

Gratitude Experience in Ten Patients with Spinal Muscular Atrophy: A Qualitative Study

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Objective: This study aimed to investigate the gratitude experience of young and middle-aged patients with spinal muscular atrophy (SMA) during hospitalisation to provide a theoretical basis for medical professionals to develop gratitude intervention programs.

Methods: Patients with SMA who were treated with nusinersen in the Department of Neurology of the First Hospital of Jilin University between April 20 and May 20, 2024 were selected using the purposive sampling method, and semi-structured interviews were conducted. The interview data were analysed using Colaizzi's 7-step analysis method.

Results: Four themes were extracted: gratitude stimulated positive emotions; gratitude enhanced life motivation; gratitude reflected the level of social support; and gratitude promoted life restructuring planning.

Conclusion: Patients with SMA have gratitude experience and their own perception of life. Strengthened feelings of gratitude could be helpful to the psychological resilience of patients, influence their view of life in a grateful way of thinking and help them actively plan all aspects of their lives, from daily living to social re-integration.

Keywords: young and middle-aged, spinal muscular atrophy, gratitude, social support, positive psychology

Introduction

Spinal muscular atrophy (SMA) is an autosomal recessive inherited disease caused by homozygous deletion or mutation of survival motor neuron (SMN) 1, leading to the degeneration of motor neurons in the anterior foot of the spinal cord and the motor nucleus of the lower brainstem and subsequent lower motor neuron disorder with weakness and atrophy of the innervated muscles.¹ The prevalence of SMA is estimated to be 1–2 per 100,000 individuals, and its incidence is approximately 8–10 per 100,000 live births.² The main symptoms of SMA include progressive symmetrical muscle weakness, loose limbs and muscle atrophy, with corresponding loss of motor function.

Currently, there is no cure for SMA. Nusinersen, the first drug indicated for the treatment of SMA, was approved by the US Food and Drug Administration on 23 December 2016. Intrathecal injection of this drug can improve the clinical symptoms of patients with SAM; however, the disease cannot be cured and requires lifelong medication.³ Therefore, patients with SMA often suffer from psychological disorders⁴ due to various factors, such as family care, the financial burden during the treatment process and personal emotions, and are prone to stigma and negative emotions, leading to a loss of life beliefs.

Positive psychology, a unique subdomain of psychology initially founded by Martin Seligman in 1998, advocates the exploration of human virtues, such as love, forgiveness, gratitude and optimism. Increasing evidence has shown that positive psychology intervention is associated with improved health of patients,^{5–7} and gratitude is an important trait (among others) of positive psychology. McCullough et al⁸ defined gratitude as an emotional experience of appreciation and pleasure in individuals when they receive external favours. It focuses on discovering potential positive emotions and transforming them into a positive force to help people cope effectively with pressure and enhance individual well-being and life satisfaction.⁹ Studies have shown that individuals with gratitude experience have strong resilience to correctly and rationally analyse problems and generate positive emotions to protect themselves from the impact of destructive

emotions, such as resentment, anxiety and depression.^{10,11} Many studies on gratitude in patients with diseases other than SMA have been reported in China and other countries, and it has been found that gratitude can correct negative thinking, improve life satisfaction and effectively control the disease of patients.^{12–14}

The psychological status of patients with SMA has been explored in relevant studies. Qian et al⁴ found that patients with SMA and their families are challenged with a high level of burden due to difficulties in treatment selection, limited social activities and premature death. A large-scale study on SMA in the United States showed that all domains of self-perceived health-related quality of life were decreased in patients compared with those in healthy controls.¹⁵ However, studies on the gratitude experience of patients with SMA have not yet been reported, and the presence of gratitude in patients with SMA and the effect of gratitude on these patients are rarely known.

On this basis, the present study is designed to understand qualitatively the life history and gratitude experience of patients with SMA and their perception of life. It aims to clarify the interpretation of the concept and connotation of gratitude by patients with SMA in the context of Chinese culture to improve their psychological resilience and provide a basis for the development of targeted nursing intervention programs. In addition, specific suggestions for the SMA family are provided to help patients return to society with a healthy physical and mental state.

Study Participants and Methods

Study Participants

Patients with SMA who were treated with nusinersen in the Department of Neurology of the First Hospital of Jilin University between April 20 and May 20, 2024 were selected using the purposive sampling method and interviewed at the second admission. The inclusion criteria included patients (1) diagnosed with SMA by doctors, (2) treated with nusinersen, (3) aged ≥16 years and (4) who knew their diagnosis. The exclusion criteria included patients (1) with impaired verbal communication, (2) with serious mental and cognitive impairment or (3) who refused to participate in the study. The study lasted for one month. Eligible participants were all qualitatively studied during the study period. The sample size was determined according to the principle of data saturation, and 10 patients were finally included in the semi-structured interviews. The general data of the patients interviewed are presented in Table 1. They were all wheelchair users and had parents or wives as full-time caregivers to help them with the activities of daily living. This

Table 1 General Data of Patients Interviewed

No.	Sex	Age(Years)	Education	SMA Types	Employment	Marital Status	Caregiver	Age of Onset
P1	F	17	Self-educated	II	Self-employed	Unmarried	Parents	6 months
P2	M	29	Self-educated	II	Online game administrator	Unmarried	Parents	12 months
P3	M	22	Senior high school	III	Self-employed	Unmarried	Parents and grandparents	19 years
P4	F	32	Elementary school	III	WeChat business	Unmarried	Parents	12 months
P5	M	25	Bachelor degree	II	Doctor of TCM	Unmarried	Parents	10 months
P6	M	33	Elementary school	II	E-commerce	Unmarried	Mother	3 months
P7	M	22	Junior high school	II	Programmer	Unmarried	Parents	18 months
P8	M	43	Elementary school	II	E-commerce	Married	Wife	12 months
P9	F	29	Self-educated	II	Customer service for shopping websites	Unmarried	Parents	12 months
P10	M	18	Self-educated	II	Customer service for shopping websites	Unmarried	Parents	6 months

study was approved by the hospital ethics committee and permitted by the nursing management department of the hospital, the head of neurology and the study participants.

Methods

Development of the Interview Outline

The interview outline was initially drafted according to the objectives of the study and literature review, as well as by expert consultation, and the final version was developed after pre-interviews with two young and middle-aged patients with SMA and further revision. The interview questions included the following: (1) “Could you please tell me about your experience during the journey of growing up and how to deal with the difficulties caused by the disease?” (2) “What help and support have you received in the process of your growth?” (3) “Could you please describe the process and feelings during your treatment?” (4) “Are there any other factors/persons that contribute to your positive attitude?” (5) “Who is the one you most want to express gratitude to, and what do you want to say to him/her?” (6) “How has gratitude changed you?”

Data Collection

Before the interview, the purpose, method and content of the interview were explained, and promises to strictly abide by the principles of confidentiality and anonymity were provided to the patients, and consent was also obtained from them. Semi-structured face-to-face in-depth interviews were conducted based on the interview outline, and the interview process of each patient was recorded with a recording pen. The interview was conducted in a single ward or nurse’s lounge in the Department of Neurology, with 30–60 minutes required for each interview.

Data Processing and Analysis

The interview recordings were transcribed into text word-for-word within 24 hours after the interview, and the data were analysed using Colaizzi’s 7-step analysis method. Briefly, (1) the original data were read carefully; (2) significant statements were extracted; (3) meanings were summarised and extracted; (4) the common characteristics or concepts of the meanings were identified to form themes, theme clusters and categories; (5) the themes were integrated with the phenomenon under study into an exhaustive description; (6) similar concepts were distinguished to sublimate the themes; and (7) the results were returned to the interviewed patients for validation. The interview data were repeatedly read, sorted and classified by two investigators, and the results were analysed and compared. Any disagreements were resolved through discussion in the research group.

Quality Control

Adequate communication with the patients was required before the interview to reduce any concerns in the patients. The whole process of the interview was objectively recorded, with no inductive intervention given to the patients. The raw data were analysed independently by two investigators. When disagreement occurred, data were compared and discussed to reach a consensus to improve the reliability of the results.

Results

Gratitude Stimulated Positive Emotions

Gratitude encouraged individuals to evaluate their lives more positively and optimistically. Patients with “grateful hearts” were more likely to be moved by the actions of others, which maximised their satisfaction with life.

Concern and love perceived by patients were the cornerstone of their optimism, which helped patients to distract their attention and avoid excessive thinking. Patient 1 (P1): “I have had no mood or mental illness because of this disease since I was young, and it is relatively normal”. P4: “In fact, my family has been really happy for so many years because my parents have never talked about divorce because of the disease I have”. P10: “There are so many joyful things, and every day is a happy day, in general”.

Most patients were very grateful to the person who gave care and said that they wanted to give something in return, which also became a potential driving force for the active recovery of patients. P1: “When there is inconvenience while going out, sometimes strangers help me, and I appreciate them”. P3: “I’m very grateful to my family. My parents are

actually quite stressful, and most of the time, it is my grandparents who take care of me”. P7: “I feel that many parents with normal children are not as good as my parents, and I am very glad to have them as my parents”.

Physical touch can be a positive psychological experience. Many patients said that many things moved them during the treatment, which reduced the psychological pressure to a certain extent. P1: “I am warmed and touched by the person who is helpful to me”. P2: “My family members actually give me the most companionship. What’s more, after coming here, the medical staff helped a lot, which makes me touched very much”. P3: (after thinking for a long time) “I am more grateful to my father. After all, I am quite heavy, and my father carries me on his back when it is inconvenient”.

Gratitude Enhanced Life Motivation

Patients hoped passionately to live independently with relatively high enthusiasm for recovery, especially regarding strengthening their limb muscles, due to the burden posed on the family. P1: “Sitting in bed for a long time makes me very tired, and my neck and spinal scoliosis are getting worse. If I were in bed, I would hope that I could use my own wheelchair in the future and use a computer to do more long-term work”. P2: “I am in my 30s, and my grandmother is in her 80s. She takes care of my daily living. I can’t express myself in language. You know, this is also part of the motivation. Otherwise, I can’t work so actively”. P3: “I am also trying to shorten the recovery time as much as possible; that is, to reduce the overall recovery time. I hope that the total time is shorter and the recovery is faster. That’s roughly what I mean”.

Gratitude Reflected the Level of Social Support

Gratitude is the most direct way for patients to repay the social support they receive. In terms of policies, nusinersen is covered by medical insurance, which greatly reduces the economic burden on patients and their families. P1: “I definitely believe that our country is particularly strong; after all, the price of such expensive drugs can remain low”.

In terms of daily living, patients with SMA generally connect with their families, medical professionals and relevant social workers; therefore, these patients mainly express their gratitude to the country and these individuals. P4: “Doctors and nurses are really enthusiastic, careful and patient with us. In fact, the most touching thing about this is that I can take the medicine without having to go far because of the effort of Dr. Yu’s team; otherwise, I have to go to Beijing for the medication. If I go to other places for the treatment, the proportion of cooperative medical reimbursement is much lower. Moreover, I have to spend time travelling, which is a big expense”. Gratitude plays an essential role in establishing favourable doctor–patient and social relationships to promote family harmony, maintain social interaction and build confidence for patients to restore their normal lives. P7: “Yes, it’s much better in Northeast China. For example, the subway staff came and helped me a lot. Once I needed to go upstairs, one staff member saw me and asked me to wait until he asked several security guards to directly carry me upstairs. They lifted me in the wheelchair directly”.

Gratitude Promoted Life Restructuring Planning

The patient’s family provided them with the greatest support. Almost all caregivers of the participants were their spouses or parents, who took on more responsibilities, including caring for patients, raising children and earning money to support their families. Patients and their families spent more time together than they did before the illness. With the care and encouragement of the families, the patients were independent and self-reliant and actively planned to reconstitute their lives after recovery from SMA. P1: “I still hope to take care of myself with surplus capacity, such as sitting in the wheelchair by myself while holding someone else”. P2: “What to do? Make more money! A further plan is that I still hope my condition will get better, for sure. I just wait for the medical breakthroughs; that is all I look forward to”. P5: “The short-term plan is to study and pass the examinations for certificates. First of all, I hope I pass the examinations next month. A long-term plan is to realise my value. Because of this disease, I can easily understand the patients’ desire for health. I will do my best to help others and realise my expectations”.

Discussion

Gene therapy has proven to be a breakthrough in the treatment of SMA in recent years. Nusinersen is an oligonucleotide therapy that promotes the production of functional SMN proteins by interacting with the precursor mRNA of the SMN2

gene.¹⁶ Clinical studies have shown the significant efficacy of nusinersen in the treatment of SMA, including improved motor function, delayed disease progression and increased survival rates.¹⁷ The price of nusinersen was as high as CNY 699,700 per unit at the beginning of its launch, and this sky-high drug price deterred patients and their families.^{18,19} In December 2021, the high price was reduced to CNY 33,000 after China's medical insurance negotiations, and nusinersen was covered by the national medical insurance. The introduction of nusinersen and its subsequent inclusion in national medical insurance have brought a ray of hope to patients with SMA, and many patients are grateful to the country and society.

Studies have shown that gratitude is an effective approach to promoting health and enhancing the subjective well-being of individuals.²⁰ The overall quality of life evaluation and future expectations of individuals are maximised, and the psychological well-being and sense of happiness are increased in those with gratitude experience. In the present study, 10 patients reported their rich gratitude experience and the effect of gratitude on their life impact after medication treatment, with the consequent psychological experience of inner growth.

It was not difficult to find in the patients' statements that SMA has posed great distress to them and their families, especially as the prognosis of the disease is uncertain.²¹ Most of the young and middle-aged people in China assume the responsibility of supporting their parents and raising their children at the same time; this is the mainstay of family and social development. In addition, young and middle-aged patients with SMA are prone to becoming anxious and depressive during hospitalisation due to the poor prognosis, slow recovery and high cost of the disease. Studies¹¹ have shown that gratitude expressed by patients is their positive feedback to the giver and also protects patients from destructive emotions, such as resentment, anxiety and depression. In the present study, patients with gratitude experience developed strong resilience and a positive, optimistic attitude when challenged by the traumatic event of SMA; they became more concerned about themselves and with better belief in recovery.

A positive treatment attitude may alleviate the symptoms of muscle weakness, and gratitude can guide a positive change of perception in patients.²² Most of the patients in the present study realised the importance of maintaining a positive attitude and held hopes for future recovery with a new understanding of health and life.

Young and middle-aged patients assume more family and social responsibilities and have higher requirements for their self-image. The sequelae of SMA, including physical, swallowing and language disorders, pose an extremely heavy burden on these patients. Therefore, young and middle-aged patients with SMA may experience strong emotional responses and even psychological crises. These patients have great potential for gratitude. The qualitative study of Farrar²³ reports on the profound emotional effects of SMA on patients and their families, consistent with the results of this study, and explores how positive emotions can prevent mental illness in children and parents.

Gratitude is essentially a complex process of inner growth in which patients think about their past and current experiences, and hospitalisation is the best time to rouse their sense of gratitude.

Therefore, medical professionals should focus their attention on evaluating the gratitude level of patients during hospitalisation and develop individual, professional, comprehensive and feasible gratitude intervention plans for young and middle-aged patients based on their special characteristics. These should include, for example, encouraging patients to express gratitude to their families orally or by letter, recalling past gratitude events by listing a gratitude list, writing a diary of gratitude and promptly recording emotional experiences when gratitude is triggered. Patients are guided to view life with a grateful mind and actively plan all aspects of their lives, from daily living to social reintegration, such as setting up a suitable family rehabilitation environment, developing healthy dietary plans and keeping a regular lifestyle, to benefit themselves in the long term.

There were limitations in this study. First, the authors were inexperienced in executing a qualitative study and lacked in some of the required skills, such as interview technique, theme extraction and summarising; results were possibly influenced by the nature of the questions. Second, although the themes of this study were thoroughly examined, it was a relative concept. The study did not investigate if there are differences in time, age or other characteristics of the participants. With the passage of time, policy adjustment and regional differences, the findings may vary. Finally, the participants interviewed in this study were patients admitted to our hospital, which may affect the findings of the study.

Conclusion

In summary, patients with SMA have gratitude experience and their own perception of life. Strengthened feelings of gratitude could be helpful to the psychological resilience of patients, influencing them to view life gratefully and helping them actively plan all aspects of their lives, from daily living to social re-integration.

Data Sharing Statement

All data generated or analyzed during this study are included in this published article.

Ethics Approval and Consent to Participate

This study was conducted in accordance with the declaration of Helsinki. The studies involving human participants were reviewed and approved by Jilin University First Hospital (ID: 24K094-001). The patients/participants provided their written informed consent included publication of anonymized responses/direct quotes to participate in this study. At the same time, We also obtained parental informed consent for patients under the age of 18.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors declare that they have no competing interests.

References

1. Rare Disease Society of Chinese Research Hospital Association. China alliance for rare diseases, Beijing society of rare disease clinical care and accessibility, Chinese expert group on clinical diagnosis and treatment guidelines for adolescent and adult spinal muscular atrophy. Clinical practice guideline for adolescent & adult patients with spinal muscular atrophy. *J Rare Dis*. 2023;2(2):231–255. doi:10.12376/j.issn.2097-0501.2023.01.010
2. Verhaart IEC, Robertson A, Wilson IJ, et al. Prevalence, incidence and carrier frequency of 5q-linked spinal muscular atrophy - A literature review. *Orphanet J Rare Dis*. 2017;12(1):124. doi:10.1186/s13023-017-0671-8
3. Corey DR, Nusinersen, an antisense oligonucleotide drug for spinal muscular atrophy. *Nat Neurosci*. 2017;20(4):497–499. doi:10.1038/nn.4508
4. Qian Y, McGraw S, Henne J, Jarecki J, Hobby K, Yeh WS. Understanding the experiences and needs of individuals with spinal muscular atrophy and their parents: a qualitative study. *BMC Neurol*. 2015;15:217–222. doi:10.1186/s12883-015-0473-3
5. Benoit V, Gabola P. Effects of positive psychology interventions on the well-being of young children: a systematic literature review. *Int J Environ Res Public Health*. 2021;18(22):12065. doi:10.3390/ijerph182212065
6. Cunha LF, Pellanda LC, Reppold CT. Positive psychology and gratitude interventions: a randomized clinical trial. *Front Psychol*. 2019;10:584. doi:10.3389/fpsyg.2019.00584
7. Pan S, Ali K, Kahathuduwa C, Baronia R, Ibrahim Y. Meta-analysis of positive psychology interventions on the treatment of depression. *Cureus*. 2022;14(2):e21933. doi:10.7759/cureus.21933
8. McCullough ME, Emmons RA, Tsang JA. The grateful disposition: a conceptual and empirical topography. *J Pers Soc Psychol*. 2002;82(1):112–127. doi:10.1037//0022-3514.82.1.112
9. Liu Q, Jin A, Fei Y, Liu XQ, Zhu XP. Application progress of gratitude theory in nursing of patients with chronic diseases. *Chin J Modern Nurs*. 2021;27(28):3916–3920. doi:10.3760/cma.j.cn115682-20210227-00886
10. Portocarrero FF, Gonzalez K, Ekem-Agbaw M. A Meta-analytic review of the relationship between dispositional gratitude and well being. *Pers Indiv Differ*. 2020;164:110101. doi:10.1016/j.paid.2020.110101
11. Tala A. Gracias por todo: una revisión sobre la gratitud desde la neurobiología a la clínica [Thanks for everything: a review on gratitude from neurobiology to clinic]. *Rev Med Chil*. 2019;147(6):755–761. Spanish. doi:10.4067/S0034-98872019000600755
12. Zhao M, Jiang W, Nong J, Lu XF. Effects of gratitude intervention on the hope level and self-efficacy of patients with compensatory phase cirrhosis. *Chin J Modern Nurs*. 2017;23(14):1880–1883. doi:10.3760/cma.j.issn.1674-2907.2017.14.013
13. Li C, Wang R, Yang F. Effects of gratitude extension-construction theory in ovarian cancer patients undergoing chemotherapy. *Chin J Modern Nurs*. 2023;12:1648–1652. doi:10.3760/cma.j.cn115682-20220324-01412
14. Liu Q, Qiu C, Fei Y, et al. Gratitude experience of young and middle-aged hospitalized stroke patients from the perspective of positive psychology: a qualitative study. *Nurs J Chin People's Liberat Army*. 2021;38(08):31–33,64. doi:10.3969/ji.ssn.1008-9993.2021.08.009

15. Iannaccone ST, Hynan LS, Morton A, et al. The PedsQL in pediatric patients with spinal muscular atrophy: feasibility, reliability, and validity of the pediatric quality of life inventory generic core scales and neuromuscular module. *Neuromuscul Disord.* **2009**;19(12):805–812. doi:10.1016/j.nmd.2009.09.009
16. Hagenacker T, Wurster CD, Günther R, et al. Nusinersen in adults with 5q spinal muscular atrophy: a non-interventional, multicentre, observational cohort study. *Lancet Neurol.* **2020**;19(4):317–325. doi:10.1016/S1474-4422(20)30037-5
17. Finkel RS, Chiriboga CA, Vajsar J, et al. Treatment of infantile-onset spinal muscular atrophy with nusinersen: final report of a Phase 2, open-label, multicentre, dose-escalation study. *Lancet Child Adolesc Health.* **2021**;5(7):491–500. doi:10.1016/S2352-4642(21)00100-0
18. Wang Q. Spinal muscular atrophy yesterday, today and tomorrow. *Chin J Difficult Complicat Cases.* **2023**;22(01):98–101,105. doi:10.3969/j.issn.1671-6450.2023.01.019
19. Yang JY. A new drug for the treatment of spinal muscular atrophy: nusinersen. *Chin J New Drugs Clin Rem.* **2018**;37(5):259–261. doi:10.14109/j.cnki.xyylc.2018.05.003
20. Maltby J, Wood AM, Day L. Personality predictors of levels of forgiveness two and a half years after the transgression. *J Res Pers.* **2008**;42(3):1088–1094. doi:10.1016/j.jrp.2007.12.008
21. Ho HM, Tseng YH, Hsin YM, Chou FH, Lin WT. Living with illness and self-transcendence: the lived experience of patients with spinal muscular atrophy. *J Adv Nurs.* **2016**;72(11):2695–2705. doi:10.1111/jan.13042
22. Sztachañska J, Krejtz I, Nezelek JB. Using a gratitude intervention to improve the lives of women with breast cancer: a daily diary study. *Front Psychol.* **2019**;10:1365. doi:10.3389/fpsyg.2019.01365
23. Farrar MA, Carey KA, Paguinto S-G. Financial, opportunity and psycho-social costs of spinal muscular atrophy: an exploratory qualitative analysis of Australian carer perspectives. *BMJ Open.* **2018**;8:e020907. doi:10.1136/bmjopen-2017-020907

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