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Original Research

Physician Workforce in Kansas: Where are the Orthopedic Surgeons?

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ABSTRACT

Introduction. Rural patients have greater need but less access to orthopedic surgical care than their urban counterparts. Previous studies have investigated rural surgical care, but this is the first to assess the Kansas orthopedic surgery workforce to identify changes over time and rurality and inform thinking about future workforce composition.

Methods. The authors analyzed 2009 and 2019 AMA MasterFile and Area Health Resource File (AHRF) data. Using frequencies, percentages, and calculations of orthopedic surgeons per capita, we assessed workforce changes by rurality (Rural Urban Continuum Codes).

Results. The dataset included 307 orthopedic surgeons; 197 were in both 2009 and 2019. Of these, 165 were in active practice in 2009 and 244 in 2019, an increase of 79 (47.9%). Kansas had smaller proportions of surgeons in rural (non-metro) versus urban (metro) counties in both years. Orthopedic surgeons per capita grew throughout the state, but the increase was smaller in rural counties. There were 11 women orthopedic surgeons in both years, 3.6% of the total 307. Among the 197 surgeons in both years, four (2.0%) were women. No women orthopedic surgeons were in non-metro counties either year.

Conclusions. Although the Kansas orthopedic surgery workforce grew from 2009 to 2019, rural Kansas remains a *surgery desert* based on orthopedic surgeons per capita. Further studies could determine whether this trend is similar to that in other rural states and how to attract orthopedic surgeons to rural practice. ***Kans J Med 2024;17:1-5***

INTRODUCTION

Rural America has long been referred to as older and sicker,[1] but the more nuanced reality is that those in rural communities have higher rates of chronic diseases, activity limitations, and chronic pain, and lower levels of physical activity than those in urban locations, while having less access to healthcare[2] and, for some, lower levels of health literacy.[3] Patients in rural areas tend to prefer receiving orthopedic care locally, especially for chronic, potentially disabling conditions such as osteoarthritis.[4] However, rural Americans have less access to orthopedic surgeons than do their urban counterparts,[5] especially a lack of access to local care.[6] While states with larger rural populations, except in the South, have a higher density of orthopedic surgeons overall,[7] there is a widening gap between urban and rural communities in the per capita number of orthopedic surgeons.[5] In addition, while more women are entering the practice of orthopedic surgery (women represented 6.0% of orthopedic surgeons in 2010 and 7.4% by 2022), available data regarding physician distribution have not looked at the gender composition of the orthopedic surgeon workforce by rurality in Kansas.[8-10]

Patient-physician gender concordance has been well-studied, but results have been divided on the benefits of concordance.[11] Outcomes studied have been related to patient-physician communication,[11-13] patient satisfaction or care rating scores,[14-18] impressions of care outcomes,[19] and care and intervention effectiveness.[20,21] While the vast majority of patients who seek orthopedic care do not express a preference for the gender of their surgeon, those who do may prefer having a woman surgeon.[22] Some studies have found that patients who receive care from women surgeons have better outcomes in terms of mortality, and that women patients, in particular, may have better outcomes when treated by a woman surgeon; however, this line of inquiry is relatively nascent.[23,24] Even if outcomes are not different based on surgeon gender, increasing the number of rural orthopedic surgeons requires making positions in these areas attractive to all potential candidates, including the increasing number of women orthopedic surgeons.

A variety of approaches to addressing the orthopedic needs of those in rural communities have been developed over the years. These include outreach clinics, telemedicine, and recruiting additional orthopedic surgeons to these communities.[4] In a survey of 145 hospital administrators, Weichel et al.[25] found that 71.0% noted a need for increased orthopedic services in their communities, and one-third of rural hospitals were actively recruiting orthopedic surgeons. However, there is limited information regarding changes in the rural orthopedic workforce, including the composition of that workforce over time.

This study used physician- and county-level data to describe the Kansas orthopedic surgery workforce, aiming to add to our knowledge of diversity issues in rural care and inform thinking about the future composition of the orthopedic surgery workforce.

METHODS

The authors used 2009 year-end and 2019 year-end American Medical Association (AMA) MasterFile data for surgical specialists in Kansas to describe the orthopedic surgery workforce by rurality, type of practice, and gender using summary statistics. This allowed the authors to describe what percentage of orthopedic surgeons were engaged in direct patient care in rural counties.

We limited our analysis to the following surgical specialties (as named in the MasterFile): orthopedic adult reconstructive surgery, orthopedics (foot and ankle), orthopedic surgery, orthopedic surgery of the spine, orthopedic surgery-trauma, orthopedic pediatric surgery, orthopedic sports medicine, orthopedic musculoskeletal oncology, and hand surgery/orthopedic surgery. In the MasterFile, surgeons identified their primary specialty; therefore, these categories do not necessarily neatly align with existing residency programs, fellowships, or board certifications. The MasterFile codebook suggests that the category "orthopedic surgery" is general orthopedic surgical practice. For our purposes, we used the term "orthopedic surgeons" as an umbrella term inclusive of all aforementioned categories.

The MasterFile contained data on sex in 2009 (using categories "M"

and "F") and gender in 2019 (also using categories "M" and "F"). We recognized gender and sex were different, and therefore the collection and treatment (use of M and F categories) of these data were problematic. However, we treated both years' data as gender to approximate how gender representation may have changed from 2009 to 2019. We have referred to the "F" category as women and the "M" category as men.

We analyzed the total number of orthopedic surgeons in 2009 and 2019, excluding trainees. We differentiated between those present in both years versus those only present in the 2019 data. We analyzed the primary type of practice (direct patient care, semi-retired, retired, not active for other reasons, or not classified) and the genders of these surgeons. We defined those in "direct patient care" as in "active practice" to avoid any confusion with the term DPC, which often stands for "direct primary care." We also included "semi-retired" in our definition of active practice, as those surgeons may still have been providing patient care, though likely on a part-time basis. We compared the presence of orthopedic surgeons in Kansas counties by using the Rural-Urban Continuum Code (RUCC) classification system.[26] We categorized RUCC's 1-3 as urban, consistent with the RUCC's description of 1, 2, and 3 as "metro" counties. We categorized 4-9 as rural, although we recognized that counties in category 4 differ greatly from counties in category 9. Counties that are a 4 may have populations of 20,000 or greater and are adjacent to metro areas. A 7 would have a population between 5,000 and 20,000 and not be adjacent to metro areas. The most rural RUCC classification, a 9, includes counties with populations under 5,000 and not adjacent to metro areas. We considered urban and rural to be interchangeable with metropolitan and non-metropolitan, respectively, although there are a variety of approaches to rural classification and terminology.

We utilized the 2020-21 release of the Health Resources and Services Administration (HRSA) Area Health Resource File (AHRF) to analyze basic population demographics for urban and rural U.S. counties, including population and percentage of population over age 65. We did not have access to the AHRF for 2009; only population variables for 2010 were available. So, comparisons between 2009 MasterFile data and 2010 AHRF data were approximate. When describing these data together, we used "2009/10."

RESULTS

In our Kansas dataset, there were 307 orthopedic surgeons. Of these, 197 were present in both 2009 and 2019, while an additional 110 were added to the MasterFile between January 1, 2010 and December 31, 2019 (Table 1). These data included fully retired surgeons. No surgeons were marked "presumed dead" in the dataset, but it should be noted that the 75th percentile of age in 2019 was 65, and there were three surgeons over age 90.

Table 1. Kansas orthopedic surgery workforce by practice type, 2009 and 2019.

Practice Type, n (%)	2009	2019	Change
Direct patient care	161 (81.7%)	234 (76.2%)	73 (66.4%)
Semi-retired	4 (2.0%)	10 (3.3%)	6 (5.5%)
*Subtotal, active practice**	*165 (83.8%)*	*244 (79.5%)*	*79 (71.8%)*
Retired	28 (14.2%)	54 (17.6%)	26 (23.6%)
Not active for other reasons	1 (0.5%)	1 (0.3%)	0 (0.0%)
No classification	3 (1.5%)	8 (2.6%)	5 (4.5%)
Total	197 (100.0%)	307 (100.0%)	110 (100.0%)

*The definition of active practice includes those classified by the MasterFile as in "direct patient care" as well as those classified as "semi-retired," as those in semi-retirement could still be providing some patient care.

Of the 197 orthopedic surgeons in the 2009 data, 165 were in active practice; 32 surgeons changed practice type from 2009 to 2019. Most practice type changes were due to retirement. In 2019, 24 moved from active practice to full retirement. Also in 2019, there were 244 orthopedic surgeons actively practicing in Kansas, representing a net gain of 79 (47.9%) surgeons over the 10-year period. Of the 79 actively practicing orthopedic surgeons acquired from 2009 to 2019, 62 (78.5%) were incorporated into urban counties, while 17 (21.5%) were added in rural counties. The increase of 17 surgeons in rural counties represented a 58.6% growth in the rural orthopedic surgery workforce. Nevertheless, this surge in rural counties did not encompass any female practitioners, nor did it substantially impact the proportion of locally available orthopedic surgeons for rural populations. Kansas experienced a slight increase in proportion of orthopedic surgeons practicing in rural counties from 2009 to 2019. Of the 165 orthopedic surgeons actively practicing in 2009, 136 (82.4%) were in urban counties, and 29 (17.6%) were in rural counties. Among the 244 orthopedic surgeons in active practice in Kansas in 2019, 198 (81.1%) practiced in urban counties, and 46 (18.9%) practiced in rural communities.

For those in practice in rural counties in 2009, orthopedic surgeons were most commonly found in RUCC 5 (n = 10). Counties in this classification have populations of 20,000 or more and are not adjacent to urban areas. There were no orthopedic surgeons in counties classified as RUCC 8 or 9. Among orthopedic surgeons practicing in rural areas in 2019, 17 (35.4%) were in RUCC 5 counties. There was one orthopedic surgeon located in a county classified as RUCC 9, but none in any RUCC 8 counties, showing little change over the preceding decade.

Four women were in active orthopedic surgery practice in 2009, comprising 2.4% of the 165 actively practicing orthopedic surgeons that year. All these women practiced in urban counties. One woman surgeon retired between 2009 and 2019. Among the 110 orthopedic surgeons who entered the workforce after 2009, there were six women who entered active practice, resulting in a total of nine women orthopedic surgeons in active practice in 2019, 3.7% of the total 244 in active practice. All nine (100.0%) women orthopedic surgeons practiced in

urban counties (Table 2).

According to the AHRF, in 2010, Kansas had a total population exceeding 2.8 million, with approximately 1.9 million people (66.5%) residing in urban counties and 956,485 (33.5%) in rural counties. By 2019, the state's population had grown to over 2.9 million, reflecting a net increase of 60,196 people. Urban counties housed 2.0 million people (68.7%), while 911,973 (31.3%) resided in rural areas. This demographic shift resulted in an increase from approximately 7.2 orthopedic surgeons per 100,000 population in urban areas in 2009/10 to 9.9 in 2019. In contrast, rural areas experienced a rise from 3.0 orthopedic surgeons per 100,000 population in 2009 to 5.0 in 2019 (Table 3).

Table 2. Kansas orthopedic surgeons in active practice by rurality, 2009 and 2019.

Active Practice by Rurality, n (%)	2009	2019	Change
Men, urban (Metro)	132 (80.0%)	189 (77.5%)	57 (72.2%)
Women, urban (Metro)	4 (2.4%)	9 (3.7%)	5 (6.3%)
Subtotal, urban (Metro)	*136 (82.4%)*	*198 (81.1%)*	*62 (78.5%)*
Men, rural (Non-metro)	29 (17.6%)	46 (18.9%)	17 (21.5%)
Women, rural (Non-metro)	0 (0.0%)	0 (0.0%)	0 (0.0%)
Subtotal, rural (Non-metro)	*29 (17.6%)*	*46 (18.9%)*	*17 (21.5%)*
Total	165 (100.0%)	244 (100.0%)	79 (100.0%)

Table 3. Kansas orthopedic surgeons in active practice per capita, 2009 and 2019.

	2009/10	2019	Change
Kansas, urban (Metro)	7.2	9.9	2.7
Kansas, rural (Non-metro)	3.0	5.0	2.0
Difference	*4.1*	*4.8*	*0.7*
Kansas	5.8	8.4	2.6

DISCUSSION

People living in rural areas undergo more orthopedic procedures, such as total hip or knee replacements, than those in urban communities, indicating the need for access to orthopedic care in rural areas.[27] However, rurality was based on where the patients resided, not where care was provided. To assess access to local care, we needed to look at databases such as those used in this study. Although our study identified a rise in the count of orthopedic surgeons practicing in Kansas, including in rural communities, there was a widening gap in the number of surgeons per 100,000 population during the specified time. This is noteworthy due to a substantial portion of the state's population residing in counties that are not adjacent to urban areas. The absence of adjacency has implications for patients, affecting their ability to travel for care, as well as for providers, impacting their ability to commute to these areas or provide periodic, elective care. This underscores the necessity to augment the number of orthopedic surgeons in rural communities.

Similar issues with concentration of surgeons, including orthopedic surgeons, in urban communities have been identified in other states. Uribe-Leitz et al.[28] found that 47% of counties in California were "surgical deserts", meaning fewer than 6 orthopedic surgeons per 100,000 population. Even with the increase in orthopedic surgeons in rural areas, the current data demonstrated that rural Kansas also could be described as a "surgical desert." This not only impacts the ability of patients in rural communities to obtain care for their chronic musculoskeletal conditions locally, but also leads to longer travel distances to access emergency orthopedic care. Weichel et al.[25] estimated that for communities without orthopedic surgeons, residents traveled an average of 50 miles for emergent orthopedic care and 45 miles for elective care. However, distances ranged up to 250 and 135 miles, respectively. This can place hardships on patients and their families and is of particular concern regarding emergent care. Although orthopedic surgeons may indeed travel to operate and provide outreach clinics, our data did not address this aspect. This represents a notable limitation in workforce research overall, as the MasterFile, a commonly used source, does not accommodate multiple practice locations.

In addition to the immediate impact on access to orthopedic care, the relative lack of orthopedic surgeons can impact the longer-term viability of rural hospitals and access to healthcare. The American Hospital Association reported that 136 rural hospitals closed in 2010 to 2021, similar to the time period of the current study, and more than half of rural hospitals in Kansas are at risk of closing.[29,30] Orthopedic surgeons can be economic drivers for local hospitals.[31,32] Hoskins et al.[31] demonstrated the significant positive impact that surgical services have on the bottom line of critical access hospitals, and Weichel[25] noted that having a full-time orthopedic surgeon significantly impact the bottom line of rural hospitals. Recruiting more orthopedic surgeons to rural Kansas would seem to be critical in maintaining overall access to care for people living in those areas.

However, it can be challenging to recruit orthopedic surgeons to rural areas due to financial constraints, as physicians are incurring greater levels of debt in completing their education and training and may seek opportunities with greater compensation, or at minimum, greater volumes of cases.[33] In addition, there may be concerns with the ability to provide subspecialty care in more rural settings. More than 90% of graduating orthopedic residents complete a subspecialty fellowship, and attracting these physicians to areas in which they may not have the resources to practice to the limits of their training have been shown to limit their interests in rural care.[34]

Given the increasing diversity in orthopedic surgery, including the number of women entering the field,[8] we need to better understand the demographic details of the orthopedic workforce and what leads to, or prevents, orthopedic surgeons from practicing in rural communities. Our analysis of MasterFile and AHRF data found that while the number of women orthopedic surgeons in active practice in Kansas more than doubled from 2009 to 2019, this translated to women comprising only 3.7% of the active orthopedic surgery workforce in the state by 2019. This was somewhat lower than the national percentage of 7.4%.[10] None of the women surgeons in Kansas practiced in rural counties; this was in contradistinction to findings by Rodgers et al.,[35] who found that there were no differences in percentages of women compared to men in urban versus rural practice. There is emerging evidence of increased risks of adverse post-operative outcomes,

including for some common orthopedic procedures, when comparing the results of women patients who were treated by men surgeons to those of women patients treated by women surgeons, although similar impacts of gender discordance were not seen when assessing outcomes of male patients treated by women or men surgeons.[23] However, even if outcomes are not dependent on surgeon gender, efforts to recruit orthopedic surgeons to rural areas need to be made to all orthopedic surgeons and to take account of needs they have related to practice and life outside of work. Women physicians face unique challenges in practice, especially having the resources needed to integrate work and home responsibilities. Women orthopedic surgeons are more impacted by issues with work-family balance and bearing the burden of home responsibilities than are men,[36] similar to women physicians in other specialties.[37] Addressing this requires that hospitals understand the needs of women physicians and provide the resources to make jobs in rural areas attractive. As noted by Ponzio et al.,[36] "the discrepancy in work-family integration must be addressed to attract, support, and retain women as successful orthopedic surgeons."

Limitations. This study was primarily limited by access to data and the observational nature of the data used. This also was a descriptive study using two cross-sectional datasets, meaning we cannot draw causal conclusions among concepts like patient population characteristics, surgeon sub-specialty, and surgeon practice location. Kansas shared demographic trends with other similarly rural states during the study period, such as rural population loss. For these reasons, it served as a reasonable example that other primarily rural and frontier states facing surgical workforce shortages might consider. Future, similar studies in other states could help inform regional and national conversations on addressing orthopedic care for rural communities. In addition, the data we used were common in the literature, and it was important to highlight what can and cannot be done using these datasets.

Future Directions. To better inform efforts to improve access to rural orthopedic surgery care, it is important for centralized institutions, such as the AMA and HRSA, to collect more granular demographic data about providers and for surgical specialty societies to identify those providing rural care in their databases. On a more local or regional level, it is important to assess data regarding the orthopedic surgery workforce in states with similar rural compositions. This would allow identification of programs that are working for those with a greater per capita rural orthopedic surgery workforce. If findings show all rural states are in similar positions, that could indicate the need for discussion and intervention at a regional or national level.

In addition, work is needed to identify and address challenges that keep orthopedic surgeons from establishing practices in rural communities, especially issues with education debt, compensation packages, and the ability to practice subspecialty care. However, efforts can start earlier in the physician pipeline. Physicians from rural communities are more inclined to eventually practice in such areas; however, the percentage of medical students from rural backgrounds has continued to decline.[38] If the rural healthcare workforce is to keep up with demand, there needs to be continued efforts to reach rural students and interest them in careers in medicine and surgery. In addition, once physicians enter orthopedic surgery training programs, efforts could be made to increase their exposure to rural practice with the goal of interesting them in this as a future career option. However, recruiting, supporting, and retaining women in rural orthopedic practice requires additional career-long efforts above and beyond those intended to initially interest them in careers in orthopedic surgery.[39] Women orthopedic surgeons are more likely to practice in larger groups than are men,[35] but these large groups are unlikely to exist outside of urban locations. We need to better understand the support that women find in these larger groups to determine whether those issues can also be addressed by smaller, non-urban hospitals and practices.

CONCLUSIONS

While the orthopedic surgery workforce in Kansas increased over a 10-year period, rural counties in Kansas still exist in a *surgical desert* for orthopedic care. This is of concern in terms of the impact this may have on meeting both acute and chronic care needs, as well as the potential impact on the bottom line of rural and critical access hospitals. Growing the rural surgical workforce requires attracting and interesting medical students and trainees in rural care, as well as identifying and addressing barriers they might face in practice. In lieu of growing the permanent workforce, rural healthcare organizations may want to explore establishing surgical outreach programs by partnering with urban surgeons. Further study is needed to determine whether other states have faced similar workforce trends and to determine how to attract orthopedic surgeons, including women, to rural practice.

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Keywords: rural health, orthopedics, orthopedic procedures, healthcare disparities, health workforce

KANSAS JOURNAL of MEDICINE

Brief Report

The Role of Telemedicine in Rural Specialty Care: Priorities and Recommendations From Rural Primary Care Physicians

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ABSTRACT

Introduction. The study goal was to understand telemedicine's role in improving access to rural specialty care. Other outcomes included assessing specialty availability and frequency of referrals at rural sites.

Methods. This mixed methods study included surveys and semi-structured interviews of rural primary care physicians (PCPs). Survey data were analyzed with summary statistics and cross-tabulations. Interview transcripts were inductively thematically analyzed.

Results. Of the 19 PCPs who completed the survey, 37% agreed/strongly agreed current telemedicine practices connected patients to better specialty care; 90% agreed/strongly agreed it had such potential. Interviews revealed telemedicine could improve care when local specialists were unavailable and provided the most benefit in acute care settings or specialist follow-ups. Most survey respondents reported outreach specialists were highly effective in addressing rural specialty care needs. Respondents reported cardiology, general surgery, orthopedic surgery, ENT/otolaryngology, and dermatology as the most frequently referred-to specialties. In-person neurology, gastroenterology, and dermatology were unavailable in many communities. Respondents identified psychiatry as a high priority for telemedicine and discussed clinic-to-clinic visits to optimize telemedicine use.

Conclusions. The perceived discrepancy between the current and potential roles of telemedicine in rural specialty care suggests that telemedicine may not fully align with the needs of rural patients and could be optimized for rural practice settings. While local, in-person access to specialists remains a priority, telemedicine can reduce patient burdens and improve care when in-person specialists are unavailable. Telemedicine proponents can identify high-priority areas for implementation through quantitative assessment of specialty care utilization and access as reported by PCPs. ***Kans J Med 2024;17:6-10***

INTRODUCTION

Telemedicine may be a way to bring specialty care and expertise closer to rural Americans while improving care continuity, costs, and interprofessional communication.[1,2] Telemedicine expansion accelerated during the COVID-19 pandemic;[3,4] however, rural providers face unique barriers to implementation, including policy regulations, financial and administrative support, and fewer participating specialists.[5] Rural utilization, perceptions, and telemedicine-related obstacles likely have changed amid a progressive technological landscape, necessitating further research.[6]

While telemedicine interventions have aimed to address *perceived* needs, there is insufficient research assessing the *actual* specialty care needs of rural patients and providers, and how telemedicine can address them. Without a comprehensive understanding of rural needs, rapid telemedicine development may bypass populations it aims to serve and disrupt, rather than enhance, rural primary care practices.

METHODS

This cross-sectional study included primary care physicians (PCPs) practicing in rural communities and participating in the University of Kansas School of Medicine's Summer Training Option in Rural Medicine (STORM) program. The research team collected data via survey and semi-structured interviews. The survey included questions about specialty care availability, frequency of specialty referrals, the perceived value of telemedicine in providing specialty care, and communication between PCPs and specialists. We conducted statistical analyses using SPSS; cross-tabulations described the relationship between specialty availability and referral frequency. Graphical representations using weighted averages show results of the cross-tabulations (Figure 1).

The research team based the semi-structured interview guide on questions from a prior study about optimizing telemedicine strategies and adapted for rural PCPs[7] (See Appendix 1 for the complete interview guide; appendix is only available online at journals.ku.edu/kjm). Research assistants conducted interviews in-person or via Zoom, recorded them, and professionals transcribed them verbatim. Braun and Clarke's guidelines informed inductive data coding and theme generation.[8] Two authors (FH, AH) coded responses independently, resolving conflicts by consensus. We used this framework to achieve the primary study goal: understanding the optimal role of telemedicine in rural specialty care from the perspective of PCPs. The University of Kansas Medical Center (KUMC) Institutional Review Board approved this study.

RESULTS

Nineteen PCPs responded to the survey, and 20 participants completed interviews. Respondents' average county populations was 1,991 to 38,972 people (mean: 15,176). Most survey respondents were male (63.2%), with an average practice duration of 7.0 years and an average of 5.8 years in their current community. All respondents reported practicing in an ambulatory setting, with 73.7% also inpatient, 63.2% emergency, 15.8% school-based, 21.1% intensive care unit, 84.2% long-term care facility, and 10.5% in other settings. More than half (57.9%) provided some obstetrical care, with 72.7% of those performing vaginal deliveries and 63.6% offering surgical obstetrics. Proximity to a tertiary care center ranged from 15 minutes to over 3 hours. See Table 1 for demographics.

Table 1. Demographic information.

Gender	Male	12	63.2%
	Female	6	31.6%
	Prefer not to say	1	5.3%
		Mean	**StDev**
Years as Practicing Physician		7.0	0.9
Years Practicing in a Rural Community		7.1	1.0
Years Practicing in Current Community		5.8	0.8
Number of Other Primary Care Providers		3.4	0.4
Number of Midlevel Providers		3.9	0.6
County Size (2020 U.S. Census)		16040.6	11973.5
Rural Code (RUCC 2013)		6.6	1.7
		N	**%**
Practice Settings[a]	Ambulatory	19	100.0%
	In-patient	14	73.7%
	Emergency	12	63.2%
	School-based	3	15.8%
	ICU	4	21.1%
	Long-term care facility	16	84.2%
	Other practice setting[b]	2	10.5%
Obstetrical Care Provided	No	8	42.1%
	Yes	11	57.9%
Obstetric Services*	Prenatal care	10	90.9%
	Vaginal delivery	8	72.7%
	Cesarean section	7	63.6%
	Postnatal care	10	90.9%
	Other obstetric services[c]	1	9.1%
Driving Time to Nearest Tertiary Care Center	15 to 30 minutes	1	5.6%
	30 to 60 minutes	6	33.3%
	1 to 2 hours	5	27.8%
	2 to 3 hours	5	27.8%
	3+ hours	1	5.6%

[a] Responses for these topics were not mutually exclusive.
[b] Other practice settings include hospice and home visits.
[c] Other OB services were not specified.

The team assessed specialist availability and utilization using participants' reports of availability and frequency of referral. The specialties with the highest referral frequencies were cardiology (68.4% frequently referred to, 21.1% very frequently), general surgery (68.4% frequently, 21.1% very), and orthopedic surgery (57.9% frequently, 15.8% very). Critical care (26.3% not at all referred to, 57.9% infrequently) and anesthesiology (26.3% not at all, 52.6% infrequently) had the lowest referral frequencies. Neurology, gastroenterology, and dermatology were the least available specialties (36.8%, 36.8%, and 31.6% reported as not available in the community, respectively), despite frequent referrals (Figure 1). Of surveyed PCPs, 37% agreed or strongly agreed that telemedicine helped connect their patients to better specialty care. Ninety percent agreed or strongly agreed that telemedicine had the potential to connect their patients to better specialty care. Ninety-five percent agreed rural provider input was necessary for telemedicine to serve rural patients effectively (See Appendix 2 for additional results; appendix is only available online at journals.ku.edu/kjm).

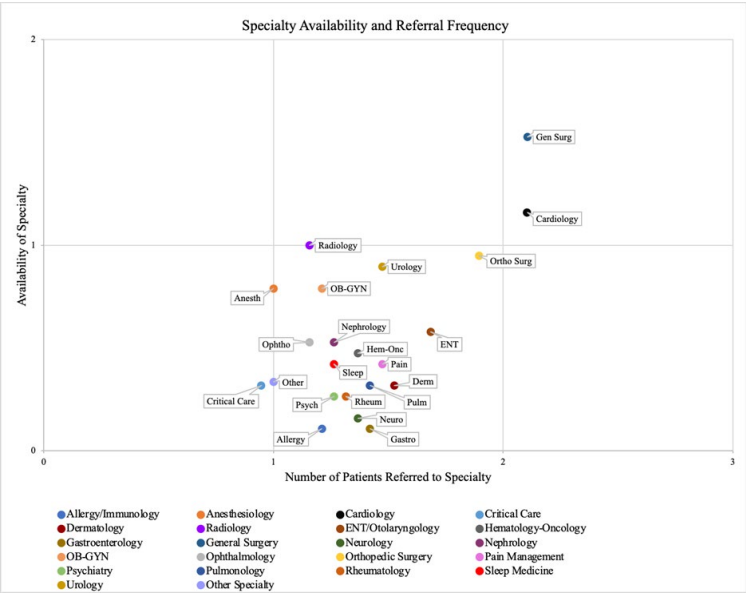

Figure 1. Specialty availability and referral frequency.
Key:
Frequency of Referrals to Specialty (x-axis)
0 = not at all (0)
1 = infrequently (1-20 patients/year)
2 = frequently (20-40 patients/year)
3 = very frequently (50+ patients/year)

Availability of Specialty (y-axis)
0 = not available in community
1 = commute to community
2 = practice primarily in community

Twenty eligible physicians participated in interviews. Interview time totaled 7.5 hours. Individual interviews averaged 22 minutes. Key themes included benefits, concerns, and other priorities related to telemedicine. Primary care physicians highlighted improved access to care and reduced travel burden for patients. They also mentioned increased support for rural providers, with the potential for a positive impact on provider recruitment and retention. Concerns included limitations of physical exams, perceived inferiority to in-person visits, reimbursement and billing complications, access to telemedicine post-pandemic, fear of telemedicine replacing outreach specialists, and patient barriers to telemedicine (e.g., internet access).

The research team subdivided priorities for the ideal use of telemedicine into general framework, visit types, and specific specialties (Table 2). Respondents identified two priorities for specialty telemedicine: 1) it should complement rather than replace in-person visits, and 2) clinic-to-clinic visits offer an ideal setup. Visit types deemed best suited for telemedicine included acute/urgent care settings and follow-up visits with specialists. Respondents highlighted psychiatry as a specific specialty in which telemedicine could have a substantial positive impact. Other high-priority specialties varied based on community needs.

Table 2. Priorities for use of telemedicine.

Theme	Priority	Selected Quotes
General framework	Secondary to in-person visits in the community	"...almost always an in-person visit is the first choice. But if an in-person visit is not going to happen because of location or cost or transportation..., then a telemedicine visit can be super helpful."
	Clinic-to-clinic visits	"[I]f I was going to design an ideal world telemedicine program, I think it would be an actual physical office that, like in the hospital or in our clinic, that the patient would come to, and all of the technology was set up and there was a nurse there that could take their vitals and then get the computer hooked up and then draw their labs if the specialist ordered labs..."
Visit types	Acute/urgent care	"In the [ER] we have telemedicine capabilities for neurology...for doing stroke assessments and things like that. Those are high priority interventions..."
	Follow-up visits	"[I]f the patient is already established with the specialist. For example, if the patient is already following that particular cardiologist, if they're just doing a follow-up for a certain condition..."
Specific specialties	Psychiatry	"I think psychiatry would be one that is really helpful..."
	Other (based on community needs)	"Neurology, rheumatology, gastroenterology, and derm(atology) are probably the big ones down here that we lack that could benefit."

DISCUSSION

Our assessment revealed that telemedicine can facilitate access to specialty care by overcoming patient barriers and minimizing provider isolation. However, those implementing telemedicine should optimize its delivery to address rural-specific limitations. Telemedicine may be considered an adjunctive tool to high-quality, in-person care. Lastly, it is possible to identify specific specialties in which telemedicine interventions may have the greatest impact.

Our data underscored a significant disconnect: only 37.0% of providers agreed that telemedicine currently helps connect their patients to better specialty care, while 90.0% agreed that it has the potential to do so. Overall, our findings echo similar concerns raised in previous studies. Namely, telemedicine does not replace an on-site physician, some fear it may disincentivize commuting specialists, and rural patients face limitations in accessing telemedicine, such as limited internet access.[1,9,10] Unexpectedly, our data suggested telemedicine could potentially improve rural provider recruitment and retention by reducing feelings of professional isolation. This is interesting given a previous study also found telemedicine to improve recruitment and retention, specifically among hospitals using telemedicine in the ED.[11]

The future availability of local and outreach specialists is uncertain, and rural workforce shortages may have an impact.[12,13] Acute/urgent settings and follow-up visits could be initial areas of focus for telemedicine implementation in communities where such services are not available.

High-priority specialty areas for telemedicine implementation could be based on perceived need (e.g., psychiatry) and a quantitative assessment of frequently utilized (e.g., cardiology, surgery) and difficult to access (e.g., neurology, gastroenterology, dermatology) specialties. The perceived need for psychiatric services is unsurprising given the ever-increasing disparity in mental health services, particularly in rural areas.[14] Psychiatric e-consultations and telehealth visits may already be implemented in several rural primary care settings. A variety of specialties are increasingly encouraging e-consultations for collaboration with rural provider collaborations, referral triage, and decreasing appointment wait times.[15-17] The specialties identified in our study as high-priority may not be generalizable to all rural communities. There are limitations to our utilization of specialist availability and referral frequency as proxies for community need. The impact of availability on referral frequency is unclear; less available specialists may be inherently less referred to. More precise individual community-based needs assessments would be appropriate.

Finally, approaches such as clinic-to-clinic visits, also called facilitated, synchronous visits, may be a strategy to overcome patient barriers and optimize telemedicine use. One study showed telemedicine visits conducted primarily as clinic-to-clinic visits had equivalent outcomes to in-person office visits for endocrinology patients.[18] The Study to Promote Innovation in Rural Integrated Telepsychiatry (SPIRIT) trial demonstrated that clinic-to-clinic psychiatry visits improved outcomes for patients in rural and underserved areas.[19] This is encouraging given our study identified psychiatry as a priority for telemedicine. Waibel, et. al.[15] reported significant patient cost savings related to travel and time off work with the use of facilitated, synchronous specialist visits. These studies support telemedicine as an effective tool to improve access to specialty care, especially when integrating facilitated, synchronous and asynchronous models.

This study had several limitations. Its sample consisted of rural Kansas physicians involved in medical education. Therefore, our conclusions may not be generalizable to all rural physicians. More patient-centered study designs would more accurately reflect patient telemedicine perspectives. Our sample size was appropriate for qualitative work, as we reached code saturation.[20] Our sample was also relatively homogenous, a factor indicating appropriateness of a smaller sample size.[21] However, our small survey sample size limited analytical power. A more comprehensive study could better assess the value of telemedicine for rural specialty care.

CONCLUSIONS

Our findings suggest that telemedicine may not fully align with the specific needs of rural areas and should be optimized to address the limitations of rural practice. While local access to specialists is preferred, most rural providers consider telemedicine fundamental in facilitating access to specialty care. It is most useful in acute settings where immediate specialist input could improve clinical outcomes and in follow-up visits that would otherwise burden rural patients. Specific areas of priority for telemedicine intervention include high-need specialties such as psychiatry, cardiology, and surgery, as well as difficult-to-access specialties such as neurology, gastroenterology, and dermatology. Further research is needed on clinical outcomes in specialty-specific telemedicine interventions, the effectiveness of clinic-to-clinic visits, and telemedicine's impact on the rural physician workforce.

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Keywords: rural health, telemedicine, access to care, consultation and referral

KANSAS JOURNAL of MEDICINE

Brief Report

Prenatal Decision-Making in Patients with Limited English Proficiency: What Factors are Involved?

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ABSTRACT

Introduction. A comprehensive definition of culture encompasses shared norms, beliefs, expectations, language, and customs, all of which are crucial considerations when working with patients with limited English proficiency (LEP). In this study, the authors examined how language, external influences, and patient-provider relational factors associated with decisional conflict in prenatal care patients.

Methods. The authors conducted a cross-sectional study to assess decisional conflict related to postpartum contraception, elective induction, and newborn feeding methods. The survey included questions about demographics, communication methods, external influences, and provider trust, and was distributed to prenatal care patients who spoke either English or Spanish. Data analysis involved using descriptive statistics and chi-square analyses.

Results. Out of the 23 respondents, 12 were Spanish-speaking and 11 were English-speaking. Spanish-speaking participants were less likely to have health insurance compared to English-speaking participants ($\chi^2(1, N = 23) = 3.67, p = 0.016$). There was no statistically significant difference in decisional conflict between English- and Spanish-speaking participants. Religion affected 11 of 23 participants' decisions, while partner expectations influenced 10 of 23 participants. Working with an interpreter and the quality of interpretation were crucial for Spanish-speaking individuals. Most participants (59%) felt that the provider's understanding of the patient's cultural background was important for decision-making.

Conclusions. While there was no association between language discordance and decisional conflict, several factors influencing prenatal decision-making were identified. The use and quality of interpretation significantly affected decision-making and should be prioritized for patients with LEP. Religion and partner expectations were found to be highly influential in decision-making. Respondents also emphasized the importance of the provider's understanding of the patient's cultural background. ***Kans J Med 2024;17:11-15***

INTRODUCTION

Shared decision making (SDM) involves multiple parties reaching a consensus about a preferred treatment and is a growing clinical practice.[1] While there is no definitive model for this approach to patient-centered care, there are guiding principles. SDM prioritizes patient autonomy and beneficence by providing patients with adequate information about their options to aid in their informed decision-making.[2] This approach has improved patient satisfaction, clinical outcomes, adherence to treatment, and lowered incidences of decisional conflict and regret.[3-5] Decisional conflict refers to an individual's uncertainty about the course of action when choices involve risk, loss, regret, or challenge personal life values.[6] As patients assume greater responsibility for their health decisions, evaluating the efficacy of SDM and its correlation with experiencing decisional conflict becomes increasingly important.

Decisional conflict is essential to evaluate not only for assessing the effectiveness of SDM and communication, but also for understanding the factors that contribute to a patient's role in decision-making. Previous studies indicate a higher incidence of poor-quality patient-clinician communication and SDM among non-White individuals, patients with limited English proficiency, families of lower socioeconomic status, and patients with lower education levels.[7,8] Providers and patients speaking different languages are associated with worse communication, which could impact SDM. Therefore, it is crucial to investigate how language discordance and culture might impact the degree of uncertainty regarding medical decisions in patients with limited English proficiency.[9]

Prenatal care is an area of medicine where SDM is particularly important, as pregnant women are required to make numerous decisions for both them and their child.[10] These decisions include choosing a birth control method, determining the mode of delivery, and selecting feeding options for the newborn. These decisions often involve SDM, as there are usually multiple options without a clear right or wrong choice.

Many prenatal decisions also are subject to cultural, familial, and societal influences.[10,11] Coast et al.[10] created a systematic mapping of interventions that have been implemented to address cultural factors that affect women's use of skilled maternity care, stating that,

> *"Childbirth, and the time around birth, is a social and cultural event that is often governed by [societal] norms. However, in most societies, the dominant culture, expressed through social institutions such as the healthcare system, regulates how health issues are both perceived and addressed."*

Understanding the impact of linguistic, cultural, familial, and social experiences is crucial for physicians to comprehend the drivers behind patient decision-making, especially in non-white individuals.[12] Providers need to consider these cultural and societal influences as integral components of the SDM process, particularly in the context of decisional conflict. This study aimed to identify factors associated with decisional conflict in prenatal care, including language proficiency, cultural influences, and patient-provider trust.

METHODS

This cross-sectional study included patients from two safety net clinics in Kansas City: The University of Kansas Medical Center's (KUMC) Maternal Options that Matter (MOM) Clinic and Jaydoc Free Clinic's Women's Health Initiative Program (WHIP). We distributed surveys to consenting patients receiving care at these clinics. Inclusion criteria were being 18 years or older, English- or Spanish-

speaking, receiving prenatal care at either clinic, and having made or being in the process of making decisions regarding postpartum birth control, elective induction versus spontaneous labor, and breastfeeding versus formula feeding. We excluded patients under 18 years old and those unable to complete a survey in English or Spanish. We distributed the surveys across various weeks of gestational age during pregnancy, but the timing of survey completion and specific weeks of gestational age were not recorded by the research team.

The survey comprised 35 items, incorporating Likert scale and binary response options. It gathered demographic data on race, ethnicity, birth country, education level, primary language, and health insurance status. To assess decisional conflict, the validated SURE (Sure of myself, Understand information, Risk-benefit ratio, Encouragement) questionnaire was included.[13,14] This 4-item checklist screens for clinically significant decisional conflict, with a cutoff score of ≤3 out of 4 indicating such conflict.[13,14] In addition to the SURE questionnaire, the survey included researcher-developed questions on external influences (seven questions) and the impact of language interpretation quality and method (three questions) on decision making. Furthermore, it featured researcher-developed inquiries on patient trust in providers, focusing on gender and racial background, cultural understanding, and perceived patient interest (four questions).

This study received approval from the KUMC Institutional Review Board. Surveys were distributed using Quick Response (QR) codes on paper within the clinic space, directing participants to individual surveys on their personal devices. Informed consent was obtained before initiating the survey, and no incentives were offered for participation. Survey completion was estimated to take 10 minutes. Data collection and management were conducted using REDCap® electronic data capture tools hosted at the University of Kansas Medical Center.[15,16] We used descriptive statistics and chi-square to analyze the data.

RESULTS

A total of 23 patients completed the surveys: 11 responded in English and 12 in Spanish. The response rate is unknown as documentation of those who did not complete the survey was not recorded, given the convenience sampling method during clinic visits. Summary of patient demographic data is presented in Table 1.

When comparing groups through chi-square analysis, a significantly higher proportion of English-speaking respondents anticipated having health insurance after birth compared to their Spanish-speaking counterparts ($\chi^2(1, N = 23) = 3.67, p = 0.0161$). English-speaking respondents were predominantly from the U.S. or Mexico, while Spanish-speakers originated from various countries in Latin America.

As shown in Table 2, there was no significant difference in decisional conflict rates between English- and Spanish-speaking participants regarding postpartum contraception, elective induction, and method of newborn feeding ($\chi^2(1, N = 23) = 4.296, p = 0.637$). The highest level of decisional conflict was observed regarding elective induction, while the lowest level was related to feeding method. However, there were no significant differences between English- and Spanish-speaking patients ($\chi^2(1, N = 23) = 3.701, p = 0.296$).

Table 1. Study population demographics.

Demographics	English Respondents (n =11)	Spanish Respondents (n =12)
Age at time of survey (mean ± SD)	23.4 ± 3.6	26.9 ± 5.6
Ethnicity (%)		
Non-Hispanic/Latinx	3 (27.3%)	0 (0%)
Hispanic/Latinx	8 (72.7%)	12 (100%)
Race (%)		
White	5 (45.5%)	4 (33.3%)
Black	1 (9.1%)	0 (0%)
Other	3 (27.3%)	6 (50%)
Unspecified	2 (18.2%)	2 (16.7%)
Country of Origin (%)		
United States	5 (45.5%)	0 (0%)
Mexico	5 (45.5%)	4 (33.3%)
El Salvador	0 (0%)	1 (8.3%)
Honduras	0 (0%)	3 (25%)
Guatemala	0 (0%)	3 (25%)
Colombia	0 (0%)	1 (8.3%
Ethiopia	1 (9.1%)	0 (0%)
Primary language (%)		
English	8 (72.7%)	0 (0%)
Spanish	2 (18.1 %)	12 (100%)
Other	1 (9.1%)	0 (0%)
Highest education level (%)		
Some high school	3 (27.3%)	4 (33.3%)
High school degree or equivalent	5 (45.5%)	7 (50%)
Some college	2 (18.2%)	0 (0%)
Bachelor's degree	0 (0%)	1 (8.3%)
Graduate/professional degree	1 (9.1%)	0 (0%)
Insurance status at time of birth (%)		
Insured	6 (54.5%)	1 (8.3%)
Uninsured	5 (45.5%)	11 (91.7%)

Table 2. Patient-reported decisional conflict regarding perinatal care coordination using the SURE questionnaire. A cutoff score ≤3 out of 4 is used to identify decisional conflict.

Reported Conflict	English Respondents (n =11)	Spanish Respondents (n =12)
Induction of labor		
Conflict	5 (45.5%)	5 (41.7%)
No Conflict	6 (54.5%)	7 (58.3%)
Postpartum contraception		
Conflict	5 (45.5%)	3 (25%)
No Conflict	6 (54.5%)	9 (75%)
Feeding methods		
Conflict	2 (18.2%)	2 (16.7%)
No Conflict	9 (81.8%)	10 (83.3%)

The most frequently cited cultural factors influencing decision-making were religious affiliation and partner expectations in both groups. Respondents most often indicated that these factors had at least "some" influence compared to others. Family and community expectations, as well as family traditions, did not significantly impact participants' decisions. Gender expectations were rarely influential, although patients may not discern the similarities between partner expectations and gender expectations.

External factors played a similar role in decision-making for both English- and Spanish-speaking participants. There was no statistically significant difference between the two groups regarding the influence of partner expectations ($\chi^2(1, N = 23) = 1.051$, $p = 0.305$) or the influence of family traditions ($\chi^2(1, N = 23) = 1.155$, $p = 0.283$).

Most participants indicated that the gender and racial background of the provider did not significantly affect their decision-making. For those for whom these factors were important, the rates were similar for both English- and Spanish-speaking participants. Thirteen respondents noted that the health care provider's understanding of their cultural background significantly impacted their decision-making, either 'very much so' or 'somewhat'. The factor most frequently chosen as having the greatest impact on decision-making ('very much so') was the provider demonstrating concern for the patient's best interest (n = 15). Among those who felt their health care provider did not have their best interest in mind, 75% (n = 3) were Spanish-speaking. Figure 1a illustrates responses to cultural influence prompts, while Figure 1b illustrates responses to health care provider factors.

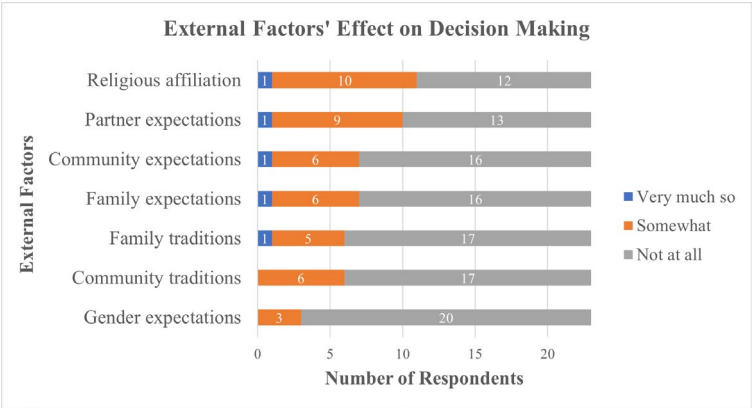

Figure 1a. Participant responses to Likert scale prompts regarding external factors that may have influenced prenatal decision-making. Participants were asked to rank the importance of the following aspects on their decision-making.

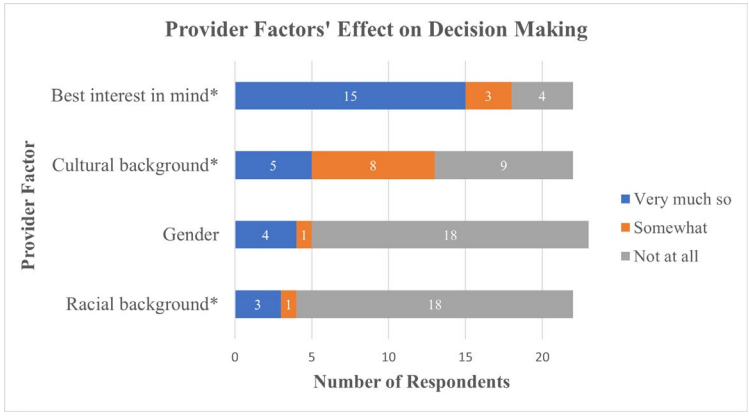

Figure 1b. Participant responses to Likert scale prompts regarding patient-provider relational factors that may have influenced prenatal decision-making. Participants were asked to rank the importance of the following aspects on their decision-making.
*Some patients chose not to answer all questions resulting in variable total values.

Seven out of the 12 Spanish-speaking participants worked with a professional in-person interpreter, two used a virtual interpreter, and three conversed directly with their provider in Spanish. Among those who worked with an in-person interpreter, the majority felt that it significantly enhanced their clinical experience (n = 5). Figure 2 depicts respondents' views on language interpretation. Both participants who used virtual interpreters during their prenatal visits emphasized the importance of the interpreter's presence and the quality of interpretation.

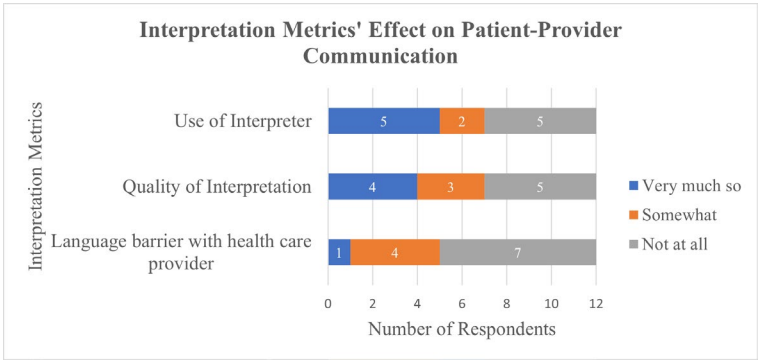

Figure 2. Responses to Likert scale questions from Spanish-speaking participants who utilized interpreting services.

DISCUSSION

When addressing the needs of LEP patients, it is crucial to embrace a comprehensive definition of culture, which includes shared norms, beliefs, expectations, language, and customs, as well as external influences.[17,18] In this study, we aimed to explore how external influences, provider attributes, and the method and quality of language interpretation contribute to decision-making in prenatal care.

Our findings revealed no significant difference in decisional conflict between English- and Spanish-speaking individuals regarding postpartum contraception, elective induction, or newborn feeding methods. This suggests that language discordance alone may not be the primary factor contributing to decisional conflict. Therefore, simply providing proper interpreting services may not fully address the reported issues of poor patient-clinician communication among non-English speakers.[7]

Beyond language, we identified other factors, such as partner expectations and religious affiliation, that may influence prenatal decision-making and contribute to feelings of decisional conflict. However,

participants who utilized interpreters indicated that interpretation was crucial to their clinical experience, emphasizing the importance of interpretation quality.

A systematic review by Flores et al.[19] highlighted that the quality of care diminishes when patients with limited English proficiency lack access to interpreters or are provided with inadequately trained interpreters. This suggests that patients without proper language interpretation may face challenges, such as poor interpersonal interactions, increased likelihood of misunderstanding, decisional conflict, and compromised care. As health care professionals facilitate discussions regarding care coordination and informed decision-making, ensuring proper interpreting services could help alleviate feelings of decisional conflict and potentially reduce adverse outcomes across patient populations, as poor interpretation cannot be overlooked as a contributing factor.

We found that religious affiliation and partner expectations were the most influential cultural factors affecting decision-making. It is not surprising that various religious beliefs can influence health decisions, especially regarding contraception.[20] Partners often play a significant role in the prenatal and postnatal journey, so their expectations can heavily influence the decisions made by expectant mothers. In the traditional SDM model, the health care provider and the patient are the main participants. However, the influence of partners may disrupt this model, potentially leading to decisional conflict if the partner's and patient's beliefs and expectations are not aligned. Moreover, SDM processes may inadvertently involve individuals outside of the patient-clinician relationship. Clinicians should be mindful of these external influences and consider them when guiding patients through their decision-making processes.

We also found that participants were influenced by their health care provider's understanding of their culture, with 13 respondents indicating that it impacted their decision-making. Coast et al.[10] published a comprehensive review mapping interventions that providers can use to address cultural factors affecting maternal care in diverse contexts. They emphasized the importance of an interdisciplinary approach and active dialogue with communities to understand their cultural systems, health beliefs, practices, and preferences. These strategies are crucial in healthcare delivery to diverse populations to enhance understanding and reduce cultural ignorance. Fisher et al.[18] refers to this practice as "cultural leverage," which describes the strategy of improving the health of communities of color by utilizing their cultural practices, products, philosophies, or environments to facilitate behavior change in both patients and practitioners. By employing "cultural leverage" with patients, providers can demonstrate acknowledgment and understanding of a patient's culture, potentially mitigating a patient's decisional conflict.

While culture typically includes aspects of religion, language, beliefs, and norms that shape an individual's or group's perspectives, it can be challenging to separate cultural factors influencing decision-making from concurrent economic and geographic constraints.[18] This was evident in our study, particularly with Spanish-speaking respondents who were significantly less likely to have health insurance compared to their English-speaking counterparts. We recognize that these differences are not solely due to language spoken, but are influenced by broader contextual factors affecting certain populations, which must be fully considered to provide appropriate care.

Additionally, most participants in our study believed that their health care providers had their best interests in mind. However, three of the four participants who disagreed were Spanish-speaking. While this study cannot pinpoint the exact source of this sentiment among these patients, it may indicate an area for improvement in how providers treat LEP patients and the need to ensure all components of culturally competent medical care are provided.

This study is limited by its cross-sectional design, conducted at varying weeks gestation during the patients' prenatal course, which was not documented. As a result, there may be variability in responses, as patients likely received differing levels of counseling throughout their pregnancy. Further studies are warranted in this context, with larger sample sizes and prospective study designs. Additionally, studies should explore the influence of other non-English languages and cultural contexts from patients' countries of origin. Future research on the practice of cultural leverage and its impact on decision outcomes for diverse communities in medicine would be a valuable addition to the existing literature on this topic.

CONCLUSIONS

While language discordance did not significantly relate with decisional conflict, we identified several factors that associated with prenatal decision-making. The presence and quality of interpretation were found to be beneficial to communication and decision-making, highlighting the importance of prioritizing these services for patients with LEP. Religious beliefs and partner expectations were the cultural aspects most frequently reported by patients as influencing their decision-making, albeit to varying degrees. Interestingly, the factor most cited was the belief that their health care provider had their best interests in mind. This underscores the need for providers to consider the broader cultural contexts and external influences that contribute to a patient's decision-making process, while also emphasizing the importance of building trust with patients.

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Keywords: prenatal care, decision making, cultural competence

KANSAS JOURNAL of MEDICINE

Case Report

Gallbladder Torsion: A Rare but Critical Diagnosis

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INTRODUCTION

Gallbladder torsion or volvulus is a rare cause of acute abdominal pain that is often difficult to diagnose and requires prompt surgical intervention.[1-2] Pre-operative diagnosis is only made in a quarter of patients with the remaining cases in the literature diagnosed at the time of cholecystectomy.[1] Non-torsion related acute cholecystitis may be treated conservatively with antibiotics or percutaneous drainage, however, a delay in diagnosis or misdiagnosis of gallbladder torsion may lead to gallbladder necrosis and subsequent perforation, biliary peritonitis, and sepsis.[3-5] Improving the rate of pre-operative diagnosis of gallbladder torsion is essential to reduce its associated morbidity and mortality.

CASE REPORT

A 68-year-old male presented to the emergency department with abdominal pain that began five days prior and progressively worsened. Abdominal pain was accompanied by nausea and anorexia. Physical examination revealed generalized abdominal tenderness without guarding or rebound tenderness. His initial lab studies demonstrated a normal white blood cell count, total bilirubin, and serum lipase.

Initial contrast-enhanced computed tomography (CT) demonstrated a distended gallbladder within the mid abdomen with the gallbladder fundus directed toward the left side of the abdomen and nodular enhancement along the right margin of the gallbladder (Figure 1). Additional findings included diffuse biliary duct dilatation, though there was no gallbladder wall thickening or significant pericholecystic stranding or fluid. Right upper quadrant ultrasound (US) demonstrated a dilated gallbladder with edematous, thickened gallbladder wall, and biliary sludge (Figure 2). No cholelithiasis was visualized, and the sonographic Murphy sign was negative.

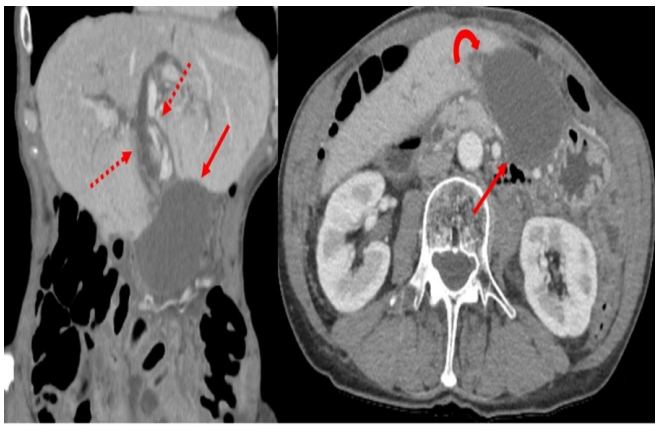

Figure 1. CT images of gallbladder torsion with dilated gallbladder located within the mid to left hemi-abdomen (solid arrow), diffuse intrahepatic biliary duct dilatation (dashed arrows), and enhancing cystic duct/vascular pedicle along the right margin of the gallbladder (curved arrow).

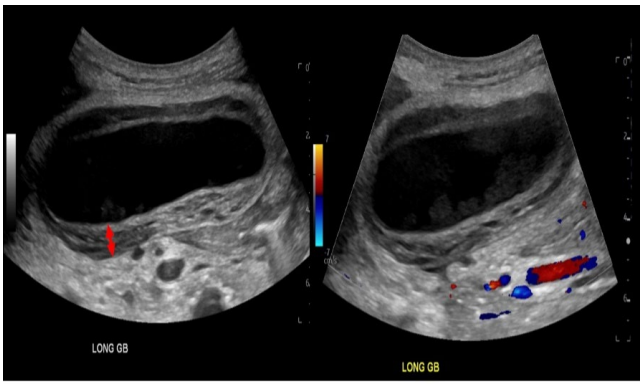

Figure 2. Grayscale and color Doppler ultrasound images of diffuse gallbladder wall thickening (double head arrow) with heterogeneous low echogenicity areas of edema within the thickened wall.

Hepatic iminodiacetic acid (HIDA) scintigraphy demonstrated adequate hepatic uptake of the radiotracer, normal biliary excretion, and normal transit to small bowel through the ampulla of Vater; however, the gallbladder was not visualized for the duration of the examination including after morphine injection (Figure 3). These findings were consistent with acute cholecystitis and cystic duct obstruction.

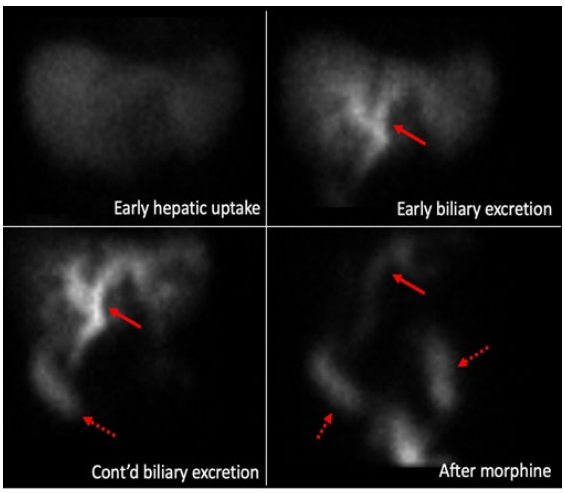

Figure 3. HIDA scan with initial, early hepatocyte uptake (upper left), progressive biliary excretion of tracer (solid arrows), and normal excretion of tracer into small bowel loops (dashed arrows). Further transit of tracer into small bowel after the administration of morphine (bottom right) though no tracer fills the gallbladder.

Non-contrast magnetic resonance cholangiopancreatography (MRCP) demonstrated marked gallbladder wall thickening and edema with diffuse T1-weighted (T1W) hyperintense signal throughout both the thickened wall and lumen of the gallbladder (Figure 4). The gallbladder orientation remained transverse with the fundus directed toward the left side of the abdomen. These imaging findings were consistent with hemorrhagic or gangrenous cholecystitis secondary to gallbladder torsion given the gallbladder orientation.

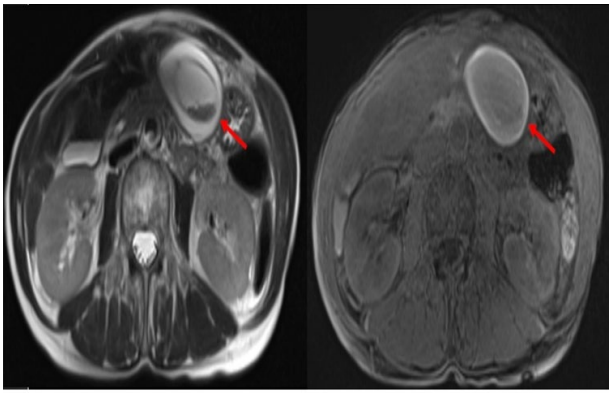

Figure 4. MRCP images of acute gangrenous/hemorrhagic cholecystitis secondary to torsion with thickened gallbladder wall (solid arrow-left) and abnormal, diffuse T1W hyperintense signal throughout the lumen and wall of the gallbladder (solid arrow-right).

The pre-operative diagnosis of gallbladder torsion was confirmed via open cholecystectomy, which demonstrated a completely gangrenous gallbladder with the gallbladder fundus located to the left of umbilicus secondary to twisting of the gallbladder along its vascular pedicle. Following cholecystectomy, the patient recovered and was discharged to home.

DISCUSSION

Gallbladder torsion, or volvulus, is a rare cause of acute abdominal pain that most commonly affects older females with a median age at presentation of 77 years old.[1] Pre-operative diagnosis is difficult though important as conservative treatments often pursued in cases of non-torsion-related acute cholecystitis in the elderly may delay surgical intervention and predispose to gallbladder necrosis, perforation, and sepsis.[1,3-5]

Torsion of the gallbladder results in obstruction of biliary outflow from the gallbladder and narrowing or occlusion of the cystic artery perfusing the gallbladder.[3] Although the direct cause of gallbladder torsion is unknown, it is thought that congenitally abnormal, incomplete, or absent mesenteric attachments of the gallbladder to the gallbladder fossa of the liver, sometimes termed *floating gallbladder* or *wandering gallbladder*, may predispose to torsion.[3,6,7] Acquired hepatic atrophy from aging and underlying liver disease may also predispose to gallbladder torsion.[3]

Imaging may aid in the pre-operative diagnosis of gallbladder torsion, though imaging findings overlap with non-torsion causes of acute cholecystitis, such as gallbladder distention, wall thickening, and pericholecystic fluid.[4] The prevalence of gallstones in gallbladder torsion is significantly less than those with acute gallbladder pathology without torsion.[8] Ultrasound and CT may also demonstrate abnormal positioning of the gallbladder, such as with a transverse orientation or a twisted vascular pedicle or cystic duct to the right of the distended gallbladder.[4] The presented case demonstrated transverse orientation of the gallbladder with the fundus directed towards the left hemiabdomen as opposed to the right. Additionally, there was an enhancing vascular pedicle and/or cystic duct along the right margin of the gallbladder.

HIDA scintigraphy can be utilized in equivocal or difficult cases of acute gallbladder pathology, however, it is not helpful in determining the cause of cystic duct obstruction in the setting of acute cholecystitis. In both cases of gallbladder torsion and obstructing calculus within the cystic duct, the gallbladder will not opacify with radiotracer despite the administration of morphine, which acts by closing the ampulla of Vater and allowing excreted radiotracer within the bile ducts to be directed towards the gallbladder lumen rather than the small bowel.[9]

MRI and/or MRCP in cases of gallbladder torsion may demonstrate areas of T1W hyperintense signal within the wall of the gallbladder, as seen in the presented case, compatible with acute necrosis and/or hemorrhage.[10] Other findings that can be seen on MRI/MRCP are similar to those seen on CT with abnormal gallbladder orientation and/or twisted pedicle.

CONCLUSIONS

In summary, gallbladder torsion is a rare though important cause of acute abdominal pain and should be a diagnostic consideration, particularly in elderly patients with acute gallbladder pathology, abnormal gallbladder orientation by imaging, and an absence of gallstones.

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Keywords: gallbladder, gallbladder disease, acute cholecystitis

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Case Report

Is it Worsening ADHD or Graves' Disease? A Case Report of Undiagnosed Graves' Disease in a Patient with ADHD

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INTRODUCTION

Attention-deficit/hyperactivity disorder (ADHD) is a common neurodevelopmental disorder affecting children and adolescents with a worldwide prevalence of 7.2%.[1] ADHD is a heterogeneous disorder consisting of symptoms of inattention and/or hyperactivity-impulsivity which cause impairment in multiple settings, including home and school.

ADHD phenomenology includes three presentations: inattentive, hyperactive-impulsive, and combined.[2] The predominantly inattentive ADHD subtype includes difficulty with listening, paying attention to details, sustaining focus, following instructions, and organizing tasks or activities. Patients with the hyperactive-impulsive presentation of ADHD often fidget, struggle to remain still or seated, are restless, talk excessively, and are often described as being "on the go" or "driven by a motor". The combined subtype of ADHD includes six or more symptoms from each of the inattentive and hyperactive-impulsive categories.[2] To receive a diagnosis of ADHD, symptoms must have been present for the last six months and must have caused difficulties in two or more settings. In addition, multiple symptoms must have been experienced prior to the age of 12 years.[2] Many children and adolescents with ADHD (24-50%) also experience symptoms of emotional dysregulation, including irritability, aggressive outbursts, and mood lability.[3]

Hyperthyroidism is defined as the excessive synthesis and secretion of thyroid hormones, while thyrotoxicosis comprises the physical signs and clinical manifestations caused by the excessive concentration and action of thyroid hormones in bodily tissues.[4] Graves' disease, which accounts for 60-80% of cases of hyperthyroidism in children and adolescents (some sources indicate more than 95% of cases),[4,5] is an autoimmune disorder in which autoantibodies (Thyroid Stimulating Immunoglobulin, TSI) activate the thyroid stimulating hormone (TSH) receptor (TSH-R), leading to hypersecretion of thyroid hormones.[6] The disease's complex immune pathogenesis also includes a cytotoxic process whereby antithyroid peroxidase (TPOAb) and antithyroglobulin (TgAb) autoantibodies attack the thyroid gland.[4,6] The clinical presentation of Graves' disease typically includes a triad of symptoms, comprising goiter, exophthalmos, and tachycardia.[4] Other symptoms include nervousness, excitability, difficulty concentrating, tremor, systolic hypertension, hyperkinesia, sweating, weight loss, difficulty sleeping, heat intolerance, muscle weakness, frequent bowel movements, polyuria, and menstrual disorders in girls.[4,7]

ADHD and thyrotoxicosis share many symptoms, such as hyperactivity, increased energy, irritability, emotional lability, and difficulty concentrating and sleeping. Hyperthyroidism/thyrotoxicosis is less common in adolescents than in adults,[4] nonetheless it is a serious condition that must be considered in the differential diagnosis of patients with a history of ADHD who present with worsening ADHD-like symptoms despite no changes in pharmacotherapy.

CASE REPORT

A 16-year-old girl in foster care for the past two years with a history of ADHD, disruptive mood dysregulation disorder, and enuresis was admitted to the adolescent behavioral health unit after medical stabilization for an intentional overdose of desvenlafaxine, lurasidone, and melatonin. A month before this admission, the patient had been hospitalized in another facility due to anger outbursts and attempts to run away from her foster home. At discharge, she had been prescribed lurasidone 60mg daily, desvenlafaxine 50mg daily, clonidine 0.2mg twice a day, and melatonin 3mg at bedtime, and had been placed with her older sibling.

During the patient's hospitalization at our facility, she reported recent stressors, such as the sudden death of her father two months prior and frequent changes in placement. The patient described her suicide attempt as an impulsive act on the wave of emotional turmoil after finding sentimental objects related to her father at the house she shared with her older sibling. The patient reported a history of unstable mood, irritability, difficulty sleeping, impulsivity, high energy, difficulty concentrating, racing thoughts, anxiety, and palpitations. On examination, she was observed to be anxious, restless, visibly jittery, and displaying rapid speech. On admission, recorded vitals indicated tachycardia (heart rate of 120 bpm) and high blood pressure (systolic 146/diastolic 84 mmHg).

Regarding the patient's prior history, her current case worker reported that the patient had experienced sexual and physical abuse in her childhood when she lived with her mother. After she was removed from her mother's care, the patient lived with her father and stepmother before entering the foster care system. According to the case worker, the patient had experienced many psychiatric hospitalizations and multiple foster placements in the past two years due to episodes of anger outbursts (one to two times per month), running away, and physical aggression toward foster parents and children. Furthermore, the patient endorsed a history of engaging in self-harm behaviors, such as cutting and scratching, marked impulsivity, and risky sexual behaviors. Based on collateral information provided by her older sibling, the patient had a history of ADHD, combined type.

As part of the diagnostic evaluation, routine laboratory tests were conducted, and results revealed low TSH <0.01 mcIU/ml (lab reference value 0.35-4.94) and elevated free T4 level of 1.7 ng/dl and repeat level next day of 1.8 ng/dl (lab reference value 0.7- 1.5). According to the patient's current case worker, elevated thyroid hormone levels were also present during the last hospitalization a month prior. However, due to a lack of stable placement, the patient had not followed up with a primary care physician for further evaluation and management.

During this admission, a pediatric endocrinologist was consulted

and an additional workup was recommended to identify the etiology of the patient's thyrotoxicosis. T3 total was found to be elevated at 334 ng/dl (lab reference value 35-193), thyroperoxidase (TPO) and thyroglobulin antibodies returned elevated at 733 IU/ml (lab reference range 0-6) and 42 IU/ml (lab reference range 0-5), respectively. Thyroid stimulating immunoglobulin (TSI) result was pending during the hospitalization. Based on the clinical features and available laboratory results, thyrotoxicosis due to Hashimoto's thyroiditis was initially suspected due to only mildly elevated free T4 level.

To address ADHD symptoms, the patient was resumed on her regimen of clonidine and the dosage was adjusted to 0.2mg at bedtime. The pediatric endocrinologist suggested adding atenolol 25mg daily for symptom control of thyrotoxicosis with the plan to continue it until the patient's free T4 would normalize or decrease in value. The endocrinologist also recommended continuous monitoring of the patient's vital signs.

During the hospitalization, the atenolol dose was titrated to 25mg in the morning and 12.5mg after dinner for a total of 37.5mg/day. Following atenolol treatment for a few days, the patient appeared less jittery and anxious, more attentive, better able to maintain eye contact, and her blood pressure values improved along with the normalization of her heart rate. Overall, the patient's symptoms ameliorated both subjectively and objectively. She denied enuresis throughout her hospital stay.

The patient was eventually discharged home with her older sibling after obtaining consent from her case worker. Her primary diagnoses at discharge included adjustment disorder with mixed disturbance of emotions and conduct, ADHD, combined type, grief reaction due to the recent loss of her father, social anxiety disorder, and thyrotoxicosis possibly due to Hashimoto's thyroiditis. Follow-up appointments were scheduled with primary care, endocrinology, and psychiatry. A recommendation was made to reassess the patient's ADHD symptoms in the outpatient setting after normalization of thyroid hormone levels and to consider using first-line pharmacotherapy (e.g., psychostimulants) if ADHD symptoms persisted despite clonidine unless contraindications were present.

According to hospital records, after discharge, the patient failed to follow up with primary care. However, the pending result of TSI came back elevated at 3.42 IU/L, which was consistent with a diagnosis of Graves' disease. The pediatric endocrinologist, who had been consulted during the patient's hospital stay and was following the patient at discharge, started methimazole 10mg daily to inhibit thyroid hormone production with the plan to repeat thyroid laboratory monitoring in two to three months.

DISCUSSION

Comorbid medical conditions can exacerbate psychiatric diagnoses and complicate their treatment, especially when they share similar symptoms. The presence of undiagnosed Graves' disease in the context of ADHD was explored in a previous case study,[8] in which the undiagnosed thyroid disease greatly impacted the treatment of a patient with Tourette's disorder and ADHD and initially led to poor clinical response and polypharmacy. This is similar to the case of our patient whose worsening symptoms were initially attributed to an exacerbation of a previous diagnosis of ADHD rather than a comorbid thyroid disease. However, after the discovery of abnormal thyroid functioning, the patient's worsening clinical presentation was largely ascribed to the co-occurring diagnosis of thyrotoxicosis given the fact that symptoms greatly improved following the initiation of treatment with atenolol.

This case warns the clinicians of how easily serious medical conditions like thyrotoxicosis could be missed and left untreated in patients with a history of ADHD if laboratory tests (in this case TSH) are not routinely ordered. For our patient, the delay in early detection and treatment of her medical condition was compounded by being in the foster care system, the lack of consistency in case management, and the numerous difficulties related to continuity of medical care associated with placement instability. Therefore, in inpatient psychiatric settings, the recommendation is to order routine laboratory tests, such as TSH, for all patients who are admitted and to start treatment (i.e., "Test and Treat,") for medical conditions impacting psychiatric presentations.

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Keywords: attention deficit disorder with hyperactivity, graves disease, thyrotoxicosis

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Case Report

The Effectiveness of Buprenorphine Transdermal Patch and Low Dose Sublingual Buprenorphine Induction to Transition to Long-Acting Subcutaneous Buprenorphine Injection in Opioid Use Disorder in Inpatient Setting

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INTRODUCTION

Buprenorphine, available in various formulations, has demonstrated safety and efficacy in treating opioid use disorder (OUD).[1] All forms of buprenorphine possess a unique pharmacological profile, functioning as a high-affinity partial agonist at μ-opioid receptors.[2] With an extended serum half-life and prolonged duration of action compared to traditional full agonists, it presents an attractive option for OUD treatment, as it induces less tolerance and poses a lower risk of respiratory depression.[1]This combination of factors makes it an appealing option for OUD as it produces less tolerance, and carries a lower risk for respiratory depression.[1] Moreover, buprenorphine can alleviate cravings and binds to the μ-receptor more strongly than other opioids such as fentanyl and heroin, thereby reducing the likelihood of overdose in cases of relapse while on buprenorphine therapy; however, it can precipitate withdrawal symptoms in the presence of an agonist. This risk can be mitigated by initiating treatment with a low dose and closely monitoring withdrawal symptoms using the Clinical Opiate Withdrawal Scale (COWS).[3]

Although FDA-approved only for pain, buprenorphine transdermal patch presents an excellent alternative for the micro induction of buprenorphine.[4] Micro induction is a novel approach that overcomes need for prolonged opioid tapers and reduces risk of precipitated withdrawals. Micro induction can be done both in inpatient and outpatient settings. The buprenorphine transdermal patch, typically initiated in the inpatient setting, helps reduce opioid withdrawal symptoms.[5-7] Its use as a rapid induction technique with transition to long-acting injectable (LAI) buprenorphine is proposed in this case study. The patch's dosage ranges from 5 to 20 micrograms per hour which is relatively low compared to oral and sustained released formulations.[4] Safety should be assured by discussing risks and benefits with patients. Ingesting or chewing the transdermal patch can pose serious risks due to the sudden release of buprenorphine, potentially resulting in a stronger high, and the potential for adverse consequences far outweighs any perceived benefits.[8]

One of the goals of this micro induction technique is sustained adherence to the OUD treatment, which could be accomplished simply by avoiding multiple daily doses of sublingual (SL) buprenorphine.[9] By giving LAI buprenorphine injection following discharge on buprenorphine transdermal patch, the authors aimed to ensure lasting adherence and reduce the likelihood of relapse.[10]

CASE REPORT

A 21-year-old male with a history of OUD and cannabis use disorder presented to the addiction clinic after initiating SL buprenorphine-naloxone 8-2 mg three times daily during an Emergency Department (ED) visit. He took SL buprenorphine for five days but had to discontinue due to precipitated opioid withdrawals (POW) from using fentanyl. Subsequently, he was reinitiated in the clinic with the standard method, abstaining from opioid use for 24 hours before initiating SL buprenorphine, but experienced POW again with a relapse to fentanyl. Despite multiple ED and office visits, attempts to transition him from fentanyl to buprenorphine were unsuccessful. As a result, the patient was admitted to the hospital for opioid withdrawal and micro induction of buprenorphine, as he declined methadone for OUD and outpatient micro induction of buprenorphine.

The patient reported using fentanyl three hours before admission, and his urine drug screen (UDS) was positive for both fentanyl and THC. Upon admission, a transdermal buprenorphine patch (10mcg/hour) was initiated, and the course of micro induction is detailed in Table 1. Medications to manage withdrawal symptoms were available as needed and administered as indicated in Table 1. The patient completed the micro induction successfully without significant events. He experienced minimal withdrawal symptoms, with his COWS[3] never exceeding 2, and expressed satisfaction with the process. Although he expressed anxiety about receiving the LAI buprenorphine injection, he received it successfully and expressed a commitment to maintaining abstinence from opioids in the future.

DISCUSSION

Buprenorphine micro induction is an increasingly utilized technique. This case study aimed to highlight the significant role of transdermal buprenorphine and SL low-dose buprenorphine induction in transitioning from high-potency synthetic opioids (HPSO) to LAI buprenorphine injection. This approach involved initiating buprenorphine at a continuous low dose, gradually increasing it until reaching a therapeutic level.[5] This facilitates the prompt initiation of SL buprenorphine at increased doses over several supervised days. Subsequently, LAI buprenorphine can be administered once the patient is opioid-free and stabilized on an oral dose of SL buprenorphine.[7]

Previous studies have utilized low doses (0.2 mg - 0.5 mg) of SL buprenorphine for induction, even though these higher doses and formulations differ notably from transdermal buprenorphine patch.[9,10] Menard et al.[11] described patients successfully being transitioned from methadone to SL buprenorphine using the patch to avoid severe withdrawal symptoms, but these patients were already established on methadone for OUD. Additionally, the efficacy of transdermal buprenorphine in noncancer chronic pain, particularly when transitioning from prescribed opioids in the outpatient setting, is proven,[12] although at very low doses. Kornfeld et al.[12] indicates the patch is efficacious in reduction of withdrawal symptoms in the transition to SL buprenorphine. These efficacies could implicate transdermal

Table 1. Dose and patient response through the course of admission until outpatient injection.

	Transdermal Buprenorphine Patch	BPN, Twice Daily	COWS	Withdrawal Symptoms	Medication, Indication	Vitals
Day 1	10 mcg/hr.	1 mg bid	1	Anxiety, fatigue, insomnia, GI upset	Clonidine 0.1 mg PO, anxiety	HR 71 RR 16 BP 145/72 T 36.8
Day 2	10 mcg/hr.	1 mg qid	2	Lower back soreness, anxiety	Hydroxyzine 25 mg PO, anxiety Trazodone 50 mg PO, insomnia	HR 48 RR 18 BP 135/72 T 37.2
Day 3	10 mcg/hr.	1 mg eight times a day	1	Rhinorrhea, sneezing	Promethazine 12.5 mg PO, nausea	HR 57 RR 18 BP 125/68 T 37.1
Day 4	10 mcg/hr.	8 mg bid	1	Anxiety	None	HR 42 RR 19 BP 107/51 T 36.8
Day 5	300 mg Buprenorphine extended-release injection, outpatient		0	None	None	HR 92 BP 108/95 T 36.8 RR 18

buprenorphine as an appropriate initial agent in the treatment of chronic pain or induction of buprenorphine in multiple settings while reducing the chronic opioid burden on patients.[13,14]

The secondary goals of this method are increased adherence and success of buprenorphine, attributed to its supervised nature. While prior research has shown the effectiveness of outpatient induction compared to supervised methods, it is important to note that not all patients may have access to inpatient care, as seen in this case study.[6,11] This inpatient technique eliminates the need for patients to manage their own induction and abstinence. It could provide a valuable option to patients who are hesitant or anxious to withdraw but willing to do so in a supervised way.

CONCLUSIONS

The need for new approaches to treat OUD is critical due to the changing and unpredictable supply of non-prescribed opiates. Transdermal buprenorphine, known for its safety, has shown effectiveness in inpatient buprenorphine induction, minimizing withdrawal symptoms. Further research is required to evaluate its efficacy in outpatient settings and different patient populations. Widespread acceptance of transdermal buprenorphine for induction could enhance access for hesitant patients and improve the success rate of buprenorphine induction.

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Keywords: buprenorphine, opioid-related disorders, substance withdrawal syndrome

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Case Report

Lemierre's Syndrome Caused by *Streptococcus pneumoniae* in a Patient with Carbimazole-Induced Severe Neutropenia

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INTRODUCTION

Lemierre's syndrome is characterized by septic thrombophlebitis of the internal jugular vein, typically originating from oropharyngeal infection and accompanied by surrounding soft tissue inflammation, septic emboli, and persistent bacteremia.[1] This rare condition had a higher incidence of 1/1,000,000 in the pre-antibiotic era, but its occurrence has significantly diminished since the introduction of antibiotics in the 1960s.[2] In recent times, there is a renewed focus on Lemierre's syndrome, possibly influenced by changes in antibiotic prescription patterns for conditions like pharyngitis or tonsillitis.[3] The primary causative agent is often *Fusobacterium necrophorum*, a normal human oropharyngeal microflora. Other implicated organisms include *Fusobacterium nucleatum*, Streptococcus species (such as *Streptococcus pyogenes* and the *Streptococcus milleri* group), *Hemophilus influenzae*, *Escherichia coli*, *Eikenella corrodens*, and *Bacteroides*.[4,5] Methicillin-resistant *Staphylococcus aureus* (MRSA) has also been identified as a rare cause of Lemierre's syndrome.[6]

We present a case involving a 22-year-old female with Graves' disease undergoing anti-thyroid drug treatment. The patient developed drug-induced neutropenia, subsequently leading to Lemierre's syndrome secondary to *Streptococcus pneumoniae* and an underlying lower respiratory tract infection.

CASE REPORT

A 22-year-old female, previously diagnosed with Graves' disease and currently on a regimen of Carbimazole 10mg twice daily for the past three months, presented to the emergency department with complaints of fever, neck pain, neck swelling, multiple oral ulcers, and odynophagia persisting for four days. Upon arrival, the patient exhibited a fever of 103°F, tachycardia (heart rate: 133/min), tachypnea, and low blood pressure measuring 60/30 mm Hg. Physical examination revealed diffuse neck swelling with induration, tenderness, and erythema.

Meeting the criteria for severe sepsis, the patient received intravenous fluid resuscitation following standard guidelines. However, as blood pressure did not respond to fluid resuscitation, she was diagnosed with septic shock, prompting the initiation of vasopressors. Two sets of blood cultures were obtained, and broad-spectrum antibiotics were promptly administered. Further investigations revealed leukopenia with an absolute neutrophil count (ANC) of 10, elevated inflammatory markers (including an increased erythrocyte sedimentation rate and C-reactive protein), and elevated procalcitonin values (Table 1).

Table 1. Results of laboratory and radiological investigations.

Investigation	Findings
Hemoglobin	10.0 g/dL (13-16 g/dL)
Total Leucocyte count	0.21 x 10^3 cells/uL (4000-11000/ul)
Absolute Neutrophil count	10 cells/ ul
Platelet count	418 x 10^3 /ul (150 x 10^3 – 450 x 10^3/ul)
Blood film morphology	RBC-Microcytic hypochromic RBC picture with many pencil cells and few target cells and elliptocytes WBC-Marked leucopenia and neutropenia DLC (%)-25 cells counted: Neutrophil = 0, Lymphocyte = 25, Monocyte = 0 Platelets- Increased in smear (5,30,000)
Urea	63 mg/dL (6-24 mg/dL)
Creatinine	0.74 mg/dl (0.7-1.4 mg/dL)
Bilirubin – Total	1.46 mg/dL (0.3-1.2 mg/dL)
Bilirubin- conjugated	0.76 mg/dl (<0.2 mg/dL)
Aspartate transaminases (AST)	45 IU/L (8-33 IU/L)
Alanine transaminases (ALT)	32 IU/L (8-48 IU/L)
Alkaline phosphatase (ALP)	96 IU/L (30-120 IU/L)
Total Protein	6.94 g/dL (6.6 -8.3 g/dL)
Serum Albumin	2.97 g/dL (3.5-5.2 g/dL)
Serum Globulin	3.97 g/dl (2.0-3.5g/dL)
Procalcitonin	20.4 ng/dl (<0.02ng/dL)
CRP	358.9 mg/dl (less than 5 mg/dL)
ESR	74 mm/hr (0-20 mm/hr)
Serum cortisol	44.38 ug/dL
Prothrombin time	22.6 seconds (11-14 seconds)
INR	1.77
Thyroid profile	TSH - 0.1mIU/L (0.35-5.50 mIU/L) FREE T3 - 1.19pg/ml (2.2-4.20 pg/ml) FREE T4 - 0.58ng/dl (0.89-1.70 ng/dL)
CECT NECK	Left IJV thrombosis with bilateral multilevel cervical lymphadenopathy and diffuse retropharyngeal and parapharyngeal edema

Abbreviations: DLC: Differential leucocyte count; RBC: Red blood cells; WBC: White blood cells; ESR: Erythrocyte sedimentation rate; CRP: C- reactive protein; INR: International Normalized Ratio; TSH: Thyroid Stimulating Hormone; CECT: Contrast-enhanced computed tomography; IJV: Internal Jugular Vein

The patient was suspected to have oropharyngeal infection, leading to the addition of metronidazole to the treatment plan. A contrast-enhanced computed tomography (CECT) of the neck with thorax was performed, revealing left internal jugular vein thrombosis with diffuse retropharyngeal and parapharyngeal edema extending from C2-C6 levels. The imaging also showed multi-level cervical lymphadenopathy and bilateral pneumonia (Figure 1).

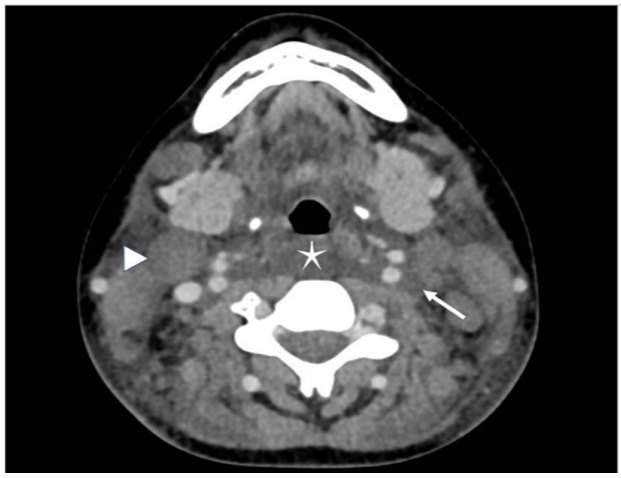

Figure 1. CECT Neck axial section. It shows diffuse hypodensity in retropharyngeal region (*), left IJV thrombosis (arrowed), enlarged right level II cervical lymph node (arrowhead).

The thyroid gland exhibited diffuse enlargement. Thyroid function tests indicated significantly low levels of thyroid-stimulating hormone, free T3, and free T4, collectively suggestive of hypothyroidism, raising the possibility of drug-induced hypothyroidism. Transthoracic echocardiography revealed a collapsed inferior vena cava alongside normal cardiac contractility.

As the patient gradually improved, vasopressors were tapered over a span of three days. On the third day, blood culture results identified *Streptococcus pneumoniae*, susceptible to Ceftriaxone, Levofloxacin, Linezolid, and Vancomycin. Ceftriaxone was continued, while vancomycin and metronidazole were discontinued on the fourth day of hospitalization. To address the low ANC, Injection Filgrastim was administered for three days. A PET-CT scan conducted revealed the presence of septic emboli in the lungs and kidneys, with no involvement observed in the brain or other organs (Figure 2).

An Ear, Nose, and Throat (ENT) surgeon was consulted and determined that surgical intervention was unnecessary. Intravenous ceftriaxone was continued for a total of 14 days. Subsequent cultures sent on day 7 and day 10 yielded sterile results. The patient's condition improved gradually with intravenous antibiotics (ceftriaxone), and no surgical intervention was required. She was discharged on day 14 of her hospital stay.

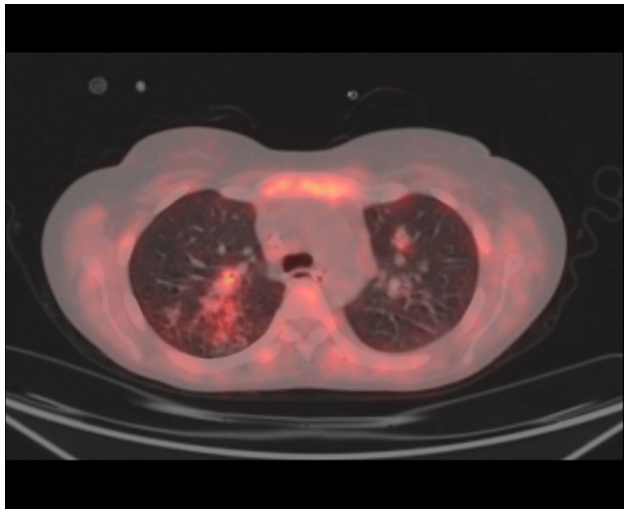

Figure 2. PET-CT with Thorax (axial section). It is suggestive of hypermetabolic lesions in parapharyngeal areas and bilateral lung fields.

DISCUSSION

Lemierre's syndrome, initially described by French physician Andre Lemierre in 1936 as post-pharyngitis anaerobic sepsis, is a rare and potentially fatal condition characterized by septic thrombophlebitis of the internal jugular vein resulting from oropharyngeal bacterial infections.[2] *Fusobacterium necrophorum* is the most commonly implicated organism, with *Fusobacterium nucleatum* following closely behind, and pharyngitis or tonsillitis serving as the causative focus in over 85% of cases.[7] Other causative organisms encompass *Staphylococcus* (including MRSA), *H. influenzae*, *Eikenella corrodens*, *Porphyromonas asaccharolytica*, *Bacteroides*, and Enterobacteriaceae. While Streptococci can contribute to Lemierre's syndrome, infections are generally associated with *Streptococcus viridans*, *Streptococcus pyogenes*, and *Streptococcus milleri* species. It is exceptionally rare for Lemierre's syndrome to be secondary to *Streptococcus pneumoniae*. In our patient, the diagnosis of Lemierre's syndrome was established based on clinical manifestations including fever, neck swelling, internal jugular vein thrombosis, and positive blood cultures.

Streptococcus pneumoniae, typically regarded as a commensal in the nasal mucosa, occasionally presents as clinically significant skin and soft tissue infections (SSTIs). These SSTIs, attributed to *Streptococcus pneumoniae*, constitute less than 2% of all streptococcal infections but can lead to severe conditions, including necrotizing fasciitis.[8,9] Such SSTIs are predominantly observed in immunocompromised individuals, encompassing those with haematological malignancies, Systemic Lupus Erythematosus, and diabetes. In our case, the patient exhibited a significant state of immunocompromise, characterized by a persistently low total leukocyte count and ANC. The diminished ANC was identified as the primary risk factor contributing to the *Streptococcus pneumoniae* infection and the subsequent development of Lemierre's syndrome in our patient.

Lemierre's syndrome can give rise to various complications, including internal jugular vein (IJV) thrombosis and septic emboli affecting organs such as the lungs, joints, and brain. Additionally, this syndrome has the potential to induce bacteremia, leading to severe conditions like osteomyelitis, acute respiratory distress syndrome, and septic shock.[10] In our patient's case, the presence of both IJV thrombosis and septic shock was observed, indicating the progression of Lemierre's syndrome.

The management of Lemierre's syndrome involves systemic administration of antibiotics and surgical intervention if necessary. Antibiotic therapy plays a pivotal role and should be guided by microbiological etiology and antimicrobial susceptibility. Effectiveness has been noted with the use of penicillin, carbapenem, or piperacillin/tazobactam, often combined with metronidazole. While the optimal treatment duration remains uncertain, a regimen of intravenous antibiotics lasting two weeks, followed by a total of four to six weeks, appears sufficient. However, the duration should be personalized considering disease severity and ongoing evaluations.[11] There is no specific recommendation regarding the use of anticoagulation therapy in the treatment of Lemierre's syndrome.

A high suspicion of disease is crucial for prompt diagnosis. Pneumonia or empyema is the most common primary site of infection in *Streptococcus pneumoniae*-associated Lemierre's syndrome.[12] Early initiation of antibiotics and the management of complications are key components in patient care and are associated with improved outcomes. While Lemierre's syndrome secondary to *Streptococcus pyogenes* and the *Streptococcus milleri* group have been reported, Lemierre's syndrome secondary to *Streptococcus pneumoniae* has not been documented in the literature. This case is likely the first reported instance. Although *Streptococcus pneumoniae* commonly causes skin, lung, and central nervous system infections, the consideration of life-threatening Lemierre's syndrome as a rare manifestation is essential. Prompt treatment with sensitive intravenous antibiotics and timely de-escalation are crucial for early recovery and to prevent antimicrobial resistance.

CONCLUSIONS

Lemierre's syndrome, marked by septic thrombophlebitis of the internal jugular vein, is a rare and potentially life-threatening condition linked to oropharyngeal infections. Our case involves a young immunocompromised female who developed Lemierre's syndrome due to an *Streptococcus pneumoniae* infection originating from the lower respiratory tract, underscoring the unusual etiology and significance of this condition. Timely diagnosis, prompt initiation of appropriate antibiotics, and effective management of complications are imperative for favorable outcomes.

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Keywords: Lemierre's syndrome, Otorhinolaryngologic infection, Streptococcus pneumoniae, Carbimazole associated neutropenia, thrombophlebitis

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