Assessing the Impact of a Telemedicine Program on Caregivers of Children with Complex Chronic Conditions

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Abstract

Background: The COVID-19 pandemic was a paradigm shift in global healthcare. This situation highlighted the role of telemedicine in adapting to the care requirements of pediatric patients and their families by facilitating remote consultations and ensuring continuity of care.

Objective: We aim to establish the usefulness of a telemedicine program for caregivers of complex chronic children

Methods: We performed a quasi-experimental pre-post intervention study of a telemedicine program, regarding healthcare system use and caregiver quality of life, and comparing two periods: before and during the COVID-19 pandemic. The study was performed on caregivers of complex chronic children followed in a specialized unit

Results: 34 families were included. The mean number of visits per year was higher in the pre-intervention period for both primary care consultations ($p<0.039$) and hospital-specialized medical consultations ($p<0.031$). The number of emergency room visits per year was lower in the pandemic period compared to the pre-pandemic period ($p<0.012$). In both groups, an improvement in caregiver quality of life at 12 months was detected ($p<0.035$). However, the Rosenberg self-esteem scale score of the primary caregiver was significantly lower at three months compared to the baseline ($p<0.030$)

Conclusions: Our study demonstrates that the use of a telemedicine program during the pandemic, resulted in a decrease in scheduled face-to-face care and a reduction in the number of emergency department visits. Regarding the caregiver quality of life, it was worse in those families who had a child affected by a neurological condition or whose child was older when the disease was diagnosed. Clinical Trial: This study was approved by the Research Ethics Committee of Malaga in May 2017 (reference: PIN-0287-2016).

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AUTHORSHIP CONTRIBUTION STATEMENT

Aurora Madrid-Rodríguez performed writing and original draft conceptualization. María José Peláez-Cantero performed formal analysis. Ana Suárez-Carrasco and Antonia Gámez-Ruiz performed investigation. Elena Godoy-Molina performed writing, review and editing. Alfonso Lendínez-Jurado and Isabel Leiva-Gea designed methodology, provided resources and revised the final manuscript.

ABSTRACT

Introduction: The COVID-19 pandemic was a paradigm shift in global healthcare. This situation highlighted the role of telemedicine in adapting to the care requirements of pediatric patients and their families by facilitating remote consultations and ensuring continuity of care.

Objetive: We aim to establish the usefulness of a telemedicine program for caregivers of complex chronic children.

Materials and Methods: We performed a quasi-experimental pre-post intervention study of a telemedicine program, regarding healthcare system use and caregiver quality of life, and comparing two periods: before and during the COVID-19 pandemic. The study was performed on caregivers of complex chronic children followed in a specialized unit.

Results: 34 families were included. The mean number of visits per year was higher in the pre-intervention period for both primary care consultations (p<0.039) and hospital-specialized medical consultations (p<0.031). The number of emergency room visits per year was lower in the pandemic period compared to the pre-pandemic period (p<0.012). In both groups, an improvement in caregiver quality of life at 12 months was detected (p<0.035). However, the Rosenberg self-esteem scale score of the primary caregiver was significantly lower at three months compared to the baseline (p<0.030).

Discussion: Our study demonstrates that the use of a telemedicine program during the pandemic, resulted in a decrease in scheduled face-to-face care and a reduction in the number of emergency department visits. Regarding the caregiver quality of life, it was worse in those families who had a child affected by a neurological condition or whose child was older when the disease was diagnosed.

Conclusions: Our study demonstrates the effectiveness of a Telemedicine program in the care of complex chronic patients and their families in the context of the COVID-19 pandemic.

Keywords: telemedicine; telehealth; complex chronic children; pediatrics complex chronic conditions; palliative care; COVID-19.

1. INTRODUCTION

Technological advances and progress in medicine have reduced infant mortality

(1,2). These survival successes have contributed to an increase in the prevalence of incurable diseases and disability, which leads us to reassess the way we approach and organize health services to adapt them to these new care needs (3,4).

In developed countries, children with a complex chronic condition (CCCs) represent 0.67-5% of the pediatric population, but consume up to 40% of total health expenditure, involving a high level of care both in and out of hospital, primary care, and non-health services (5-9).

The prevalence of life-threatening or life-limiting illness has increased in recent decades, with an estimated 61.1 per 10,000 children aged 1-19 years in the United Kingdom. In all studies, approximately 30% have cancer; the remaining 70% comprise a combination of conditions, mainly neurodegenerative, metabolic, and genetic. Of these, 50% will require the specialized care of a pediatric palliative care unit (10,11).

A new strategy is therefore needed to transform the current care model, which is focused on treatment and in which the patient is a passive subject, into a proactive model more centered on the needs of patients and their caregivers, in which they have more information and are given more autonomy to take an active role in managing their disease (12,13).

In this sense, new information and communication technologies (ICTs) provide tools that enable access to a wide range of resources. In 2020, the COVID-19 pandemic transformed medical care through telemedicine with a demand for remote health care and telehealth (14,15). While the world faced an unprecedented pandemic, patients with chronic diseases needed special attention on an ongoing basis and, if warranted, an adaptation of their usual care (16).

In complex chronic patient and pediatric palliative care units, telemedicine provides an accessible and equitable tool. With these tools, families can remain in contact with professionals establishing a schedule for specific services in a time-effective manner and adapted to the stage of the disease (17,18).

Due to the COVID-19 pandemic, and considering the vulnerability and fragility of these patients, all the tools offered by ICTs in the continuity of care have become even more useful, since they allow non-face-to-face visits; but with a capacity comparable to face-to-face visits, minimizing the risk of contagion and reducing the need to travel to a medical center (19).

This study aimed to develop and evaluate a Telemedicine program for the care of complex chronic children or those receiving palliative care, in a complex chronic and pediatric palliative care unit, during the COVID-19 pandemic.

2. MATERIALS AND METHODS

We design a quasi-experimental pre-post intervention study in a Complex Chronic Patient and Palliative Care Unit of a pediatric tertiary hospital.

The study population comprised complex chronic children or children needing palliative care and their families. As inclusion criteria, children must meet the definition of the medical complexity algorithm defined by the Seattle Children's Hospital Group and score at least 6,5 points on the PedCom Scale (20,21). They also had to be followed in the Complex Chronic Patient and Pediatric Palliative Care Unit of the hospital and had a high level of complexity, defined not only by the identification of the child as a complex chronic patient but also by the need of high consumption of resources: two or more hospital admissions; at least one admission to intensive care or six or more visits to the emergency department in the last twelve months) (22). Patients older than 18 years of age or those families not signing the informed consent where excluded.

2.1. Telemedicine Program Description

The Telemedicine program has remote management and control from the hospital and the technical support center. It has three main functionalities:

1. Communication: The technology used was an Android-based smart TV platform

connected to a television, which allows access to advanced telecommunication and medical device management services. It allows videoconferencing between patient (through the TV) and hospital (via PC) or patient and medical professional (via PC, tablet or Smartphone), based on SIP technology (market standard).

2. Information/Training: by the platform families could access a menu with audiovisual contents. The contents were developed by the professionals of the unit and includes:
    - Information including any telephone numbers of interest for caregivers.
    - Access to videos with recommendations, procedures, and theoretical and practical workshops including specific training videos and Power point presentations on techniques and care.
    - Information on the current vaccination schedule.

3. Health care: allows clinical assessment via video call, with the option of requesting tests (analytical and radiological), modification of technological support parameters, and electronic medical prescriptions.

During recruitment, to avoid selection bias caused by the digital divide in cases where families did not have internet at home, internet was provided through project funding.

2.2 Evaluation of the Program

The evaluation of the program was undertaken through the comparison of two one-year time periods, before the pandemic (March 2019 to February 2020) and during the pandemic (March 2020 to February 2021).

Sociodemographic variables described are: age, gender, primary caregiver (parent and/or both), number of siblings, place of residence, and country of origin.

1. Evaluation of face-to-face health care variables:
    a. Scheduled:
        - Primary care consultations.
        - Hospital outpatient consultations per year.
        - Number of outpatient consultations in the Unit.
        - Number of home visits made by the Unit.
        - Number of scheduled hospital admissions.
    b. Urgent:
        - Number of emergency room visits.
        - Number of urgent hospital admissions.

2. Evaluation of telephone health care variables:
    - Number of calls from the Unit to caregivers.
    - Number of calls from caregivers to the Unit.

3. Caregiver reported outcomes:
    - The Rosenberg Self-Esteem Scale (23,24).
    - The Positive and Negative Affect Schedule: PANAS (25,26).
    - Health-related quality of life of primary caregivers: EuroQol-5D-5L (27).
    - Short Zarit scale to assess primary caregiver burden (28,29).

The evaluation of the Telemedicine program was carried out with periodic assessments (baseline, 3, 6 and 12 months) of the health care variables and caregiver reported outcomes. The last assessment was carried out 12 months after the start of the intervention. The health care data was collected by telephone and the tests by mail and email.

2.3 Data analysis

Data analysis was performed using free R 4.0.2 software (R-CoreTeam 2020) (https://www.r-projetc.org/). A Shapiro-Wilk test analysis was performed to determine the normality of the study variables. Results are presented as mean±SD values in normal distributions or as median (IQR) in non-normal distributions. For independent quantitative variables, Student's *t*-test was used, if they followed a normal distribution or the Mann-Whitney U test if they were not normal. A Wilcoxon signed-rank test was performed to analyze differences in the non-normal distributions, and the paired t-test was used in the normal distributions. To compare qualitative variables, a chi-square test was used in independent samples and McNemar's test in related samples. A P-value <0.05 was considered statistically significant. P-values were adjusted using the Benjamini-Hochberg correction for multiple comparisons.

3. RESULTS

3.1. Study Population Characteristics

A total of 34 patients were included, ranging in age from 3 months to 14.2 years, with a median of 4.9 years, 52.9% (18/34) of whom were female. The CCC that most affected our patients was neurological in 73.5% (25/34), followed by gastrointestinal in 70.6% (24/34), respiratory in 50% (17/34), cardiovascular in 41.2% (14/34), and oncological in 20.6% (7/34). In addition, neurological CCCs were also the most frequent main CCC in 47.06% (16/34), with cerebral palsy being the most prevalent neurological disease (68.75%, 11/16).

The primary caregiver was the mother in 82.4% (28/34) and 47.1% (16/34) of the families received social assistance; 91.2% of the families (31/34) received psychological care. 29.4% of the families (10/34) were from a foreign country and 41.2% (14/34) had no other children, with a maximum of 5 children and a median of 1 child.

3.2 Impact of the Telemedicine program on health care activity during the pandemic

Regarding scheduled care, bivariate analysis showed that the mean number of visits per year to primary care consultations was higher in the pre-pandemic period ($p<0.039$), as were hospital specialty consultations ($p<0.031$) (Table 1).

Table 1. Analysis of the variables associated with face-to-face health care activity.

TYPE OF CARE ACTIVITY		Pre-Telemedicine period	Pandemic period with Telemedicine	*P* value
		Mean (SD)	Mean (SD)	
SCHEDULED	PRIMARY CARE CONSULTATIONS	9.2 (9.9)	6.6 (5.9)	.039
	SCHEDULED CONSULTATIONS HOSPITAL SPECIALTIES	29.6 (192)	23.4 (19.2)	.031
	NURSE CONSULTATIONS CC AND PPC UNIT	8.3 (6.8)	10.1 (9.5)	.545
	PEDIATRICIAN CONSULTATIONS CC AND PPC UNIT	6.7 (5.6)	7.2 (5.6)	.772
	NURSING HOME VISITS CC AND PPC UNIT	3.1 (5.3)	2.4 (4.8)	.209
	PEDIATRICIAN HOME VISITS CC AND PPC UNIT	2.8 (5.0)	2.1 (4.4)	.097
	SCHEDULED ADMISSIONS	0.6 (1.0)	0.6 (0.8)	.603
URGENT	URGENT - EMERGENCY HOSPITAL VISITS	3.6 (4.0)	2.1 (2.1)	.012
	URGENT HOSPITAL ADMISSIONS	1.4 (1.5)	1.2 (1.5)	.199

SD: Standard deviation. CC: complex chronic; PPC: pediatric palliative care.

In terms of emergency care, there were significant differences in the number of emergency department visits per year when comparing the pre-pandemic and pandemic periods with fewer visits during the pandemic ($p<0.012$). No significant differences were found between periods for emergency hospital admissions (Table 1).

With respect to before and during pandemic telephone care carried out by the Unit, we noted a non-significant greater number of calls made by the Unit during the pandemic ($p<0.059$). There was a significant increase in telephone calls made by caregivers to the Unit ($p<0.003$) when comparing the two periods of care (Table 2).

Table 2. Analysis of the variables in relation to telephone health care activity.

TYPE OF CARE ACTIVITY	Pre-Telemedicine period	Pandemic period with Telemedicine	*P* value

	Mean (SD)	Mean (SD)	
UNIT CALLS TO CAREGIVERS	36.4 (34)	48 (26)	.059
CAREGIVERS CALLS TO UNIT	6.2 (7.8)	10.5 (9.5)	.003

SD: Standard deviation

There were no significant differences between the mean length of stay measured in days of hospital admission before and during the pandemic.

Tables 1 and 2 display the variables related to the health care provided.

3.3 Impact of the Telemedicine program on the caregiver reported outcomes

Significant differences were seen in the VAS values of the EQ-5D quality of life test of the primary caregivers at 12 months compared to baseline with an increase in VAS scores at 12 months (*P* .035).

The Rosenberg self-esteem scale scores of the primary caregivers were significantly lower at 3 months than at baseline (*P* .030).

There was a significant negative correlation between the short Zarit scale for primary caregiver burden at 6 months with the number of siblings (adjusted R^2=37%, *P* .009).

The VAS value of the EQ-5D quality of life scale of the primary caregivers at 12 months correlated negatively and significantly with age at diagnosis of the child's illness (adjusted $R^{2=}$24%, *P* .049).

The variable difference of the index value of the EQ-5D quality of life scale of the primary caregivers from baseline to 3 months was negatively correlated with age at patient admission (adjusted R^2= 97%, *P* .007).

In the analysis of the relationship between caregiver quality of life and the different types of CCCs, the variable difference from baseline to the 3-month index value of the EQ-5D quality of life scale of the primary caregivers was negatively correlated with neurological disease (adjusted R^2 =89%, *P* .039).

Table 3 shows the results of the tests at the start of the intervention and at 3, 6, and 12 months.

Table 3. Analysis of test results

		0 months Mean (SD)	**3 months** Mean (SD)	**6 months** Mean (SD)	**12 months** Mean (SD)	***P* value**
Rosenberg self-esteem scale		29.3 (5.2)	27.3 (6.4)	31.0 (4.2)	33.0 (4.8)	.030
Positive and negative affectivity scales PANAS	Positive affect	29.1 (7.4)	29.5 (7.9)	33.1 (9.4)	33.8 (7.5)	.413
	Negative affect	26.8 (8.8)	23.8 (6.8)	24.3 (7.1)	22.8 (6.2)	.348
Health-related quality of life EuroQol-5D-5L	EQ5D VALUE	0.814 (0.180)	0.766 (0.151)	0.838 (0.153)	0.838 (0.142)	.053
	EQ5D VAS	76 (18)	75 (15)	78 (14)	75 (21)	.035
Short ZARIT scale		20.1 (7.0)	24.3 (5.0)	20.8 (5.9)	19.8 (5.7)	.440

SD: Standard deviation

4. DISCUSSION

Our study demonstrates the effectiveness of a Telemedicine program in the care of these patients and their families in the context of the COVID-19 pandemic, with a decrease in both scheduled and urgent health care services.

In our study, the main condition of the patient was neurological; being the most frequent condition as it has been widely described in the literature that neurological patients are the most prevalent, our results are in accordance with this, with neurological CCCs being the most frequent, followed by gastrointestinal and respiratory CCCs. In our sample, also coinciding with published findings, the mother fulfills the role of primary caregiver (13,30-34).

During the pandemic, in line with other publications, there was evidence of a decrease in scheduled care in children with CCCs as well as in urgent care, which occurred both in the general pediatric population, and in children with CCCs or with special health care needs (35-42).

The increase in telephone care during the pandemic, both by the Unit, but especially by the increase in calls from caregivers, may be explained by the decrease in face-to-face care in primary care and in specialized hospital care during this period. This finding highlights the telephone support provided by the Unit to minimize the impact of the pandemic on children with CCCs, as stated in several publications on specific care strategies aimed at this particularly vulnerable population (43).

In our study, the absence of significant differences between hospital admissions, both scheduled and urgent, has been described in other studies on telemedicine in children with CCCs (18) and may be due to the high complexity of the patients included in this study, where pandemic-related factors did not influence either the number of hospital admissions or the length of hospital stay.

Several studies have described the importance of the psychosocial needs of children with CCCs and their families and how these needs, especially about mental health, increased during the pandemic (44-45). Regarding the impact of telemedicine programs on quality of life, the publications to date have not demonstrated an improvement in the quality of life of caregivers, although studies of the interventions are usually cross-sectional, without multiple cut-off points. Our work therefore highlights the importance of conducting studies that monitor the impact and benefits of these programs throughout the intervention to verify their usefulness (46-48).

In our study, a greater burden on primary caregivers was detected in families with more children, a demographic factor not previously described that should be considered to reinforce support for these families.

The quality of life of the primary caregivers measured by the VAS value of the EQ-5D scale at 12 months was poorer in those with children with CCCs diagnosed at an older age. Similarly, we found a negative correlation between the difference in the index value of the 5Q-5D quality of life scale of the primary caregivers at 3 months compared to baseline in those with children with CCCs who were older at admission to the Unit. We observed that an older age of the child at diagnosis and at admission to the Unit negatively influenced the quality of life of the parents with a nadir at 3 months and at 12 months during the pandemic. The influence of the age of the child with a CCC on the quality of life of the primary caregivers during the pandemic has not been examined. We should therefore analyze the factor of child age as a modulator of the quality of life of the parents.

Studies published on the impact of neurological diseases have shown an increased family, work, and economic burden on primary caregivers, specifically on caregivers of children with cerebral palsy (49). In our study, a negative correlation was found between the index value of the 5Q-5D quality of life scale of the primary caregiver at 3 months compared to baseline with neurological CCCs, which was also the most frequent CCC in our study.

The findings of lower self-esteem in the primary caregiver with a nadir at 3 months of the Telemedicine program in the pandemic setting could be due both to being in full confinement with less psychosocial and family support, such as the adaptation of home care with a change of care support, and the care burden that this may have entailed. This nadir in terms of quality of life of the primary caregiver at 3 months has not been evaluated in other studies and may be useful for reinforcing psychosocial support to families, considering this period as a turning point in caregiving. On the other hand, the fact that the VAS of the EQ-5D quality of life test revealed an increased quality of life at 12 months could be attributed to adaptation to the situation, as well as decreased isolation measures one year after the pandemic.

The use of telemedicine prior to the pandemic was demonstrated to be effective in children with CCCs, both in reducing the number of emergency department visits and in reducing costs (48,50-54). In our study, however, it was difficult to determine with certainty whether the decrease in urgent or unscheduled care was due to the COVID-19 pandemic or to the intervention of the Telemedicine program. It was found that during the pandemic there was a decrease in emergency department visits, as many caregivers were afraid to go to hospitals to avoid their children becoming infected (36,38). In addition, we must consider the decrease in respiratory symptoms due to the confinement and isolation measures described in several studies (38,40,42). Studies with larger and more homogeneous samples comparing the Telemedicine intervention after the pandemic should be carried out to assess whether its effectiveness in terms of care is maintained without the interference caused by the pandemic.

It is important to leverage the growth of the ICTs that emerged during the pandemic for the benefit of patients, especially those most vulnerable and with specific care needs such as children with CCCs. Telemedicine provides equity in care. Efforts should be made to evaluate the results of telemedicine programs in patients, primary caregivers, and healthcare teams, bearing in mind health care, economic, and quality of life variables in patients and their families monitored over time.

5. CONCLUSIONS

In our study, the use of a telemedicine program a pandemic setting resulted in a decrease in scheduled face-to-face care and a reduction in the number of emergency department visits compared to the pre-pandemic period. On the other hand, we saw an increase in telephone support without any impact on hospital admissions. We identified the most vulnerable families in this group of patients as those with a child affected by a neurological condition, those who had an older child when the disease was diagnosed and families with a greater number of children, these allows us to consider the redirection of resources in case of being limited. Our results indicate that the Telemedicine program was effective in the care of children with CCC and their families during the COVID-19 pandemic.

6. ACKNOWLEDGEMENTS

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7. CONFLICT OF INTEREST

The Authors declare no conflict of interest.

8. FUNDING

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9. ETHICS

This study was approved by the Research Ethics Committee of Malaga in May 2017 (reference: PIN-0287-2016).

REFERENCES

1. Cooter R, Pickstone J. Medicine in the Twentieth Century. Amsterdam: Taylor & Francis; 2000.
2. Porter R. The Greatest Benefit to Mankind: A Medical History of Humanity from Antiquity to the Present. London: Harper Collins;1997.
3. Cohen E, Friedman J, Nicholas DB, et al. A home for medically complex children: The role of hospital programs. J Healthc Qual. 2008;30:7-15. [Doi: 10.1111/j.1945-1474.2008.tb01137.x].
4. WHO. Busse R, Blümel M, Scheller-Kreinsen D, et al. Tackling Chronic Disease in Europe: Strategies Interventions and Challenges: No. 20 (Observatory Studies Series). The European Observatory on Health Systems and Policies. World Health Organization, 2010. Available from : https://www.euro.who.int/__data/assets/pdf_file/0008/96632/E93736.pdf [Last accesed: 07-02-2022].
5. Berry JG, Hall M, Neff J, et al. Children with medical complexity and Medicaid: Spending and cost savings. Health Aff (Millwood). 2014;33:2199-206. [Doi: 10.1377/hlthaff.2014.0828].
6. Braddock ME, Leutgeb V, Zhang L, et al. Factors influencing recurrent admissions among children with disabilities in a specialty children's hospital. J Pediatr Rehabil Med. 2015;8:131-9. [Doi: 10.3233/PRM-150326].
7. Dunbar P, Hall M, Gay JC, et al. Hospital Readmission of Adolescents and Young Adults With Complex Chronic Disease. JAMA Netw Open. 2019;2:e197613. [Doi: 10.1001/jamanetworkopen.2019.7613].
8. Simon TD, Berry J, Feudtner C, et al. Children with complex chronic conditions in inpatient hospital settings in the United States. Pediatrics. 2010;126(4):647-655. [Doi: 10.1542/peds.2009-3266].
9. Cohen E, Berry JG, Camacho X, et al. Patterns and costs of health care use of children with medical complexity. Pediatrics. 2012;130(6):e1463-70. [Doi: 10.1542/peds.2012-0175].
10. Fraser LK, Gibson-Smith D, Jarvis S, et al. Estimating the current and future prevalence of life-limiting conditions in children in England. Palliat Med. 2021 Oct;35(9):1641-1651. [Doi: 10.1177/0269216320975308].
11. IMPaCCT: standards for paediatric palliative care in Europe. European Journal of

Palliative Care. 2007;14(3):109–114.

12. Institute of Medicine. Crossing the quality chasm: a new health system for the 21st century. Washington, D.C.: The National Academies Press; 2001.
13. Mead N, Bower P. Patient-centredness: a conceptual framework and review of the empirical literature. Soc Sci Med. 2000;51(7):1087–1110. [Doi: 10.1016/s0277-9536(00)00098-8].
14. Wosik J, Fudim M, Cameron B, et al. Telehealth transformation: COVID-19 and the rise of virtual care. J Am Med Inform Assoc. 2020;27(6):957-962. [Doi: 10.1093/jamia/ocaa067].
15. Barney A, Buckelew S, Mesheriakova V, et al. The COVID-19 pandemic and rapid implementation of adolescent and young adult telemedicine: challenges and opportunities for innovation. J Adolesc Health. 2020;67(2):164–171. [Doi: 10.1016/j.jadohealth.2020.05.006].
16. Low JK, Manias E. Use of Technology-Based Tools to Support Adolescents and Young Adults With Chronic Disease: Systematic Review and Meta-Analysis. JMIR Mhealth Uhealth. 2019;7(7):e12042. [Doi: 10.2196/12042].
17. Bird M, Li L, Ouellette C, et al. Use of Synchronous Digital Health Technologies for the Care of Children With Special Health Care Needs and Their Families: Scoping Review. JMIR Pediatr Parent. 2019;2:e15106. [Doi: 10.2196/15106].
18. Braun L, Steurer M, Henry D. Healthcare Utilization of Complex Chronically Ill Children Managed by a Telehealth-Based Team. Front. Pediatr. 2021;9:689572. [Doi: 10.3389/fped.2021.689572].
19. Shah AC, Badawy SM. Telemedicine in Pediatrics: Systematic Review of Randomized Controlled Trials. JMIR Pediatr Parent. 2021;4(1):e22696. [Doi:10.2196/22696].
20. Simon TD, Cawthon ML, Stanford S, et al. Center of Excellence on Quality of Care Measures for Children with Complex Needs (COE4CCN) Medical Complexity Working Group. Pediatric medical complexity algorithm: a new method to stratify children by medical complexity. Pediatrics. 2014;133(6):e1647-1654. [Doi: 10.1542/peds.2013-3875].
21. Godoy-Molina E, Fernández-Ferrández T, Ruiz-Sánchez JM, et al. A scale for the identification of the complex chronic pediatric patient (PedCom Scale): A pilot Study. An Pediatr (Engl Ed). 2022;97(3):155-160. [DOI: 10.1016/j.anpede.2021.10.004].

22. Parente V, Parnell L, Childers J, et al. Point-of-Care Complexity Screening Algorithm to Identify Children With Medical Complexity. Hosp Pediatr. 2021;11(1):44-51. [Doi: 10.1542/hpeds.2020-0066].
23. Rosenberg, M. Society and the adolescent self-image. Princeton, NJ: Princeton University Press,1965.
24. Atienza L, Moreno-Balaguer I. An analysis of the dimensionality of the Rosenberg self-esteem scale in a sample of Valencian adolescents. Universitas Tarraconensis. 2000;22:34-45.
25. Watson D, Clark LA, Tellegen A. Development and validation of brief measures of positive and negative affect: the PANAS scales. J Pers Soc Psychol. 1988;54(6):1063-1070. [Doi: 10.1037//0022-3514.54.6.1063].
26. Joinar TE, Sandín B, Chorot P, et al. Development and factor-analytic validation of the SPANAS among women in Spain: (More) cross-cultural convergence in the structure of mood. Journal of Personality Assessment, 1997:68(3):600-615. [Doi: 10.1207/s15327752jpa6803_8].
27. Herdman M, Gudex C, Lloyd A, et al. Development and preliminary testing of the new five-level version of EQ-5D (EQ-5D-5L). Qual Life Res. 2011;20(10):1727-1736. [Doi: 10.1007/s11136-011-9903-x].
28. Bédard M, Molloy DW, Squire L, et al. The Zarit Burden Interview: a new short version and screening version. Gerontologist. 2001;41(5):652-657. [Doi: 10.1093/geront/41.5.652].
29. Gort AM, March J, Gómez X, et al. Escala de Zarit reducida en cuidados paliativos. Med Clin (Barc). 2005;124: 651-653. [Doi: 10.1157/13074742].
30. Climent FJ, García M, Escosa L, et al. Children’s medically complex diseases unit. A model required in all our hospitals. An Pediatr (Engl Ed). 2018;88(1):12-18. [Doi: 10.1016/j.anpedi.2017.04.002].
31. Gordon JB, Colby HH, Bartelt T, et al. A tertiary care-primary care partnership model for medically complex and fragile children and youth with special health care needs. Arch Pediatr Adolesc Med. 2007;161(10):937-944. [Doi: 10.1001/archpedi.161.10.937].
32. Berry JG, Agrawal R, Kuo DZ, et al. Characteristics of hospitalizations for patients who use a structured clinical care program for children with medical complexity. J Pediatr. 2011;159(2):284-290. [Doi: 10.1016/j.jpeds.2011.02.002].

33. Rehm RS. Nursing's contribution to research about parenting children with complex chronic conditions An integrative review, 2002 to 2012. Nurs Outlook. 2013;61:266-290. [Doi: 10.1016/j.outlook.2013.03.008].
34. Macedo EC, da Silva LR, Paiva MS, et al. Burden and quality of life of mothers of children and adolescents with chronic illnesses: an integrative review. Rev Lat Am Enfermagem. 2015;23(4):769-777. [Doi: 10.1590/0104-1169.0196.2613].
35. Onofri A, Pavone M, De Santis S, et al. Telemedicine in children with medical complexity on home ventilation during the COVID-19 pandemic. Pediatr Pulmonol. 2021;56(6):1395-1400. [Doi: 10.1002/ppul.25289].
36. Baumbusch J, Lloyd JEV, Lamden-Bennett SR, et al. The unintended consequences of COVID-19 public health measures on health care for children with medical complexity. Child Care Health. 2022;48(6):970-978. [Doi: 10.1111/cch.12968].
37. Saqib MAN, Siddiqui S, Qasim M, et al. Effect of COVID-19 lockdown on patients with chronic diseases. Diabetes Metab Syndr. 2020;14(6):1621-1623. [Doi: 10.1016/j.dsx.2020.08.028].
38. Kadambari S, Abo YN, Phuong LK, et al. Decrease in Infection-related Hospital Admissions During COVID-19: Why Are Parents Avoiding the Doctor? Pediatr Infect Dis J. 2020;39(11):e385-e386. [Doi: 10.1097/INF.0000000000002870].
39. Wilder JL, Parsons CR, Growdon AS, et al. Pediatric Hospitalizations During the COVID-19 Pandemic. Pediatrics. 2020;146(6):e2020005983. [Doi: 10.1542/peds.2020-005983].
40. Markham JL, Richardson T, DePorre A, et al. Inpatient Use and Outcomes at Children's Hospitals During the Early COVID-19 Pandemic. Pediatrics. 2021;147(6):e2020044735. [Doi: 10.1542/peds.2020-044735].
41. Gavish R, Levinsky Y, Dizitzer Y, et al. The COVID-19 pandemic dramatically reduced admissions of children with and without chronic conditions to general paediatric wards. Acta Paediatr. 2021;110(7):2212-2217. [Doi: 10.1111/apa.15792].
42. Markham JL, Richardson T, Teufel RJ, et al. Impact of COVID-19 on Admissions and Outcomes for Children With Complex Chronic Conditions. Hosp Pediatr. 2022;8:e2021006334. [Doi: 10.1542/hpeds.2021-006334].
43. Wong CA, Ming D, Maslow G, et al. Mitigating the Impacts of the COVID-19 Pandemic Response on At-Risk Children. Pediatrics. 2020;146(1): e20200973. [Doi: 10.1542/peds.2020-0973].

44. Geweniger A, Barth M, Haddad AD, et al. Impact of the COVID-19 Pandemic on Mental Health Outcomes of Healthy Children, Children With Special Health Care Needs and Their Caregivers-Results of a Cross-Sectional Study. Front Pediatr. 2022;10:759066. [Doi: 10.3389/fped.2022.759066].
45. von Schulz J, Serrano V, Buchholz M, et al. Increased behavioral health needs and continued psychosocial stress among children with medical complexity and their families during the COVID-19 pandemic. Infant Ment Health J. 2022;43(1):111-126. [Doi: 10.1002/imhj.21962].
46. Looman WS, Hullsiek RL, Pryor L, et al. Health-Related Quality of Life Outcomes of a Telehealth Care Coordination Intervention for Children With Medical Complexity: A Randomized Controlled Trial. J Pediatr Health Care. 2018;32(1):63-75. [Doi: 10.1016/j.pedhc.2017.07.007].
47. Perpiñá-Galvañ J, Orts-Beneito N, Fernández-Alcántara M, et al. Level of Burden and Health-Related Quality of Life in Caregivers of Palliative Care Patients. Int J Environ Res Public Health. 2019;16(23):4806. [Doi: 10.3390/ijerph16234806].
48. Ferro F, Tozzi AE, Erba I, Dall'Oglio I, et al. Impact of telemedicine on health outcomes in children with medical complexity: an integrative review. Eur J Pediatr. 2021;180(8):2389-2400. [Doi: 10.1007/s00431-021-04164-2].
49. Brehaut JC, Kohen DE, Raina P, et al. The health of primary caregivers of children with cerebral palsy: how does it compare with that of other Canadian caregivers? Pediatrics. 2004;114(2):e182-91. [Doi: 10.1542/peds.114.2.e182].
50. Mosquera RA, Avritscher EBC, Pedroza C, et al. Telemedicine for Children With Medical Complexity: A Randomized Clinical Trial. Pediatrics. 2021;148(3):e2021050400. [Doi: 10.1542/peds.2021-050400].
51. McConnochie KM, Ronis SD, Wood NE, et al. Effectiveness and safety of acute care telemedicine for children with regular and special healthcare needs. Telemed e-Health. 2015;21(8):611–621. [Doi: 10.1089/tmj.2014.0175].
52. McKissick HD, Cady RG, Looman WS, et al. The Impact of Telehealth and Care Coordination on the Number and Type of Clinical Visits for Children With Medical Complexity. J Pediatr Health Care. 2017;31(4):452-458. [Doi: 10.1016/j.pedhc.2016.11.006].
53. Notario PM, Gentile E, Amidon M, et al. Home-Based Telemedicine for Children with Medical Complexity. Telemed J E Health. 2019;25(11):1123-1132. [Doi:

10.1089/tmj.2018.0186].

54. Wenderlich AM, Herendeen N. Telehealth in pediatric primary care. Curr Probl Pediatr Adolesc Health Care. 2021;51(1):100951. [Doi: 10.1016/j.cppeds.2021.100951].

ABREVIATIONS

CCCs: complex chronic conditions

ICT: Information and Communication Technologies