



ORIGINAL ARTICLE

Impact of the coronavirus pandemic on mental health and health care in adults with neurofibromatosis: Patient perspectives from an online survey

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Abstract

The coronavirus pandemic increased anxiety and stress and prevented access to health care worldwide; it is unclear how COVID-19 affected adults with a multi-system genetic disorder such as neurofibromatosis (NF). An anonymous online survey was distributed through an international registry and foundations to adults with NF (June–August 2020) to assess the impact of the pandemic on mental health and NF health care. Six hundred and thirteen adults (18–81 years; $M = 45.7$) with NF1 (77.8%), NF2 (14.2%), and schwannomatosis (7.8%) provided complete responses. Respondents rated moderate-to-high amounts of worry about the impact of COVID-19 on their emotional (46.3%) and physical health (46.7%), and 54.8% endorsed moderate-to-high pandemic-related stress. Adults with diagnosed/suspected mental health disorders or moderate-to-severe NF symptom impact as well as females endorsed higher COVID-19 stress ($p < 0.01$). Less than half who missed a doctor's appointment for their NF care (43.4%) used telehealth. Of these, 33.3% and 46.2% reported that telehealth met their needs to a moderate or high degree, respectively. Results indicated that subgroups of adults with NF experience higher COVID-19-related worries and stress and may need additional support. Furthermore, telehealth is under-utilized and could help NF providers connect with patients, although improved delivery and patient training may facilitate expanded use of these services.

KEYWORDS

coronavirus, COVID-19, neurofibromatosis, psychosocial, telehealth

1 | INTRODUCTION

The severe acute respiratory syndrome coronavirus (SARS-CoV-2) infection causing coronavirus disease (COVID-19) has profoundly impacted the care of patients and health care delivery globally

(Dopfer et al., 2020). Countries responded to COVID-19 by issuing lockdowns, reallocating health care resources for affected patients, and delaying nonessential care, resulting in a significant reduction in health care access and utilization (Giamello et al., 2020; Tartari et al., 2020). The COVID-19 pandemic also exacerbated disparities in

health care, with racial and ethnic minority groups and low income families being disproportionately affected (Millett et al., 2020; van Dorn et al., 2020). Furthermore, the pandemic negatively impacted the multidisciplinary care and mental health outcomes of adults with chronic health conditions such as diabetes, hypertension, and pulmonary diseases (Chudasama et al., 2020). In addition, various stressors, ranging from concerns about the illness to social isolation, caused significant emotional distress and mental health difficulties in the general public and health care professionals (Hossain et al., 2020; Sani et al., 2020). However, no research has examined these issues among individuals with complex genetic conditions (Radtke et al., 2021). Understanding the unique mental health concerns and stressors associated with the COVID-19 pandemic as well as changes in routine care among individuals with genetic conditions is pivotal to prioritizing resources and providing appropriate care.

The neurofibromatoses (NF) are a group of complex genetic conditions that include neurofibromatosis 1 (NF1), neurofibromatosis 2 (NF2), and schwannomatosis, which can affect nearly every organ system in the body. Although the three types of NF have significantly different symptoms and presentations, all have the predisposition to develop nerve sheath tumors. Tumor-related complications may include headache, pain, hearing and vision loss, gait problems, and seizures while nontumor manifestations may include neurologic, cognitive, skin, and bone abnormalities (Ardern-Holmes et al., 2017; Riccardi, 2010). Adults with NF also may exhibit more symptoms of depression and anxiety, higher levels of stress, and poorer self-esteem than the general population (D. Wang et al., 2012). Thus, routine care and close monitoring by a multidisciplinary team of professionals is required to diagnose NF-related manifestations, reduce disease-related symptoms, and minimize more serious complications. Additionally, these patients may undergo surgery to remove tumors or participate in clinical trials to evaluate the effects of a new drug on the growth of their neurofibromas and schwannomas (Ly & Blakeley, 2019; Plotkin & Wick, 2018). However, with the advent of the pandemic, a significant proportion of NF clinics reported reduced patient volumes or a delay in patients accessing novel therapies (e.g., selumetinib) followed by an increase in telehealth utilization (Radtke et al., 2021).

Although telehealth services were available prior to the COVID-19 pandemic (Lin et al., 2018), rates of use varied across the United States due to reimbursement, state, and federal policies (Adler-Milstein et al., 2014). However, telehealth expanded rapidly during the pandemic (Hollander & Carr, 2020) because of increased demand, eased restrictions, and expanded coverage (Weigel et al., 2020), which enabled the provision of health services without in-person visits (Monaghesh & Hajizadeh, 2020). Nevertheless, various barriers to telehealth implementation continue to exist, including cost, reimbursement policies, privacy and security concerns, technical difficulties, limited internet access, and poor computer literacy skills (Scott Kruse et al., 2018; Weigel et al., 2020). These barriers may be more pronounced in rural communities, low-income families, and racial and ethnic minorities (Katzow et al., 2020). A recent study identified a significant acceleration in telehealth for NF care during the COVID-19 pandemic and generally positive experiences endorsed by health care

clinicians (Radtke et al., 2021); however, studies have yet to assess patients' own perspectives about the transition to telehealth care.

1.1 | Aims

The aims of the current study were to assess the perspectives of adults with NF regarding the impact of COVID-19 on their mental health and NF health care. First, we aimed to characterize rates of COVID-19-related worries and overall stress and identify the main pandemic-related stressors endorsed by this population. Second, we investigated group differences in worries and overall stress associated with COVID-19, based on demographic and clinical characteristics (e.g., gender, NF type, symptom impact, and preexisting mental health diagnoses or difficulties). Finally, we examined patients' reported use of telehealth during the pandemic and perceptions about whether telehealth services met their health care needs adequately.

2 | METHODS

2.1 | Participants

Eligible participants included adults ≥ 18 years of age with a self-reported diagnosis of NF1, NF2, or schwannomatosis and the ability to read and understand English.

2.2 | Measures

A multidisciplinary, global group of NF clinical researchers and patient representatives from the Response Evaluation in Neurofibromatosis and Schwannomatosis (REiNS) International Collaboration iteratively developed an anonymous survey for adults (≥ 18 years) with NF, who read and understand English, to assess their perspectives about the design and conduct of patient-centered psychosocial trials. The initial survey consisted of four sections: demographic characteristics, NF-related clinical complications, psychosocial well-being, and the development of psychosocial trials. Due to the widespread lockdowns in March 2020, the group added another section consisting of 11 COVID-19-related questions, which were adapted from the NIMH CoRonavirus Health Impact Survey (CRISIS; Nikolaidis et al., 2021) with permission from the authors (personal communication, Argyris Stringaris). The final items were programmed into the REDcap (Research Electronic Data Capture) survey format. The NIH IRB reviewed the protocol and determined the anonymous adult survey was exempt. The current study analyzed data from the following three sections of the survey.

Demographic characteristics. Items utilized in this study include age, gender identity, race, ethnicity, country of residence (United States or other), highest level of education, and employment status before and during COVID-19.

Clinical characteristics. Items assessing NF-related clinical characteristics used in this report include: (1) type of NF, (2) current mental health diagnosis, (3) suspected mental health problems, (4) whether

respondents had seen a mental health professional or taken medication for a mental health problem recently, (5) the extent to which mental health difficulties affected their everyday functioning (four response options from “not at all” to “a lot”), and (6) whether or not they had experienced learning difficulties when growing up. The respondents also answered a question about “how much all their NF symptoms affected their everyday life,” from four options (“no effect,” “mild degree,” “moderate degree,” and “severe degree”), which was used as a perceived measure of NF symptom impact.

COVID-19 questions. In this section, four items assessed different COVID-19-related worries (being infected, friends or family being infected, physical health effects, and emotional health effects), and one item assessed how stressful the pandemic had been for them. These five items were answered on an ordinal scale with five response options from “not at all” to “extremely.” Another item asked participants to select the coronavirus-related stressors they had experienced from a list of 15 options (e.g., unemployment, loss of health insurance, caring for a family member with COVID-19). Two items assessed whether the pandemic affected NF-specific care by asking whether: (1) an in-person doctor’s appointment for NF care or (2) an in-person NF treatment was canceled or delayed because of the coronavirus pandemic, and if yes, how much missing that appointment or treatment concerned them (five responses options from “not at all” to “extremely”). Two items assessed their use and satisfaction with telehealth by asking (1) if they used telehealth instead of an in-person visit for NF care because of the pandemic (yes/no), and (2) how much the telehealth appointment met their needs (five responses options from “not at all” to “extremely”). Finally, an open-ended question asked if there were other ways the coronavirus pandemic and stay-at-home order had affected them. For a complete copy of the Adult NF Psychosocial Survey, see Appendix S1 or the REINS website at <https://ccrod.cancer.gov/confluence/display/REINS/Patient+Reported+Outcomes>.

2.3 | Procedures

On June 19, 2020, the Children’s Tumor Foundation (CTF) distributed the REDcap survey internationally through their online registry to 4745 adults with NF1, NF2, or schwannomatosis (Seidlin et al., 2017). In addition, NF foundations in the United States (NF Network, NF Northeast, NF Midwest, NF Michigan, Littlest Tumor Foundation, and Texas NF Foundation) further disseminated the survey by email and social media platforms. It was described to potential participants as an online survey to help researchers design psychosocial trials for adults with NF and to learn about how the coronavirus pandemic was affecting them. There was no compensation for completing the survey. The CTF registry sent a reminder in July and August, and the survey was closed on August 10, 2020.

2.4 | Data analysis

Quantitative and ordinal data were analyzed with descriptive and Chi-square statistics. When there were missing responses for particular items, the valid percent was presented. Significant omnibus Chi-

square tests were followed by post hoc analyses using Bonferroni correction to determine which comparisons were statistically significant. For ease of interpretation, responses to the items about how much NF symptoms and mental health difficulties affect everyday life were grouped: “not at all” and “a little” were combined to reflect mild impact and “somewhat” and “a lot” were combined to reflect moderate-to-severe impact. For various questions that asked about worry, stress, and concerns about events related to COVID-19, as well as satisfaction with health care during the pandemic, response options “not at all” and “slightly” were categorized as having a low amount; “moderately” was categorized as having a moderate amount; and “very much” and “extremely” were coded as having a high amount. Qualitative self-report data was summarized using inductive content analysis (Elo & Kyngäs, 2008) by two investigators, and disagreements were discussed and resolved with a third investigator to achieve 100% agreement.

3 | RESULTS

3.1 | Characteristics of the total sample

3.1.1 | Demographic characteristics

Of the 886 adults with NF who checked that they agreed to participate in the study, we obtained 613 complete surveys (69.2% completion rate). As shown in Table 1, respondents had a mean age of 45.7 (range 18–81; $SD = 14.2$), were predominately white (85.9%), not Hispanic or Latino (92.9%), female (70.6%), US residents (78.0%), and had a college degree (56.3%). The non-US participants ($n = 134$) were from 29 different countries (Table S1). Prior to the coronavirus pandemic, 60.5% of respondents were employed; of these, the majority were working full time (77.2%). In the subset of adults working prior to the pandemic, 72.6% reported still being employed at the time they completed the survey.

3.1.2 | Clinical characteristics

Respondents were diagnosed with NF1 (77.8%), NF2 (14.2%), and schwannomatosis (7.8%), reflecting the varied prevalence of the three conditions. Slightly over half (53.8%) of the total sample reported a history of learning difficulties (Table 2), with a higher percent of adults with NF1 reporting learning difficulties (63.7%) than those with NF2 (19.5%) or schwannomatosis (16.7%). Approximately half of all respondents (51.3%) indicated that they have a mental health diagnosis; of these, 56.3% reported that they had seen a mental health professional recently and 71.5% had taken medication for it recently. Among the respondents who had not been diagnosed or were unsure of their diagnostic status, 27.2% thought that they have mental health difficulties. The majority (63.5%) of respondents who endorsed having mental health diagnoses or difficulties rated that these problems had a moderate-to-severe impact on their everyday life. In the total sample, over half (55.1%) rated that their NF symptoms impacted their daily life from a moderate-to-severe degree.

TABLE 1 Demographic characteristics of the total sample

| Characteristic | Total patient sample | | NF1 | | NF2 | | Schwannomatosis | |
|------------------------------------|----------------------|------|----------|------|----------|------|-----------------|------|
| | <i>n</i> | % | <i>n</i> | % | <i>n</i> | % | <i>n</i> | % |
| Total N | 613 | 100 | 477 | 77.8 | 87 | 14.2 | 48 | 7.8 |
| Age | | | | | | | | |
| <i>M</i> | 45.7 | | 45.6 | | 45.2 | | 47.8 | |
| <i>SD</i> | 14.2 | | 14.1 | | 15.6 | | 12 | |
| Gender ^a | | | | | | | | |
| <i>n</i> | 612 | | 477 | | 87 | | 48 | |
| % Female | 432 | 70.6 | 339 | 71.1 | 61 | 70.1 | 32 | 66.7 |
| % Male | 174 | 28.4 | 132 | 27.7 | 26 | 29.9 | 16 | 33.3 |
| % Nonbinary | 3 | 0.5 | 3 | 0.6 | 0 | 0 | 0 | 0 |
| % Transgender male | 2 | 0.3 | 2 | 0.4 | 0 | 0 | 0 | 0 |
| Race | | | | | | | | |
| <i>n</i> | 609 | | 474 | | 87 | | 48 | |
| % White | 523 | 85.9 | 403 | 85 | 74 | 85.1 | 46 | 95.8 |
| % Asian | 24 | 3.9 | 21 | 4.4 | 2 | 2.3 | 1 | 2.1 |
| % Black or African American | 22 | 3.6 | 18 | 3.8 | 3 | 3.4 | 1 | 2.1 |
| % American Indian or Alaska Native | 4 | 0.7 | 2 | 0.4 | 2 | 2.3 | 0 | 0 |
| Ethnicity | | | | | | | | |
| <i>n</i> | 594 | | 460 | | 86 | | 48 | |
| % Not Hispanic or Latino | 552 | 92.9 | 426 | 92.6 | 78 | 90.7 | 48 | 100 |
| % Hispanic or Latino | 42 | 7.1 | 34 | 7.4 | 8 | 9.3 | 0 | 0 |
| US residency | | | | | | | | |
| <i>n</i> | 609 | | 474 | | 87 | | 48 | |
| % Resident | 475 | 78.0 | 368 | 77.6 | 66 | 75.9 | 41 | 85.4 |
| % Nonresident | 134 | 22.0 | 106 | 22.4 | 21 | 24.1 | 7 | 14.6 |
| Education levels | | | | | | | | |
| <i>n</i> | 613 | | 476 | | 87 | | 48 | |
| % Did not complete high school | 11 | 1.8 | 10 | 2.1 | 1 | 1.1 | 0 | 0 |
| % High school degree or equivalent | 111 | 18.2 | 94 | 19.7 | 12 | 13.8 | 5 | 10.4 |
| % Some college—no degree | 144 | 23.6 | 116 | 24.4 | 17 | 19.5 | 11 | 22.9 |
| % Associate's degree | 68 | 11.1 | 58 | 12.2 | 7 | 8.0 | 3 | 6.3 |
| % Bachelor's degree | 170 | 27.7 | 125 | 26.3 | 27 | 31.0 | 18 | 37.5 |
| % Graduate degree | 107 | 17.5 | 73 | 15.3 | 23 | 26.4 | 11 | 22.9 |
| Employment status | | | | | | | | |
| <i>n</i> | 611 | | 476 | | 87 | | 48 | |
| % Unemployed pre-COVID | 242 | 39.6 | 170 | 35.7 | 46 | 52.9 | 26 | 54.2 |
| % Still working | 268 | 43.9 | 221 | 46.4 | 29 | 33.3 | 18 | 37.5 |
| % Laid off/furloughed | 58 | 9.5 | 50 | 10.5 | 5 | 5.7 | 3 | 6.3 |
| % Work status otherwise changed | 43 | 7.0 | 35 | 7.4 | 7 | 8.1 | 1 | 2.1 |

^aOne respondent indicated that their gender identity was not listed.

3.2 | COVID-19-related worries and stress

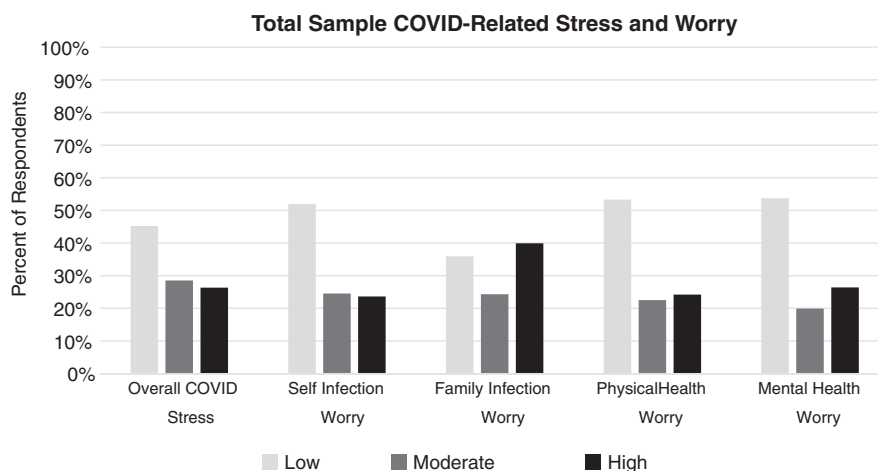
When examining specific worries related to the pandemic in the total sample (Figure 1), 48.1% reported moderate-to-high levels of worry about being infected themselves, 64.1% about their family members or friends being infected, 46.7% about their physical health, and 46.3% about their emotional health. In addition, 84.3% of all

participants reported some level of overall stress due to the coronavirus pandemic with 54.8% reporting moderate-to-high levels (moderate = 28.5%; high = 26.3%).

The top three endorsed stressors related to the coronavirus pandemic were unemployment of self (14.5%; *n* = 89), significant reduction in/loss of main source of income (12.1%; *n* = 74), and the inability to quarantine due to work deemed “essential” (11.4%;

TABLE 2 Clinical characteristics of the total sample

| Characteristic | Total patient sample | | NF1 | | NF2 | | Schwannomatosis | |
|--|----------------------|------|-----|------|-----|------|-----------------|------|
| | n | % | n | % | n | % | n | % |
| Mental health diagnosis | | | | | | | | |
| n | 610 | | 476 | | 86 | | 48 | |
| % Diagnosed | 313 | 51.3 | 242 | 50.8 | 44 | 51.2 | 27 | 56.3 |
| % Not diagnosed | 277 | 45.4 | 216 | 45.4 | 41 | 47.7 | 20 | 41.7 |
| % Unsure | 20 | 3.3 | 18 | 3.8 | 1 | 1.2 | 1 | 2.1 |
| Suspected mental health diagnosis | | | | | | | | |
| n | 294 | | 232 | | 41 | | 21 | |
| % Believe to have a mental illness but not diagnosed | 80 | 27.2 | 64 | 27.6 | 11 | 26.8 | 5 | 23.8 |
| % Do not believe | 163 | 55.4 | 123 | 53.0 | 27 | 65.9 | 13 | 61.9 |
| % Unsure | 51 | 17.3 | 45 | 19.4 | 3 | 7.3 | 3 | 14.3 |
| Learning disability (LD) | | | | | | | | |
| n | 612 | | 477 | | 87 | | 48 | |
| % Reported LD history | 329 | 53.8 | 304 | 63.7 | 17 | 19.5 | 8 | 16.7 |
| % No reported LD history | 220 | 35.9 | 125 | 26.2 | 62 | 71.3 | 33 | 68.8 |
| % Unsure | 63 | 10.3 | 48 | 10.1 | 8 | 9.2 | 7 | 14.6 |

FIGURE 1 Ratings of the total sample regarding overall COVID-19-related stress and worries about infection of oneself, infection of a family member or friend, one's physical health, and one's emotional health

$n = 70$). For the open-ended question about any other ways that the coronavirus pandemic and stay-at-home orders affected them, participants ($n = 198$) wrote in a response; the three most frequent themes not previously noted were all negative experiences: social isolation, disruption to routines and plans, and unmet medical needs. A few respondents reported a positive outcome: less work stress, more time with family, saving money, and being able to cover a tumor-related facial disfigurement with a mask.

3.3 | COVID-19-related worries and stress by demographic and clinical characteristics

Group differences in COVID-19-related worries and overall stress were examined based on various demographic and clinical characteristics (Figure 2) as described below.

3.3.1 | Demographic characteristics

Age groups. The three age groups (18–39, 40–60, >60 years) did not differ in overall COVID-19-related stress. However, there were age group differences in reported levels of worry regarding the impact of COVID-19 on emotional health [$\chi^2(4) = 10.212$; $p < 0.05$]. Post hoc testing revealed that adults who were ages 18–39 years were more likely to report high levels of worry about the pandemic's influence on their emotional health than those older than 60 ($p < 0.01$). Age group was not significantly related to the other worry items.

Gender identity. Due to small sample sizes, adults indicating gender identities besides male and female (<1% nonbinary/transgender) were excluded from these analyses. Chi-Square omnibus tests revealed a significant gender identity by stress interaction [$\chi^2(2) = 29.359$, $p < 0.01$] such that females were more likely to report higher levels of COVID-19-related stress than males ($p < 0.01$).

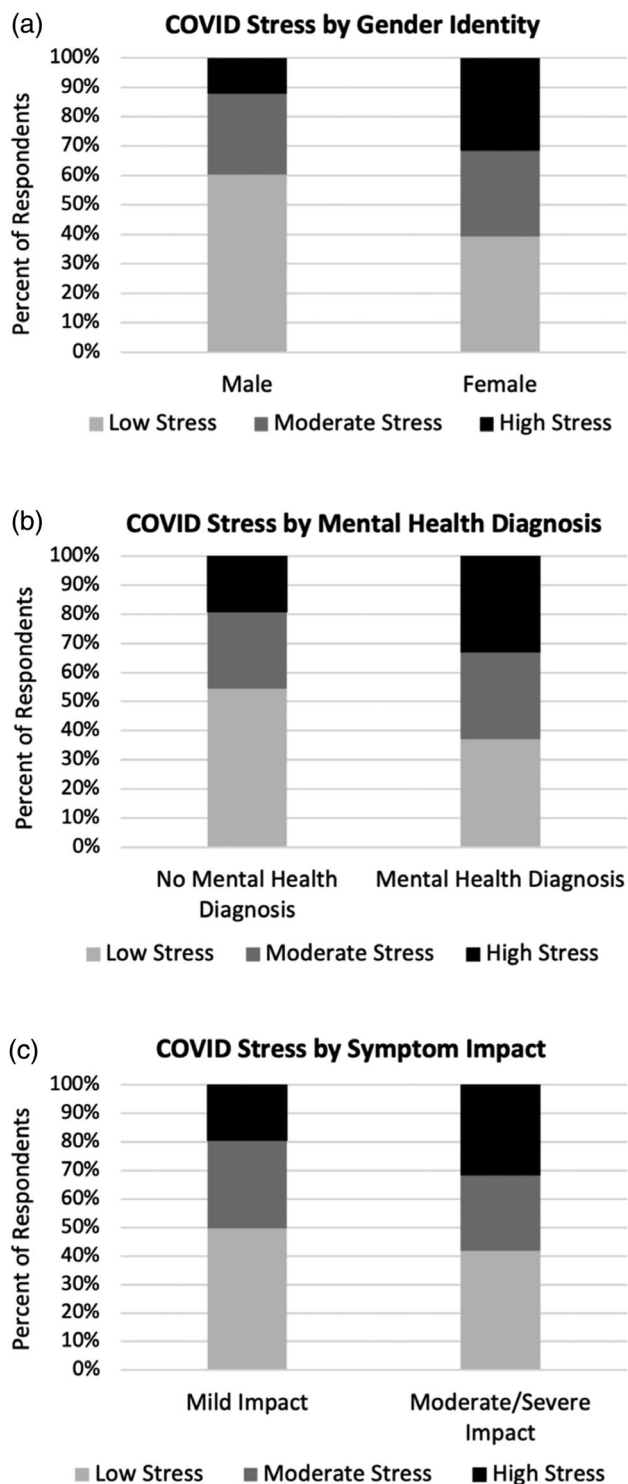


FIGURE 2 Group differences in ratings of overall COVID-19-related stress by (a) gender identity, (b) mental health diagnosis, and (c) NF symptom impact on daily life

Similar gender response patterns were found regarding worry about being infected with coronavirus [$\chi^2(2) = 12.906, p < 0.01$], worry about friends or family being infected with coronavirus [$\chi^2(2) = 13.317, p < 0.01$], and worry about one's physical health [$\chi^2(2) = 7.746, p < 0.05$] and emotional health [$\chi^2(2) = 12.464,$

$p < 0.01$] being impacted by the pandemic. Females also were more likely to report having a mental health diagnosis than males [$\chi^2(1) = 20.830, p < 0.01$]. However, gender differences on NF symptom impact and mental health impact were not significant.

Racial groups. No significant racial differences were observed in COVID-19-related worries or overall stress, but we could only compare White, Black, and Asian participants due to low numbers of respondents in the other racial categories.

US residents. Adults with NF residing in the United States did not report any significant differences in COVID-19-related worries or stress compared to those residing outside of the United States.

Education level. No significant differences were found in COVID-19-related worries or overall stress among individuals with a college degree compared those without a college degree.

Unemployment due to COVID-19. Being laid-off or furloughed during the pandemic was associated with overall levels of stress [$\chi^2(2) = 12.461, p < 0.01$]; those who were unemployed were more likely to report moderate stress levels ($p < 0.05$) and less likely to report low stress levels ($p < 0.01$) than those who were still working. There were no differences in the specific worries assessed between employed or unemployed respondents.

3.3.2 | Clinical characteristics

NF diagnosis. There were no differences in overall COVID-19-related stress or in level of worry about physical or emotional health being affected by the pandemic among the three NF diagnoses. However, adults with NF2 ($p = 0.01$) and schwannomatosis ($p < 0.05$) both were more likely than those with NF1 to report high levels of worry about becoming infected with COVID-19 [$\chi^2(4) = 21.001, p < 0.01$], and adults with NF2 were more likely than those with NF1 ($p = 0.01$) to report high levels of worry regarding their friends or family being infected with COVID-19 [$\chi^2(4) = 19.515, p < 0.01$].

Mental health diagnoses. Omnibus chi-square tests revealed a relationship between a reported mental health diagnosis and level of overall stress due to the coronavirus pandemic [$\chi^2(2) = 20.654, p < 0.01$] as well as reported worry regarding the pandemic's impact on emotional health [$\chi^2(2) = 17.417, p < 0.01$] and physical health [$\chi^2(2) = 6.229; p < 0.05$]. Post hoc comparisons revealed that adults with NF who were diagnosed with a mental illness were significantly more likely to report high stress than those who were not ($p < 0.01$). In addition, participants with a mental health diagnosis were more likely to report high worry regarding the pandemic's impact on their emotional health ($p < 0.01$) and less likely to report low levels of worry about the pandemic's impact on their physical health ($p < 0.05$). A mental health diagnosis also was associated with degree of NF symptom impact on daily life [$\chi^2(1) = 11.655, p < 0.01$], indicating that adults who rated their NF symptoms as having a moderate-to-severe impact on their daily life were more likely to report a mental health diagnosis than those who rated a mild degree of NF symptom impact.

Among those who were not formally diagnosed with a mental illness, the belief that one had mental health issues was associated with

reported stress due to coronavirus [$X^2(2) = 13.895, p < 0.01$] as well as worry related to its impact on emotional health [$X^2(2) = 21.557, p < 0.01$]. Post-hoc comparisons revealed a similar pattern, with those believing they suffered from mental illness being more likely to report high stress than those who did not ($p < 0.05$); these participants also were more likely to report high worry regarding the pandemic's impact on their emotional health ($p < 0.01$).

Mental health impact. Mental health impact on daily functioning was significantly related to reported levels of COVID-19-related stress [$X^2(2) = 8.183; p < 0.05$]; adults who reported that their mental health difficulties had a mild impact on their everyday life were more likely to report low levels of COVID-19-related stress than those who rated that their mental health difficulties had a moderate-to-severe impact ($p < 0.01$). Mental health impact on daily functioning also was related to reported worry regarding the pandemic's impact on physical [$X^2(2) = 7.845; p < 0.05$] and emotional health [$X^2(2) = 8.559; p < 0.05$] such that adults reporting mild mental health impact were more likely to rate low worry on both items than those reporting a moderate-to-severe mental health impact ($p < 0.05; p < 0.01$; respectively).

NF symptom impact. In the total sample, NF symptom impact was associated with reported worry about one's physical health being influenced by coronavirus [$X^2(2) = 6.127, p < 0.05$] and reported stress due to the coronavirus pandemic [$X^2(2) = 11.313, p < 0.01$]. Post-hoc comparisons revealed that adults who reported moderate-to-severe levels of NF symptom impact were more likely to report high levels of worry regarding the pandemic's effect on their physical health ($p < 0.05$) as well as high levels of coronavirus-related stress compared to those who reported mild NF symptom impact ($p < 0.01$).

3.4 | Access to NF health care and satisfaction with telehealth

Due to the COVID-19 pandemic, 30.2% of participants reported that they had missed an in-person doctor's appointment for their NF care and 18.7% missed an in-person NF treatment because it was canceled or delayed. Those with moderate-to-severe NF symptom impact were more likely than those with mild symptom impact to have missed an in-person doctor's appointment [$X^2(1) = 22.834, p < 0.01$]. Of the adults who missed an appointment for their NF care, almost half (48.3%) reported that they were moderately to highly concerned about missing it. Of those who missed a NF treatment, the majority (64.2%) expressed moderate-to-high concern.

Missing a NF-related medical appointment due to the pandemic was associated with significantly more worry about being infected with coronavirus [$X^2(2) = 11.440, p < 0.01$], friends and family being infected with coronavirus [$X^2(2) = 7.278, p < 0.05$], the influence of the pandemic on physical health [$X^2(2) = 19.015, p < 0.01$], and emotional health [$X^2(2) = 13.775, p < 0.01$] as well as higher overall coronavirus-related stress [$X^2(2) = 15.344, p < 0.01$]. Post-hoc comparisons revealed that those who had missed an appointment were

more likely to report high levels of coronavirus-related worry (all four items; each $p < 0.01$) and stress ($p < 0.01$).

Out of all respondents, 21.4% used telehealth to meet with their NF providers during the coronavirus pandemic, with just over half (51.2%) reporting that it met their needs "very much" or "extremely." Telehealth was used by less than half (43.4%) of the respondents who reported having an in-person appointment for their NF care canceled or delayed due to the pandemic. Of those respondents, 33.3% reported that the telehealth appointment moderately met their needs while 46.2% reported that it highly met their needs (combined ratings of "very much" or "extremely"; Figure 3).

In examining characteristics that may be related to the use of telehealth services, we found that respondents who reported a history of learning difficulties were less likely to use telehealth than those without learning difficulties [$X^2(1) = 3.97, p < 0.05$]. Finally, respondents who reported moderate-to-severe NF symptom impact were significantly more likely to use telehealth than those reporting mild

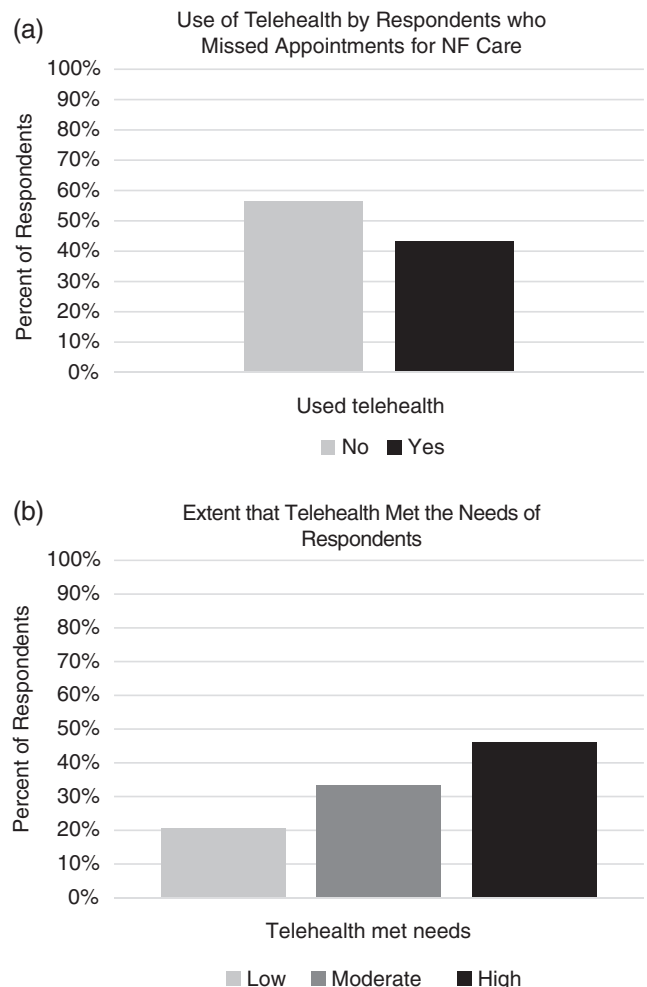


FIGURE 3 (a) Less than half of the respondents who missed a medical appointment for their NF care used telehealth, and (b) one-third of these respondents rated that the telehealth appointment met their needs to a moderate degree while approximately one-half rated that it met their needs to a high degree ("very much" to "extremely")

symptom impact [$X^2(1) = 6.71, p < 0.05$]. Other factors, such as age group, gender identity, mental health diagnosis, and education level were not significantly related to telehealth use.

4 | DISCUSSION

To our knowledge, this paper is the first to assess the perspectives of adults with NF1, NF2, and schwannomatosis regarding the impact of COVID-19 on their mental health and NF-related medical care. Over half of the survey respondents endorsed moderate-to-high levels of overall COVID-19-related stress with the most common stressors being related to finances (unemployment of self, reduced/loss of income) and health concerns (inability to quarantine as an “essential” worker). The survey also found that respondents who were unemployed were more stressed than those who were continuing to work. These COVID-19-related stressors are not unique to those with NF or even those with a chronic medical condition. For example, financial concerns from COVID-19 were rated as highly stressful in a community survey of adults in the United States (Park et al., 2020). Furthermore, previous research has revealed links between unemployment and depression (Zuelke et al., 2018), with longer periods of unemployment being particularly impactful on mental well-being (Latsou & Geitona, 2018). However, past research have shown that many adults with NF1 report delays in completing their education (Doser et al., 2019), likely due in part to the cognitive and attention problems that are common among these individuals (Descheemaeker et al., 2013; X. Wang et al., 2019). These educational delays and cognitive impairments may limit employment opportunities, making the possibility of losing income even more concerning. Indeed, research has documented lower employment rates than the general population in both NF1 and NF2 (Hamoy-Jimenez et al., 2020). Related to the health concerns reported during the pandemic, group differences were evident when examining respondents with varying levels of NF symptom impact. Specifically, those with moderate-to-severe NF symptom impact reported more overall stress and worries about their physical health being impacted by COVID-19 compared with those with mild NF symptom impact. Perhaps individuals with greater NF symptom impact feel susceptible to other medical conditions and tend to worry more about their health since they may already experience heightened concern about their day-to-day physical well-being.

Notable gender differences emerged from the survey data. Women were more likely than men to report high levels of coronavirus-related stress, worry about self or others being infected with coronavirus, and worry about their physical and emotional health being impacted by the pandemic. Women with NF are not alone in their tendency to report higher stress during the current pandemic. Female gender conveyed more risk for psychological distress or poorer well-being due to COVID-19 in numerous other studies and across various countries (González-Sanguino et al., 2020; Liu, Zhang, Wei, et al., 2020; Mazza et al., 2020; Park et al., 2020). This finding is not surprising given previous research documenting higher rates of depression and anxiety in females than those in males (Seedat

et al., 2009) as well as poorer mental health in female caregivers compared to that in male caregivers (Edwards et al., 2017). Women may experience higher stress associated with the coronavirus pandemic due to an increased burden of domestic responsibilities, including childcare, homeschooling, and the increase in housework with many family members at home, as well as the addition of remote work or loss of employment outside the home (Farré et al., 2020).

Slightly over half of survey respondents endorsed having a diagnosed mental health disorder while almost an additional third reported having a mental health problem that was undiagnosed. These respondents reported higher levels of COVID-19-related stress than those without a known or suspected mental health diagnosis; however, only about half of those with a diagnosis reported seeing a mental health professional. Similarly, in the general US population, rates of generalized anxiety disorders increased during the early stages of the COVID-19 pandemic and were related to higher levels of stress (Cordaro et al., 2021). In the United States, it is estimated that about one in five adults experience mental illness in a given year, while less than half receive treatment (SAMHSA, 2020). Since many respondents in our survey reported untreated mental health problems, they may be more susceptible to the stressors that arose with the COVID-19 pandemic. Our survey data also indicated that young adults with NF were more likely to report high levels of worry about the pandemic's influence on their emotional health than older adults, which is consistent with current findings in the general population (Czeisler et al., 2020; Liu, Zhang, Wong, et al., 2020). In addition, mental health diagnoses were associated with moderate-to-severe NF symptom impact. In the general population, research has identified an interaction between mental and physical health such that adults with mental health difficulties are more likely to develop physical conditions and vice versa (Doherty & Gaughran, 2014). Furthermore, individuals with both mental and physical difficulties are at an increased risk of having other morbidities, poorer health outcomes, and reduced quality of life (Doherty & Gaughran, 2014). Thus, routine appointments for multidisciplinary NF care should incorporate mental health screenings to facilitate referrals for psychiatric needs. Such integrated care would allow providers to follow both the physical and mental health of their patients closely, including to monitor and support their COVID-19-related psychosocial concerns (Pfefferbaum & North, 2020) and educate them about current COVID-19 guidelines.

During the first 6 months of the coronavirus pandemic, a third of respondents reported missing an in-person doctor visit for their routine NF care and slightly under a quarter endorsed missing a scheduled NF treatment that had been delayed or canceled at the time of survey participation. Missed doctors' appointments also may delay obtaining an initial diagnosis of NF and starting treatments. It is likely that missing these appointments were influenced by both clinic cancellation policies and patient factors. Notably, patients who reported higher impact of their NF disease symptoms were more likely to report missing an appointment, possibly because they generally have more frequent health care visits. Missing an appointment also was related to more COVID-19 stress in general and more worry about being infected. Thus, some of these individuals may have decided to

forgo an appointment due to concerns about possible infection when traveling to or visiting a clinic. However, slightly under half of the respondents who had missed an appointment were engaging in telehealth, and only about half of those said that telehealth appointments highly met their needs. In particular, individuals were less likely to use telehealth if they had a history of learning difficulties and only mild NF symptoms that impacted their daily life. After the majority of NF clinics moved to telehealth, NF clinicians reported high satisfaction with the use of telehealth but had concerns about not being able to conduct physical exams (Radtke et al., 2021).

Barriers to telehealth among care providers in general included concerns about reimbursement (Scott Kruse et al., 2018; Sisk et al., 2020) and the technology (Helleman et al., 2020), which was similar among NF clinicians (Radtke et al., 2021). Obvious benefits of telehealth for patients include eliminating travel time and increased access among those in rural or underserved communities (Madigan et al., 2020). However, previous research have documented that patients are less likely to use telehealth if they are older, of lower socioeconomic status (SES), less educated, and less computer-literate (Scott Kruse et al., 2018) so challenges remain to the widespread adoption of such technology. Importantly, when patients defer examinations or treatments it can cause “downstream consequences,” leading to more serious health care needs in the future (Wosik et al., 2020).

Results of this survey should be interpreted in the context of some limitations. First, the survey was cross-sectional and causality of relationships cannot be implied. Second, the survey sample had limited diversity and some potential bias affecting the generalization and interpretation of results. The sample consisted primarily of White respondents. This outcome is due to the generally White representation of the international NF Registry (Seidlin et al., 2017), which was the largest source used for recruitment. Furthermore, all respondents were English-speaking, and the majority were from the United States and female. As such, the results reflect the perspectives of the adults with NF enrolled in the registry but do not represent the racial, ethnic, and sex distribution of the NF population in the United States or other countries (Anderson & Gutmann, 2015; Le & Bedocs, 2020). In addition, those with mental health concerns may have been more likely to complete the psychosocial survey, which may result in some ascertainment bias in the sample that could have influenced the outcomes. Third, we did not collect information about SES. We have data on education level, which often is used as a proxy for SES, but income levels may be more informative for directly understanding stressors faced by individuals who have lost income due to COVID-19. Fourth, the survey was conducted within the first 6 months during the initial lockdowns that caused an abrupt stop to clinical services followed by a transition to telehealth. It is unclear whether the same patient barriers noted in this study are still relevant, as telehealth medical visits are more routinely being implemented across the country even as in-person services are beginning to return. Finally, the level of COVID-19-related restrictions (e.g., quarantine, businesses closing) differed across the United States and other countries as well as specific areas

within a country, making it difficult to make sweeping statements about the impact of the coronavirus across regions. However, respondents were asked to answer the survey questions considering “the worst of the coronavirus pandemic in your area.” Thus, this report offers a revealing snapshot of how COVID-19 was impacting adults with NF during the most restrictive time periods.

Results of this survey have important implications for future research. First, due to the significant disparities in health care across racial and ethnic groups, research with a more diverse sample is critical for understanding the full impact of the coronavirus pandemic on emotional and physical health as well as barriers to health care access and telehealth delivery. A top priority is to improve the racial and ethnic representation as well as international participation in future studies to learn more about and address the health disparities related to COVID-19 and in general. A critical next step to help meet this priority is to disseminate surveys written in languages other than English. Second, more research is needed to investigate specific barriers and facilitators of telehealth to promote wide implementation across various groups for medical care. While prior qualitative and quantitative studies have shown that telehealth interventions are feasible and associated with improvements in depression, anxiety, pain, coping skills, and quality of life (Carter et al., 2020; Greenberg et al., 2019; Lester et al., 2020), studies should explore the patients' perceptions of telehealth for medical appointments. Given the documented burden of medical care for NF and limited specialized NF clinics, using telehealth for some medical visits may be more convenient and may increase the engagement of patients in their routine NF care. Medical providers should consider discussing the option of telehealth with their patients with NF and problem solve potential barriers through education and training. For patients with learning difficulties, who were the least likely to use telehealth, teaching them how to set up and use telehealth services may help with engagement. Finally, a large body of research documents the complex psychosocial factors associated with NF and the importance of assessing and addressing these comorbidities. However, future surveys should include questions about the particular type of mental health issues diagnosed in individuals with NF and more about the therapeutic services they receive to identify more specific needs. The current survey results support closely monitoring the stress and concerns of adults with NF, particularly women and those with preexisting mental health difficulties or greater NF symptom impact, to assess for difficulties coping with the coronavirus pandemic, and to facilitate mental health referrals when necessary.

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CONFLICT OF INTEREST

None of the authors have any conflicts of interest to report.

AUTHOR CONTRIBUTIONS

All authors have agreed to the order in which their names are listed in the manuscript. They all have contributed significantly to the work per the journal's criteria as listed below. All authors have given final approval of the version to be published and agree to be accountable for all aspects of the work. Contributions from others who does not meet the criteria for authorship are listed in the Acknowledgments section with permission from the contributor. Pamela L. Wolters: Design and conceptualization of the study; acquisition, analysis, and interpretation of the data; drafting and revising the manuscript. Stephanie Reda: Acquisition, analysis, and interpretation of the data; drafting and revising the manuscript. Staci Martin: Design and conceptualization of the study; interpretation of the data; drafting and revising the manuscript. Nour Al Ghriwati: Interpretation of the data; drafting and revising the manuscript. Melissa Baker: Acquisition, analysis, and interpretation of the data, revising the manuscript. Dale Berg: Design and conceptualization of the study; interpretation of the data; revising the manuscript. Greg Erickson: Design and conceptualization of the study; interpretation of the data; revising the manuscript. Barbara Franklin: Design and conceptualization of the study; interpretation of the data; revising the manuscript. Vanessa L. Merker: Design and conceptualization of the study; interpretation of the data; revising the manuscript. Beverly Oberlander: Design and conceptualization of the study; interpretation of the data; revising the manuscript. Stephanie Reeve: Design and conceptualization of the study; interpretation of the data; revising the manuscript. Claas Röhl: Design and conceptualization of the study; interpretation of the data; revising the manuscript. Tena Rosser: Design and conceptualization of the study; interpretation of the data; revising the manuscript. Mary Anne Toledo-Tamula: Acquisition, analysis, and interpretation of the data; revising the manuscript. Ana-Maria Vranceanu: Design and conceptualization of the study; acquisition and interpretation of the data, revising the manuscript.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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