motor vehicle accident. Magnetic resonance imaging (MRI) revealed distal quadriceps tendinosis and a partial tear of the vastus intermedius tendon. She failed extensive conservative management. A PRP injection was performed at the distal quadriceps tendon. In combination with physical therapy, there was substantial improvement of symptoms and corresponding decreased tendinosis on follow-up MRI.

Discussion: Successful use of PRP has been documented in the literature for a variety of chronic orthopaedic conditions including, but not limited to, patellar tendinopathy, lateral epicondylosis, and gluteal tendinopathy. To our knowledge, this is the first reported case of the treatment of chronic quadriceps tendinopathy with a PRP injection.

Conclusion: This case expands the treatment options for chronic quadriceps tendinopathy and highlights another use of PRP within the field of regenerative orthopaedics.

INTRODUCTION

The advent of nonsurgical cellular therapy has altered the approach and treatment of common musculoskeletal pathologies, such as chronic tendinopathy and osteoarthritis. Conservative treatment with physical therapy, activity modification, anti-inflammatory medications, and rest remain beneficial and effective.[1] Despite compliance, some patients continue to experience symptoms, including pain, loss of range of motion, and limited function. Platelet-rich plasma (PRP) has emerged as an option to treat damaged tendon and cartilage, which are at times refractory to healing because of hypovascularity and various other factors.[2] Comprised of an increased platelet concentration, PRP contains an abundance of growth factors (ie, TGF-β1, PDGF, bFGF, VEGF, EGF, IGF-1) that, when introduced at the site of injury, complement the natural healing process.[3]

Current literature describes the efficacious use of PRP for lateral epicondylosis,[1-4] knee osteoarthritis,[3] patellar tendinopathy,[3] and other various tendinopathies; however, there remains an absence of reported use in quadriceps tendinopathy. We report what we believe is the first case of chronic distal quadriceps tendinopathy treated with an ultrasound-guided PRP injection in conjunction with a specific physical therapy program. There were clinical and radiographic improvements at 2 months and 19 months post-procedure, respectively.

CASE PRESENTATION

A 47-year-old woman was referred to our sports medicine clinic for consultation of chronic left knee pain. Her symptoms began 4 years prior from injuries sustained in a rollover motor vehicle crash. She reported persistent deep and aching generalized left knee pain. Ascending stairs and squats aggravated the pain. At its worst, she reported her knee pain at an 8 out of 10 on the visual analog scale (VAS). She denied any associated numbness or tingling in the leg. Initial treatment by an outside physician included analgesics, over-the-counter knee braces, and physical 4

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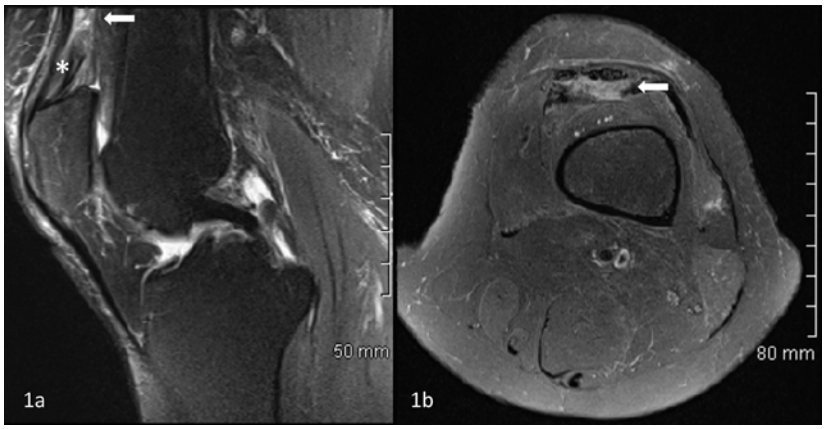
months totaling 31 visits. Upon review of the physical therapy notes, the patient was treated for both chronic knee and low back pain. She reported compliance with her home exercise program. On physical examination, inspection of her left knee revealed no joint effusion. She had no tenderness to palpation over the medial or lateral joint line or the popliteal fossa. Patellar compression and distal quadriceps palpation reproduced pain. All ligamentous testing revealed a firm endpoint. McMurray's test was negative. Manual muscle testing of the left lower extremity was significant for weak hip abduction, 4/5, and knee extension, 4+/5. No neurovascular deficits were noted. The contralateral knee exam was normal. The patient had a nonantalgic gait. Knee radiographs obtained were unremarkable for any significant degenerative joint narrowing.

Figure 1. Sagittal (a) and Axial (b) T2-Weighted Fat Saturated Magnetic Resonance Imaging of the Left Knee

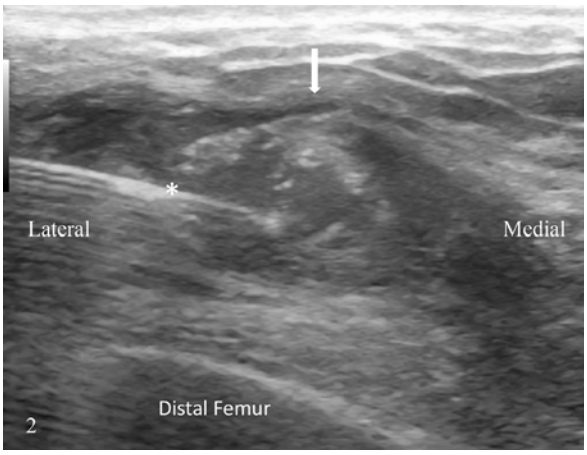


Images demonstrate thickening and hyperintense signal in the distal quadriceps tendon (asterisk). Vastus intermedius contribution of the tendon is torn approximately 2 cm from the patellar attachment (arrow).

Due to persistent pain and weakness despite extensive conservative care, magnetic resonance imaging (MRI) of the left knee was obtained. Increased T2 signal intensity and thickening throughout the distal portion of the quadriceps tendon was compatible with tendinosis. The deep portion of the quadriceps tendon (vastus intermedius) was partially torn 2.0 cm (0.02 m) proximal from the patellar attachment (Figure 1). The patella was positioned normally within the femoral groove. All ligamentous, menisci, and osseous structures were intact. Results were discussed with the patient, and physical therapy for 1 day per week with a home exercise program 5 days per week focusing on quadriceps and hip abductor strengthening was recommended.

The patient followed up in clinic 3 months later after completion of her physical therapy regimen. She reported a minimal improvement in symptoms with continued pain in the distal quadricep region, limiting her function. Physical examination remained consistent as described above. Treatment options were discussed with the patient, including continued physical therapy, PRP injection, and surgical consultation. She elected to proceed with a PRP injection and was instructed to hold anti-inflammatories for 7 days prior to the procedure. Whole blood (60 cc) was collected and centrifuged for 17 minutes using the Arthrex Angel system: spin 1 at 3500 rpm and spin 2 at 3000 rpm. Three cc of 1% lidocaine was used to anesthetize the local skin only. Under sterile technique and ultrasound guidance, 3 cc of leukocyte (neutrophil)-rich PRP (LR-PRP) (7% hematocrit) was injected into the pathological portion of the left distal quadriceps tendon (Figure 2). A tenotomy was performed with a 22-gauge needle in this area simultaneously. She was recommended to be non-weight bearing on the left lower extremity for the next 3 days and then slowly wean off the crutches as pain tolerated over the following week. She was instructed to avoid anti-inflammatory medications for 6 weeks. Physical therapy was started at 2 weeks with isometric quadriceps strengthening, followed by eccentric strengthening at 6 weeks with weekly physical therapy appointments.

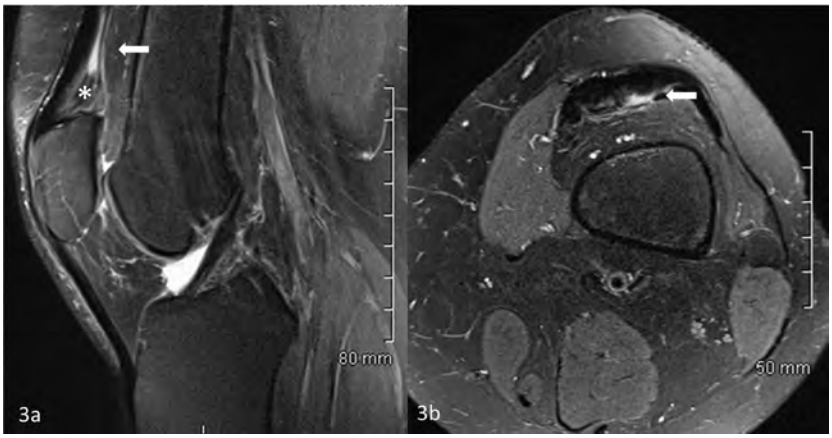
Figure 2. Transverse Plane Ultrasound Image of the Distal Quadriceps Tendon With a 22-Gauge Needle (asterisk) in the Vastus Intermedius Tendon (arrow)



Two months following the PRP injection, the patient presented for follow-up in the clinic. She reported a significant improvement of her knee pain—at worse a 2 out of 10 on the VAS. On exam, she had less pain with palpation of the distal quadriceps tendon. Her knee range of motion improved. Initial knee flexion was 130 degrees; post-procedure it was 140 degrees. She was advised to continue her home exercise program and follow up as needed.

The patient presented to the clinic 19 months after her PRP procedure with a new complaint of acute left knee pain following

Figure 3. Sagittal (a) and Axial (b) T2-Weighted Fat Saturated Magnetic Resonance Imaging of the Left Knee



Images demonstrate decreased thickening and hyperintense signal in the distal quadriceps tendon (asterisk) compared to prior imaging (Figure 1). The vastus intermedius tear (arrow) remains visible, but with a more chronic appearance compared to Figure 1.

a mechanical fall. Tenderness to palpation over the medial joint line with a positive McMurray's test was remarkable on physical examination. A knee MRI was obtained to evaluate for a medial meniscus tear. MRI imaging confirmed a nondisplaced, horizontal tear involving the posterior horn of the medial meniscus with an intrameniscal cyst of the left knee. Interestingly, in comparison to the left knee MRI done 21 months prior (as described above), there was marked improvement in the T2 signal of the MRI in the area of the quadriceps tendon insertion (Figure 3).

DISCUSSION

We describe the first known published case of PRP for quadriceps tendinosis, thereby expanding the scope of PRP as it relates to the field of orthopaedics. Musculoskeletal pathology involving ligaments and tendons can significantly affect an individual's quality of life. Both conservative management and surgical intervention have proven to provide relief; however, refractory pain and discomfort can endure.[4] The drive to bridge this gap has resulted in the exponential use of orthobiologics as a regenerative option for orthopaedic care. Prepared by centrifugation of whole blood, PRP is comprised of an autologous increased concentration of platelets containing growth factors and cytokines with the ability to regeneratively heal and decrease inflammation related to ulcers, burns, bone diseases, and tendinopathies.[2,4-6] Systematic reviews and meta-analyses have documented pain relief with PRP in lateral epicondylosis,[1-4] osteoarthritis of the knee,[3] patellar tendinopathy,[3] plantar fasciitis,[3] and rotator cuff injuries.[4]

Clinical evaluation of PRP has focused on the treatment of tendon injuries and tendinopathies. Healing of tendon and ligaments occurs through a complex series of stages involving inflammation, proliferation and tissue remodeling.[3,6] Numerous cytokines found in PRP play pivotal roles in the signaling pathways that occur during the healing process. PRP also may stimulate neovascularization, which may not only increase the nutrients and blood supply required for cells to regenerate damaged tissue, but also remove debris from injured tissue.[3,6] Such characteristics are particularly advantageous in chronic tendinopathies, where the biological conditions make tissue healing challenging.[3,6]

Despite its efficacy, there remains no general consensus regarding the optimal cellular composition of PRP preparations. Variability in collection protocols and concentration of blood components, including leukocyte content, has created challenges in interpretation of the literature.[6] Current literature suggests that PRP with a higher leukocyte (neutrophil) concentration—LR-PRP—is associated with pro-inflammatory effects.[3,6] Commercial systems are diverse in their platelet capture efficiency, speed of centrifugation and the type of collection tube mechanism.[3,6] Patient specific factors, such as medications taken, also influence the specific anatomy of PRP injectate.[3,6] Determination of the optimal PRP composition for tendinopathies remains a focus of current research.

The dominant extensor mechanism of the lower extremity—the quadriceps tendon—is composed of the superficial rectus femoris, the deep vastus intermedius, vastus medialis, and vastus lateralis. Though uncommon and generally occurring in patients older than 40 years, injuries to the quadriceps tendon typically illicit the triad of anterior knee pain, weakened active extension, and a suprapatellar gap.[7,8] Despite careful physical examination, quadriceps injuries are sometimes overlooked. Nonsurgical management is the mainstay of treatment for incomplete or partial quadriceps tendon ruptures.[9] Ilan et al recommend management of partial quadriceps tears with knee immobilization for 6 weeks in full extension, followed by range of motion and strengthening exercises.[9] Once the straight-leg raise is pain-free and the quadriceps muscle becomes more stable, the immobilizer can gradually be removed and the patient can return to training.[9] In comparison, our post-PRP protocol weans the patient from crutches at day 3 and initiates isometric quadriceps strengthening at week 2. Early movement, instead of prolonged immobilization, is advantageous for the patient, allowing for quicker rehabilitation and decreased atrophy and stiffness.

Our case focuses specifically on the vastus intermedius, which originates on the anterior femoral shaft and inserts on the superior border of the patella. Our literature review found only 3 cases describing an isolated vastus intermedius partial tear or rupture.[10-12] Two of the cases describe successful, nonsurgical treat-

ment with 4 weeks of immobilized, progressive graduated knee flexion followed by a stretching and proprioception therapy program in weeks 5 and 6.[10,11] A third case series outlines both conservative and operative management.[12] In contrast, our patient continued to report knee pain and weak knee extension despite bracing and focused physical therapy. Due to the persistent nature of her symptoms, PRP was recommended. Within 2 months following PRP, she reported significant clinical improvement, with noticeably increased knee extensor strength. Repeat MRI obtained 19 months after PRP injection revealed decreased tendinosis in the distal quadriceps tendon.

CONCLUSION

Within the past decade, cell-based therapies for orthopaedic conditions have increased considerably in the United States.[13] Incorporating these products into clinical practice requires a thorough understanding of the indications for their use.[4,14] While the results are encouraging, further studies, such as randomized controlled trials, are needed for PRP and its role in the treatment of chronic tendinopathy.

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Recurrent Stroke and Fatal Ruptured Mycotic Aneurysm Caused by Invasive *Aspergillus fumigatus* Infection

Istiaq Mian, MD; Sam Ives, MD; Garry Jean-Louis, MD; Andrew Laczniak, MD

ABSTRACT

Introduction: *Aspergillus* species are ubiquitous fungi that may cause invasive infection, particularly in immunocompromised patients. Invasive aspergillosis most commonly affects the lungs but can also disseminate to the central nervous system (CNS). Manifestations of CNS aspergillosis include abscesses and, rarely, mycotic aneurysm leading to subarachnoid hemorrhage (SAH).

Case Presentation: A 48-year-old man undergoing treatment for squamous cell cancer of the larynx with chemotherapy and steroids presented with dysarthria and weakness. He was found to have both lung and CNS infection secondary to *Aspergillus* species. While receiving intravenous antifungal treatment after biopsy-proven *Aspergillus* infection, he developed a fatal SAH caused by a mycotic aneurysm.

Discussion: Intracranial mycotic aneurysms are uncommon. However, mycotic aneurysm leading to a fatal SAH is a well-documented sequela of CNS aspergillosis. Mortality rates for CNS aspergillosis are extremely high.

Conclusion: In immunosuppressed patients with neutropenia or using chronic steroids who have concurrent pulmonary and CNS infection, there should be a low threshold to treat empirically for fungal infections prior to confirmation of diagnosis.

INTRODUCTION

Aspergillus spores are regularly inhaled from soil or decaying vegetation. While *Aspergillus* infections occur throughout the United States, Wisconsin has among the highest number of overall cases.[1] In an immunocompetent host, these spores rarely cause infection, but in patients with neutropenia or other immunosuppression, lung infection can occur. Dissemination via bloodstream can affect virtually any organ, including the skin, eyes, liver, kidneys, bone, and brain.[2]

CASE REPORT

A 48-year-old man presented to the hospital after coworkers noticed dysarthria with left-sided weakness. He was reportedly driving on the wrong side of the road the previous day. His past medical history was notable for head and neck squamous cell cancer, and he had completed 3 weeks of cisplatin and radiation treatment. He was taking oral dexamethasone daily for soft tissue facial edema and was on trimethoprim/sulfamethoxazole for *Pneumocystis* prophylaxis. He had been discharged from another hospital 3 days prior to presentation with *Pseudomonas aeruginosa* pneumonia, diagnosed via sputum culture and chest x-ray.

Physical exam was notable for an ill-appearing drowsy male with mild left-sided weakness. Computed tomography (CT) chest showed numerous new solid and cavitary lesions throughout both lungs, the largest of which measured 5 cm in the right hilum and 4.6 cm in the left lower lung (Figure 1).

Initial brain magnetic resonance imaging (MRI) showed multiple ring-enhancing abscesses throughout the bilateral hemispheres and his cerebellum. The patient was empirically started on vancomycin, cefepime, and metronidazole. On hospital day 4, he developed acute aphasia and new transient right-sided weakness. Repeat brain MRI with magnetic resonance angiography (MRA) showed stable diffuse abscesses with a new infarct in the left thalamus without intracranial aneurysm. The patient was started on

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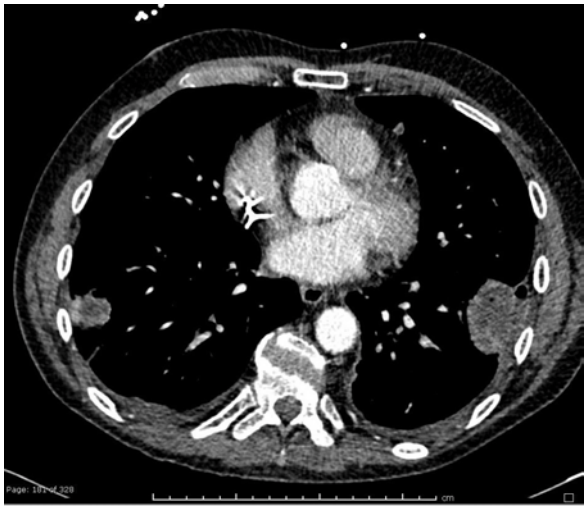
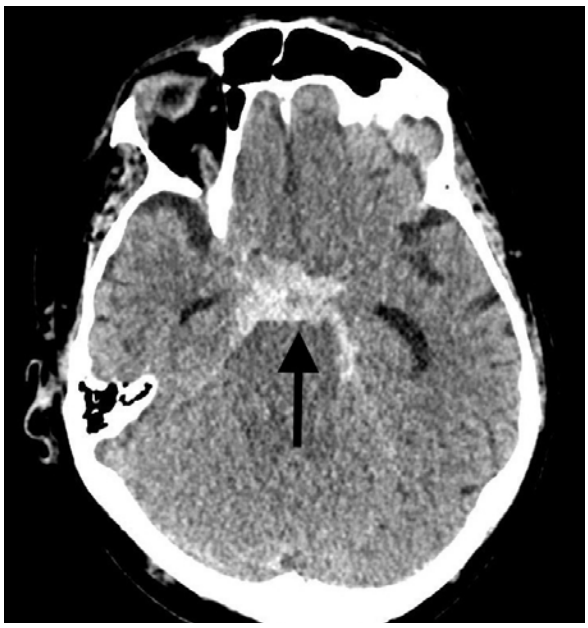
Figure 1. Chest CT Showing Cavitary Lesions



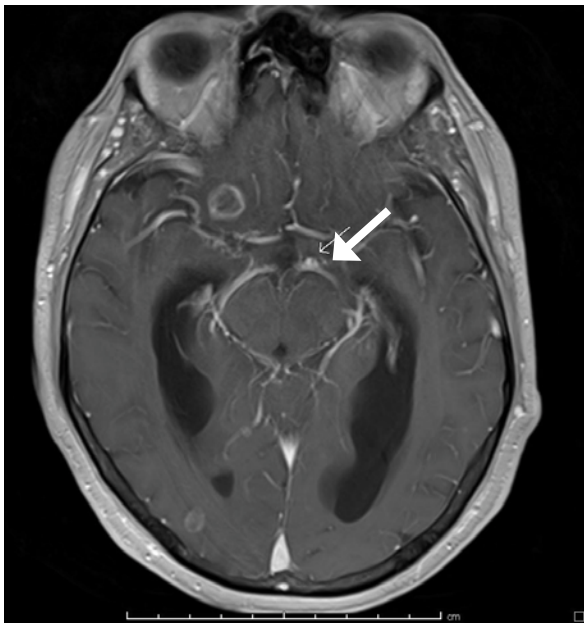
The left-sided cavitary lesion contains gas and a rim of necrosis, also known as the "halo sign."

Figure 2. Computed Tomography Head



Arrow points to subarachnoid blood in the basal cisterns.

Figure 3. Brain Magnetic Resonance Angiography



Arrow points to mycotic aneurysm.

intravenous amphotericin B to broadly cover possible fungal infections, including mucormycosis. Neurosurgery consultation recommended pulmonary biopsy rather than brain biopsy due to the higher risks of the latter intervention. The biopsy of one of the pulmonary nodules grew *Aspergillus fumigatus*.

After the biopsy results returned positive for *Aspergillus* on hospital day 8, intravenous micafungin and voriconazole were started. He continued to be intermittently somnolent throughout his hospital course. On hospital day 12, medical emergency was called as the patient was unresponsive and apneic. His pupils were dilated and nonreactive to light. Emergent head CT showed acute subarachnoid hemorrhage (SAH) with moderate hydrocephalus. (Figure 2). MRA of the brain showed enlarging abscesses with a 2 mm mycotic aneurysm in the posterior cerebral artery, consistent with the location of his SAH (Figure 3).

After a family care conference with the neurosurgery team, family decided to pursue comfort care and the patient died 3 days later.

DISCUSSION

Due in part to an increasing number of patients receiving immunosuppression, *Aspergillus* infections are becoming more prevalent. Both the number of hospitalizations and deaths from *Aspergillus* infection have increased greatly over the past 20 years.[3]

Aspergillus infection typically starts in the lung or sinuses due to inhalation of spores and can spread either contiguously or disseminate via the bloodstream. Central nervous system (CNS) aspergillosis due to abscess formation presents with variable symptoms, including mental status changes, headaches, or more acute presentations such as stroke or seizure.[4,5]

Once invasive *Aspergillus* disseminates to the brain, the mortality rate increases from 58% to 88%.[6] Mycotic aneurysm leading to SAH is a particularly devastating event in CNS *Aspergillus* infection. Intracranial mycotic aneurysms are uncommon in general but are a known sequela of CNS aspergillosis. Thirty-three patients with intracranial mycotic aneurysms due to *Aspergillus*

have been reported in the literature from 1990 to 2005.[7] This is certainly underdiagnosed, as a review of 92 cases of CNS aspergillosis showed that only 56% of cases were diagnosed during life.[8]

Mycotic aneurysm is a focal abnormal dilation of an artery due to infection. Sir William Osler first used this term in 1885 to describe an aneurysm caused by bacterial endocarditis. The term "mycotic" was applied because of the resemblance of the aneurysm to fungal vegetation. Mycotic, however, is a misnomer as most mycotic aneurysms are bacterial.[9]

The main treatment for mycotic aneurysm involves antimicrobial therapy, open neurosurgery, endovascular approach, and/or a combination of them. Given the lack of randomized controlled trials, there are no definitive guidelines to direct clinical decision-making. Overall management depends on whether the aneurysm has ruptured, the aneurysm characteristics, and the patient's overall health status. In those without high surgical risk, endovascular or surgical treatment is advised given the risk of rupture.[10] To date, there are no trials comparing endovascular versus neurosurgical approach; however, endovascular is becoming more popular. In a retrospective review of patients with intracranial mycotic aneurysm (mostly from infective endocarditis) in the *American Journal of Neuroradiology* (*AJNR)*, overall mortality was lower in the intervention group (13%) versus those who received antibiotics alone (40%).[11]

Patients deemed too high risk for surgery should undergo antimicrobial treatment for at least 4 to 6 weeks followed by repeat angiography. In the *AJNR* study, of the 22 patients treated with antibiotics alone, 64% had resolution of their mycotic aneurysm, although this was limited by a small sample size and variable angiographic follow-up.[11] It should be noted that this study had only 1 patient with a mycotic aneurysm caused by *Aspergillus*.

CONCLUSION

In immunosuppressed patients who have concurrent pulmonary and CNS findings, there should be a high suspicion for *Aspergillus* infection and a low threshold to treat empirically prior to confirmation of diagnosis. Despite antimicrobial treatment, CNS aspergillosis mortality remains high. Neurosurgery consultation is necessary in patients with mycotic aneurysm, as endovascular or neurosurgical repair can be effective in those without high surgical risk.

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