

Evaluating the Cost-Effectiveness of Home-Based Palliative Care for Children with Special Health Care Needs: A Review Study

Azam Eshaghian-dorcheh¹, *Mitra Zandi², Maryam Rassouli³, Mamak Tahmasebi⁴, Fatemeh Esmaeilzadeh⁵

¹Ph.D Candidate in Nursing, School of Nursing and midwifery, Shahid Beheshti University of Medical Sciences, Tehran, Iran. ²Post Doc in Nursing, Assistant Professor, School of Nursing and Midwifery, Shahid Beheshti University of Medical Sciences, Tehran, Iran. ³PhD in Nursing, Associate Professor, Cancer Research Center, Shahid Beheshti University of Medical Sciences, Tehran, Iran. ⁴MD, Associate Professor, Cancer Institute Palliative Medicine Fellowship, Tehran University of Medical Sciences, Tehran, Iran. ⁵Ph.D. Candidate in Nursing, School of Nursing and Midwifery, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

Abstract

Background

Due to the increased survival of children with chronic and complex diseases, transferring a part of care provision to home and harnessing the family's potential with the aim of shortening the length of stay and returning the child to the safe and familiar atmosphere of the family can affect the health system and the family. This study aimed to evaluate the cost-effectiveness of home-based palliative care for children with complex health care needs based on the literature review.

Materials and Methods: After selecting the keywords related to the research question, by examining MeSH, Emtree and the experts' opinions mentioned in articles, searching the databases PubMed/Medline, EMBASE, Scopus and Web of Science was done through a combination of them.

Results: Finally, 11 studies met the inclusion criteria, which were investigated in three groups of methodological features, economic outcomes and indirect outcomes. The reviewed studies were of medium to high quality. Eight were cohort studies, one pre-post study, and 2 economic evaluation studies. Studies showed that the decrease in the costs of home-based palliative care for the children in need of special health care, although little, was the result of either reduced hospital stay or the decreased number of hospitalizations.

Conclusion

In order to set up pediatric palliative care services in Iran, considering the cultural atmosphere and the limited resources of the health system, attention should be paid to the cost effectiveness of care settings and the provided services, and measures should be taken in order to improve the situation of the children with special care needs, based on social conditions.

Key Words: Children, Chronic Disease, Cost Effectiveness, Home Care Services.

*Please cite this article as: Eshaghian Dorcheh A, Zandi M, Rassouli M, Tahmasebi M, Esmaeilzadeh F. Evaluating the Cost-Effectiveness of Home-Based Palliative Care for Children with Special Health Care Needs: A Review Study. Int J Pediatr 2020; 8(11): 12381-395. DOI: **10.22038/ijp.2020.45949.3745**

*Corresponding Author:

Mitra Zandi, Nursing & Midwifery School, Shahid Beheshti University of Medical Sciences, Vali-e Asr Street, Niyayesh cross, Tehran, Iran. Postal code: 1985717443

Email: mitra.zandi@yahoo.com

Received date: Mar.23, 2020; Accepted date: Jul. 22, 2020

1- INTRODUCTION

Chronic disease management is one of the most important health issues all around the world (1). Childhood chronic and life-threatening diseases, with the need for palliative care, include a broad range of curable and recurrent diseases from cancer and organ failure to diseases such as cerebral palsy which are unpredictable and non-progressive (2). In addition to the primary disease, ill children are faced with undesirable levels of care due to severe disabilities, comorbid conditions, fragmented cares, receiving inadequate information, different care plans, over medicalization, high treatment costs, and the inadequate participation of family caregivers (3). The increased survival rate in children with chronic diseases who need palliative care through the use of high technology interventions and equipment designed to be feasible outside the hospital has drawn the policymakers' attention towards home-care. Home-based care, developed under the title "care closer to home", aims to provide proper care for children and adolescents with chronic diseases and reduce their length of stay (4, 5), and guarantees the child's return to the safe and familiar atmosphere of the family (6). Children with special care needs are technology-dependent and require frequent and prolonged hospitalizations and ongoing care (7). Since families have to go to hospitals for receiving services, the transportation and commuting costs increase. Given the families' desire to provide care for children at home (8), this type of care, if cost-effective, is considered as an alternative approach in long-term care (9). According to an American study, palliative care provision for the hospitalized children was less costly by performing fewer radiologic procedures and effective pharmacological services for symptoms (10). Another study estimated the end-of-life palliative care costs to be annually \$110,000 for infants and \$62,000

for children, which indicates the need of special financial resources in children and infants' care, in comparison with the \$28,000 cost for adults (11). In adults, home-based palliative care has a positive effect on the quality of life in patients and reduces end-of-life care costs (12) as well as resulting in the families' satisfaction (12). Therefore, investigating the costs and the outcomes of palliative care plays an important role in establishing these services for children with special care needs, in different settings of service provision. Shirinabadi-Farahani et al. (2018) have regarded financial problems as one of the barriers to establishing home-based palliative care for cancer patients in Iran (13). On the other hand, the unfair distribution of pediatric palliative care services has made families unable to provide these services for their children (8). The lack of proper insurance coverage for these services is also one of the major challenges in using them (14).

However, families and patients experience more convenience and fewer burdens when they receive services at home (15). Facilitating the access to services, from primary to advanced level, has been a challenge for health policymakers in regard to these children (16). It is of great importance to explore strategies to provide and maintain sustainable services with the lowest amount of resource usage (17). Economic policies seek to improve the quality of health services, to reduce healthcare costs, and to avoid unnecessary hospitalization and imposed costs (18). Therefore, due to limited financial resources, it is necessary to conduct economic evaluation studies, with the aim of providing the best evidence to make decisions on choosing proper services for the allocation of limited funds (17). By utilizing more desirable care approaches, the allocation of limited funds to ineffective interventions is reduced, and while providing appropriate services to

service-receiver, their satisfaction and service quality is met. Because the pediatric palliative care.../As the pediatric palliative care in Iran is just emerging and these studies can be helpful in setting up services in different areas of healthcare service delivery, this review study was carried out based on literature review, with the aim of evaluating the cost effectiveness of home-based palliative care for children with special care needs.

2- MATERIALS AND METHODS

2-1. Method

This study aimed to review cost-effectiveness studies on home-based palliative care in children with chronic and life-threatening diseases. The search was done based on the keywords pediatric palliative care, home care services, chronic

and threatening illness, and cost effectiveness. Similar keywords were extracted from MeSH and Emtree and also through manual search by reviewing the titles and the abstracts of the articles. Keyword combinations in Medline are shown in **Table.1**. After keywords were determined, two separate researchers searched the electronic databases Scopus, Web of Science, EMBASE and Medline via PubMed, in the period of time from January 2000 to May 2019. In order to access the rest of the papers, the articles references were reviewed by hand searching. Moreover, the Google and Google Scholar search engines were also checked for more assurance. After the searches were done by two researchers, the articles obtained from each database were imported into EndNote X8.

Table 1: Combined words with synonyms in PubMed.

Database	Key words
PubMed/ Medline	Search (((((((domestic health care[tiab] OR home care agencies[tiab] OR home care program[tiab] OR home care programme[tiab] OR home care services, hospital-based[tiab] OR home health care[tiab] OR home health nursing[tiab] OR home nursing[tiab] OR home service[tiab] OR home treatment[tiab] OR homecare[tiab] OR homemaker services[tiab]))) OR ((Palliative home based care[tiab] OR Pediatric palliative home care[tiab] OR Palliative home care[tiab] OR Children palliative home care[tiab] OR Home health care[tiab] OR pediatric home care[tiab] OR children home care[tiab])) OR (("Home Care Service"[tiab] OR "Service, Home Care"[tiab] OR "Care Services, Home"[tiab] OR "Domiciliary Care"[tiab] OR "Care, Domiciliary"[tiab] OR "Services, Home Care"[tiab] OR "Home Care"[tiab] OR "Care, Home"[tiab] OR "Hospital Home Care Services"[tiab] OR "Hospital-Based Home Care"[tiab] OR "Hospital Based Home Care"[tiab] OR "Home Care Services, Hospital Based"[tiab] OR "Hospital-Based Home Care Services"[tiab] OR "Hospital Based Home Care Services"[tiab] OR "Hospital Based Home Cares"[tiab] OR "Hospital Based Home Cares"[tiab])) OR ((("Home Care Service"[mh] OR "Service, Home Care"[mh] OR "Care Services, Home"[mh] OR "Domiciliary Care"[mh] OR "Care, Domiciliary"[mh] OR "Services, Home Care"[mh] OR "Home Care"[mh] OR "Care, Home"[mh] OR "Hospital Home Care Services"[mh] OR "Hospital-Based Home Care"[mh] OR "Hospital Based Home Care"[mh] OR "Home Care Services, Hospital Based"[mh] OR "Hospital-Based Home Care Services"[mh] OR "Hospital Based Home Care Services"[mh] OR "Hospital-Based Home Cares"[mh] OR "Hospital Based Home Cares"[mh])) OR (((("Palliative care"[tiab] OR "Care, Palliative"[tiab] OR "Palliative Treatment"[tiab] OR "Palliative Treatments"[tiab] OR "Treatment, Palliative"[tiab] OR "Treatments, Palliative"[tiab] OR "Therapy, Palliative"[tiab] OR "Palliative Therapy"[tiab] OR "Palliative Supportive Care"[tiab] OR "Supportive Care, Palliative"[tiab])) OR ((("Palliative care"[mh] OR "Care, Palliative"[mh] OR "Palliative Treatment"[mh] OR "Palliative Treatments"[mh] OR "Treatment, Palliative"[mh] OR "Treatments, Palliative"[mh] OR "Therapy, Palliative"[mh] OR "Palliative Therapy"[mh] OR "Palliative Supportive Care"[mh] OR "Supportive Care, Palliative"[mh])) OR ((End of life care[tiab] OR Terminal care[tiab] OR Pediatric Palliative care[tiab] OR Palliative care nursing[tiab] OR Children palliative nursing care[tiab])) OR ((palliation[tiab] OR palliative care[tiab] OR palliative consultation[tiab] OR palliative medicine[tiab] OR symptomatic treatment[tiab])) AND (((((chronic illness[tiab] OR (End of life disease[tiab] OR Life Threatening condition[tiab] OR Life Limiting condition[tiab] OR Terminal illness[tiab])) OR ((("Chronic Disease"[tiab] OR "Disease, Chronic"[tiab] OR "Diseases, Chronic"[tiab] OR "Chronic Illness"[tiab] OR "Chronic Illnesses"[tiab] OR "Illness, Chronic"[tiab] OR "Illnesses, Chronic"[tiab] OR "Chronically Ill"[tiab])) OR ((("Chronic Disease"[mh] OR "Disease, Chronic"[mh] OR "Diseases, Chronic"[mh] OR "Chronic Illness"[mh] OR "Chronic Illnesses"[mh] OR "Illness, Chronic"[mh] OR "Illnesses, Chronic"[mh] OR "Chronically Ill"[mh])) OR (((("pediatrics"[tiab] OR "pediatrics"[mh] OR (Pediatric[tiab] OR Children[tiab] OR Infants[tiab] OR Adolescence[tiab] OR Young children[tiab] OR Neonates[tiab])) OR ((Community paediatrics[tiab] OR community pediatrics[tiab] OR paediatric aspect[tiab] OR paediatric care[tiab] OR paediatric practice[tiab] OR paediatric service[tiab] OR paediatrics[tiab] OR paediatrics[tiab] OR pediatric aspect[tiab] OR pediatric care[tiab] OR pediatric practice[tiab] OR pediatric service[tiab])) AND (((("Analyses, Cost-Benefit"[mh] OR "Analysis, Cost-Benefit"[mh] OR "Cost-Benefit Analyses"[mh] OR "Cost Benefit Analysis"[mh] OR "Analyses, Cost Benefit"[mh] OR "Analysis, Cost Benefit"[mh] OR "Cost Benefit Analyses"[mh] OR "Cost Effectiveness"[mh] OR "Effectiveness, Cost"[mh] OR "Cost-Benefit Data"[mh] OR "Cost Benefit Data"[mh] OR "Data, Cost-Benefit"[mh] OR "Cost-Utility Analysis"[mh] OR "Analyses, Cost-Utility"[mh] OR "Analysis, Cost-Utility"[mh] OR "Cost Utility Analysis"[mh] OR "Cost-Utility Analyses"[mh] OR "Economic Evaluation"[mh] OR "Economic Evaluations"[mh] OR "Evaluation, Economic"[mh] OR "Evaluations, Economic"[mh] OR "Marginal Analysis"[mh] OR "Analyses, Marginal"[mh] OR "Analysis, Marginal"[mh] OR "Marginal Analyses"[mh] OR "Cost Benefit"[mh] OR "Costs and Benefits"[mh] OR "Benefits and Costs"[mh] OR "Cost-Effectiveness Analysis"[mh] OR "Analysis, Cost-Effectiveness"[mh] OR "Cost Effectiveness Analysis"[mh])) OR ((("Analyses, Cost-Benefit"[tiab] OR "Analysis, Cost-Benefit"[tiab] OR "Cost-Benefit Analyses"[tiab] OR "Cost Benefit Analysis"[tiab] OR "Analyses, Cost Benefit"[tiab] OR "Analysis, Cost Benefit"[tiab] OR "Cost Benefit Analyses"[tiab] OR "Cost Effectiveness"[tiab] OR "Effectiveness, Cost"[tiab] OR "Cost-Benefit Data"[tiab] OR "Cost Benefit Data"[tiab] OR "Data, Cost-Benefit"[tiab] OR "Cost-Utility Analysis"[tiab] OR "Analyses, Cost-Utility"[tiab] OR "Analysis, Cost-Utility"[tiab] OR "Cost Utility Analysis"[tiab] OR "Cost-Utility Analyses"[tiab] OR "Economic Evaluation"[tiab] OR "Economic Evaluations"[tiab] OR "Evaluation, Economic"[tiab] OR "Evaluations, Economic"[tiab] OR "Marginal Analysis"[tiab] OR "Analyses, Marginal"[tiab] OR "Analysis, Marginal"[tiab] OR "Marginal Analyses"[tiab] OR "Cost Benefit"[tiab] OR "Costs and Benefits"[tiab] OR "Benefits and Costs"[tiab] OR "Cost-Effectiveness Analysis"[tiab] OR "Analysis, Cost-Effectiveness"[tiab] OR "Cost Effectiveness Analysis"[tiab])) OR ((Cost effectiveness[tiab] OR cost effectiveness ratio[tiab] OR cost efficiency analysis[tiab])) OR Healthcare cost[tiab]))

2-2. Data Extraction

Quantitative studies published from the from the end of May 2019 to the beginning of 2000, focus on the concept of palliative care for the children from one month to 21 years of age, clinical trial studies, cohort and economic evaluation, feasibility of home-based child care and access to full text articles constituted the inclusion criteria. Neonatal and prenatal home-care, care plans with no quantitative data, chronic diseases such as type I diabetes and childhood asthma did not meet the criteria for life-threatening or limiting illnesses. Evaluating the effectiveness of home care in adults and children altogether, acute care at home (acute respiratory problems, diarrhea and nausea) and gray literature (thesis, editorial) were excluded, too. There were no language restrictions for articles. After determining the inclusion and exclusion criteria, two researchers reviewed the papers. The cases of disagreement were discussed. In the first stage of study screening, duplicate items were removed using EndNote X8. Then the list of articles and their abstracts were imported into Word. At this stage, the titles and abstracts of the articles were reviewed by two independent researchers. In the next step, the full texts of articles were reviewed and then, based on the inclusion criteria, the articles were selected. The selected articles were reported using the PRISMA guideline for

systematic review studies (19). The baseline characteristics of the articles (first author's name, the year of publication, and country), purpose, setting, design, taken measures, and data extraction time were collected and presented as a table by two research partners after the complete review of the studies. A summary of the characteristics of the retrieved studies is displayed in **Table.2**.

2-3. Appraising the Articles' Quality

The retrieved studies were evaluated based on the criterion developed by Hawker et al. (2002) for appraising the studies with different designs. This criterion consisted of the following nine parts: Abstract and title, introduction and aims, method and data, sampling, data analysis, ethics and bias, results, transferability or generalizability, and implications and usefulness. Each item was assigned a score between 1 (very poor, minimum score), and 4 (good, maximum score for each item). In total, a score between 9 and 36 will be obtained from all items (20). By reviewing each item, researchers aimed to examine the articles' conformity with the highest quality. The studies were separately appraised by two researchers and disagreements were discussed and a consensus was reached. The examined studies had a score of 17 to 35 based on the criterion. In general, the reviewed articles were of average quality.

Table-2: General characteristics of included studies.

First Author, Year, (Reference)	Country	Study design	Study Population (n)	Sample characteristic	Quality assessment score (20)/ total score= 36
Baker, 2015, (21)	USA	Pre-post	Total= 48 pre- intervention= 18 post-intervention =30	Pre- intervention age 0.3 (0–10.8) year, Post- intervention age 0.5 (0–25.2) year, Age at discharge, median (range), year: pre 1.1 (0.6–11.2); post 1.0 (0.4–25.3), Children with: BPD, Congenital diaphragmatic hernia, Neuromuscular disease, Spinal cord injury/trauma, Congenital heart disease, Genetic/chromosomal anomaly, Oncology/bone marrow transplant,	30

				Airway malacia, Pulmonary hypertension, Upper airway obstruction, Pulmonary interstitial glycogenesis, Seizure disorder/infantile spasms, Postoperative extubation failure.	
Miano, 2002, (22)	Italy	Prospective study	n= 45	Children with cancer 29 males, 16 females 1 month–19 years.	30
Gans, 2016, (23)	USA	Retrospective cohort (preservice/post-service enrollment)	n= 132	Children under 20, with life-threatening diseases, at least 60 days of presence in the program.	33
Gans, 2012, (24)	USA	Retrospective cohort before–after study with no control	n= 123	Children from 1 to 20 years of age with life-threatening conditions, Neurologic, cancer, pulmonary, neuromuscular, gastrointestinal, cardiac.	17
Sharif Azar, 2015, (25)	Iran	Cost effectiveness research; The random assignment of children to two groups	n=56	Children with CP under 12 years.	34
Bradford, 2014, (17)	Australia	Cost Minimization Analysis	-	-	28
Chong, 2018, (26)	Singapore	Cohort, retrospective and prospective	n= 71, Palliative care group, n= 67 Control group	Children with life threatening condition.	35
Postier, 2014, (27)	USA	Retrospective cohort before–after study	Total=425 Patients with cancer= 225, Patients without cancer= 200	200 children with cancer and 225 children with non-malignant diseases.	33
Berg, 2007, (28)	USA	Prospective	n= 14	Children 6 to 18-years-old with a mean age of 14.5, with CHF and a need for continuous inotropic support.	24
Madden, 2015, (29)	USA	Retrospective chart review	n= 45	(3 to 27-year-old patients with a mean age of 8), with medulloblastoma under treatment, taking Cisplatin as chemotherapy agent.	29
Hassani, 2019, (30)	Iran	Retrospective cohort study without control group	n= 67	67 children (32 girls, 47.7%). The average: 5.2 ± 4.9 (2-15) years congenital airway malformations; cystic fibrosis; Severe laryngomalacia, polyneuropathy syndrome in mitochondrial myopathy; hypoxic ischemic, encephalopathy, and CP.	32

3- RESULTS

Of the 1,739 studies retrieved from databases and the article references, 377 duplicate studies were excluded in the first step. The titles and abstracts of the remaining 1,359 papers were reviewed, and 1,212 articles were excluded because of their irrelevance to the purpose of the research. Then the full texts of 147 articles were reviewed. At this stage, 136 studies

were excluded as a result of not meeting the inclusion criteria. Finally, 11 articles were included in the study. This process is displayed in **Figure.1**. The characteristics of the articles are presented in **Table.2** (17, 21-30). The results of the studies are divided into the following three classes: methodological features of the articles, economic outcomes and indirect outcomes. The summary of the results is presented in **Table.3**.

Table-3: Summary of key findings.

First Author, Year	Summary of Key Findings
Baker, 2015 (21)	Reduced length of stay (LOS) in hospital. Reduced LOS in the respiratory care unit. Reduced mean of direct costs per hospitalization. Reduced readmission rate during the 12-month period with no statistical difference. No significant differences in ER visits during the 6-month period, and readmission rate within the 30-day period.
Miano, 2002 (22)	A significant decrease in the home care costs per patient in comparison with hospital costs.
Gans, 2016 (23)	A decrease in the length of hospitalization from 4.2 to 2.3 days per month. Decrease in the length of stay in hospital from 16.7 to 6.5 days after entering the care plan. Reduced number of hospitalizations and readmission during the 30-day period with no statistically significant difference. Reduced care costs per patient, reduced hospitalization costs per child.
Gans, 2012 (24)	Reduced costs. A 1/3 decrease in the length of hospitalization (from 4 to 2.8 days per month for each child). Improved quality of life in the child, as well as higher satisfaction of the family and the staff.
Sharif Azar, 2015 (25)	Significant decreased was observed between home and clinic rehabilitation.
Bradford, 2014 (17)	The reduced cost of video consultation in palliative home care compared with home and outpatient visits
Chong, 2018 (26)	Reduced hospital admission rate in children compared with the control group (2.46 times more in the control group). Reduced length of stay in hospital. No difference in ER visits in comparison with the control group. Significant difference in care costs compared with the control group, one year prior to death. Improved quality of life in children and decreased parental care burden.
Postier, 2014 (27)	No difference in the number of hospitalizations in cancer children before and after entering the care plan. Reduced LOS in cancer and non-cancer patients with at least 6 months of palliative care experience. Changes in length in the group of cancer children by a mean of 2 weeks and in the group of children with non-cancer conditions, 38 days. Reduced care costs in both groups of cancer and non-cancer samples. Increased ER visits in cancer children.
Berg, 2007 (28)	A significant difference between home care and hospital care costs.
Madden, 2015 (29)	A significant decrease in treatment costs at home (chemotherapy with cisplatin, a 2-hour hydration and a 5-hour drug monitoring) compared with the costs of similar procedures in the hospital.
Hassani, 2019 (30)	The cost-effectiveness of home mechanical ventilation compared with hospital care.

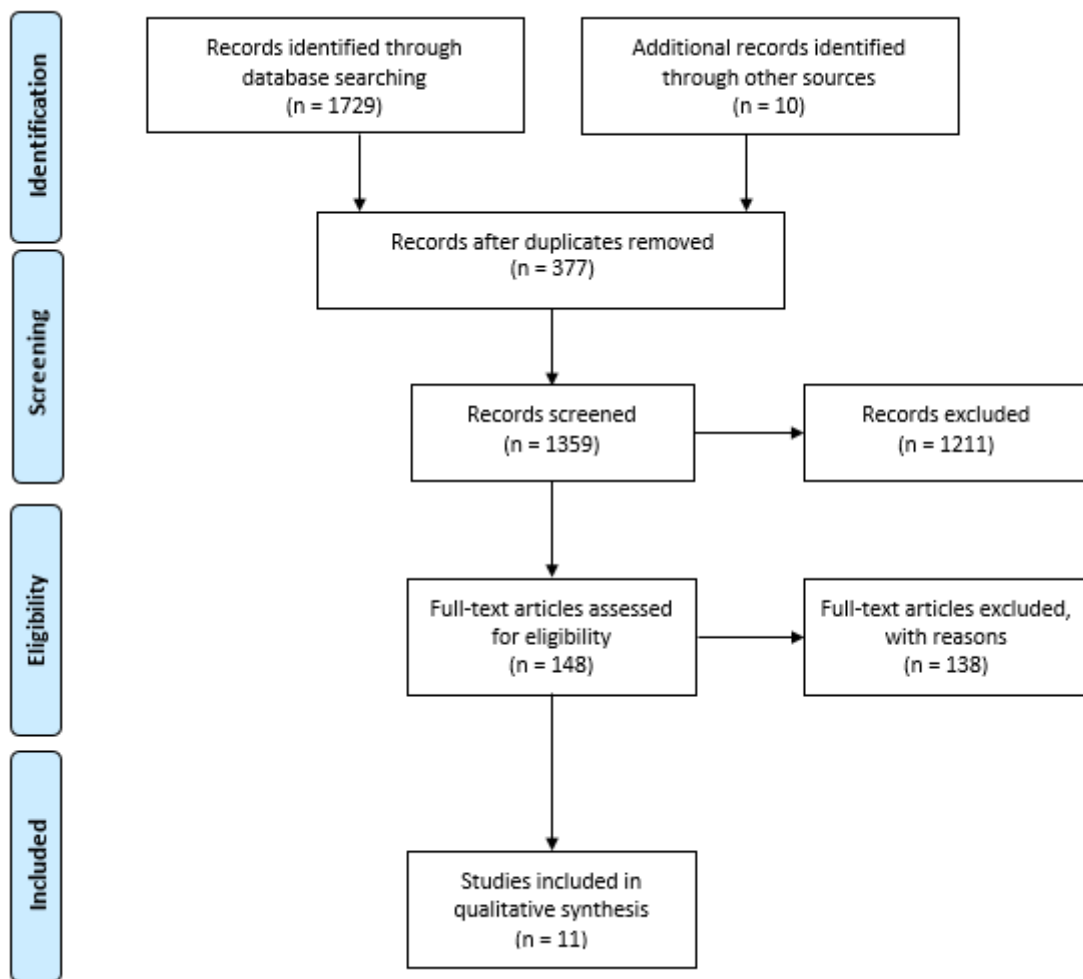


Fig.1: Combined words with synonyms in PubMed.

3-1. Methodological Features of the Articles

Of the 11 reviewed studies (total population=1,739), 6 were conducted in the USA (21, 23, 24, 27-29), 2 in Iran (25, 30), and one in Italy (22), Singapore (26), and Australia (17) each. Eight were cohort studies (retrospective cohort and prospective cohort), one pre-post study (21), and 2 economic evaluation studies (17, 25). No clinical trials have been conducted in this area. Just one of the economic evaluation studies randomly assigned the samples into two groups (25). There were two prospective studies (17, 22-28), four retrospective studies (23, 24,

27, 30), a retrospective chart review (29) and one partly retrospective, partly prospective study (26). Two of the studies investigated children under mechanical ventilation (21, 30). A home rehabilitation study examined children with cerebral palsy (25), and another one, children with Congestive Heart Failure (CHF) (28). One study specifically investigated children with medulloblastoma who received chemotherapy at home (29). A research studied cancer children (22), while in another one, a comparison was made between cancer and non-cancer children (27). In four studies, children with life-threatening illnesses were investigated (17, 23, 24, 26).

The sample size of the studies varied from 14 to 425 (two groups with 200 and 225 samples) and the total number of children in all studies was 981, the majority of which were male (approximately 65%). In the Cost Minimization Analysis (CMA) study, only the total number of video consultations ($n = 95$), and the number of trips ($n = 24$) were reported, and the children's demographic information was not explained in detail (17). The age of the patients ranged from 1 month to 21 years. In a retrospective chart review, over a 17-year period (1995-2013), the maximum age was 27 years, with a mean of 8 years, and no information was available on the age of children at the time of inclusion in the program, or at the end of it (29). Several pediatric palliative care programs were referred to in the studies, such as Partners for Children (PFC) (23, 24), Ventilator Care Program (VCP) (21), the pediatric neuro-oncology program in the US (29), the star pals in Singapore (26), and a care plan for cancer children in a children's hospital in Italy (22).

3-2. Economic Outcomes

Changes in costs were one of the economic outcomes of home care for children with chronic diseases. Eleven studies provided evidence of changes in costs (17, 21-29). This change mainly includes decreases (21-29), and in the Cost Minimization Analysis (CMA) the costs of home-based palliative care provided in 3 forms of home visit, outpatient visit and video consultation were compared. This study, with focus on cost saving in specialized home-based palliative care, reported increases in the costs of home visits and outpatient visits, with additional costs for staff transportation in home visits as well as the child and family visits to health care centers (17). In other studies, that have reported decreases in costs, the main reasons of decreases were reduced number of hospitalizations (23, 26), and length of stay in hospital (21, 23, 24, 26, 27).

Decreases in the number of children's hospitalizations have been reported by Chong et al. (26). Gans et al. (2016) considered this change as statistically insignificant (23). Postier et al. reported no significant differences in the number of hospitalizations of children with cancer, before and after attending palliative care programs (27). A change was reported by Baker et al. (2016), and Gans et al. (2016), in unplanned readmissions during the 30-day period following discharge that, despite the decrease, was not statistically significant (21, 23). Berg et al. (2007) reported only the total number of readmissions of children with heart failure due to the absence of a comparison group before and after, according to which, 57% of children had experienced readmission at least one time (28). In Baker et al.'s study on ED visits, no significant differences were observed after 30 days and after 6 months, either before or after the implementation of discharge and home care program (21). In addition, Chong et al. reported no significant changes in this indicator either before or after the program (26). According to Postier et al., there would be an increase in hospital/ER admissions with the prolonged presence of cancer children in the palliative care program (27). Reduced length of stay in hospital (21, 23, 24, 26, 27) was another indicator. Baker et al. reported a decrease of almost 60 days in the length of stay in the ICU (21). The decrease in length of stay from at least 10 (23) to a maximum 150 days (21, 27) has been reported in these studies.

3-3. Indirect Outcomes

The quality of life and satisfaction of family and healthcare staff have also been investigated in several studies as outcomes of home-based palliative care. Three studies focused on the quality of life (24-26). One study investigated the children's quality of life (25, 26). Two other studies studied the improvement of care ability

and better quality of life in families and caregivers (24, 26). In two studies, the satisfaction of family and hospital staff was studied (21, 24); no significant difference was reported by Baker et al in this regard (21). Chong et al. examined the caregivers' care burden, which was significantly different compared with the baseline measurement in 3-month and 6-month follow-up. In addition, this study also referred to a better control of children's pain at home with a higher odds ratio for pain free within 3 months compared with that of the baseline, indicating a better pain control and an improved quality of life in children (26). Studies by Madden et al. and Gans et al. showed a better symptom management at home (23, 29). In-home chemotherapy injection was considered and recommended as the early stages of palliative care (29). Providing nursing services at home was of great importance. The members of home-based pediatric palliative home care team were nurses. Sufficient home care nursing services and a 24/7 access to these services were correlated with better coordination and lower use of inpatient services (21-23, 29).

4- DISCUSSION

This study was conducted to review the cost-effectiveness of providing palliative home care for children with chronic and life-threatening illnesses. The financial challenges and costs of pediatric palliative care have been considered as the third priority among the 10 global research priorities, which have a major role in determining care policies and procedures (31). Despite the importance of providing palliative care for children with chronic and complex diseases, one of the barriers to accessing integrated palliative care in many countries is the lack of transparency of the effectiveness and economic aspects of this type of care for health policymakers (11). In setting up palliative care services for children, attention should be paid to the

different settings of service, and the benefits and effectiveness of each setting, a care model appropriate for the situation of the child, the family and the culture of society should be considered. As a general result, pediatric palliative home care helps reduce the costs and the use of inpatient services by decreasing the length of hospital stay (21, 23, 24, 26, 27), and the number of hospitalizations (23, 26). Developed countries with advanced pediatric palliative care services, such as the United States (21, 23, 24, 27-29), Australia (17), Singapore (26), and Italy (22) have considered home care in long-term care (17). Global evidence puts emphasis on home care provision for children with life-threatening illnesses, especially at the end of life, and recommends family-based care (32-35).

Two Iranian studies examined the cost-effectiveness of home-based rehabilitation in children with CP (25), and home care in children under mechanical ventilation (30), according to which, providing rehabilitation services for children is more effective at home than in rehabilitation clinics, ICUs and hospitals. However, this study has only focused on the physical conditions of the child and the psychological aspects and burden are ignored. Since rehabilitation services, especially occupational therapy, are costly and they are only partially covered by insurance services, family's out-of-pocket costs will increase and transportation costs will be imposed on the family, too (25). The costs of care and long-term treatment have been a barrier to receiving services that leads to the family giving up the treatment, the increased severity of the disease and the incidence of complications (36), and imposes additional costs on the family and the health system. Therefore, providing insurance coverage for rehabilitation and home care services will help decrease family's financial burden and out-of-pocket costs to some extent.

According to 10 studies, the cost of home-based palliative care for children with life-threatening diseases was reduced (21-27). This is partly the result of reducing the need for inpatient services (23, 24); other influential factors were better care coordination and resource management (28), caregiver training, discharge readiness (21), and reduced invasive and high-tech interventions for children, especially in end-of-life care (26). The need for medicines and consumable equipment in these children remains constant at home and in the hospital. The difference in costs is a result of hoteling, and the costs of overnight beds and hospital admission services, which would partly be eliminated at home. It is therefore logical that cost reduction is small given the complexity of the child's condition.

As an example, Gans et al. (2012) reported an 11 percent reduction in costs (24). The study by Bradford et al. compared the costs of palliative care services in home visits, outpatient visits, and video consultation. In home visits, more money was spent due to the additional costs of trips and the increased spent time, and the video counseling saved more money in care provision per child. In this study, the child and the family are tele-handled at home by the treatment team via communication technology, which reduces the extra costs of care (17). The use of tele-health systems in chronic patients has also had a positive impact on disease management and outcomes (37). In setting up pediatric palliative care, it is necessary to consider the need to use a technology tailored to fit the culture and the condition of the society and to provide part of the services through modern approaches, despite the fact that visiting the child and face-to-face communication is the gold standard of care (38). With regard to the internet coverage and available equipment, using these systems in Iran will save the time and resources of family and health

system. Reduced length of stay (LOS) in children with chronic diseases is another outcome of pediatric palliative home care. This decrease can be due to the children's enrollment in a comprehensive care plan, in which the timely discharge of children from hospitals is done by ensuring follow-up, and families avoid the confusion and anxiety. Family's readiness for child's discharge reduced length of stay in the hospital from 10 to 150 days. In the ICU, the reduction was even higher, more than 2 months. Therefore, a reduced LOS can contribute to lower costs (21, 23, 24, 26, 27). In this regard, the study conducted by Gold et al. (2016) on children with medical complexity (CMC) showed that long hospitalization increases costs, and that in these children, reduced LOS can prevent prolonged bed occupancy, especially in the ICU, which consequently decreases the costs of care and treatment (39), and helps increase the admission of children with higher chances of survival which will contribute to justice in the provision of health care services. In Iran, due to the unavailability of pediatric palliative care services (6), families have to visit the ER and hospitalize the child in the ICU (40).

Therefore, due to the high costs of ICU stays (\$700 in public hospital, based on the tariffs issued by government in 2019), the timely discharge of the child results in saving huge amounts of money on hospital service provision. The main challenges of establishing home care in Iran are the lack of proper insurance coverage and the limited number of home nursing services approved by the High Council of Insurance (14). Therefore, choosing home care is associated with increased out-of-pocket medical expenses for addressing care needs (41), and dealing with the stress caused by unmet health care needs (42). By implementing the health transformation plan in Iran and reducing out-of-pocket payments for inpatient services, using available insurance funds (43), compared

with home care which is considered a luxury and an expensive service, people are inclined to use hospital services. However, as long as these services are not properly covered by insurance, they are not welcome by people, and the burden of chronic diseases would still be on the government and insurers' shoulders. It is therefore logical to consider the experiences of developed countries such as Germany and Japan with regard to the insurance coverage of home care and long-term care that reduces the use of hospital services with the timely discharge of the patient (44). Additionally, in Malaysia these costs are partly funded by the government and insurance companies and partly by charities and NGOs (45).

In Iran, regarding the lack of proper insurance coverage, charities play an important role in providing these services. No statistically significant change was observed in the number of ER visits in this study (21, 26), which could be due to the unpredictability and uncertainty of children's diseases, the child's critical condition, and also the families' sensitivity over children, as they tend to visit health centers even with minor changes in their children's condition. On the other hand, observing the child's condition, especially at the end of life and the severity of symptoms may force families to visit the ER to have the child examined. Inadequate training on child care and the lack of follow-up examinations of the child and the family by the home care team are also of the reasons for ER visits by these children. The quality of life, satisfaction and burden had been examined through studies, as indirect outcomes. The improvement of the quality of life in CP children as a result of home rehabilitation services was significant (25). After setting up specialized palliative home care for German children, an improved quality of life and symptom relief, and reduced psychological distress and parental burden

were reported after 8 weeks (15). This finding is consistent with the results of the reviewed studies (24, 26). Pain control and symptom management have also been investigated in several studies. Better pain control and the comfort of the child and the family, during the 3-month period, explain the enhanced health-related quality of life in these children (26). Home visits and nursing services assures the family of appropriate child care (29), and plays an important role in identifying and coordinating patient and family needs (46). Studies have emphasized the role of general nurses and nursing practitioners in providing services for children and adolescents with life-threatening diseases (47). There is a correlation between the end-of-life home nursing services and the decrease in acute hospitalization (48). It is recommended to implement nurses' knowledge and skills in responding to patients' needs and support the services available in nursing home visits (49).

In Iran, due to the lack of home care centers specialized for children and a shortage of specialist pediatric nurses, services for children are provided by general nurses. Due to the differences in the characteristics of disease and symptom control between children and adults, it is necessary to establish home care centers for children and to train nurses for this purpose. Retraining courses on the child and family relationship, children's procedures and problems in home care can help families to trust in home care nurses and encourage them to use these services. The establishment of pediatric palliative care in Iran, as a need perceived by the health system and families of children with special care needs, requires the planning of health managers and policymakers and the provision of financial resources (8). The limited financial resources for the health system have made the use of family potential and home care in children with chronic diseases inevitable. The shortage

of health care providers to provide home care, the lack of policies and guidelines, insufficient funding, and the lack of end-of-life palliative care services are among the infrastructure barriers to the establishment of palliative care in developing countries. Home care is associated with reduced hospitalizations, comfort for patients, and an increased number of deaths at home as requested by many patients and families, avoiding costly and futile treatments, and using public resources to help reduce health care costs (50). Facilitating the integration of home care into child health services as family-centered care and the implementation of comprehensive care plans would make links between first level services, hospitals, child rehabilitation services, hospice services and respite care. Policy makers' acceptance of pediatric palliative care and appropriate insurance coverage for these services will ensure the implementation of the pediatric home care plan.

5- CONCLUSION

Studies have shown a decrease in the length of stay in children with chronic diseases, a reduced number of hospitalizations and consequently reduced care costs. As a result of differences in study types, the population under study and the duration of care, and in general, due to the lack of homogeneity in the studies, the conditions for meta-analysis were not met. The wide range of pediatric palliative care and subsequently, the various needs of children and families can explain the different research methods and results. Therefore, it is recommended to conduct clinical trial studies in order to assess the cost-effectiveness of home care and compare it with other care settings. According to 2017 statistics in Iran (51), there are a total of 505 PICU beds with a bed occupancy rate of approximately 100 percent; while some patients are waiting to be admitted to PICU. Regarding the high

costs of establishment and maintenance of ICU beds, the management of specialty beds and the adoption of appropriate strategies, as well as the timely discharge of the child and his/her transference to home after being ensured of the family's readiness are necessary due to bed restrictions. Higher access to palliative care services at the first level of service provision system and the use of appropriate communication facilities with regard to the internet coverage can help reduce hospital services in children. Moreover, concerning the important role of NGOs and charitable organizations in providing services for patients, the proper ground for their activities in this field should be provided. According to the results of this review study, the establishment of pediatric home care centers and training the staff in performing pediatric procedures can prevent the abandonment of children with these diseases after discharge, and reduce their frequent hospitalizations by entrusting the children and their families to knowledgeable personnel.

6- ACKNOWLEDGMENTS

Thanks are due to the instructions of the respectful professors in developing this article.

7- CONFLICT OF INTEREST: None.

8- REFERENCES

1. Paré G, Poba-Nzaou P, Sicotte C. Home telemonitoring for chronic disease management: An economic assessment. *International Journal of Technology Assessment in Health Care*.2013;29(2):155-61.
2. Chambers L, Dodd W, McCulloch R, McNamara-Goodger K, Thompson A, Widdas D, et al. A guide to the development of children's palliative care services. Bristol: Association for Children's Palliative Care. 2009.
3. Bogetz J, Ullrich C, Berry J. Pediatric hospital care for children with life-threatening

illness and the role of palliative care. *Pediatric Clinics*. 2014;61(4):719-33.

4. Wager KA, Lee FW, Bradford WD, Jones W, Kilpatrick AO. Qualitative evaluation of South Carolina's Postpartum/Infant Home Visit program. *Public Health Nursing*. 2004;21(6):541-6.

5. Parker G, Spiers G, Gridley K, Atkin K, Birks Y, Lawson K, et al. Systematic review of international evidence on the effectiveness and costs of paediatric home care for children and young people who are ill. *Child: Care, Health and Development*. 2013;39(1):1-19.

6. Khanali-Mojen L, Rassouli M, Eshghi P, Zendedel K, Akbari- Sari A, Heravi-Karimooi M, et al. Pediatric Palliative Care in Iran: Applying Regionalization of Health Care Systems. *Asian Pacific journal of cancer prevention: APJCP*. 2018;19(5):1303.

7. Simon TD, Berry J, Feudtner C, Stone BL, Sheng X, Bratton SL, et al. Children with complex chronic conditions in inpatient hospital settings in the United States. *Pediatrics*. 2010;126(4):647-55.

8. Rassouli M, Salmani N, Mandegari Z, Dadgari A, Fallah Tafti B, Bagheri I. Challenges of palliative care for children with cancer in Iran: a review. *Iranian Journal of Pediatric Hematology and Oncology*. 2019;9(1):48-62.

9. Geyer J, Korfhage T. Long-term Care Insurance and Carers' Labor Supply—A Structural Model. *Health Economics*. 2015;24(9):1178-91.

10. Ward-Smith P, Korphage R, Hutto C. Where health care dollars are spent when pediatric palliative care is provided. *Nursing Economics*. 2008;26(3):175.

11. Knapp C, Thompson L, Vogel W, Madden V, Shenkman E. Developing a pediatric palliative care program: addressing the lack of baseline expenditure information. *American Journal of Hospice and Palliative Medicine®*. 2009;26(1):40-6.

12. Maetens A, Beernaert k, De Schreye R, Faes K, Annemans L, Pardon K, et al. Impact of palliative home care support on the quality and costs of care at the end of life: a

population-level matched cohort study. *BMJ Open*. 2019;9(1):e025180.

13. Shirinabadi-Farahani A, Rassouli M, Mojen L, Ansari M, Ebadinejad Z, Tabatabaee A, et al. The Feasibility of Home Palliative Care for Cancer Patients: The Perspective of Iranian Nurses. *International Journal of Cancer Management*. 2018;11(8): e80114.

14. Rassouli M, ShirinAbadi-Farahani A, Khanali-Mojen L. Palliative Care Perspectives and Practices in the Islamic Republic of Iran, and their Implication on Patients' Quality of Life. In: Silbermann, editor. *Palliative Care: Perspectives, Practices and Impact on Quality of Life A global view*. New York: Nova Scientific Publisher; 2017.

15. Groh G, Borasio G, Nickolay C, Bender H, von Lüttichau I, Führer M. Specialized pediatric palliative home care: a prospective evaluation. *Journal of Palliative Medicine*. 2013;16(12):1588-94.

16. Inkelas M, Ahn P, Larson K. Access to Health Care for California's Children with Special Health Care Needs. 2004.

17. Bradford NK, Armfield NR, Young J, Smith AC. Paediatric palliative care by video consultation at home: a cost minimisation analysis. *BMC Health Serv Res*. 2014 Jul 28;14:328. doi: 10.1186/1472-6963-14-328.

18. Lorentz M. Telenursing and home healthcare: the many facets of technology. *Home healthcare now*. 2008;26(4):237-43.

19. Moher D, Liberati A, Tetzlaff J, Altman DG, Group atP. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *Annals of Internal Medicine*. 2009;151(4):264-9.

20. Hawker S, Payne S, Kerr C, Hardey M, Powell J. Appraising the evidence: reviewing disparate data systematically. *Qualitative Health Research*. 2002;12(9):1284-99.

21. Baker C, Martin S, Thrasher J, Moore H, Baker J, Abman S, et al. A standardized discharge process decreases length of stay for ventilator-dependent children. *Pediatrics*. 2016;137(4): e20150637.

22. Miano M, Manfredini L, Garaventa A, Haupt R, Fieramosca S, Tanasini R, et al.

Home care program in a paediatric haematology and oncology department. Results of the first year of activity. *Haematologica*. 2001;33(3):218-23.

23. Gans D, Hadler M, Chen X, Wu S, Dimand R, Abramson J, et al. Cost Analysis and Policy Implications of a Pediatric Palliative Care Program. *Journal of Pain and Symptom Management*. 2016;52(3):329-35.

24. Gans D, Kominski G, Roby D, Diamant A, Chen X, Lin W, et al. Better outcomes, lower costs: palliative care program reduces stress, costs of care for children with life-threatening conditions. *UCLA Center for Health Policy Research*. 2012.

25. Sharif Azar E, Ravanbakhsh M, Torabipour A, Amiri E, Haghighizade M. Home-based versus center-based care in children with cerebral palsy: a cost-effectiveness analysis. *Journal of Medicine and Life*. 2015;8(Spec Iss 4):245-51.

26. Chong P, De Castro Molina J, Teo K, Tan W. Paediatric palliative care improves patient outcomes and reduces healthcare costs: Evaluation of a home-based program. *BMC Palliative Care*. 2018;17(1).

27. Postier A, Chrastek J, Nugent S, Osenga K, Friedrichsdorf S. Exposure to home-based pediatric palliative and hospice care and its impact on hospital and emergency care charges at a single institution. *Journal of Palliative Medicine*. 2014;17(2):183-8.

28. Berg A, Snell L, Mahle W. Home Inotropic Therapy in Children. *Journal of Heart and Lung Transplantation*. 2007;26(5):453-7.

29. Madden J, Northouse B, Tong S, Dorneman L, Foreman N, Rush S, et al. Benefits of using an early palliative care intervention in pediatric oncology: Quality of life and cost savings related to home delivery of cisplatin. *Journal of Hospice and Palliative Nursing*. 2015;17(4):319-24.

30. Hassani S, Navaei S, Shirzadi R, Rafiemanesh H, Masiha F, Keivanfar M, et al. Cost-effectiveness of home mechanical ventilation in children living in a developing country. *Anaesthesiology Intensive Therapy*. 2019;51(1):35-40.

31. Downing J, Knapp C, Muckaden M, Fowler-Kerry S, Marston J. Priorities for global research into children's palliative care: results of an International Delphi Study. *BMC Palliative Care*. 2015;14:36.

32. Berry J, Hall M, Dumas H, Simpser E, Whitford K, Wilson K, et al. Pediatric Hospital Discharges to Home Health and Postacute Facility Care: A National Study. *JAMA Pediatr*. 2016;170(4):326-33.

33. Caicedo C. Health and Functioning of Families of Children With Special Health Care Needs Cared for in Home Care, Long-term Care, and Medical Day Care Settings. *Journal of Developmental and Behavioral Pediatrics*. 2015;36(5):352-61.

34. Vickers J, Thompson A, Collins GS, Childs M, Hain R. Place and provision of palliative care for children with progressive cancer: a study by the Paediatric Oncology Nurses' Forum/United Kingdom Children's Cancer Study Group Palliative Care Working Group. *Journal of Clinical Oncology*. 2007;25(28):4472-6.

35. Vollenbroich R, Duroux A, Grasser M, Brandstätter M, Borasio G, Führer M. Effectiveness of a Pediatric Palliative Home Care Team as Experienced by Parents and Health Care Professionals. *Journal of Palliative Medicine*. 2012;15:294-300.

36. Emamgholipour S, Akbari Sari A, Geravandi S, Mazrae H. Estimation of out-of-pocket and catastrophic expenditures among patients with cardiovascular diseases in Khuzestan. *Journal of Payavard Salamat*. 2017;11(3):297-307.

37. Cabán P. The Use of Home Telemonitoring for Heart Failure Management Among Hispanics, Non-Hispanic Blacks, and Non-Hispanic Whites. 2019;37(6):345-9.

38. Bradford N, Herbert A, Walker R, Pedersen L, Hallahan A, Irving H, et al. Home telemedicine for paediatric palliative care. *Stud Health Technol Inform*. 2010;161:10-9.

39. Gold J, Hall M, Shah S, Thomson J, Subramony A, Mahant S, et al. Long length of hospital stay in children with medical complexity. *Journal of Hospital Medicine*. 2016;11(11):750-6.

40. Rassouli M, Sajjadi M. Palliative Care to the Cancer Patient. In: silbermann m, editor. The Middle East as a Model for Emerging Countries. USA: Nova Science; 2014.
41. Khandelwal N, Curtis J. Economic implications of end-of-life care in the ICU. *Current Opinion in Critical Care*. 2014;20(6):656-61.
42. Kuo D, Goudie A, Cohen E, Houtrow A, Agrawal R, Carle A, et al. Inequities in health care needs for children with medical complexity. *Health Aff (Millwood)*. 2014;33(12):2190-8.
43. Zarei E, Pouragha B, Khodakarim S. Out of pocket payment by inpatients of public hospitals after health sector evolution plan a cross-sectional study in Tehran City. *Journal of Hospital*. 2017;16(3):9-17.
44. Tamiya N, Yamaoka K, Yano E. Use of home health services covered by new public long-term care insurance in Japan: impact of the presence and kinship of family caregivers. *International Journal for Quality in Health Care*. 2002;14(4):295-303.
45. Hospice Malaysia. A Needs Assessment of Palliative Care in Malaysia: 2016 2016. Available at: <https://www.hospismalaysia.org/wp-content/uploads/2016/10/Palliative-Care-Needs-Assessment-Malaysia-2016.pdf>.
46. Ferrell B, Coyle N, Paice J. Oxford textbook of palliative nursing: Oxford University Press; 2014.
47. Kremeike K, Reinhardt D, Sander A. The role of nursing in pediatric palliative home care—Experiences from Lower Saxony, Germany. *Journal of Nursing Education and Practice*. 2014;4(2):243-52.
48. Chitnis X, Georghiou T, Steventon A, Bardsley M. Effect of a home-based end-of-life nursing service on hospital use at the end of life and place of death: a study using administrative data and matched controls. *BMJ supportive & palliative care*. 2013;3(4):422-30.
49. Strano A, Briggs A, Powell N, Brockman J, Taylor J, Butler A, et al. Home Healthcare Visits Following Hospital Discharge: Does the Timing of Visits Affect 30-Day Hospital Readmission Rates for Heart Failure Patients? *Home healthcare now*. 2019;37(3):152-7.
50. Brant J, Fink R, Thompson C, Li Y, Rassouli M, Majima T, et al. Global Survey of the Roles, Satisfaction, and Barriers of Home Health Care Nurses on the Provision of Palliative Care. *Journal of Palliative Medicine*. 2019;22(8):945-60.