

## **A Multiple-Stakeholder Perspective of Patient–Provider Communication Among Families With Rare Diseases in Taiwan Through a Cross-Cultural Lens**

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Using a cultural lens and engaging all stakeholders' perspectives (e.g., patients, family caregivers, and health care and community providers), this study aimed to understand risk factors that hinder effective patient–provider communication (PPC) among families experiencing rare diseases in Taiwan. Findings from five focus group discussions with 40 participants recruited using purposive sampling suggest various risk factors related to

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effective PPC, including high regard for authority, face-saving communication, keeping harmonious relationships with providers, low socioeconomic status, patients' and caregivers' limited disease literacy, symptom-oriented communication habits, limited channels for timely PPC, discordant information from different sources, forgetting to keep daily records of the disease, and confusion about seeking specialists' assistance. Families with rare diseases in Asian communities would benefit from culturally tailored services and educational programs to enhance their disease-specific health literacy, motivation, engagement, and communication skills. Structural barriers could be tackled by providing integrated care coordinating cross-departmental communication between medical and social services.

*Keywords: patient–provider communication, rare diseases, culture, focus group, qualitative research*

Rare diseases are medical conditions with an extremely low prevalence rate. Even though the prevalence of each rare disease is low, the total number of rare diseases is estimated to exceed 10,000, affecting as much as 10% of the global population (Haendel et al., 2020). The threshold that distinguishes a rare disease varies by country. In the United States, rare diseases are defined as any disease affecting fewer than 200,000 people in the country (Herder, 2017), whereas the European Union's threshold is fewer than 5 per 10,000 people (Richter et al., 2015). Taiwan's threshold is fewer than 1 in 10,000 people, which is stricter than most other countries (Song, Gao, Inagaki, Kokudo, & Tang, 2012). These patients face not only unique challenges relevant to quality of care (von der Lippe, Diesen, & Feragen, 2017) but also different patient–provider relationships compared with those with more common medical conditions (Liuccio, Belotti, Comune, Zambito, & Schulz, 2015).

Patient–provider communication (PPC) in the process of treating rare diseases is characterized by nontraditional dynamics. There are various types of rare diseases; however, the challenges experienced by patients with rare diseases are similar, including inexperience of most clinicians with diagnosing or treating most rare diseases, resulting in delayed or wrongful diagnoses and suboptimal clinical management (Haendel et al., 2020). Western literature shows that health care providers usually lack sufficient expertise relevant to rare diseases, and patients are often forced to become an expert regarding the disease, leading to nonconventional role discrepancies between providers and patients and subsequently malfunctioning PPC patterns (Budysh, Helms, & Schultz, 2012; Liuccio et al., 2015). PPC has been recognized as a substantial determinant of patients' health outcomes and quality of care (Bensing, 1991; Duberstein, Meldrum, Fiscella, Shields, & Epstein, 2007).

In recent years, the occurrence of rare diseases has increased in Taiwan, as has expenditures associated with the treatment of rare diseases. Specifically, from 2003 to 2014, the prevalence of rare diseases increased from 10.57 to 33.21 per 100,000, associated total health expenditures increased from \$18.65 million to \$137.44 million, and associated drug expenditures increased from \$13.24 million to \$121.98 million (Hsu et al., 2018; Lin, Lin, & Hung, 2013). The increasing burden of rare diseases is further complicated by ecological factors such as uneven distribution of resources, slower flow of information, and

relatively insufficient medical resources in rural areas in Taiwan (Fan & Chiu, 2016). These patients often miss medical appointments or do not get necessary care in time, thus affecting treatment efficacy and their quality of life. Previous studies on rare diseases mostly involved in-person interviews from the patient perspective only (Budysh et al., 2012; Liuccio et al., 2015), overlooking diverse insights provided by multiple stakeholders such as medical providers, community service providers, and caregivers regarding PPC. In addition, PPC has been found to be influenced by cultural norms (Lwin & Salmon, 2015; Pun, Chan, Wang, & Slade, 2018). However, research on PPC has mostly been conducted in Western societies and rarely examined communication barriers or needs experienced by patients with rare diseases in Eastern countries characterized by a collectivist culture, such as Taiwan (Schieppati, Henter, Daina, & Aperia, 2008).

Hence, to fill the gap in the literature, this exploratory study examined barriers affecting PPC experienced by families with rare diseases in Taiwan through a culture-specific lens by conducting focus groups with diverse stakeholders. The current study gained in-depth perspectives from 40 stakeholders, including patients with rare diseases (i.e., epidermolysis bullosa, phenylketonuria, Prader-Willi syndrome, Huntington's disease, maple syrup urine disease, Angelman syndrome, achondroplasia), caregivers, health care providers, social workers, and frontline workers at community-based agencies relevant to rare disease services. This study is the first to provide a comprehensive view of barriers to PPC at different levels among families with rare diseases in a collectivist culture. Findings from this study provide important implications for professionals from multiple disciplines regarding providing culturally tailored clinical practices and programs for families with rare diseases in Asian societies with collectivist cultures to improve their PCC literacy and quality of life.

## **Literature Review**

### ***Challenges of Patient–Provider Communication***

#### *Role Discrepancies*

In contrast to the traditional patient–provider dynamic in the process of seeking treatment, patients with rare diseases have unique experiences with multiple providers in the health care system. The most common barrier in PCC identified in previous studies is role discrepancies between patients and professionals (Budysh et al., 2012; Liuccio et al., 2015). Rare diseases often require highly specialized treatment, yet health care providers usually lack expertise relevant to specific rare diseases. This insufficient knowledge can lead to delayed diagnosis, mistreatment, or denial of medical services among patients. As a result, patients with rare disease are left with no choice but to become experts and gain control over the communication process, which represents a shift from traditional patient and professional roles in which physicians dominate the medical encounter (Budysh et al., 2012).

In a systematic review of qualitative studies regarding the experiences of patients with rare diseases in the health system, von der Lippe et al. (2017) found that they acted as advocates for their health and acquired access to information about their diseases on the Internet and through support groups. Patients and their caregivers often felt the need to exercise self-agency and ensure the quality of treatment they receive. They also described unmet needs regarding a holistic treatment perspective and the

importance of coordinated services among health professionals (von der Lippe et al., 2017). Hence, both patients with rare diseases and providers need to comprehend and accept the role expectations of patients as “informed, engaged, and interactive” (Liuccio et al., 2015, p. 4) partners in the therapeutic process, rather than passive recipients of health care services.

Although these studies shed light on PPC in the context of rare diseases, their findings were limited to communication between patients and physicians, leaving out constructive inputs from other medical professionals who are prominent in the treatment of rare diseases such as nurses and social workers. As such, Budysh et al. (2012) suggested that future studies should examine both patients’ and physicians’ perspectives and explore the decision-making style of providers. Taking a multiple-stakeholder approach, Pun et al. (2018) reviewed extant research on roles and expectations of nurses, clinicians, patients, and family members to learn about PPC in the East Asian cultural context. However, their review study excluded patients with rare diseases.

#### *Health Literacy and Socioeconomic Status*

Health literacy is defined as “the degree to which individuals have the ability to find, understand, and use information and services to inform health-related decisions and actions for themselves and others” (Centers for Disease Control and Prevention [CDC], 2022, p. 2). Using qualitative methods, Leung, Bo, Hsiao, Wang, and Chi (2014) explored possible reasons why Chinese patients have difficulty with health literacy regarding diabetes-related information and identified three barriers: cultural, structural, and personal factors. Examining the prevalence of inadequate health literacy in the United States, Williams, Davis, Parker, and Weiss (2002) found that it affected PPC through compliance with recommended treatments, understanding the physician’s vocabulary, validity of the medical history, and validity of medical tests, which may affect health outcomes. In the context of rare diseases, scholars suggested that health literacy and capacity building is a key arena to empower patients living with rare diseases and contribute to a sustainable and resilient health care system (de Santis, Hervas, Weinman, Bosi, & Bottarelli, 2019).

Low socioeconomic status (SES) seems to be associated with low levels of disease knowledge and health literacy, which influences the quality of PPC, whereas patients with higher SES ask more questions and receive more medical information from providers (Hironaka & Paasche-Orlow, 2008; Verlinde, De Laender, De Maesschalck, Deveugele, & Willems, 2012). It was also found that patients from low-income families received more directive and less informative care and participated less in consultations than patients of higher SES (Willems, De Maesschalck, Deveugele, Derese, & De Maeseneer, 2005).

#### ***Asian Collectivist Culture in Patient–Provider Communication***

Previous studies in Western societies found that current models of health care communication propagated a patient-centered approach to delivering health care (McCarthy et al., 2013; Naughton, 2018; Pun et al., 2018). Western cultures emphasize individuals’ needs and rights when making a treatment decision. In contrast, in Asian collectivist cultures, a patient is part of a larger social unit, suggesting that Asian culture favors a family-centered model that places a high value on family decision-making (Ishikawa & Yamazaki, 2005). Pun et al. (2018) found that both collectivist and individualist values exist in East Asian

health care contexts. There has been a gradual change toward more recognition of patient autonomy, the right to be active participants in shared decision-making, and the right to be fully informed, as explicitly or implicitly indicated by many studies. Pun et al. (2018) suggested a need to consider local culture in understanding and interpreting medical encounters in East Asia, especially the need for a specific culturally appropriate model of health communication in East Asia. Leung et al. (2014) identified three cultural factors that might affect different components of health literacy among Chinese immigrants with diabetes in the United States: high regard for authority, a desire to avoid being burdensome to others, and a desire to be together or follow a collective approach. These factors characterize collectivism, a core element of Chinese culture (Wang & Liu, 2010).

PPC, as a form of communication, should be influenced by cultural characteristics of general communication. Rooted in traditional Confucian cultural values and a yin-yang perspective, scholars theorize that Chinese communication is marked by implicit communication (i.e., communicating in a reserved, indirect, and suggestive manner rather than being direct and articulate), listening-centered communication (i.e., entitlement to speak is deeply associated with seniority, hierarchy, and expertise), polite communication (i.e., showing respect, which is rooted in the Confucian notion of harmony), insider communication (i.e., readiness to communicate with individuals in a network and hesitance or avoidance related to communicating outside network), and face-directed communication (i.e., emphasis on personal reputation and prestige; Fang & Faure, 2011; Gao & Ting-Toomey, 2012). How these Chinese communication characteristics affect clinical PCC deserves special attention.

This study is the first to examine PPC among families with rare diseases in a collectivist culture (Taiwan) through in-depth discussions involving 40 diverse participants: patients with rare diseases, caregivers, health care providers, social workers, and frontline workers at community-based health agencies relevant to rare diseases.

## **Method**

### ***Recruitment Procedure***

Using a purposive qualitative sampling method, service recipients (patients and family caregivers) and service providers (physicians, genetic counselors, nurses, dietitians, occupational therapists, behavioral therapists, social workers, and frontline service providers at community-based rare disease agencies) were referred by research collaborators such as medical providers at a local hospital's genetic counseling center and staff members working in rare disease nongovernmental organizations. Eligible patients needed to be officially diagnosed by a physician as having a rare disease. The types of rare diseases among patient participants included epidermolysis bullosa, phenylketonuria, Prader-Willi syndrome, Huntington's disease, maple syrup urine disease, Angelman syndrome, and achondroplasia. Eligible service providers needed to have provided services for patients with rare diseases for more than one year to ensure adequate knowledge on the topic. Those who did not meet inclusion criteria or had (a) inadequate Chinese listening and speaking skills or (b) physical or mental conditions that prevented participation in focus group discussions were excluded from the study. Eligible referred individuals were informed of the purpose of the study. Forty eligible participants consented to participate in the study, including 6 patients with rare diseases, 7 family

caregivers, 9 medical providers, and 18 community service providers. Table 1 shows the demographic characteristics of all participants.

**Table 1. Demographic Characteristics of Participants (N = 40).**

	Patients (n = 6)	Family Caregivers (n = 7)	Medical Service Providers (n = 9)	Community Service Providers (n = 18)
Age in years, <i>M</i> ( <i>SD</i> )	25.3 (10.6)	49.7 (7.4)	38.4 (7.2)	40.0 (7.1)
Gender				
Male	6 (100)	1 (14)	2 (22)	0 (0)
Female	0 (0)	6 (86)	7 (78)	18 (100)
Education level				
High school or less	3 (50)	5 (71)	0 (0)	2 (11)
College degree	3 (50)	2 (29)	4 (44)	10 (56)
Master's degree or beyond	0 (0)	0 (0)	5 (56)	6 (33)
Marital status				
Single	5 (83)	0 (0)	1 (11)	6 (33)
Married	1 (17)	5 (72)	8 (89)	11 (61)
Live alone	0 (0)	1 (14)	0 (0)	0 (0)
Divorced	0 (0)	1 (14)	0 (0)	1 (6)
Years in practice				
1–3			1 (11)	1 (6)
4–6			1 (11)	1 (6)
7–9			0 (0)	3 (16)
10 or more			7 (78)	13 (72)

Note. Figures reflect *n* (%).

### Data Collection

Five focus group discussions were moderated by the principal investigator between March and April 2018 at local coffee shops, meeting rooms in the library of a hospital, and community agencies in Taiwan. Participants filled out a questionnaire that collected demographic data before focus group discussions started. Each focus group interview lasted 90 minutes and was conducted in Mandarin Chinese. The moderator guide included open-ended, nonleading questions, allowing any topic to surface freely in the discussion. Participants discussed status quo, challenges, and needs related to PPC and quality of care experienced by families with rare diseases in Taiwan. The wording and language of the interview prompts were adjusted in each role-specific focus group in response to the participants' varying literacy level. An example focus group question for patients and family caregivers is: "How do doctors and other health care workers usually talk with you?" An example question for physicians and nurses is: "What challenges do you face in communicating with your patients with rare diseases?" The moderator allowed the discussion to

cover any topics and used soliciting probes such as “Can you tell me more about that?” and “What does everyone else think about that?” to encourage elaboration by participants and input from other members in the focus group. The institutional review board at Hualien Tzu Chi Hospital approved the study in 2017.

### ***Data Analysis***

All focus groups were audio recorded and transcribed verbatim for analysis. Transcripts were imported into NVivo qualitative software for analysis. Thematic content analysis, a method that portrays the thematic content of interview transcripts by identifying common themes (Anderson, 2007), was used to analyze the qualitative data. Thematic content analysis is capable of analyzing the richness of individuals’ personal experiences in novel areas (Griffith et al., 2011). An inductive approach was adopted because the population of patients with rare diseases in Taiwan was previously unexplored and knowledge about factors that affect their PPC and quality of care is fragmented (Elo & Kyngäs, 2008). In a bottom-up process, inductive thematic content analysis derives themes from the data without predetermined ideas (Elo & Kyngäs, 2008).

The coding process was informed by the steps outlined by Anderson (2007) and Elo and Kyngäs (2008). In the initial open-coding stage, three coders first independently reviewed all transcripts to become immersed in the content (Lawrence & Tar, 2013). The coders then examined the transcripts line by line, highlighting all meaningful descriptions relevant to the topic of inquiry, marking each distinct unit of meaning, and writing notes and headings to describe all aspects of the content.

In the next stage, they separately compared notes and headings and grouped similar units into higher-order themes. The coders modified the themes as they integrated more units. Once they incorporated all units, they reviewed, compared, and redistributed all meaning units in each category as appropriate. They also relabeled, collapsed, or subdivided themes in this process. They repeated this process until each coder felt all possible themes had been identified. Once all three coders generated their individual set of themes based on personal interpretation, they had weekly meetings to review their themes and resolve discrepancies. They repeated the process of redistributing units, reorganizing themes, and relabeling themes. If unresolvable discrepancies arose, the principal investigator was consulted. Three coders also conducted the abstraction process, which involved formulating a general description of the research topic through generating categories. Through this process, the three coders collectively worked toward a finalized codebook when satisfied that data saturation had been achieved. To ensure intercoder agreement, the coders independently applied the codebook to sections of data. The coders obtained a Cohen’s kappa score of .86, which indicates high interrater reliability. They analyzed the data in the original language, Mandarin Chinese. After they finalized the codebook, a bilingual research assistant translated the data related to key themes into English for publication purposes. A senior researcher oversaw the accuracy of the translation.

### **Results**

Themes relevant to barriers to quality of PPC emerged from focus group discussions with patients with rare diseases, family caregivers, medical professionals, community service providers, and other staff members in rare disease organizations. Table 2 shows the overarching themes, subthemes, source roles, and exemplar excerpts.

**Table 2. Themes and Quotes.**

Theme	Role of Participant		Example Quotes
	Medical Provider	Patient or Caregiver	
<i>Barriers to quality communication</i>			
High regard to authority	✓		They just follow medical providers' instructions and do whatever [they are asked to do].
Face-saving communication	✓		At the time of explanation, they did not make us feel that they had any confusion. On the contrary, they make you feel that they know it very well.
Keeping harmonious relationships with providers	✓		They have doubts about your words, but they won't express them at the moment.
Low socioeconomic status of the patient's family	✓		The parents do not have a good grasp of the medical concept. . . . This is especially difficult for children of unprivileged families living in Hualien County.
Limited disease knowledge and health literacy	✓		They just rambled on all kinds of symptoms patients have. However, they didn't know which of these symptoms were actually related to the disease.
Symptom-oriented communication habits		✓	I only come back when there is a [medical] issue.
Limited channels for timely PPC		✓	Sometimes he will have [medical] issues. . . . We want to ask doctors immediately how to deal with this issue.



Consolidation of disagreeing information obtained within and outside of patient-provider communication.		✓		The book was somehow different from what the doctor told me.
Forgetting to keep daily records of the disease	✓		✓	We forget to bring up questions that we had been wanting to ask.
Confusion about seeking specialists' assistance	✓		✓	I don't know which specialist to consult, what to consult and what kind of questions I could ask.
Passivity of patients and caregivers in circulating clinical information across medical encounters	✓			Parents need to take the initiative to inform us about the medical information they receive from other providers or channels . . . However, some of the parents won't tell you, or maybe they just forget.

Eleven themes referenced the effectiveness of PPC. Medical providers discussed (a) high regard for authority, (b) face-saving communication, (c) keeping harmonious relationships with providers, (d) low SES of the patient's family, (e) patients' and caregivers' limited disease knowledge and health literacy, and (f) passivity of patients and caregivers in circulating clinical information across medical encounters. Patients and family caregivers reported (g) symptom-oriented communication habits, (h) limited channels for timely PPC, and (i) discordant information from different sources. Both parties mentioned (j) forgetting to keep daily records of the disease and (k) confusion about seeking specialists' assistance. Below, these themes are discussed with direct quotes from the participants. Direct quotes are noted with participants' roles, case numbers, and genders in the following parentheses.

### ***High Regard for Authority***

Medical providers reported that patients often played passive roles in the power dynamic of PPC. Rather than clarifying points of confusion, patients considered medical providers as authoritative figures positioned high in the social hierarchy and tended to defer to medical instructions and decisions from providers. "Patients and caregivers don't express their feelings. Even after a diagnosis is made, family caregivers still know nothing about the disease. They just follow medical providers' instructions and do whatever [they are asked to do]" (Nurse, #19, female). High regard for authority led to obedience among patients and their caregivers, which undermined the quality of PPC, resulting in a lack of understanding of advice regarding medical instructions and treatment for patients with rare diseases.

### ***Face-Saving Communication***

Most medical providers reported that patients and their caregivers seemed to pretend that they had a good grasp of medical information or advice during PPC out of a desire to save face. Providers had the misleading impression that their patients and caregivers fully comprehended the information they communicated, whereas in fact they did not. "In terms of educating patient families of disease knowledge, at the time of explanation, they did not make us feel that they had any confusion. On the contrary, they make you feel that they know it very well" (Nurse, #19, female). Face-saving communication approach led to a gap in perceptions between patients and their caregivers relative to medical providers.

### ***Keeping Harmonious Relationships With Providers***

Patients and their caregivers were hesitant to express their doubts about providers' clinical advice or decisions out of a desire to maintain interpersonal harmony. They often concealed their doubts and suspicions by adopting an agreeable attitude in PPC, unless they had evidence from other sources to support their alternative viewpoints. "They have doubts about your words, but they won't express them on the spot. However, later, when they get a second opinion from other places, they will come back to question you" (Nurse, #19, female).

When patients and family caregivers had questions, they were inclined to consult multiple providers to avoid confronting any of the providers. "Our patients do want to ask doctors questions. However, most of the time, they consult many doctors and then decide by themselves which doctor's points make the most sense" (Physician, #1, male). Interpersonal harmony with providers seemed to be a major concern of patients and their family caregivers, who were cautious about being perceived as critical of providers' medical knowledge in PPC.

### ***Low Socioeconomic Status of the Patient's Family***

Many medical providers pointed out that families' low SES affected the quality of PPC, preventing help-seeking behaviors. Low SES was also associated with patients' and caregivers' inability to comprehend medical instructions from providers. "Parents of children with diseases do not have good medical concepts, so they never think that they should seek medical help immediately. This is especially difficult for children of underprivileged families" (Nurse, #3, female). Another participant said, "Even after we explain the situation of the disease, you will find out that they don't understand you and that they still do not follow your advice for caregiving. Communication with patients has something to do with their socioeconomic status" (Nurse, #19, female).

Patients and family caregivers with low SES were less likely to initiate communication, and even if they did, they were less likely to fully comprehend the medical conversation. Low SES compromised the fidelity of treatment because of poor PCC.

### ***Patients' and Caregivers' Limited Disease Knowledge and Health Literacy***

Almost all families affected by rare diseases had no prior knowledge about the disease because of its rarity. Therefore, communication was challenging during the first few medical encounters. Patients and caregivers tended to overlook the severity of the rare disease and were less motivated to engage in conversations if they felt they had low disease-specific health literacy. "Patients are referred to us for diagnosis by rare disease nonprofit organizations. However, family members themselves don't think there is a need for diagnosing the disease, so when you try to explain to them, they don't take it seriously" (Nurse, #19, female).

Because of limited disease knowledge and health literacy, patients and caregivers had difficulty differentiating crucial symptoms from less relevant symptoms. As a result, they were inclined to report symptoms indiscriminately, which compromised the efficiency of PPC.

Because the disease is rare, there are many areas that the patients and caregivers don't understand. When they called us, they rambled on about all kinds of symptoms. They didn't know which of these symptoms were actually related to the disease. It is critical to educate them at the very beginning. (Nurse, #11, female)

Limited disease knowledge and health literacy should be addressed as early in the treatment as possible because it can weaken families' motivation, engagement, and efficacy in the process of communication.

### ***Passivity of Patients and Caregivers in Circulating Clinical Information Across Medical Encounters***

Because patients and caregivers often received care from multiple providers simultaneously, providers expected updates on clinical information that patients and caregivers received from other medical encounters. To the providers' frustration, some patients and caregivers took a passive attitude when it came to disclosure of clinical information.

Parents need to take the initiative to inform us about the medical information they receive from other providers or channels, otherwise we have no way of knowing it. For example, my patient would let me know what specialties they visited last week and what the doctor said. However, some of the parents won't tell you, or maybe they just forget. We end up not knowing what they have done clinically and what things we should be aware of, according to other doctors. If they don't tell us, we simply won't know. (Nurse, #13, female)

For young patients, the responsibility largely rested with family caregivers.

[Regarding a specific child patient,] the child's dad takes the child to see the doctor by himself. We don't follow them for returning doctor visits. The dad never let us know more about those medical visits. He never informed us of what exact medical exams they have done. (Social Worker, #5, female)

The circulation of medical information was critical for comprehensive and integrated care for patients. Providers affiliated with different departments and institutions were disconnected, so they heavily relied on the self-agency of the patients to ensure the flow of information. Providers emphasized that the quality of care would be at risk and their hands would be tied if patients and caregivers did not exercise their due diligence in relaying relevant medical information.

### ***Symptom-Oriented Communication Habits***

Patients and caregivers reported that they tended to visit the hospital only when patients had symptoms that required immediate medical attention. When patients' symptoms were well managed by them without degradation, they thought there was no need for medical consultation. "I only come back when there is a [medical] issue" (Caregiver, #4, female). "I only visit medical providers when I felt uncomfortable, such as when I don't know how to deal with a certain medical issue I have" (Patient, #3, male). Patients and caregivers took a reactive rather than proactive approach to hospital visits. As a result, patients visited doctors irregularly, which may have delayed early diagnosis, treatment, and detection of changes in symptoms.

### ***Limited Channels for Timely Patient–Provider Communication***

Many family caregivers mentioned that timely PPC is critical for dealing with medical urgency in outpatient settings. Without timely medical consultation, family caregivers often needed to send patients to urgent care at local hospitals. Prompt medical consultation has the potential to reduce unnecessary visits to urgent care.

If my child suddenly begins to vomit badly, then we want to ask doctors immediately for solutions. If timely communication with providers over the phone or mobile apps solves the problem, then maybe we don't have to send our kid to the emergency care at the hospital. (Caregiver, #12, female)

Family caregivers expressed the desire for a channel enabling prompt communication with providers whenever patients had symptoms at home.

### ***Discordant Information From Different Sources***

Patients with rare diseases and family caregivers tended to supplement doctors' professional opinions with informal research through channels like the Internet, archived materials, and the rare disease community. "Sometimes I just wanted to consult the doctor on something that I read from the book, because the book was somehow different from what the doctor told me" (Caregiver, #14, female). Conflicting clinical information occasionally arose from different informational resources. Patients and family caregivers expressed the need to address these discrepancies with providers through PPC.

### ***Forgetting to Keep Daily Records of the Disease***

Some patients and caregivers reported that they were not used to keeping daily medical records. Without the habit of taking notes, they often forgot to ask questions they had at home during medical encounters. "Sometimes when we meet with doctors, we forget to bring up questions that we had been wanting to ask. We recall the questions after we have returned home" (Caregiver, #2, male).

Medical providers also mentioned that caregivers, particularly those with low SES, had difficulty tracking patients' symptoms. "Parents with low socioeconomic status living in rural areas have difficulty in documenting patients' progress" (Nurse, #19, female). Although providers might perceive medical recipients as passive and reserved in medical encounters, in fact patients and family caregivers often struggle to recall a great amount of information in a short period.

### ***Confusion About Seeking Specialists' Assistance***

Rare diseases often came with complex symptoms that needed multidisciplinary care from multiple specialists. Many patients and caregivers expressed frustration about communicating with different physicians across departments and specialties. They sometimes felt confused about which specialist was the right one for diagnosis or treatment, especially for uncommon symptoms. "Recently, my child had an issue of chewing on his hand. For issues like this, I don't know which specialist to consult, what to consult, and what kind of questions I could ask" (Caregiver, #2, male). "It is like a process of meeting the right physician for medical advice. I have been seeing pulmonologists, child pulmonologists, orthopedists, and the rehabilitation department" (Caregiver, #9, female).

Providers commented that confusion about multidisciplinary communication might cause negative treatment consequences that endanger the patient's life.

Last time, there was a patient who needed to communicate with the pediatrician, but he did not meet with the pediatrician. He thought the surgery was rather simple normally, so he went directly to the surgical department. After the surgery, the patient was sent to the ICU [intensive care unit]. My major point is that rare diseases rarely involve only one specialist. (Social Worker, #20, female)

The overwhelming burden of cross-departmental communication with multiple specialists and the high stakes involved may suggest the importance of integrated and patient-centered care for patients with rare diseases.

## **Discussion**

The objective of this exploratory study was to provide multi-informant and culturally specific evidence of barriers to quality of PPC among patients with rare diseases and their family caregivers in Taiwan. Using qualitative data collected from focus group discussions involving medical service recipients and providers with different specialties, the current study identified risk factors at multiple levels affecting

PPC and quality of care. These findings contribute to the extant literature by identifying unique challenges relevant to nontraditional PPC and quality of care experienced by patients with rare diseases in a collectivist culture (Liuccio et al., 2015).

The current study corroborated the well-documented impacts of culture on PPC in the population of patients with rare diseases in Eastern countries. Three emerging themes—high regard for authority, face-saving, and maintaining harmonious relationships with providers—reflect characteristics of Asian collectivist culture. Our findings show that patients with rare diseases and their caregivers in Taiwan followed the cultural script of having high regard for medical professionals, believing open communication would challenge providers' authority, and tending to leave treatment decisions to providers, which is consistent with findings of a cross-national study conducted among Asian immigrants with diabetes in the United States (Leung et al., 2014). Patient–provider relationships in Asia tend to be hierarchical (Claramita & Susilo, 2014; Lin, Huang, Chiang, & Chen, 2013), which hinders patients from discussing their concerns with their providers. This one-way, hierarchical, and paternalistic communication was the norm among our participants, with providers making clinical decisions and patients following them without open discussion. Our patient participants had limited disease knowledge, because indiscriminate compliance with providers prevented them from obtaining and processing information about their treatment plans (Leung et al., 2014).

The current study also identified face-saving practices in PPC among families with rare diseases in Taiwan, which is consistent with extant knowledge indicating that efforts to save face influence communication in Eastern culture (Fang & Faure, 2011). Admitting to miscommunication in PPC would not only embarrass providers but also make patients lose face. Patients with rare diseases and their caregivers often convince providers that they understood PPC well. Last, the tendency to maintain harmonious relationships is another cultural risk factor that threatens effective communication. Patients and family caregivers adopted a nonconfrontational approach to communication when they had questions, doubts, or concerns regarding providers' opinions. With belief in harmony as the ultimate goal of human communication (Chen, 2011), Chinese people are used to communicating indirectly and politely (Fang & Faure, 2011). Some patients or family caregivers chose not to ask any questions because asking questions is deemed inappropriate, taking time and energy away from medical providers (Leung et al., 2014). They would rather leave some questions unanswered to ensure a harmonious relationship with providers (Susilo, Marjadi, van Dalen, & Scherpier, 2019). Culture is a well-documented factor that affects PPC in Eastern countries (Lwin & Salmon, 2015; Pun et al., 2018). Because of cultural influences, PPC regarding rare diseases in Eastern societies remains largely directed by providers, and patients have a difficult time adapting to the role transition expected of them.

Consistent with extant evidence that patients with lower SES tend to have lower communication satisfaction, providers in our study associated ineffective communication with families' low SES (DeVoe, Wallace, & Fryer, 2009). The current study indicated that patients' low SES led to low communication quality through two other processes: patients' inability to comprehend PPC and weakened motivation to initiate PPC. Low levels of disease-specific knowledge and self-perceived health literacy led to resistance to PPC in the diagnosis phase. Rare diseases often involve complicated medical information, and an individual's ability to obtain, process, and understand information is tied to the complexity of the information presented (Hironaka & Paasche-Orlow, 2008). This explains why our participants struggled with communicating

disease-specific, uncommon, and complex symptoms. Scholars have suggested that patients highly value the provision of understandable information by providers (Garrino et al., 2015). Therefore, providers who serve families with rare diseases in Taiwan should tailor their communication to the complexity of the rare disease and service recipients' SES, disease knowledge, and health literacy.

Because most patients with rare diseases receive care from relatives in the same household, families in this study emphasized their need for prompt communication with providers and strategies for documenting the disease in an outpatient setting. Patients with rare diseases value experts who provide timely assistance and help when needed (Brodin, Sunnerhagen, Baghaei, & Törnbohm, 2015; Diesen, 2016). Our participants mentioned that prompt PPC could reduce their burden of unnecessary in-person visits to health care centers. Providers play an indispensable role in reminding patients and caregivers to document the disease and improving their relevant skills. For example, encouraging patients to write down questions in preparation for appointments and providing tools for this purpose can facilitate PPC (Harrington, Noble, & Newman, 2004). In addition, patients and caregivers expressed the need to consolidate information from personal research and providers. Because providers sometimes share insufficient knowledge relevant to rare diseases (Huyard, 2009), patients often become experts by educating themselves through the Internet and support groups (Crowe, McKnight, & McAneney, 2019).

Because patients and caregivers obtain information from different resources, the inconsistency of medical information should be appropriately addressed in PPC; otherwise, it may induce negative emotions in patients. For example, perceiving health professionals as lacking knowledge generates mistrust in doctors or the health care system and feelings of insecurity, fear, or anger (Barlow, Stapley, & Ellard, 2007; Grut & Kvam, 2013). Patak et al. (2009) called for assessment of patient communication needs during routine care in health care organizations. By identifying the communication needs of patients with rare diseases, the current study hopes to provoke a collaborative effort between providers and patients to address these unmet needs collectively, because failure to do so could contribute to ineffective PCC (Patak et al., 2009).

Our patients and caregivers also exhibited symptom-oriented communication habits, which affected the consistency of PPC and led to delayed diagnosis and medical care. Most rare diseases are genetic and chronic in nature (Waldboth, Patch, Mahrer-Imhof, & Metcalfe, 2016). Without an immediate cure available (Field & Boat, 2011), patients' and family caregivers' vigilance regarding the medical condition tends to taper off during the long-term treatment process of symptom management and rehabilitation work. Consequently, patients and caregivers sought PPC at a lower frequency than prescribed, jeopardizing the consistency of medical surveillance and consultation needed for monitoring and treating the disease. Enhancing patient engagement is critical to the quality of PPC by ensuring regular communication. Patient engagement can be improved through specialized positions such as rare disease coordinators who provide access to multidisciplinary services and through nontraditional forms of PPC such as public meetings, rare disease community consultations, and social media engagement (Crowe et al., 2019).

Our patient and caregiver participants were burdened by communication with multiple specialists. Because the complexity of rare diseases necessitates multidisciplinary care (Elliott & Zurynski, 2015), our participants had to communicate with multiple providers with different specialties (Crowe et al., 2019). Occasional slips in multidisciplinary communication compromised the integrity of the treatment plan. Better

coordination between health professionals will improve the quality of PPC by sharing the responsibility currently placed on patients of managing cross-provider communication.

Measures should be taken to facilitate the flow of clinical information among providers. For example, providing patients with access to their medical records such as tests, medical history, and communication from medical professionals could help them coordinate care across medical specialties (Crowe et al., 2019). Patient-centered, interdisciplinary, and holistic treatment for rare disease care characterized by better coordinated actions between health professionals has been supported by both patients (Jaeger, Røjvik, & Berglund, 2015) and health care providers (Reimann, Bend, & Dembski, 2007). As such, rare disease health care organizations and systems should consider implementing integrated care, which would address fragmentation in patient services and enable better coordinated and more continuous care (Scobie, 2021).

### ***Strengths and Limitations***

The present study has strengths that bolster its contributions to the literature. First, it took an all-inclusive approach by incorporating the perspectives of all stakeholders involved in the treatment of rare diseases. To our best knowledge, prior research tended to focus on a single perspective, either patients or physicians, whereas our study united and contrasted opinions from different roles. Patients, caregivers, physicians, nurses, dieticians, genetic counselors, social workers, and institution workers provided a coherent and collective account of barriers to PPC and quality of care. As evidenced by our findings, this multi-informant approach is critical for a panoramic view of the issue. Second, most previous studies conducted individual interviews. Findings of our study fill a methodological gap in the literature on rare diseases by conducting focus group discussions to provide in-depth perspectives on PPC. The interactive process in focus groups can unveil aspects of phenomena that are otherwise less accessible (Duggleby, 2005). Last, our study used an emic approach to understand culture-specific factors relevant to PPC among families with rare diseases in Eastern societies. The data were collected in the participants' native language and analyzed by culturally congruent researchers. The emic approach captures cultural nuances and complexities that might be overlooked by an etic approach (Peterson & Pike, 2002).

Nevertheless, some limitations should be noted. First, the study used a purposive sampling method, with patients and their family caregivers being referred, rather than randomly selected, by collaborating with medical providers and social workers at regional hospitals and organizations in Taiwan. Generalization of our study findings to regions with different health care delivery models should be made with caution. Second, our study had a small sample size. Because of societal stigma, many families with rare diseases are reluctant to participate in research and present their rare disease experience to people outside their family. The inconvenience of public transportation in rural areas was another barrier to participation of families with low SES. These factors partially explain the small sample size of patients and family caregivers in this study compared with medical providers (i.e., physicians, nurses, dietitians) from hospitals and service providers (i.e., social workers) from community-based agencies providing services for patients with rare diseases. Third, the patients with rare diseases were all men, whereas participants in the caregiver, medical, and service provider groups were predominantly women, which may have biased certain findings. The lack



of gender representation may be caused by the imbalanced gender makeup of the population of caretaking, nursing, and social work professionals in Taiwan.

### **Conclusion and Practical Implications**

Patients with rare diseases face unique challenges compared with those with more common diseases. Individual, interpersonal, and organizational factors (McLeroy, Bibeau, Steckler, & Glanz, 1988) pose barriers to the effectiveness of PPC for this population. Clinical practices should be culturally tailored to address risk factors that prevent effective PPC. To promote communication, behavioral change services and educational programs that improve patients' and caregivers' disease-specific health literacy, motivation and engagement regarding communication, level of medical compliance, and disease-documenting skills are urgently needed for families with rare diseases. Moreover, communication relevant to the needs of families with rare diseases should be conducted and collectively addressed by service providers and patients' families. A patient-centered, integrated care center (i.e., genetic counseling center) could facilitate cross-departmental communication, coordinate treatment plans for patients with rare diseases, remove structural barriers, and promote quality of life among families with rare diseases.

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