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TITOLO TESI

The Special Needs Kids (SpeNK) Project. Lessons learned and future perspectives.

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«Se la metodologia non dà
risultati, occorre cambiarla»
– Dr . Plonk –

citazione dal film "Dr. Plonk" (2007)

THESIS ABSTRACT

AIMS. To assess integrated healthcare pathways for children with special health care needs after hospital discharge from St. Orsola Malpighi University Hospital of Bologna and Local Health Authorities of Bologna and Imola.

MATERIALS AND METHODS. Existing procedures were assessed through interviews with key informants and collection of written procedures. During 24 months, children meeting inclusion criteria as incident cases were recruited at discharge with their families. During 9 months of follow-up parents' perspective was assessed with qualitative methods (SpeNK-I) and the administration of a 20-item questionnaire (SpeNK-Q), after its validation in a sample of parents of 101 preterm newborns. During follow-up, Family Pediatricians (FPs) recorded care coordination activities for subjects with an 8-item data collection tool (SpeNK-FP). Utilization of healthcare resources was examined through data-linkage with administrative databases.

RESULTS AND DISCUSSION. Different practices and procedures exist, with different level of standardization and formalization within Bologna province. A sample of 82 children was recruited, with a majority of extremely low birth weight newborns (42.6%). To 16 parents involved in SpeNK-I, continuity of care and empowerment were important, with different issues from hospitalization to home care. The SpeNK-Q administered to parents of 67 children reported high levels of continuity of care, with items referring to the informational continuity endorsed with the lowest frequency. Forty FPs recorded 382 encounters for 49 children, showing some difficulty to record these activities and a potential for care coordination. Administrative data analyses showed a higher level of utilization of healthcare services for hospitalizations, some differences in specialty outpatient care use with almost none in emergency room contacts.

CONCLUSIONS. In Bologna province, children with special health care needs run a continuous and coordinated integrated healthcare pathway although with some weaknesses. Connecting users', professionals' and system's viewpoints may facilitate the identification of improvement areas to reduce risk of fragmentation.

KEYWORDS: *children with special health care needs, continuity of care, care coordination, hospitalization, discharge, preterm birth, integrated care, family support, parents experience, pediatric primary care, family pediatrician, assessment, healthcare resources utilization*.

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Finally, this study would not have been possible without the participation of families, parents and children, and family pediatricians who consented to share their experience with us, to help in the assessment of health care for such an important although numerically minor part of pediatric population.

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NOTE ON THE AUTHOR'S CONTRIBUTION

I participated in the SpeNK Project from 2013 to 2015 during my PhD, under the supervision of my tutor, Prof. Maria Pia Fantini. I dealt personally with the research activities dedicated to explore the family’s perspective, with the development of qualitative and quantitative research tools, the data collection and analyses and writing scientific papers about the studies. From 2014 I was introduced in the study about the Family Pediatricians’ activities and the administrative data about SpeNK children, by collaborating in quality data control, statistical analyses and writing scientific papers.

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Introduction

This thesis describes objectives, methods and results of the SpeNK (Special Needs Kids) Project, conducted in Bologna province from 2012 to 2015, with the participation of University of Bologna, the St. Orsola Malpighi University Hospital of Bologna, and the Local Health Authorities of Bologna and Imola, in the framework of Region-University Research Program 2010-2012, Area 2 - Clinical governance. The study aimed to describe and review the implementation of existing sheltered hospital discharge procedures and integrated clinical pathways in Bologna province for children with complex or chronic health conditions and special healthcare needs, and to assess their utilization of healthcare resources, the family's perspective on continuity of care, the role of family pediatrician in care coordination. To achieve these goals, the study was divided in different branches, investigating different themes, which are presented as follows.

Chapter 1 illustrates synthetically the whole Project, from the background (rationale and context) to methods and main results of the Project.

Chapter 2 presents the qualitative study (SpeNK-I) conducted to investigate parents' experiences and perceptions in the interaction with healthcare services and providers during first months after their child's hospital discharge, as published by Zanello et al. (2015).

Chapter 3 describes the part of SpeNK Project aiming to develop a quantitative measure of continuity of care from parents' perspective, on the basis of SpeNK-I Results. Section 3.1 illustrates the study conducted to develop and validate a 20-item questionnaire (SpeNK-Q) in a population of parents of preterm infants, as published by Rucci et al. (2015). Section 3.2 describes methods and results of the SpeNK-Q administration to parents of children enrolled in SpeNK Project .

Chapter 4 presents the study conducted to investigate the role of Family Pediatrician (FP) in care coordination for children enrolled in SpeNK Project during 9 months after hospital discharge.

Chapter 5 illustrates the retrospective study of utilization of healthcare resources by children recruited to SpeNK Project through the data-linkage with Regional Healthcare Service databases.

Finally the Conclusion paragraph attempts to draw conclusions of the whole Project, connecting the findings presented in each Chapter, in order to summarize "lessons learned and future perspectives" for care of children with special health care needs.

Chapter 1. The SpeNK (Special Needs Kids) Project

The SpeNK (Special Needs Kids) Project is a cohort prospective study focusing on the implementation and assessment of existing sheltered discharge procedures and integrated healthcare pathways for newborns and children with special health care needs in Bologna province. It was funded by the Regional Agency for Health and Social Care, Emilia Romagna Region, grant DGR 2042/2011, in the framework of Region-University Program 2010-2012, Area 2 - Clinical governance. Ethical approval for the study was obtained from Ethics Committees of the University Hospital of Bologna and Local Health Authorities of Bologna and Imola.

1.1. Rationale

Children with special health care needs represent a growing although numerically minor category of pediatric population. There are multiple definitions of "children with special health care needs" and the estimates of prevalence and incidence depend on the assumed definition.

A review of definition and measurement of chronic health conditions in childhood by van der Lee, Mokkink, Grootenhuis, Heymans, and Offringa (2007) identified a large range of definitions in use and various operationalization of the concepts. Table 1 illustrates the most frequently cited definitions of chronic health conditions in childhood as presented in the review.

Table 1 - Most frequently cited definitions of chronic health conditions in childhood (van der Lee et al., 2007)

Source	Location	Concept	Proposed Definition	Operationalization	Prevalence
Pless and Douglas (1971)	England, Wales, and Scotland	Chronic illness	A physical, usually nonfatal condition that has lasted longer than 3 mo in a given year or necessitated a period of continuous hospitalization of more than 1 mo; of sufficient severity to interfere with the child's ordinary activities to some degree	National Child Development Study, a longitudinal survey of all children born in 1 week in England, Scotland, and Wales	11% aged <16 y
Perrin et al. (1993)	United States	Chronic health condition	A condition is considered chronic if (1) it has lasted or is expected to last more than 3 mo and (2) the definition takes into account the impact of the condition on the child, eg, level of functional impairment or medical need greater than expected for a child of that age	NHIS, 1989	31% aged <18 y; added: 5.3%; unable to conduct major activity: 0.6%
Stein, Bauman, Westbrook, Coupey, and Ireys (1993)	United States	Chronic health conditions	Conditions must have a biological, psychological, or cognitive basis; have lasted or are virtually certain to last for 1 y; and produce _1 of the following sequelae: (1) limitations of function, activities, or social role in comparison with healthy age peers in the general areas of physical, cognitive, emotional, and social growth and development; (2) dependency on 1 of the following to compensate for or minimize limitations of function, activities or social role: medications, special diet, medical technology, assistive device, or personal assistance; and (3) need for medical care or related services, psychological services, or educational services above the usual for the child's age or for special ongoing treatments, interventions, or accommodations at home or in school	NA	NA

Stein and Silver (1999)	United States	Chronic health conditions	Same as for Stein et al. (1993)	NHIS household telephone interview, 1994	14.8% aged <18 y
McPherson et al. (1998)	United States	Children with special health care needs	Children who have or are at increased risk of a chronic physical, developmental, behavioral, or emotional condition and who also require health care and related services of a type or amount beyond that required by children generally	NA	NA
Newacheck et al. (1998)	United States	Children with special health care needs	Same as for McPherson et al. (1998)	NHIS-D, 1994	12% aged <18 y; additional 6% had a presumed need for health care or related services

Abbreviations: NA, not applicable; NHIS, National Health Interview Survey; NHIS-D, National Health Interview Survey on Disability.

The earliest definitions focused on the presence of a specific condition, on the duration of the disease and its impact on daily activities (Perrin et al., 1993; Stein et al., 1993), while the most recent definition includes the risk for a chronic condition and focuses the impact on the need for health and related services (McPherson et al., 1998). According to this review, the prevalence estimate ranges from 0.22% to 44% depending on considered operationalization (van der Lee et al., 2007).

More recently, an estimate of the National Survey of Children with Special Health Care Needs Chartbook 2009–2010 in United States indicated that about 15% of children under 18 years of age have special health care needs and 23% of households with children have at least one child with special health care needs (US Dept of Health and Human Services, 2013).

In Italy, information about prevalence and incidence of children with special health care needs is limited to specific health conditions, e.g. disease requiring palliative care, and disability. A document by the Italian Health Ministry (Ministero della Salute, 2008) reports an estimated prevalence of 10 per 10,000 minors requiring palliative care with a mortality rate of 0.8-1/10,000. As for complex disabilities and genetic diseases, Mastroiacovo and Costantino (2007) estimates a prevalence of 0.5% of in pediatric population (0-17 years). Elsewhere, the prevalence of “special health care needs”, referring to a broader definition, is estimated to range from 13 to 16% (Zampino, 2010). In Emilia Romagna, a study conducted at the Local Health Authority of Bologna indicates that 1 child/teenager below 18 years of 5882 has a chronic disease requiring high-complexity care with invasive medical devices.

Similarly to adult patients with chronic conditions, children with special health care needs generally require long-term health care provided by an array of professionals and clinicians in multiple outpatient and inpatient settings. Because of the complexity of their needs, they are at risk of fragmented care and vulnerable to issues of continuity and coordination of care in their healthcare pathway. In pediatric as well as adult population, care for chronic and complex conditions requires the integration of services and the collaboration among professionals in different settings (i.e. hospital, primary care, etc.). The care process for patients with special health care needs is frequently poorly coordinated. Higher levels of patient satisfaction, less fragmentation and fewer recorded episodes of medical errors were found in the contexts where the doctors and the medical staff know patient's history and cooperate to provide coordinated care (Berry et al., 2011; Gulliford, Cowie, & Morgan, 2011; Uijen et al., 2011). Anyway, as suggested by Miller et al. (2009), situation and needs of pediatric patients are different, because of their developmental status and change, the critical mediating role played by parents in children's health care, and the impact of school context on children's social development.

Two key elements for care of children with special health care needs are continuity of care and coordination of care, with the first meaning the degree to which the patients experience their perceived care over time as coherent (Reid, Haggerty, & McKendry, 2002) and the second the intentional organization of care activities among providers and with the patient to facilitate the appropriate delivery of health care services (McDonald et al., 2007).

Continuity of care can be defined as "*the degree to which a series of discrete healthcare events is experienced as coherent and connected and consistent with the patient's medical needs and personal context*" (Haggerty et al., 2003). According to the American Academy of Pediatrics, comprehensive healthcare should ensure *inter alia* continuity, providing care over an extended period of time and planning and organizing transitions, to other pediatric providers or into adult health care services, with the child and family (American Academy of Pediatrics, 2002).

In the conceptualization developed by Reid and Haggerty (Haggerty et al., 2003; Reid et al., 2002), three types of continuity of care can be identified across healthcare settings:

1. Informational continuity: the use of information on past events and personal circumstances to make current care appropriate for each individual among providers and among healthcare events;
2. Management continuity: a consistent and coherent approach to the management of a health condition that is responsive to a patient's changing needs which is especially important in chronic or clinically complex diseases;
3. Relational continuity: an ongoing therapeutic relationship between a patient and one or more providers which bridges past to current care and provides a link to future care.

The theme of continuity of care has been widely explored in health care for adult patients with chronic conditions and multiple diseases, while less attention has been paid to pediatric patients. A review of the instruments to measure continuity of care showed that most available instruments on continuity of care from patients' perspective are designed to assess this construct in specific adult populations and settings (e.g. patients with diabetes, cancer, mental health problems, etc.) (Uijen et al., 2012), with the exception of a questionnaire to measure continuity in mental health care for children from family's perspective (Tobon, Reid, & Goffin, 2014). Miller et al. (2009) conducted a qualitative study to explore the extent to which parents' experiences and perceptions fit with the academic and service providers' perspectives about continuity of care. Results indicated that the concepts of relational, informational and management continuity were all discernible in parents' narratives (Miller et al., 2009).

According to McDonald et al. (2007), care coordination is "*the deliberate organization of patient care activities between two or more participants (including the patient) involved in a patient's care to facilitate the appropriate delivery of health care services.*" It has been widely recognized as important to achieve a high-quality, high-value, patient-centered healthcare system (Schultz & McDonald, 2014). The goal of care coordination is to support patients and their families requiring healthcare in their interaction with an increasingly complex healthcare system. As for children with special health care needs, American Academy of Pediatrics (2005) defines care coordination as "*a process that links children and youth with special health care needs and their families with appropriate services and resources in a coordinated effort to achieve good health*". It must be measurable, auditable, and amenable to continuous quality improvement (Antonelli, Stille, & Antonelli, 2008). A key role in care coordination for children with special health care needs should be played by the primary care provider (Stille & Antonelli, 2004; Stille, Jerant, Bell, Meltzer, & Elmore, 2005), and some Authors identify the clinic of the Family Pediatrician as the best place for the care coordination (Antonelli et al., 2008; Starfield, 2003). However, some studies underlined the scant involvement of primary care providers in managing care of children with chronic conditions (Palfrey, Levy, & Gilbert, 1980; Rowland, 1989).

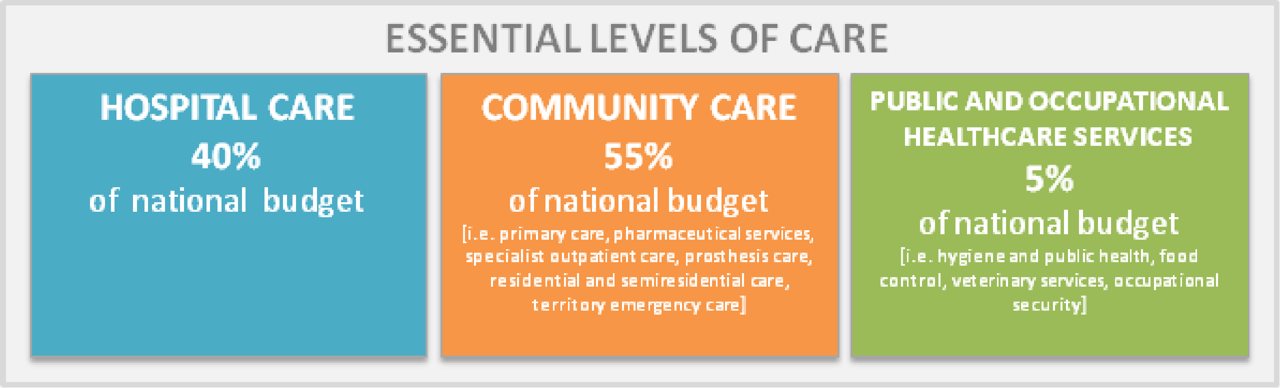
1.2. Context

1.2.1. Healthcare services in Italy

Italy is a country located in south-central Europe with a population of 60,795,612 inhabitants (source: ISTAT, January, 1 2015). With a percentage of about 8% of foreign citizen, Italy is one of the most populous countries in Europe, characterized by a low birth rate (8.4 per 1,000) and a high aging index (157,3). At administrative level, the country is subdivided into 21 regions with five regions having a special autonomous status that enables them to enact legislation on some of their local matters (e.g. health, education etc.).

Article 32 of Italian Constitution enshrines the “right to health” of all individuals, which is exerted through the National Health Service (NHS), instituted by law in 1978 (law 23 December 1978, n. 833). The NHS guarantees health care to all citizens through a public system, based on principles of universalism and comprehensiveness, financed by the State through general taxation and direct revenues by performance fees and prescription charges (i.e. “health tickets”, meaning the shares in which the patient contribute to the costs). The NHS consists of the Ministry of Health, which coordinates the National Health Plan, a number of organizations and institutions at national level, and the Regional Health Services (RHS), providing health care to citizens in each Region. The State has the responsibility to ensure all citizens the right to health with a strong system of safeguards, through the Essential Levels of Care (i.e. *LEA, Livelli Essenziali di Assistenza*), whereas the Regions have the responsibility for government and spending for achieving the national health goals. The Regions have virtually exclusive power over regulation, organization, administration and funding of health care in their territory, which is exerted through Local Health Authorities (LHAs) or Trusts (LHTs) and Hospital Trusts (HTs) on the domains of hospital care, community-primary care, and prevention and public health, in accordance with LEAs (see Figure 1).

Figure 1 - The partition of national budget for LEAs



In addition to the services included in LEAs, individual Regions may establish supplementary services to be provided to their citizens, using their own funds; in case of need, Regions and LHAs can also purchase the services to be provided to citizens from private (accredited) hospitals or clinics. In Italy, citizens can receive health care without costs or with a marginal participation in spending through the payment of a “health ticket” for each service received.

1.2.2. Healthcare services in Emilia-Romagna

Emilia-Romagna is a region in Northern Italy with around 4.4 million inhabitants. According to the Regional Report of Facilities, Expenditure, Activities 2010-2013, the Regional Health Service (RHS) comprises:

- 8 Local Health Trusts (LHTs): Piacenza, Parma, Reggio Emilia, Modena, Bologna, Imola, Ferrara, and Romagna. The LHT of Romagna was instituted on January 1st, 2014 and gathers the facilities and services of the LHTs of Cesena, Forlì, Ravenna and Rimini which starting from that date ceased to be. The other LHTs usually cover the entire provincial area, with the exception of Bologna, covered by Bologna LHT and Imola LHT;
- 4 University Hospitals (UHs): Parma (Maggiore Hospital), Modena (Policlinico Hospital), Bologna (St. Orsola Malpighi Policlinico Hospital) and Ferrara (S. Anna di Cona Hospital);
- 1 Hospital Trust: Reggio Emilia Hospital (Santa Maria Nuova Main Hospital);
- 4 Research Hospitals (IRCCS): the Rizzoli Orthopedic Institute of Bologna, the Bologna Institute of Neurological Sciences (within Bologna Local Health Trust) the Reggio Emilia Institute of Advanced Technologies and Care Models in Oncology (within the Reggio Emilia Hospital), the Romagna Institute for Cancer Research and Care in Meldola, acknowledged as Research Hospital in advanced therapies for medical oncology on May 2012.

For planning, organizational and allocation reasons, three Care Vast Areas referring to major geographical areas were created: Emilia Nord-AVEN (1,975,763 pop. – including Piacenza, Parma, Reggio Emilia, Modena), Emilia Centro-AVEC (1,358,617 pop. – including Bologna, Imola, Ferrara) and Romagna-AVR (1,124,866 pop. – including Ravenna, Forlì, Cesena and Rimini), which do not possess a legal status and consist in a functional grouping of the Health Trusts.

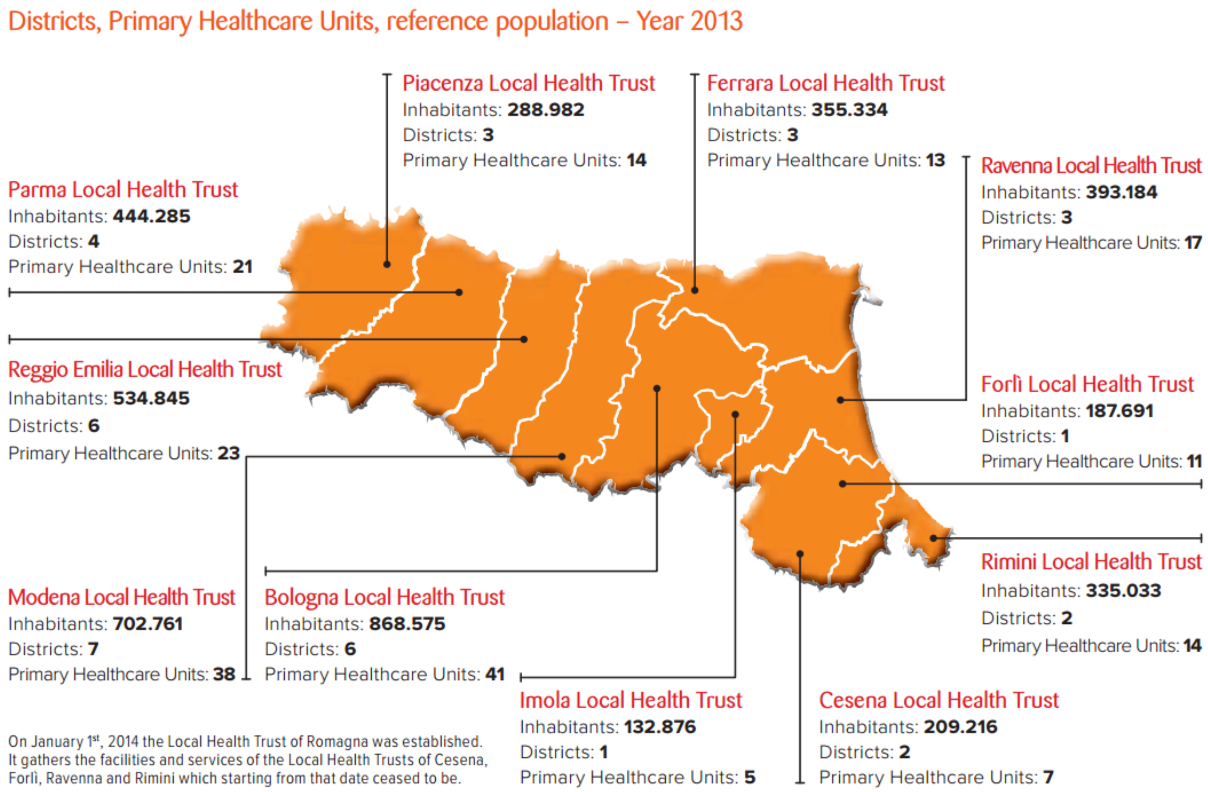
Each LHT has a number of health districts, territorial departments (i.e. Primary Care Department, Public Health Department, Mental Health and Pathological Addictions Department) and hospital departments. The Health District is the territorial organization of the LHT guarantor for the delivery of LEAs, by commissioning services to territorial and hospital departments.

The Primary Care Departments, which are in turn organized in Primary Healthcare Units (i.e. *NCP, Nuclei Cure Primarie*), work in integration with the hospital departments and the network of health and social services to ensure continuity of care, as reported in the web site of the Emilia Romagna RHS (http://salute.regione.emilia-romagna.it/). They provide: primary care (assistance of

General Practitioners and Family Pediatricians, emergency out-of-hours medical care), outpatient specialist care, home care services, healthcare for women, families, couples (Family Advisory Health Centers, community pediatricians), care for foreigners, assistance to AIDS patients, social and health care in nursing homes and at home, pharmaceutical care, and procedures for assistance abroad.

According to the above-mentioned Regional Report, as of 31 December 2013, Emilia Romagna RHS encompasses 38 Health Districts and 204 Primary Care Units (see Figure 2).

Figure 2 - Health Districts, Primary Healthcare Units, reference population (2013)



Districts, Primary Healthcare Units, reference population – Year 2013

Piacenza Local Health Trust
Inhabitants: **288.982**
Districts: **3**
Primary Healthcare Units: **14**

Ferrara Local Health Trust
Inhabitants: **355.334**
Districts: **3**
Primary Healthcare Units: **13**

Ravenna Local Health Trust
Inhabitants: **393.184**
Districts: **3**
Primary Healthcare Units: **17**

Parma Local Health Trust
Inhabitants: **444.285**
Districts: **4**
Primary Healthcare Units: **21**

Reggio Emilia Local Health Trust
Inhabitants: **534.845**
Districts: **6**
Primary Healthcare Units: **23**

Forlì Local Health Trust
Inhabitants: **187.691**
Districts: **1**
Primary Healthcare Units: **11**

Rimini Local Health Trust
Inhabitants: **335.033**
Districts: **2**
Primary Healthcare Units: **14**

Modena Local Health Trust
Inhabitants: **702.761**
Districts: **7**
Primary Healthcare Units: **38**

Bologna Local Health Trust
Inhabitants: **868.575**
Districts: **6**
Primary Healthcare Units: **41**

Imola Local Health Trust
Inhabitants: **132.876**
Districts: **1**
Primary Healthcare Units: **5**

Cesena Local Health Trust
Inhabitants: **209.216**
Districts: **2**
Primary Healthcare Units: **7**

On January 1st, 2014 the Local Health Trust of Romagna was established. It gathers the facilities and services of the Local Health Trusts of Cesena, Forlì, Ravenna and Rimini which starting from that date ceased to be.

The Districts entail the **Proximity Outpatient Clinics** (i.e. *Case della Salute*), healthcare facilities envisaged by the Resolution of the Regional Council 291/2010. They are designed to represent a reference point for citizens' access to primary care and structured as integrated services that take care of people from the moment of access, promoting the collaboration between professionals and the sharing of care pathways. The Clinics receive patients and direct them towards services, but also provide ongoing care, the management of chronic diseases and the completion of the main diagnostic pathways that do not require hospitalization. Their management is assigned to the Primary Care Department, which coordinates the care provided and interfaces with other departments. The Clinics can differ in terms of complexity and offer different services, in relation to the population density of the reference area and their geographical location. According to a regional Report on Proximity outpatient clinics of march 2015, in Emilia-Romagna there are 67 functioning and 55 projects for these facilities, for a total of 122 planned clinics.

1.2.3. The Family Pediatricians

Since 1978, primary care in Italy is provided by General Practitioners (GPs) and Family Pediatricians (FPs), independent professionals working under government contract for the NHS. LHAs pay GPS and FPs on a capitation basis depending on the number of patients enrolled in the physician's list, with a maximum of 1,500 patients for GPs and 800 for FPs.

FPs provide pediatric primary care to all children aged 0-14 years. All children from 0 to 14 years are required to register with a FP, who can follow the child up to 16 years under certain circumstances (i.e. chronic conditions or special health needs). For children over 6 years, anyway, the family can choose a GP to care for their child. Differently from GPs with expertise or interest in pediatrics, FPs are trained specialists working at the primary care level, in solo or group practices. During weekday working hours, FPs provide children with acute outpatient and home care, coordination of care for patients with chronic conditions, consults with subspecialties. Afterhours, on weekends or holidays, primary care is provided by non-pediatrician physicians, who guarantee night and weekend phone coverage as well as urgent homecare to all patients, free of charge. FPs can receive additional allowances paid by the LHAs (agreed at regional level) for the delivery of planned care to specific patients, such as home care for chronically ill and disabled children. As for primary care, families incur no costs by visits to the FP, whereas the provision of specialist outpatient services and medical devices by the RHS contemplates a partial or total sharing of costs by patients, except for low income families and families with children with chronic conditions, who are exempt from such charges. Outpatient care is provided either directly by LHA's services or by accredited public or private facilities under official agreements with LHAs.

In recent years, similar to their colleagues in other European countries, Italian FPs are changing their practice from solo towards groups, in order to improve quality and continuity of care. Models of "collaborative" practice include groups, network and associations among physicians. These models are intended to maximize accessibility for patients belonging to a geographically defined area (i.e. Health District in most of cases), by improving services delivery with longer office hours, continuity of care and wider range of services. These forms of grouping have been promoted by the NHS since 2000 through economic incentives but the current percentage is still lower than the average of other similarly oriented countries.

FPs, as well as other primary care physicians (e.g. GPs), are given the possibility to participate in a network with other physicians keeping their own practice, or to work in a group practice sharing offices and patient electronic health record system.

In Emilia Romagna, the FPs activity is entailed in Primary Care Departments. The Primary Care Units gather GPs and FPs, and other professionals in the area referring to the same Unit, in order to integrate them in the organization of LHAs. These Units are designed to ensure shared clinical pathways and continuity of care, to enhance access to outpatient visits by extending the opening hours, to improve home care for dependent people or people at risk of dependency treated at home, to improve the management of chronic conditions, and to hold waiting lists for specialist appointments and tests. Their objectives and organizational structure are defined by Agreements between the Region and the trade unions of GPs and FPs. As reported in the Regional Report of Facilities, Expenditure, Activities referring to 2010-2013, as of 31 December 2013, Emilia Romagna RHS encompasses 38 Health Districts and 204 Primary Care Units, with 3,086 General practice physicians (i.e. GPs) and 628 primary care pediatricians (i.e. FPs). GPs and FPs are given the possibility to work in association with other colleagues within the Proximity outpatient clinics. A recent regional Report on Proximity outpatient clinics (March 2015), accounts that only 45% of GPs and 23% of FPs who operate in the reference catchment area of a clinic, work in association with other colleagues within it (486 GPs of 1083, and 52 FPs of 228, respectively).

1.2.4. Regulatory Framework

The regulatory framework of primary care for pediatric population include a number of recommendations and guidelines at national and regional level.

According to the National Health Program (i.e. *Piano Sanitario Nazionale*) 2006-2008, the local and community services are identified as key elements for the organization of the health response, the integration of social and health services and the government of healthcare pathways. In particular, they are in charge of: (1) defining appropriate diagnostic/ therapeutic /rehabilitation pathways for congenital hereditary pathologies and rare diseases, through better organization of the centers of reference at regional and interregional level and implementation of care networks; (2) improving care for children and adolescents with chronic diseases developing integrated models with specialist centers, hospitals, local community services and activities, such as psychological and social care, school, patients' associations and non-profit private.

The outline of the National Health Program (i.e. *Schema di Piano Sanitario Nazionale*) 2011-2013, states that the continuity among the different healthcare settings within the network should be ensured by a coordination unit, in order to ensure the most appropriate responses to the physical, psychological, social and spiritual needs of the patient and his/her family. Special attention should be paid to the specific development of a healthcare network for palliative care and pain therapy for children with incurable or terminal diseases. Such care should be provided by teams of experts in specialist pediatric palliative care from the closest community and hospital service. The home care is the service of choice for pediatric patients: for this purpose, the appropriate actions should be promoted to activate protocols for sheltered discharge procedures from the Intensive Care Units and other hospital units, and to activate residential solutions for particular transitional conditions, when requested by families.

The National Collective Agreement (i.e. *Accordo Collettivo Nazionale*) (July 2010), which governs the relationship of the National Health Service with FPs, indicates as guidelines for the Regional Integrative Agreements with FPs, the implementation of an integrated network of services, the improvement of taking care of children with chronic conditions and the government of shared healthcare pathways.

The Integrative Regional Agreement for FPs of Emilia Romagna (2011) enhances the development of the initiative medicine and the improvement of the integrated taking in charge of children with chronic illness or conditions of psychological and social distress. In this context, the Family Pediatrician represents a key figure, with the function of "process owner" as regards the management of the health conditions of the child, particularly with special needs.

Finally, other guidelines and recommendations concerning the activities of the Family Pediatrician has been issued at regional and national level, for example the Resolution of Regional Government of Emilia Romagna DGR 2011/2007 (i.e. *Direttiva alle Aziende sanitarie per l'adozione dell'atto aziendale: indirizzi per l'organizzazione dei Dipartimenti di cure primarie, di salute mentale e dipendenze patologiche di sanità pubblica*) and the national law 38/2010 about the instructions to ensure access to palliative care and pain therapy.

1.2.5. The Region-University research and innovation programs

The Region-University Research Program is one of the main activities through which Emilia Romagna Region aims to support and encourage research within its Regional Health Service. It is mainly addressed to University-Hospital Trusts and IRCCS, as pivotal figures of integration and collaboration between health services and academia. Research Program activities aim to: encourage potentially most promising research areas, capable of meeting the informative and operative needs of health services; deepen the knowledge on emerging technologies; verify the degree of diffusion and adoption of technologies and their implications for the internal organization of Trusts. The Program includes 3 research areas: “area 1” for innovative research, “area 2” for clinical governance, “area 3” for research training and creation of research networks. After the first two successful editions (2007- 2009; 2010-2012), the Research Program in 2013 was refunded by the Region with 5 millions of Euros (Resolution 199 of 25 February 2013).

1.3. Aims

The **primary aim** of SpeNK Project was to describe and review the implementation of existing sheltered hospital discharge procedures and integrated clinical pathways in Bologna province for children with complex or chronic health conditions and special healthcare needs.

The **secondary aims** of SpeNK Project were:

- To describe existing sheltered hospital discharge procedures, degree of coordination among different healthcare settings and different providers, resource utilization for newborns and children with special health care needs living in Bologna province;
- To quantify the time spent by the family pediatrician in care-coordination activities;
- To assess experiences and perceptions of parents of children with special health care needs related to the interaction with the health care system for their children’s care, including continuity of care, shared-decision making, proactive care received, involvement of other care providers after discharge.

1.4. Collaborating Centers

The study involved the University of Bologna, the St. Orsola Malpighi University Hospital (UH) of Bologna and the Local Health Authorities (LHAs) of Bologna and Imola as collaborating centers, with a total of 11 participating units, as follows:

1. Neonatology Unit, St. Orsola Malpighi UH of Bologna, with Prof. Giacomo Faldella as Principal Investigator, and collaboration of Dr. Rosina Alessandroni and Dr. Silvia Vandini;
2. Unit of Hygiene, Public Health and Biostatistics, Department of Biomedical and Neuromotor Sciences, University of Bologna, as Methodology Unit coordinated by Prof. Maria Pia Fantini;
3. Anesthesiology and Intensive Care Unit, St. Orsola Malpighi UH of Bologna (Prof. Simonetta Baroncini and Dr. Andrea Gentili);
4. Pediatric Surgery Unit, St. Orsola Malpighi UH of Bologna (Prof. Mario Lima and Dr. Claudio Antonellini);
5. Child Neuropsychiatric Unit, St. Orsola Malpighi UH of Bologna (Prof. Emilio Franzoni and Dr. Valentina Marchiani);
6. Pediatrics Unit, St. Orsola Malpighi UH of Bologna (Prof. Andrea Pession and Dr. Giuseppina Paone);
7. Pediatric Onco-Hematology Unit and Pediatric Emergency Room, St. Orsola Malpighi UH of Bologna (Prof. Filippo Bernardi and Dr. Rosalba Bergamaschi);
8. Neonatology and Neonatal Intensive Care Unit, Maggiore Hospital of LHA of Bologna (Dr. Fabrizio Sandri and Dr. Silvia Soffritti);
9. Neonatology Unit and Primary Care Department, Hospital of LHA of Imola (Dr. Marcello Lanari, Dr. Deborah Silvestrini);
10. Primary Care Department, LHA of Bologna (Dr. Mara Morini).

1.5. Methods

The SpeNK Project is based on a cohort prospective study design. The study started in 2012 and ended in 2015; the recruitment lasted from 1st October, 2012 to 30th September, 2014. The Project included three steps: (1) the **preliminary phase** (3 months), to review of procedures and pathways in the collaborating centers, to define the study procedures, and to develop the research tools; (2) the **recruitment phase** (24 months), to enroll incident cases at hospital discharge from the participating units; (3) the **follow-up phase** (9 months for each recruited subject), to collect information about family's perspectives on the healthcare provided to their child and about activities performed by the Family Pediatrician for care coordination.

1.5.1. Preliminary phase

1.5.1.1. Review of procedures and practices

Existing procedures for sheltered hospital discharges were explored by interviewing key informants and collecting and reviewing the formalized procedures and practices approved and implemented by the collaborating centers of the University Hospital of Bologna and the two local Health Authorities of Bologna and Imola.

1.5.1.2. Definition of study procedures

A coordination group was formed consisting of representatives of participating units, who met to define the study procedures. On this basis, the Methodology Unit of the University of Bologna developed two written procedures: the “Recruitment Procedure”, targeted to persons responsible for the subjects enrollment at each unit, describing inclusion criteria and recruitment activities, and the “Follow-up Procedure”, targeted to Family Pediatricians, providing a guide line for the research activities for the follow-up of recruited subjects.

1.5.1.3. Development of research tools

The Methodology Unit of Bologna University developed the research tools for different purposes:

(1) The Recruitment Data Collection Tool used at the recruitment of patients and families to record the information about clinical conditions, hospital admission and discharge, care plan, etc. (see Appendix - SpeNK Recruitment);

(2) the Interview and focus Group (SpeNK-I) used in the qualitative study to explore family’s perspectives on continuity of care (see Appendix - SpeNK-I);

(3) A 20-item Questionnaire (SpeNK-Q) developed as quantitative measure of family’s perspective on continuity of care, on the basis of SpeNK-I results (see Appendix - SpeNK-Q);

(4) the Data Collection Tool (SpeNK-FP) to record care coordination activities of Family Pediatricians during the follow-up (see Appendix - SpeNK-FP).

Appendixes include the research tools in their original form (Italian language). The use of the research tools is described in the chapter concerning the single study.

1.5.2. Recruitment phase

1.5.2.1. Children and Families

Children and families were recruited to SpeNK Project at hospital discharge of the child from the participating units of the hospital facilities at the University Hospital of Bologna (St. Orsola Malpighi) and Local Health Authorities of Bologna and Imola.

The recruitment was conducted from 1st October 2012 to 30th September 2014 on incident cases meeting the following inclusion criteria: age from 0 to 16 years, residence in Bologna province, and the presence of at least one of the following conditions:

- Birth weight <1000 gr.;
- Complex and/or chronic health conditions defined as:
    - Need for technological assistance,
    - Acute neurological deficit,
    - Severe endocrinopathy,
    - Complex genetic malformative pathology;
- Children with oncological diseases who need palliative care or particular community care;
- Newborns with mothers in contact with mental health services or drug addiction

Only first ever hospitalizations for the condition of interest were included (incident cases). Written informed consent was obtained at recruitment from each parent to collect clinical data on children and to contact parents during follow-up period. The unique exclusion criterion was the refusal to participate in the study.

The Recruitment Data Collection Tool was used to collect and record information about the child, his/her clinical conditions and care plan, etc., and contact details about parents (see Appendix - SpeNK Recruitment).

1.5.2.2. Family Pediatricians (FPs)

Family Pediatricians (FPs) in charge of children enrolled were invited to participate in the study. The Methodology Unit arranged their involvement in the research project in coordination with the Primary Care Departments of Local Health Authorities of Bologna and Imola. Research objectives and instruments were shared with Unions of Pediatricians, first, and with FPs, in person, later. FPs received an incentive for their participation in the study, as part of a contractual agreement between the Primary Care Departments and the Unions of Pediatricians. Information and contact details about each FP were recorded in an Excel File by the Methodology Unit, whose members were in charge of contacting and supporting FPs for research activities.

1.5.3. Follow-up phase

1.5.3.1. Involvement of Families

During follow-up, families of enrolled children were involved to explore their experiences and perceptions in the interaction with healthcare services and providers who took care of their children after hospital discharge.

The first step was **a qualitative study** (SpeNK-I), involving families of first children recruited to SpeNK Project. Families were selected according to a maximum variation sampling method (Hsieh & Shannon, 2005) with regard to the child's diagnosis and the hospital of discharge and excluding parents with an inadequate level of knowledge of Italian language. On the basis of literature about continuity of care (Haggerty et al., 2003; Miller et al., 2009), the Methodology Unit developed a semi-structured interview (SpeNK-I) which was used to conduct face-to-face and phone interviews and a focus group with selected families after 1-6 months after discharge (see Appendix - SpeNK-I). A directed approach to the qualitative content analysis, as described by Hsieh and Shannon (2005), was used to identify emergent themes in parents' narratives.

The second step concerned the development and use of **a quantitative measure** of continuity of care for children with special healthcare needs from parents' perspective. To this end, the Methodology Unit developed a 20-item questionnaire (SpeNK-Q) on the basis of SpeNK-I results and referring to Haggerty's constructs of informational, management and relational continuity (Haggerty et al., 2003; Haggerty, Roberge, Freeman, Beaulieu, & Breton, 2012). The SpeNK-Q validation was conducted at St. Orsola Malpighi University Hospital of Bologna on a similar population, i.e. parents of preterm newborns accessing to the Preterm Infant Follow-up/Day-Hospital Clinic of the Neonatology Unit (PIFC) (see Chapter 3 – Section 3.1. Development and validation of the SpeNK-Q Questionnaire). After its successful validation, SpeNK-Q was **administered** to parents enrolled on SpeNK Project at 8-15 months after hospital discharge of children and results were analyzed to identify differences in continuity of care according to parents' perspective (see Chapter 3 – Section 3.2. Administration of SpeNK-Q to families recruited in SpeNK Project).

1.5.3.2. Activities of FPs

FPs who were in charge of children recruited to SpeNK Project were invited to participate in the study, during 9 months of follow-up after hospital discharge of each subject. In order to collect information about patients (e.g. complexity level) and encounters (e.g. time spent, patients' needs, etc.), they had to complete at each activity for the patient an 8-item questionnaire (SpeNK-FP). developed by the Methodology Unit by adapting the Care Coordination Management Tool (CCMT$^{©}$) by Antonelli et al. (2008) to Italian organizational context (see Appendix - SpeNK-FP) (see "Chapter 4. The role of Family Pediatrician in care coordination").

1.5.3.3. Administrative data

At the end of follow-up, data collected for each subject at recruitment were linked to the administrative data extracted from Regional Healthcare System databases. Administrative data included hospital discharge records (SDO), specialty outpatient services (ASA), emergency room contacts (PS), home care services (ADI), certificates of birth attendance (CeDAP), death certificates (REM). Data were extracted referring to a 6-9 months period after the hospital discharge and analyzed to assess use of healthcare services and resources by the population of interest. Moreover, the Methodology Unit developed an algorithm (SpeNK-A) to identify children "with special healthcare needs" referring to data included in the hospital discharge records (see "Chapter 5. Utilization of healthcare resources").

1.6. Main results

1.6.1. Review of procedures and practices

The review of existing procedures implemented at University Hospital of Bologna and Local Health Authorities of Bologna and Imola, allowed to identify the "sheltered discharge" as a specific hospital procedure for children with complex social and/or healthcare needs aiming to promote the integration of healthcare services in the transition from hospital to home. This procedure encompasses the activation of community services and primary care providers who take care of the child after hospital discharge.

At formal level, in 2003 the Local Health Authority of Imola implemented the procedure "*Sheltered Discharges - Organizational process*", replaced in 2012 with "*Hospital-Community Continuity: Sheltered Discharge*". In 2010, St.Orsola Malpighi University Hospital of Bologna and Bologna Local Health Authority implemented an inter-agency procedure for "*Assistance for pregnant women with emotional disorders in first year of a child's life. Assistance for women with prevalent social risk. Sheltered Discharge of Newborns with social or health issues*". The Protocol of Neonatology

Units of Maggiore Hospital (Local Health Authority) and St.Orsola Malpighi University Hospital was implemented in 2002 and has been officially included in the 2010 Procedure. In 2013 the Local Health Authority of Bologna implemented the PDTA (i.e. *Percorso Diagnostico Terapeutico Assistenziale*, or Diagnostic Therapeutic Healthcare Pathway) for the management of children with chronic conditions at risk of developing multiple disabilities.

On the whole, these procedures aim to define, formalize and regulate the collaboration among services, organizations and providers. Through this review, different practices and procedures with different level of standardization and formalization were identified in Bologna province for the management of the hospital discharge of children with special health care needs.

1.6.2. Characteristics of the sample

At the end of a 24-months period, a sample of 82 children was recruited, including a majority of preterm newborns with birth weight lower than 1000 gr. (42.7%) or children with other complicating conditions (12.2%). Children were male in 53.7% (n=44) of cases, had a mean age of 6.2 (±13.6, range 0-78) months at the time of recruitment, and in 78% of cases (n=64) were recruited within 28 days of age for an hospital admission related to birth event. A sheltered hospital discharge was attended to for about 65% of children (see “Chapter 5. Utilization of healthcare resources”).

1.6.3. Family’s perspectives about continuity of care

1.6.3.1. The SpeNK-I Qualitative Study

SpeNK-I involved 16 families who participated in face-to-face and phone interviews and in a focus group, within 1-6 months after children’s hospital discharge. Analysis of parents’ narratives allowed to identify emergent themes referring to informational, management and relational continuity of care (Haggerty et al., 2003), with different key elements during hospitalization, at discharge and after discharge. Moreover, empowerment emerged as essential to help parents cope with transition from hospital to home. Parents expressed different perceptions about FP regarding his/her centrality in the activation and coordination of healthcare network and exhibited different attitudes towards involvement in decision making (see “Chapter 2. A qualitative study on families’ perspectives about continuity of care”).

1.6.3.2. The SpeNK-Q Questionnaire

In SpeNK-Q validation study, 101 questionnaires were completed by parents of preterm newborns, accessing to the Preterm Infant Follow-up/Day-Hospital Clinic of the Neonatology Unit (PIFC) at St.

Orsola Malpighi University Hospital of Bologna. Principal Component Analysis of questionnaires allowed to identify 5 factors explaining 60.2% of item variance: informational continuity; coordination of care; continuity of family-pediatrician relationship; family support; information on care plan. Lower levels of continuity concerned the role of the main coordinator and the informational continuity. SpeNK-Q was found to be a psychometrically promising instrument, with a potential for use in pediatric population with special healthcare needs, to evaluate continuity of care (see Chapter 3 – Section 3.1. Development and validation of the SpeNK-Q Questionnaire).

SpeNK-Q was then administered to parents of 67 children enrolled in SpeNK Project. Item responses indicated high levels of continuity of care (mean=4.3), with lower rates about the informational continuity and the maintenance of contacts of main care coordinator with parents about care received by the child from others. Responses to open-ended questions indicated a health care network as taking care of the most of child's care in over 40% of cases, with a central role of the FPs, whereas the main coordinator was mainly represented by the hospital (65.7%) (see Chapter 3 – Section 3.2. Administration of SpeNK-Q to families recruited in SpeNK Project).

1.6.4. FPs and care coordination

Forty FPs completed 382 questionnaires for 49 patients. The majority of patients (71.4%) were patients with special health care needs, without complicating familiar or social issues. The focus of encounter included in the majority of cases clinical issues. FPs reported "no need for care coordination" in more than 40% of records about patient's needs requiring care coordination. In 51.8% of cases the FP was alone in performing the care coordination activity, and in 25% of cases in collaboration with another clinician. Activities implemented to meet the patient's needs included contacts with contacts with healthcare professionals and services in more than half of cases. According to FPs' subjective appraisal, 79.9% of encounters prevented an inappropriate services use. In general, the study shows some difficulty for FPs to record their activities and their improvement potential as care coordinators for children with special health care needs (see "Chapter 4. The role of Family Pediatrician in care coordination").

1.6.5. Utilization of healthcare resources

The analysis of administrative data showed a higher level of utilization of health services compared with pediatric population (SpeNK newborns with birth weight < 1000 gr. vs. other newborns admitted in the three hospital facilities in the recruitment period), including a significant higher number of hospital days for readmissions, a more frequent use of specialty outpatient services for diagnostics, therapy and visits and almost no difference in the number of emergency room contacts. The SpeNK-A algorithm allowed to classify 16.5% of children admitted at birth and discharged from the regional hospital facilities in the interval of interest as having “special health care needs”, referring to 6 groups: (1) Newborns with birth weight lower than 1000 gr., (2) Conditions associated with the extreme prematurity of newborn (e.g. encephalopathy, NEC), (3) Malformations and other congenital diseases (e.g. heart disease, renal abnormalities with organ failure), (4) Irreversible diseases (e.g. cerebral palsy, disability for brain damage and / or bone marrow), (5) Neurological and metabolic degenerative diseases, (6), neoplasms (see “Chapter 5. Utilization of healthcare resources”).

Chapter 2. A qualitative study on families' perspectives about continuity of care

This chapter replicates the paper "*Continuity of care in children with special healthcare needs: a qualitative study of family's perspectives*" by Zanello et al. (2015), as part of SpeNK Project.

Abstract

Background, to explore parents' experiences and perceptions on informational, management and relational continuity of care for children with special health care needs from hospitalization to the first months after discharge to the home. **Methods,** semi-structured interviews and a focus group were carried out to capture parents' experiences and perceptions. Transcripts were analyzed using a directed approach to the qualitative content analysis. **Results,** 16 families participated to this study: 13 were involved in interviews (10 face-to-face and 3 by phone) and 3 in a focus group, within 1-6 months after discharge from the University Hospital of Bologna (S.Orsola Malpighi) and from hospitals of Bologna Province. To parents of children with special health care needs, the three domains of continuity of care were relevant in a whole but with different key elements during hospitalization, at discharge and after discharge. Moreover, empowerment emerged from parents' narratives as essential to help parents cope with the transition from the hospital setting to the new responsibilities connected with the home care of their child. Parent's perceptions about the family pediatrician concerned his/her centrality in the activation and coordination of the healthcare network. Moreover, parents exhibited different attitudes towards involvement in decision making: some wished and expected to be involved, others preferred not to be involved. **Conclusions,** care coordination for children with special care needs is a complex process that need to be attended to during the hospitalization phase and after discharge to the community. The findings of this study may contribute to elucidating the perceptions and experiences of parents with children with special health care needs about the continuity of care from hospital to community care.

Keywords: *continuity of care, children with special health care needs, hospitalization, discharge, community, qualitative study, empowerment, patient engagement, parents' perspective, interview.*

2.1. Background

Children with special health care needs were defined by the Maternal and Child Health Bureau as those “who have a chronic physical, developmental, behavioral, or emotional conditions and who also require health and related services of a type or amount beyond that generally required by children” (McPherson et al., 1998).

Although this population represents a category with a low prevalence and incidence, a large proportion of them require long-term treatments, including both inpatient and outpatient health care, with high economic impact on the healthcare system.

Medical care for children with special healthcare needs often requires a variety of services, providers and programs to implement complex care plans. Compared with adults, for children with complex chronic conditions the developmental status and the critical mediating role played by parents in the interaction between the child and the healthcare services and providers must be taken into account (Miller et al., 2009). Strategies to connect patients, families, and providers with services and resources are needed to support coordinated, continuous care (Taylor et al., 2013).

Continuity of care represents a key issue for children with special healthcare needs. According to the American Academy of Pediatrics, comprehensive healthcare should ensure *inter alia* continuity, providing care over an extended period of time and planning and organizing transitions, including those to other pediatric providers or into the adult health care system, with the child and family (American Academy of Pediatrics, 2002). In the conceptualization developed by Reid and Haggerty, three types of continuity of care can be identified across healthcare settings, i.e. informational, management and relational (Haggerty et al., 2012; Reid et al., 2002).

However, to date little attention has been paid to the perspectives of families of children with special healthcare needs (Bellin, Osteen, Heffernan, Levy, & Snyder-Vogel, 2011; Diaz-Caneja, Gledhill, Weaver, Nadel, & Garralda, 2005; Knapp, Madden, Sloyer, & Shenkman, 2012). To our knowledge, only a qualitative study was conducted by Miller et al. (2009) about continuity of care in which parents were interviewed to explore the extent to which their experiences and perceptions fit with the academic and service providers’ perspectives.

In Italy, information on prevalence of this condition and on services and resources activated for this population is scanty. Discharge procedures and care plans for children with special healthcare needs have been defined in some areas, with the aim to ensure comprehensive care and avoid the risk of fragmentation. In this context the family pediatrician plays an essential role as primary care provider for children up to 16 years of age and should ensure the coordination and continuity of care among healthcare services and providers. In Italy, where universal health care is provided, financed by the government through tax payments, pediatric primary health care is provided by family pediatricians that are remunerated on a capitation basis; they are in charge of providing care and assess patients' needs, order diagnostic procedures, prescribe drugs, and refer patients to specialists and hospitals (Luciano et al., 2014). Thus, they act as 'gatekeepers' for the system.

Still, research supporting the content and use of care plans for children with chronic diseases and the family pediatrician's role is limited as well as research about the family's perspective lacks.

In Emilia-Romagna Region the SpeNK Project (Special Needs Kids) has been designed to describe the implementation of existing sheltered hospital discharge procedures and integrated clinical pathways for children with complex or chronic health conditions and special healthcare needs and to assess the family's perspective on continuity of care and the role of family pediatrician.

Family perspectives on continuity of care have been explored with a qualitative research approach, referring to Reid and Haggerty's constructs of informational, management and relational continuity of care (Haggerty et al., 2003; Reid et al., 2002) as conceptual basis, and using semi-structured methods. The aim of the present study is to examine the perceptions and experiences of families of children with special health care needs about these three constructs.

2.2. Materials and Methods

2.2.1. Study design

This qualitative study is part of the SpeNK Project. Children and families were recruited for SpeNK Project at hospital discharge of the child from the participating hospital facilities at the University Hospital of Bologna (S.Orsola Malpighi) and the two local Health Authorities of Bologna and Imola. The recruitment of children was conducted from October 1st 2012 to September 30th 2014 on incident cases meeting the following inclusion criteria: age from 0 to 16 years, residence in Bologna province, and the presence of at least one of the following conditions:

- Birth weight <1000 g;
- Complex and/or chronic health conditions defined as:
  - Need for technological assistance,
  - Acute neurological deficit,
  - Severe endocrinopathy,
  - Complex genetic malformative pathology;

- Children with oncological diseases who need palliative care or particular community care;
- Newborns with mothers in contact with mental health services or drug addiction

Only first ever hospitalizations for the condition of interest were included.

Written informed consent was obtained at recruitment from each parent to collect clinical data on children and to contact them during follow-up period (9 months from hospital discharge).

We selected the first families recruited in SpeNK Project according to a maximum variation sampling method (Hsieh & Shannon, 2005) with regard to the child's diagnosis and the hospital of discharge and excluding parents with an inadequate level of knowledge of Italian language. We contacted one of the parents by phone and invited both, whenever possible, to participate in interviews or focus group.

2.2.2. SpeNK-I Interview and Focus Group

A semi-structured interview (SpeNK-I) was developed by the authors (see Appendix - SpeNK-I). A selection of items, questions and probes was picked out from the international literature to explore a number of aspects of child care which are particularly relevant in continuity of care (Haggerty et al., 2003; Miller et al., 2009). Specifically, we examined parents' experiences and perceptions about their child's clinical condition and care plan (knowledge, communication, shared information and shared decision making), about service providers and clinicians involved in child's care (who, how and why were involved, their availability and their information exchange) to explore relational and informational continuity, and about management continuity of care within the network of hospital and community service providers and clinicians. The SpeNK-I was administered face-to-face or by phone at 1-6 months after discharge, lasted 60 minutes on average and was audiotape-recorded in the majority of cases. Where it was not possible (phone interviews), responses were documented in writing. The SpeNK-I was then used to define the topics to be discussed in a focus group with parents, including (1) discharge, (2) coordination of care and overall organization of care provided both by hospital services and by community ones, (3) communication and shared information and decision making, (4) empowerment and proactive care received for their child's care management during the hospital stay and after discharge. The focus group lasted 150 minutes and was audiotape-recorded. All audio tapes were transcribed verbatim for analysis.

2.2.3. Analysis

The semi-structured interviews were analyzed using a directed approach to the qualitative content analysis, as described by Hsieh and Shannon (2005). Qualitative content analysis is a research method for the subjective interpretation of the content of text data through the systematic classification process of coding and identifying themes or patterns (Hsieh & Shannon, 2005). In this study the directed approach to qualitative content analysis was chosen and used to validate or extend conceptually the theoretical framework of continuity of care.

The transcripts were read several times and all text related to the parents' experiences and perceptions about the continuity of care was highlighted. Based on operational definitions of the three types of continuity of care (Haggerty et al., 2003; Reid et al., 2002), category codes were defined *a priori* and applied to the relevant text. *Informational continuity of care* addresses "the use of information on past events and personal circumstances to make current care appropriate for each individual" among providers and among healthcare events. *Management continuity of care* refers to "a consistent and coherent approach to the management of a health condition that is responsive to a patient's changing needs", which is especially important in chronic or clinically complex diseases. *Relational continuity of care* addresses "an ongoing therapeutic relationship between a patient and one or more providers", which bridges past to current care and provides a link to future care (Haggerty et al., 2003).

All text that could not be coded within the predetermined coding scheme was identified and analyzed later for the attribution to a new category or a subcategory of an existing code. Therefore, some codes were developed inductively given their repeated appearance in the parents' narratives. The entire process of reading and classification was conducted jointly by two investigators of SpeNK Team, and in case of disagreement the research team met to discuss the coding scheme and the attribution of issues to categories.

2.3. Results

2.3.1. Participants

Sixteen families (i.e. 15 mothers and 8 fathers) of 17 children participated in the study.

The characteristics of parents and of children are shown in Table 2.

Table 2 – SpeNK-I: Characteristics of participants (n=16) and children (n=17)

Study participants (n=16); n (%)	Mother only Father only Both parents	8 (50.0 %) 1 (6.2%) 7 (43.8%)
Parental citizenship; n (%)	Both Italian parents One Italian parent No Italian parents	6 (37.5%) 3 (18.8%) 7 (43.7%)

Gender of children (n=17); n (%)	Male Female	8 (47.1%) 9 (52.9%)
Age of children, months		Mean=14.5, SD=16.6; Median=9.1; Range: 3.3-67.9
Time from discharge, months		Mean=3.8, SD=2.4; Median=3.5; Range: 0.7-8.3
Children's diagnosed health condition at discharge; n (%)	Prematurity < 1000 gr Encephalopathy Hydrocephalus Myopathy Malformation	9 (52.9%) 5 (29.4%) 1 (5.9%) 1 (5.9%) 1 (5.9%)
Hospital Unit; n (%)	Pediatric and Nursery Unit LHA Imola Neonatology and NICU LHA Bologna Pediatric Unit LHA Bologna Pediatric Surgery Unit UH Bologna Neonatology Unit UH Bologna Pediatric Emergency Room UH Bologna	2 (11.8%) 1 (5.9%) 3 (17.6%) 3 (17.6%) 7 (41.2%) 1 (5.9%)

Thirteen families were involved in interviews: most of them (10/13) were interviewed in their homes, while three were interviewed by phone at their request. The person interviewed was the mother in 6 cases, the father in one case and both in 6 cases. In 8 families at least one parent was immigrant. Three families (i.e. 3 mothers and 1 father) participated to the focus group and were represented by both parents in one case (both immigrant) and by the mother in the other two cases (Italian).

The interviews and the focus group were conducted at a median time of 3 months (range 1-11 months) from hospital discharge.

The median age of children was 7 months (range 1-68 months); 9/17 (52.9%) were preterm, 5/17 (29.4%) had a diagnosis of encephalopathy, 1/17 (5.9%) of hydrocephalus, 1/17 (5.9%) of myopathy, 1/17 (5.9%) of malformation.

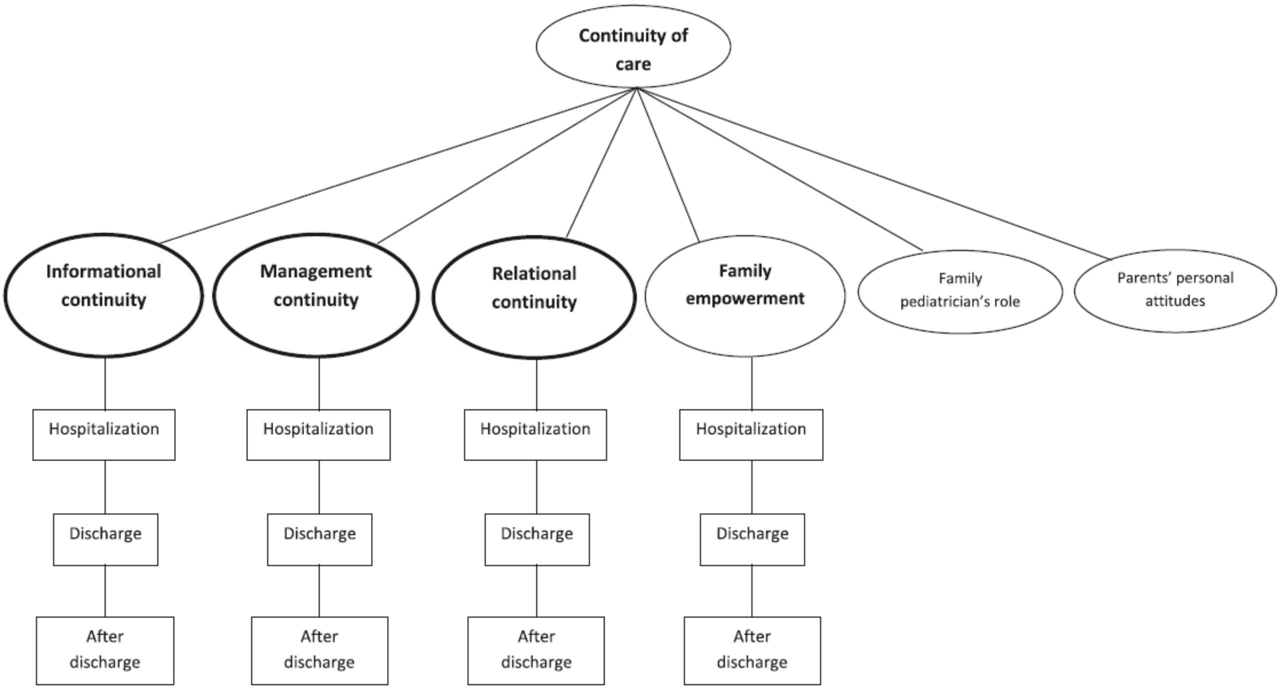
11/17 (64.7%) children were recruited at discharge from the University Hospital of Bologna, 4/17 (23.5%) from the hospital of Bologna Local Health Authority and 2/17 (11.8%) from the hospital of Imola Local Health Authority (Table 1).

2.3.2. Themes

The three categories of informational, relational, and management continuity of care, developed *a priori*, were confirmed by parents' narratives. Moreover, the family empowerment was detected as new theme. Within these four major themes, three different phases were discernible (i.e. hospitalization, discharge, after discharge). Furthermore, two more themes, not *a priori* defined, were detected in parents' narratives, referring to the role of family pediatrician and to the parents' different attitudes about the wished level of involvement in decision making and information exchange. Figure 3 provides a visual representation of the themes and the phases aforementioned.

Figure 3 - SpeNK-I: Continuity of care for children with special health care needs in parents' narratives.

The bold boxes represents the three a priori themes based on the literature, the other boxes represent the new emerged themes.



Informational continuity

Communication and exchange of information represented critical functions of the interacting system of parents, child and clinicians, necessary for the informational continuity of care.

The hospitalization

Especially during the hospitalization, most of parents perceived the information exchange between them and clinicians and among clinicians or services as essential.

> *"During hospitalization, information arrived or did not arrive. [Interview #13]*
>
> *"There's an efficient communication network, it works and sometimes it works too much: they know things before you do." [Interview #08]*

> *"If the parent, as it should be, is always or often there, he/she can help the nurse and vice versa." [Interview #01]*

Explaining clinical and medical issues in an accessible way and with a comprehensible language to unskilled people, influenced significantly the understanding and learning possibilities of half of all parents about their child's care.

> *"The information is always… I mean ... relative. You are completely unskilled and what you are told, are empty words, almost, they are not yet meaningful" "The recommendations are more prescriptive than descriptive" "The skilled people do not realize that, as unskilled person, you need to know the reason why you have to do something, because in that way, if nothing else, you internalize it" [Interview #08]*

The discharge

In few cases, the Discharge Letter filled by the hospital represented an essential information tool both for professionals and for parents in the transition from hospital to the home. For professionals it was a key element of the informational continuity of care because it ensured the information exchange among clinicians and services. For parents it represented a double-edged sword because it provided them with detailed medical information about the child's hospitalization history, but was a source of concern if not well explained and understood.

> *"What I didn't like, was to find out many thing I didn't know about my child from the discharge letter. It bothered us, and increased our fears, too." [Interview #07]*

After discharge

After discharge, the parents' experiences on the informational continuity of care in the healthcare pathway varied. In most cases, it was up to parents to ensure the informational continuity across health care settings and service providers, physically carrying medical reports, or orally communicating updates about child's conditions.

> *"The letter of discharge… they gave it to us at the hospital, but none of the wards could access it (from the computer), so we always had to bring a printed copy for them" [Interview #02]*

Management continuity

Elements of management (dis)continuity of care in parents' narratives were related to the consistency of the care management by different providers and to the adaptation to child's needs.

The hospitalization

During hospitalization, half of all parents perceived clearly the level of coherence and variability in the management of their child's care among hospital services and also within the same service staff.

> *"The team of nurses (…) The team of doctors (…) They work the same way, in a team, and I liked it a lot" [Interview #06]*

> *“They don’t have a common method, everyone does his/her own way [Interview #07].*

The discharge

The sheltered discharge procedure almost every time involved the family pediatrician, community services and parents in a meeting taking place in the hospital about a few days before discharge. It promoted the perception of being included in a care plan, within a network of health professionals, and in most cases ensured the continuity of care also by providing parents with all the contact details of the hospital, for any further need (information, doubts, worries, etc.).

> *“We got all the contact information: they gave us a beautiful discharge folder, where you find all the numbers, contacts details of all the physicians of the Ward” [Interview #01]*

After discharge

The perception of management continuity was related in most cases to the presence or absence of a comprehensive care plan and in few cases also to the level of coherence between clinicians’ methods and protocols.

> *“We were given a prospect of first steps to do, an agenda. (…) There's the follow-up of preterm newborns, then the pediatrician, the neurologist, the psychotherapist, they follow up us because we are under the kilo.” [Interview #07]*
>
> *“The “post” (discharge) is a no man’s land” [FG]*
>
> *“The problem is that everyone [every clinician] has his/her own way to do things. And now the community nurse has another method too (different from what we were used to during hospitalization).” [Interview #02]*

Relational continuity

Elements of relational continuity of care appeared in the parents’ narratives relating to the maintenance over time of an ongoing therapeutic relationship between the child and parents, and the health care providers.

The hospitalization

In the hospital setting, the continuity of the relationship between parents/child and care providers in the majority of cases related to the perception of familiarity, constant support and humanity of the professionals involved, essential for both children and parents.

> *“The nurses are very sweet, kind, more than their job would require. They get to you heart… They do their work from the heart. There, they are all my child’s angels (…) They do not only ask me about my child, but also about me. They ask me: “how are you”?” [Interview #04]*
>
> *“I had a good time there, with the head physician, the nurses, all the staff… the social health care operators were almost like family. After that, you consider them as relatives.”[Interview #11]*

The discharge

To a quarter of all parents, the transition from the hospital facility to home entailed a change in the relationships with professionals and service providers, and in the responsibilities connected to the child’s care.

> *“I think that it is important to make parents feel that they’re are supported in the transition. We had the first meeting with all those professionals and we thought: “That’s great! Finally, they will really accompany us all the way”. Then, that’s not true. Probably the organization of the health care system doesn’t allow either a real communication or a well organized network making families feel supported, especially families like ours, living so far away” [Interview #08]*
>
> *“All the mothers going home, after being many months at the hospital feel bad and scared. At home you’re 100% responsible: if something happens, what do I do?” [Interview #07]*

After discharge

When the network of hospital and community professionals works, it has an important function of reference point after discharge in most cases, for the follow-up visits and for any doubts or worries.

> *“I always had the perception of being followed up (…) I always knew to whom I had to refer for specific problems.” [Interview #11]*

Family empowerment

In the narratives of all the parents, the “empowerment” emerged as characterizing their experiences and perceptions in the interaction with the health care system. The family empowerment appeared as a process aiming to raise the parents’ ability to care their children, started during hospitalization, with the information and training provided by professionals.

The hospitalization

Half of all parents reported that during hospitalization they were involved in a specific training to manage every day and special needs of their child. The parents acknowledged the importance of being provided with adequate training and information, not only for the care activities but also for the relationship with their child.

> *“In the hospital you learn (…) They teach you a lot of things, so when you are at home, you can manage you baby” [Interview #05]*

The discharge

In the experiences of one third of parents, the discharge appeared well planned and organized, with a specific focus on their training about their child’s care management at home.

> *“From 20 days prior the hospital discharge, the nurses started to train us on practices and operations to do on the child with the medical devices” [Interview #02]*

After discharge

The empowerment of parents continued in half of cases involving them personally in the child's care (e.g. daily physiotherapy exercises) after discharge.

> *"With the physiotherapy, we are all involved (…) The large part (of rehabilitation activity) is done by us (…) The training she does in one hour of physiotherapy, doesn't stop there." [Interview #08]*

The family pediatrician's role

The role played by the family pediatrician raised mixed perceptions in parents. In few cases the pediatrician was a rarely seen and contacted person. In most cases, he/she played a pivotal mediating and coordinating role, activating the healthcare network or taking into account clinical aspects important to the parents and not included in the care plan.

> *"The (family) pediatrician really made me smile. Before the hospitalization we didn't see her. Now she makes home visits. In the previous 4 years, where was she?" [Interview #09]*
>
> *"The (family) pediatrician is very present; whenever we need her , we call and she comes. (…)." [Interview #12]*
>
> *"The family pediatrician does not coordinate our child's care. We coordinate everything […]. It. We prefer to do so. We want to care for our child." [Interview #12]*

Parents' personal attitudes

In all cases, parents perceived differently the elements of (dis)continuity of care, based on their preferences about the wished level of information and involvement in decision making, during hospitalization, or of coordination in the healthcare management of their children, after discharge.

> *"We are not physician, but we want do make decisions." [Interview #12]*
>
> *We didn't ask much because we didn't know what to ask" [Interview #03]*

2.4. Discussion

This study is the first Italian study to explore parents' perspective on the continuity of care for children with special health care needs during hospital stay, at discharge and in the first months after discharge.

Our results suggest that continuity of care issues varies from experiences related to interactions with a single professional and service during hospitalization to a global perception of being included in a comprehensive care plan within an integrated network of healthcare professionals and services, at discharge and after discharge.

Informational continuity during hospitalization concerns, in the majority of cases, the information exchange among parents, professionals and services. Compared to the after discharge experience, the hospital admission and stay emerged as the dominant theme in parents' narratives, probably because it represents a tough experience, involving the whole family and the sick child. The hospitalization is an event characterized by high uncertainty, low predictability and high level of distress and emotional burden for families (Ames, Rennick, & Baillargeon, 2011; Haggerty et al., 2003; Latour et al., 2011). We found that communication skills of professionals are particularly relevant in determining the outcome of the information flow in such situation. In contrast, the parents' perceptions and experiences about the interaction and information exchange with the healthcare system at discharge and after discharge referred mainly to the availability of written documents for sharing medical information and to their commitment in transferring the information.

The **relational continuity** is expressed as a perception of familiarity and support, mainly during the hospitalization, when it refers to the "human" quality of relationships with the staff, whereas at discharge and after discharge it refers to the activation of a network of service providers and clinicians as reference points for the family and the child's care.

The **management continuity** was perceived in terms of coherence and adaptability of the care provided to the child by multiple professionals and services within the hospital facility, whereas at discharge and after discharge referred mainly to the definition of a shared care plan with different professionals. Miller and colleagues found that compartmentalization (i.e. management discontinuity) was more likely among teams working in different settings and service sectors in parents' narratives about the children with complex chronic health conditions (Miller et al., 2009). On the contrary, we found that elements of management discontinuity of care occur also within the same healthcare setting, and this can be due to the lack of communication among the staff members.

These three conceptual categories of continuity of care in adult patients with chronic disease proved to be useful to describe parents' experiences of children with special healthcare needs. However, several conceptual overlaps can be found in the narratives of parents and in many cases informational, relational and management continuity are not easily discernible and sometimes redundant. This indicates that the continuity of care is a complex theoretical construct that requires further investigation through qualitative and quantitative studies.

Concerning parents' education and training about their own child's care, we found that the **empowerment** process aiming to provide specific skills to manage the child both during hospitalization and at home is another theme extremely relevant from parents' perspective. These particular activities should be scheduled and standardized with the special commitment of a component of the staff. When scheduled, this process was started by the hospital professionals shortly before the discharge and supported by the community professionals after discharge. These findings indicated that parents' empowerment is an important issue when dealing with special healthcare needs children, throughout the entire process of care, form hospital to home. Special efforts should be made to achieve an effective alliance with parents and families in order to sustain and ameliorate children care.

In our study we also found two other relevant themes about the continuity of care: family pediatrician's role and parents' personal attitudes.

The **role of family pediatricians** varied a lot in the ways and in the extent of their participation in the child's care after discharge, from a key person with a pivotal mediating and coordinating role in a network of clinicians and service providers, to a rarely seen and contacted person. These perceived differences could suggest a lack of clarity about the role of family pediatrician in ensuring the continuity of care for children with special healthcare needs. However, these differences could be ascribed to a different level of preparation or familiarity of the family pediatrician to care for children with various conditions. To our knowledge, only one study, investigating the willingness and ability of pediatricians to accept children and youth with special healthcare needs into their practices, was conducted in the U.S. by Agrawal et al. (2013). The results indicated that pediatricians do not feel prepared to care for all types of conditions and this problem reduces the ability to implement effective medical home care.

Our findings also indicated that parents reported different perceptions and experiences of continuity of care according to their **preferences, attitudes and behaviors**. In general, we found that the perception of a positive outcome or improvement of the child's condition affected significantly parents' satisfaction and appeared to reduce their willingness to express any criticism and to identify any unmet need. Furthermore, parents reported different preferences about the level of information and shared decision making especially during hospitalization, whereby someone wished to be asked for, involved and informed about any treatment provided to the child, someone else felt to have neither the competences nor the role to participate in decision making, and felt not to be able to understand medical issues. This specific finding suggests that, as for other medical fields (i.e. oncology), the willingness and capability to be informed and involved should be tested (Colville et al., 2009; Davidson, Brundage, & Feldman-Stewart, 1999). The engagement of the patient as care partner was identified by Haggerty et al. (2012) as an independent dimension emerging from the factor analysis of a generic measure of continuity of care in adult patients. In a study conducted by Stille et al. (2013), parent partnership in communication and decision making about subspecialty referrals for children with special needs was endorsed both by parents and clinicians, “though relatively greater enthusiasm from parents may signal the need for work in implementing this partnership”. The use of a care plan could be helpful to support parent engagement and build a partnership between parents and clinicians.

The literature about the overall **parental experience during children hospitalization** reports themes and issues similar to those emerged from our study. Interviewing parents with children admitted to 8 PICUs (Pediatric Intensive Care Units) in university medical centers in the Netherlands, Latour et al. (2011) identified some major themes, most of which recurring also in our interviews: coordination of care, information management, parent participation, attitude of professionals, emotional intensity. Another qualitative study in a tertiary care Canadian university affiliated hospital’s PICU identified three dimensions of the parental role perceived by parents, including being present and participating in the child’s care, forming a partnership of trust with the PICU health care team, and being informed of the child’s progress and treatment plan (Ames et al., 2011). Similarly to our findings, significant themes including the vividness of parents’ memories of admission, the intensity of distress associated with times of transition and the lasting impact of the experience were reported by Colville et al. (2009) in a study assessing the impact on parents of a child’s admission to intensive care in an English teaching hospital’s PICU. Similarly, a systematic review by Cleveland (2008) identified the following six needs for parents with an infant in the Neonatal Intensive Care Unit (NICU): (a) accurate information and inclusion in the infant’s care, (b) vigilant watching-over and protecting the infant, (c) contact with the infant, (d) being positively perceived by the nursery staff, (e) individualized care, (f) a therapeutic relationship with the nursing staff. Moreover, four nursing behaviors were identified as meeting parents’ needs: (a) emotional support, (b) parent empowerment, (c) a welcoming environment with supportive unit policies, and (d) parent education with an opportunity to practice new skills through guided participation (Cleveland, 2008).

Given these evidences, we can conclude that parents’ experiences and perceptions about intensive care admission of their children are similar across different geographical, cultural, and organizational contexts.

These results should be interpreted keeping in mind some **limitations**. First, this study reflected the experiences of a small number of participants, all from the same district (i.e. Bologna province) though referring to three different hospital facilities of discharge (i.e. University Hospital of Bologna, Bologna Local Health Authority hospital, Imola Local Health Authority hospital). This may limit the generalizability of findings to other contexts with different service organizations in other healthcare systems. Second, this study excluded non-Italian speaking parents. Compared to Italian speaking parents, they could have different expectations and report different experiences about the interaction with the healthcare system, related to different cultural backgrounds and to difficulties in the language comprehension.

2.5. Conclusion

In summary, the findings of this study suggest that a continuous and coordinated care should be targeted to the treatment phase (hospital vs. community), to take into account children's changing needs. Moreover, the information provided and shared decision making in the healthcare services should be personalized according to the preferences of patients/families. The development of easy-to-use instruments measuring the preferred level of engagement could help to improve the quality of healthcare services.

The findings of this study contribute to deeper understanding the complexity of perceptions and experiences of parents with children with special health care needs about the continuity of care from hospital admission to home care, given the sparse available evidence on these themes. In particular, these findings may provide knowledge to clinicians and providers working with special health care needs children, and to policy makers in order to redesign services and to improve the quality of the care provided. The involvement of patients as co-designer of healthcare services has been recently promoted by the NHS Institute for Innovation and Improvement in the UK using the "Experience Based Design" approach, a new way of bringing patients and staff together to share the role of improving care and re-designing services (Carr, Sangiorgi, Buscher, Junginger, & Cooper, 2011).

Further research is needed to examine the generalizability and transferability of our results to clinical practice and to deeper understand the role that the family pediatrician should play in coordinating and ensuring the continuity of care for children with special health care needs. Moreover, the issues raised by this study may provide the background for developing self-report instruments to assess continuity of care for children with special health care needs from parents' perspective, in order to improve and promote family-centered care.

Chapter 3. Measuring parents' perspectives on continuity of care

3.1. Development and validation of the SpeNK-Q Questionnaire

This section replicates the paper "*Measuring parents' perspective on continuity of care in children with special health care needs*" published by Rucci et al. (2015), as part of SpeNK Project.

Abstract

Introduction. Children with special health care needs are an exponentially growing population needing integrated health care programs that involve primary, community, hospital and tertiary care services. The aims of the study are (1) to develop and validate the Special Needs Kids Questionnaire (SpeNK-Q) designed to measure parents' perspective on continuity of care for children with special healthcare needs and (2) to evaluate the continuity of care based on parental experiences in this population. **Methods**. SpeNK-Q was derived from a previous qualitative study and was based on Haggerty's constructs of informational, management and relational continuity. Parents of preterm birth children completed the SpeNK-Q 20 item questionnaire at the second or subsequent planned follow-up visit after the child's hospital discharge. Principal component analysis (PCA) was used to examine the structure of the instrument. **Results**. PCA of 101 questionnaires administered allowed us to identify 5 factors explaining 60.2% of item variance: informational continuity; coordination of care; continuity of family-pediatrician relationship; family support; information on care plan. **Conclusions and discussion**. SpeNK-Q proved to be a psychometrically promising instrument. Its utilization could improve the identification of areas for service development, the delivery of coordinated care and support policy makers in redesigning integrated services.

Keywords: *special healthcare needs, children with preterm birth, factor analysis, integrated care, family support, parents experience*.

3.1.1. Background

Children with special health care needs are a highly vulnerable subset of the child population (Newacheck, Kim, Blumberg, & Rising, 2008). According to the definition of the Maternal and Child Health Bureau, children with special health care needs are those who "*have, or are at an increased risk for, a chronic physical, developmental, behavioral, or emotional conditions and who also require health and related services of a type or amount beyond that generally required by children*" (McPherson et al., 1998) (p.138).

The prevalence of non-institutionalized children with special health care needs aged 0-17 has been estimated as 12% in 1999-2000 in U.S. (Davidoff, 2004) and has been growing exponentially in the past decades due to novel treatments in life-threatening pediatric conditions that increase the survival of children with serious congenital or acquired diseases. This success factor within pediatrics has considerable societal costs and important financial and organizational consequences for health care planning (van der Lee et al., 2007).

Low birth weight newborns (<2500 g) constitute about 6% of all newborns (Kowlessar, Jiang, & Steiner, 2006). Very low birth weight (<1500 g) infants are at increased risk of chronic conditions and of poor neurodevelopment and can be considered a specific subgroup of children with special health care needs (McPherson et al., 1998). In high-income countries, progress in medical care has led to improved survival and long-term outcome among preterm infants with very low birth weight, but considerable risks for child health and development remain a matter of concern (Gibertoni et al., 2015; Saigal & Doyle, 2008).

Similar to adult patients, children with chronic or complex health conditions require the implementation and coordination of a variety of health care services and providers at different levels, from primary care to hospital care, over an extended period of time. In this context, continuity of care, meaning the degree to which the patients experience their perceived care over time as coherent (Reid et al., 2002), represents a key element of health care provision. In recent years, continuity of care has received more attention as a result of changes in healthcare systems, due to the increase in patients with chronic and multiple diseases and the increasing complexity of the health care services (Aller et al., 2013). Despite the recognized importance of continuity of health care for patients with chronic or complex conditions, the main research focus has been on adults and elderly, while less attention has been paid to children with special health care needs and their families.

To assess continuity of care for these children, the critical role of parents, mediating between the child's needs and health care services and professionals, must be taken into account (Miller et al., 2009). Parents should be involved in the assessment in order to measure and improve continuity of care for their children. Recently, Patient Reported Experience Measures (PREMs) have garnered attention for measuring experience of patients interacting with an array of professionals and services within a complex healthcare system. PREMs proved to provide more information than patient satisfaction questionnaires by encouraging the users to describe their actual experience of the care received (Whelan P.J., 2011). A review of the instruments measuring continuity of care showed that most available instruments on continuity of care from patients' perspective are designed to assess this construct in specific adult populations and settings, such as patients with diabetes, cancer, mental health problems, previously hospitalized patients, complex and chronic diseases, people being treated in primary care settings or patients in general regardless of morbidity or care setting (Uijen et al., 2012). To our knowledge, only one questionnaire has been developed to measure continuity of care in child population from the family's perspective, but it applies only to mental health care (Tobon et al., 2014).

Recently we performed a qualitative study examining the perceptions and experiences of parents of children with special health care needs while interacting with various health care services and providers (Zanello et al., 2015). Continuity of care was found to be important to parents, and several key elements were useful to develop a quantitative measure of this construct.

The conceptual framework underlying our instrument development refers to the definitions of continuity of care provided by Haggerty and colleagues (2003). This refers to three types of continuity of care. *Informational continuity of care* addresses "the use of information on past events and personal circumstances to make current care appropriate for each individual" (p.1220) among providers and among healthcare events. *Management continuity of care* addresses "a consistent and coherent approach to the management of a health condition that is responsive to a patient's changing needs" (p.1220) which is especially important for in chronic or complex clinical diseases. *Relational continuity of care* refers to "an ongoing therapeutic relationship between a patient and one or more providers" (p.1220) which bridges past to current care and provides a link to future care (Haggerty et al., 2003). Valid measures of continuity of care for children with special healthcare needs must involve parents in order to identify the areas of improvement potential and gaps in care coordination from user's perspective, in a systematic and reliable way. The aims of the present study were to develop and validate an instrument to measure continuity of care for children with special needs from the perspective of parents with preterm infants and to evaluate the continuity of care in the population assessed.

3.1.2. Materials and Methods

Setting

The study was performed at the University Hospital of Bologna (St. Orsola Malpighi) in the Preterm Infant Follow-up/Day-Hospital Clinic of the Neonatology Unit (PIFC). For the preterm infants the Unit activates a standardized follow-up procedure at the Clinic, after hospital discharge. The follow-up procedure includes planned visits from 3 until 42 months of the child's corrected age (every 3 months in the first year, every 6 months in the second year and every 12 months later). Additional visits may be scheduled for any further clinical needs.

Participants

Study participants were recruited from parents of children with preterm birth requiring integrated healthcare programs at the PIFC. Inclusion criteria were: (i) access to the PIFC for the second or subsequent follow-up visit of the child, (ii) adequate level of knowledge of Italian language. The ascertainment of inclusion criteria was made by the PIFC personnel, who invited the eligible parents to participate in the study. Parents at the first follow-up visit (three months of corrected child's age) and parents who were not sufficiently fluent in the Italian language were excluded. All parents meeting inclusion criteria accepted participation in the study.

Eighty-one parents of 101 children with preterm birth were recruited during a 4-month period (November 2013 – March 2014) and completed the questionnaire.

The Ethics Committee of the Bologna University Hospital Authority approved the study procedures and all parents consented to participate in the study.

Instrument development

The Special Needs Kids (SpeNK) questionnaire (SpeNK-Q) was developed in the framework of the SpeNK study (Zanello et al., 2015). The Emilia-Romagna Region SpeNK Project was designed to describe the implementation of existing sheltered hospital discharge procedures and integrated clinical pathways for children with complex or chronic health conditions and special healthcare needs and to assess the family's perspective on continuity of care and the role of family pediatrician. The "sheltered" discharge is a specific hospital procedure for children with complex social and/or healthcare needs that includes the activation of community services and primary care providers, who take care of the child after hospital discharge.

SpeNK-Q was derived from the results of the SpeNK-I qualitative study (Zanello et al., 2015) and was based on Haggerty's constructs of informational, management and relational continuity (Haggerty et al., 2003; Haggerty et al., 2012). In the SpeNK-I study, 16 families of children with special health care needs were interviewed to explore their experiences and perceptions on informational, management and relational continuity of care from hospitalization to the first months after discharge to the home. We found that the three domains of continuity of care were relevant to parents, with different key elements related to the treatment phase (i.e. hospitalization, discharge, after discharge) (Zanello et al., 2015).

The item development of the questionnaire was carried out through several steps. First, we reviewed the literature about measures of continuity of care (Aller et al., 2013) and found that no measures for continuity of care specific for children. Thus, we chose to refer to Haggerty's generic measure of continuity of care and Miller's study (Haggerty et al., 2003; Haggerty et al., 2012; Miller et al., 2009) to generate item statements about continuity of care for children from parents' perspective. Second, we adapted 36 items from Haggerty's generic measure about care received by adult patients to parents' perspective on their child's care and to the Italian healthcare organizational context. Third, we selected the final 20 item statements, by retaining the items which occurred most frequently in parents' narratives about continuity of care in SpeNK-I Study (Zanello et al., 2015). Lastly, we attributed to each of the 20 SpeNK-Q item statements a 5-point response option, to measure agreement or frequency. We decided to use a 5 point Likert-type scale because, using four response categories, people who see both positive and negative aspects of their perceptions would be forced to lean either towards the positive or the negative; "uncertain" would give them an option they feel comfortable with. There is also some evidence that the absence of a mid-point on an importance scale produces distortions in the results obtained. It has been reported that the lack of a mid-point has resulted in more negative ratings than would be achieved when a mid-point was available (Garland, 1991).

The item statements explore parents' perspective about their relationship and interactions with: (1) the family pediatrician (knowledge of the child's medical history, partnership and confidence); (2) the main coordinator (knowledge of the child's health needs, continuity with other providers, services and clinicians); (3) the network of healthcare providers and services involving child care such as care provision, coherence and availability of information, parents' involvement and engagement, knowledge of the child, experiences of receiving advice, and healthcare systems. SpeNK-Q includes two open questions aiming at identifying: (1) the person who is in charge of most of the child’s health; (2) the person who coordinates the child’s health care (i.e. main coordinator: for example, PIFC physician, Family Pediatrician, nurse, etc.). The two questions were used to facilitate understanding of the following items and were not included in the analysis (Appendix - SpeNK-Q Questionnaire).

Statistical analysis

Principal component analysis with orthogonal (varimax) and oblique (promax) rotation was used to analyze the construct validity of the instrument (Tabachnick & Fidell, 2013). Kaiser-Meier-Olkin (KMO) was used to assess the sampling adequacy. The sampling was considered adequate if KMO was higher than 0.5.

The number of questionnaires to be administered was determined in advance as N=100, to ensure a 5:1 subject to item ratio, as recommended for principal component analysis (Gorusch, 1983). We used the child as the unit of analysis.

The number of factors to be extracted was defined by inspecting the scree plot and considering their interpretability and consistency with the criteria that guided the construction of the instrument.

After determining the number of factors, Cronbach’s alpha was calculated for each factor to evaluate the internal consistency. Cronbach’s alpha was assumed to be satisfactory when it was ≥0.70 (Terwee et al., 2007). We computed the factor scores using the regression method (Harman, 1976). These scores are expressed as Z scores (mean=0, Standard Deviation=1) and are an estimate of the score each subject would have on each factor, if it were measured directly.

Because the PCA is based on the assumption that items are continuous variables with a normal distribution and that observations are independent, we took the log-transform of the variables and replicated the PCA using Mplus 7 software, that includes analytic procedures suitable for ordinal-level variables, with a skewed distribution, and for non-independent observations (twins). Factors were estimated using a robust weighted least squares estimator.

Using Mann-Whitney test we assessed the association between clinical characteristics of the children (i.e. clinical complications, birth weight <1500g, intensity of the healthcare services received, parity) and the factor scores of the SpeNK-Q.

To take into account the presence of twins, we also conducted secondary mixed effects analyses in which factors were regressed on children characteristics and children were nested into their family.

We calculated the percentage of parents responding to the answer options ‘disagree and strongly disagree’ or ‘never and sometimes’ in order to identify lower levels of continuity of care according to parents’ perspective.

The significance level was set at $p < 0.05$. IBM SPSS Statistics (version 20, Chicago, USA) and Mplus Version 7 (http://www.statmodel.com) were used for the analyses.

3.1.3. Results

Eighty-one parents of 101 children with preterm birth (including 20 twins) participated in the study. Parents of twins completed one questionnaire for each child. The total number of completed questionnaires was 101. The SpeNK-Q took about 10 minutes to be completed by parents and was acceptable and easy to administer.

Parents’ and children’s characteristics are presented in Table 1. Over half of the parents were mothers, with a mean age of 34.2 (±6.3; range: 18-51) years. Children were female in 52.5% (n=53) of cases, had a mean gestational age of 30.1 (±2.3, range 23.1-35.3) weeks and a mean birth weight of 1280.6 (±352.9, range: 498-2499) grams. At the time of SpeNK-Q administration, children had a mean age of 20.7 (±9.9; range: 6-43) months and had been discharged from the hospital about 19 months before. The questionnaire was completed by parents of 32 (31.7%) children within one year from hospital discharge and by parents of 69 (68.3%) children one year after hospital discharge.

Table 3 – SpeNK-Q Validation: Characteristics of parents (n=81) and infants (n=101)

	Parents (n=81)	N (%)
Respondents	Mother only Father only Both parents Grandparent	44 (54.3%) 10 (12.3%) 26 (32.1%) 1 (1.2%)
Parental Citizenship	Both Italian parents One Italian parent No Italian parents Missing	60 (74.1%) 9 (11.1%) 11 (13.6%) 1 (1.2%)
		Mean (±SD); range
Age	Mother Father	34.2 (±6.3); range: 18-51 37.8 (±7.1); range: 19-55

	Infants (n=101)	N (%)
Infants	Singletons Twins	81 (80.2%) 20 (19.8%)
Gender	Male Female	48 (47.5%) 53 (52.5%)
Clinical complications [a]	Yes	34 (33.7%)
Birth weight <1500g	Yes	82 (81.2%)
Intensity of the healthcare services received [b]	Yes	58 (57.4%)
First-born	Yes	25 (24.8%)
		Mean (±SD); range
Gestational age (weeks)		30.1 (±2.3); range: 23.1-35.3
Birth weight (grams)		1280.6 (±352.9); range: 498-2499
Age (months)		20.7 (±9.9); range: 6-43
Time from discharge (months)		19.1 (±9.8); range: 4-43

[a] At least one complication during the hospitalization at birth

[b] Presence of at least one of the following: sheltered discharge; more than 3 follow-up visits; at least 1 re-hospitalization.

3.1.3.1. Principal Component Analysis

The principal component analysis was carried out with varimax and promax rotation. KMO was 0.64, indicating that the 20 items of the SpeNK-Q were appropriate for principal component analysis. By inspecting the scree plot, a change in the curvature was observed after the 6th factors, suggesting that 6 factors were sufficient to summarize the variance of the items in a parsimonious way and that the subsequent factors were nuisance factors. After comparing the 5 and 6 factor solutions, a 5-factor varimax (orthogonal) solution was selected as the best in terms of interpretability. This solution accounted overall for 60.2% of item variance. The first factor identified was (1) 'informational continuity' that included seven items and accounted for 21.4% of the variance, followed by (2) 'care coordination' with four items accounting for 12.3% of the variance, (3) 'continuity of family pediatrician relationship' with three items accounting for 10.4% of the variance, (4) 'family support' with four items accounting for 8.7% of the variance and (5) 'information on care plan' with two items accounting for 7.4% of the variance. All item loadings were greater than 0.47, except for the item 8 (loading 0.31) (Table 4).

Table 4 - SpeNK-Q Validation: Principal Component Analysis with orthogonal rotation

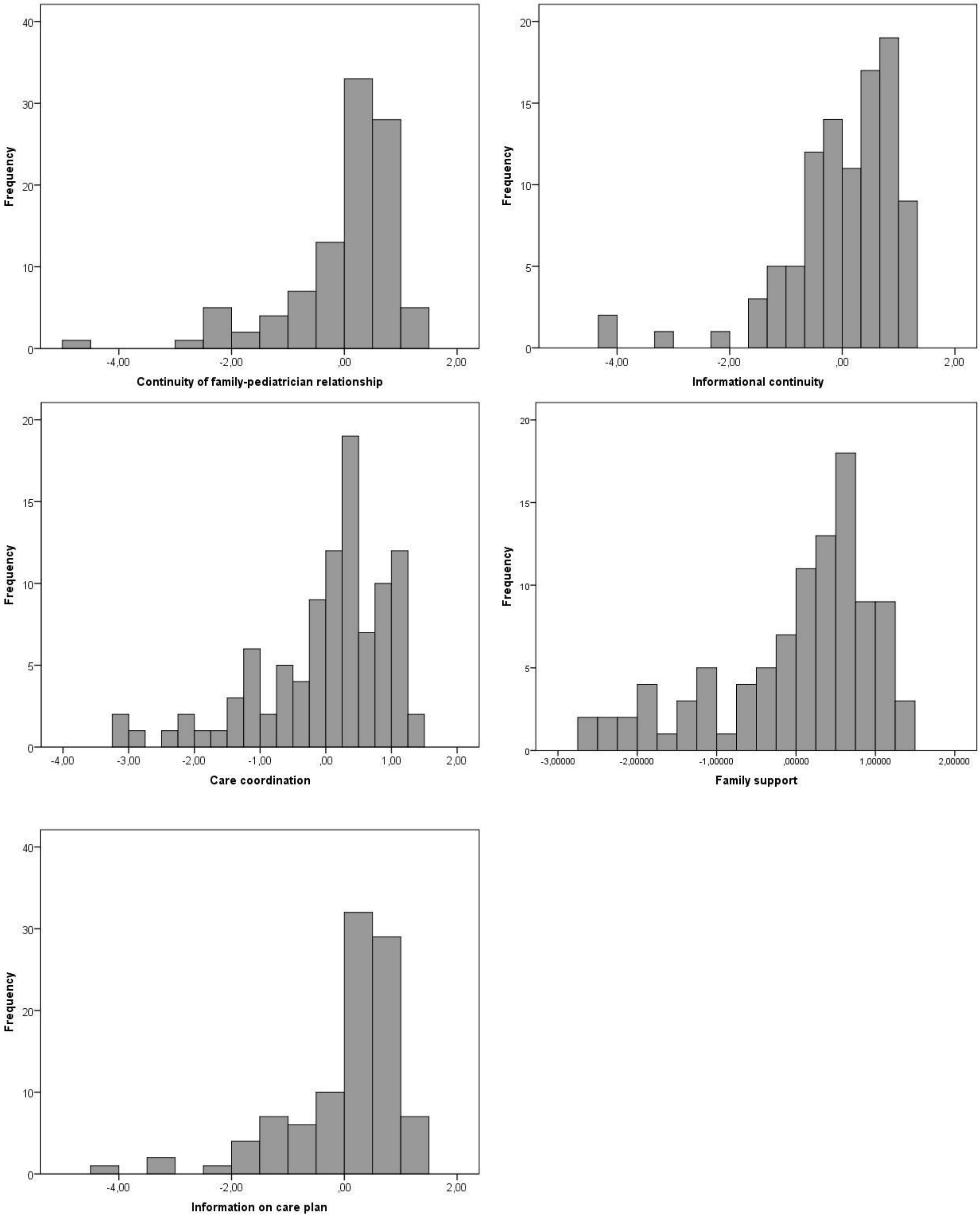
Item Number/ Item Text	**Component**				
	Informational continuity	**Care coordination**	**Relationship with FP**	**Family Support**	**Care plan**
18. I felt my child was “well known” by the people who took care of him/her (pediatrician, specialists, nurses, physiotherapists, etc.)	0.763				
20*. I felt abandoned by the healthcare system about the management of my child’s healthcare	0.733				
13*. The person who was seeing my child hadn’t the results of last check, visit or test	0.729				
14*. I had to repeat information about my child’s health which should be in his/her medical record	0.650				
12*. The person who was seeing my child ignored his/her recent medical history	0.529				
15*. I had to provide the results of a specialist’s visit to the person who was seeing my child	0.472				
8*. The persons who took care of my child told me different things about his/her health	0.312				
4. The main coordinator knows all my child’s health needs		0.865			
5. The main coordinator is always up-to-date about healthcare given by others		0.802			
6. The main coordinator contacts other clinicians about healthcare received by my child		0.729			
7. The main coordinator keeps in contact with me even when my child receives healthcare by others		0.578			
3. I feel comfortable discussing with the pediatrician all the problems related to my child’s health condition			0.859		
1. The pediatrician knows about the medical history of my child			0.811		
2. The pediatrician takes into account what worries me most about my child’s health			0.795		
19. When things changed or went wrong, I could get answers or advice~~s~~ quickly				0.742	
16. People who took care of my child gave me adequate information to take care of him/her at home				0.586	
17. People who took care of my child gave me adequate information to cope with minor problems or complications				0.573	
9. Someone explained to me the consequences of my child’s clinical conditions on his/her health				0.478	
10. Someone explained to me which treatments were made for my child and why					0.887
11. Someone explained to me the plan of tests, visits and checks that my child should do					0.830
Eigenvalue	**4.28**	**2.46**	**2.08**	**1.74**	**1.47**
Cronbach’s Alpha	**0.70**	**0.72**	**0.80**	**0.63**	**0.81**

* Reverse scored item, calculated by subtracting the item score from 6

Cronbach’s alpha for each factor is included in Table 4. Values were adequate (>0.70), except for the factor ‘family support’ (0.63).

Figure 4 provides the distribution of factor scores. Each factor showed a sufficient variability, confirming the ability to discriminate between high and low levels of continuity of care.

Figure 4 - SpeNK-Q Validation: Frequency distribution of factor scores



SpeNK-Q factor scores were unrelated with clinical characteristics and intensity of care received by the children in univariate and multivariate analyses, and in multilevel analyses adjusted for the presence of twins.

When PCA was replicated on log-transformed variables, using an estimation method suitable for ordinal-level variables and taking into account the presence of twins in the sample, results were unchanged, and the factor structure and items loadings were the same (results not reported).

3.1.3.2. Item responses and levels of continuity of care

Lastly, we calculated the percentages of the item responses to identify areas with different levels of continuity of care according to parents' perspective (Table 5). Items endorsed with the lowest frequency were 'the main coordinator contacts other clinicians about healthcare received by their child' (61.6%) and 'he/she keeps in contact with parents even when the child receives healthcare by others' (35.4%). Furthermore, over 70% of the parents reported that they had to provide the results of a specialist's visit to the person who was seeing their child. Over 20% of the parents indicated that the people who took care of their child told them different things about his/her health and over 40% had to repeat information about their child's health that should have been in his/her medical record.

Table 5 - SpeNK-Q: Responses to the item statements

	N	%	N	%	N	%
Factor 1: Informational continuity	**Never or Rarely**		**Sometimes**		**Always or Often**	
18. I felt my child was "well known" by the people who took care of him/her (pediatrician, specialists, nurses, physiotherapists, etc.)	5	5.0%	11	10.8%	85	84.2%
20*. I felt abandoned by the healthcare system about the management of my child's healthcare	92	91.1%	6	5.9%	3	3.0%
13*. The person who was seeing my child hadn't the results of last check, visit or test	81	80.2%	15	14.9%	5	5.0%
14*. I had to repeat information about my child's health which should be in his/her medical record	78	77.2%	21	20.8%	2	2.0%
12*. The person who was seeing my child ignored his/her recent medical history	88	87.1%	12	11.9%	1	1.0%
15*. I had to provide the results of a specialist's visit to the person who was seeing my child a	28	28.0%	30	30.0%	42	42.0%
8*. The people who took care of my child told me different things about his/her health	79	78.2%	14	13.9%	8	7.9%

	N	%	N	%	N	%
Factor 2: Care coordination	**Strongly disagree or Disagree**		**Uncertain**		**Strongly agree or Agree**	
4. The main coordinator knows all my child's health needs a	5	5.0%	-	-	95	95.0%
5. The main coordinator is always up-to-date about healthcare given by others a	3	3.0%	11	11.0%	86	86.0%
6. The main coordinator contacts other clinicians about healthcare received by my child b	19	19.2%	19	19.2%	61	61.6%
7. The main coordinator keeps in contact with me even when my child receives healthcare by others b	54	54.5%	10	10.1%	35	35.4%
Factor 3: Continuity of family-pediatrician relationship	**Strongly disagree or Disagree**		**Uncertain**		**Strongly agree or Agree**	
3. I feel comfortable discussing with the pediatrician all the problems related to my child's health condition	5	5.0%	10	9.9%	86	85.1%
2. The pediatrician takes into account what worries me most about my child's health	5	5.0%	11	10.9%	85	84.2%
1. The pediatrician knows about the medical history of my child	1	1.0%	5	5.0%	95	94.1%
Factor 4: Family support	**Never or Rarely**		**Sometimes**		**Always or Often**	
19. When things changed or went wrong, I could get answers or advices quickly	3	3.0%	6	5.9%	92	91.1%
16. People who took care of my child gave me adequate information to take care of him/her at home	1	1.0%	3	3.0%	97	96.0%
17. People who took care of my child gave me adequate information to cope with minor problems or complications	2	2.0%	17	16.8%	82	81.2%
9. Someone explained to me the consequences of my child's clinical conditions on his/her health	3	3.0%	11	10.9%	87	86.1%
Factor 5: Information on care plan	**Never or Rarely**		**Sometimes**		**Always or Often**	
10. Someone explained to me which treatments were made for my child and why	-	-	7	6.9%	94	93.1%
11. Someone explained to me the plan of tests, visits and checks that my child should do	1	1.0%	4	4.0%	96	95.0%

[a] Missing data n=1 [b] Missing data n=2 * Reverse scored item, calculated by subtracting the item score from 6

3.1.4. Discussion

To ensure continuity of care and to identify gaps in care coordination for children with special healthcare needs, it is essential to develop valid measures for the assessment of perceptions and experiences of parents interacting with multiple services and providers that are involved in their child's care. The SpeNK-Q proved to be a psychometrically promising instrument to measure continuity of care in children with special healthcare needs and easy to administer to parents. It may facilitate the identification of improvement potential into care for these children and help reduce the risk of fragmentation and discontinuity within the healthcare pathway.

The 5 SpeNK-Q factors identified encompassed several relevant aspects of the continuity of care and a broad spectrum of information related with the parents' perspective. In particular, Factor 1 'informational continuity' focused on the consistency of the information shared between clinicians and the feeling of being "well known" versus "abandoned" by healthcare providers. The items referred to the experience of a "common thread linking care from one provider to another and from one healthcare event to another" (i.e. informational continuity) (Haggerty et al., 2003). Only item 8 had low factor loading and could be considered for removal.

The Factor 2 'care coordination' was related to the role played by the care coordinator, identified by the parent as the professional who is in charge of most of the child's healthcare. It consisted of items assessing how well the coordinator knows all healthcare needs, maintains regular contact with the family of children and with other clinicians, and is updated about care provided by other clinicians. This factor was consistent with the "coordinator role" dimension of the questionnaire "Patient Perceived Continuity from Multiple Clinicians" developed and validated by Haggerty et al. (2012).

Items composing Factor 3 'continuity of family-pediatrician relationship' reflected the experience of an ongoing therapeutic relationship between the child and the family pediatrician. The fact that we identified these two factors~~,~~ reflecting the informational continuity, on the one hand, and the relational continuity, on the other hand, is consistent with recent studies about continuity of care (Aller et al., 2013; Gulliford et al., 2011). Aller and colleagues (2013) underlined the distinction between relational continuity, referring to the patient-provider relationship, and "seamless care" considered as continuity across care levels, which includes both transfer of medical information and care coherence.

The Factor 4 'family support' concerned the information given to the family about the child's conditions, for taking care of the child at home; coping with minor complications and the possibility of getting answers or advice quickly when necessary. Recently we published a study on parents' experiences and perceptions of the continuity of care provided to their children with special healthcare needs after hospital discharge (Zanello et al., 2015). We found that, according to parents, the support received through the information and training provided by healthcare professionals was essential to make them able to care for their children. It was crucial to help parents cope with the transition from the hospital setting to the new responsibilities connected with the home care of their child.

The Factor 5 ‘Information on care plan’ concerned the explanations given to the family about the care provided and planned for the child such as treatments, plan of tests and examinations. This is a new and specific dimension, distinct from the “informational” continuity of care, and deserves further investigations to examine whether it represents an independent aspect of the continuity of care.

The 5 SpeNK-Q factors encompassed different issues compared with the other unique instrument measuring the continuity of care in child population developed by Tobon et al. (2014). This difference could be explained by the diverse demographic characteristics (i.e. newborns vs. adolescents) and healthcare needs of samples (i.e. special health care needs vs. mental health), requiring a different array of services in different settings. Moreover Tobon et al. used an *a priori* approach to develop their sub-scales.

The issues addressed by SpeNK-Q factors are similar to themes emerged in our previous qualitative study (Zanello et al., 2015), indicating that our instrument is able to detect significant areas of continuity of care that are relevant to parents of children with special needs in different settings, such as communication, information exchange and parent involvement in the child’s care (Aujoulat, d'Hoore, & Deccache, 2007). On the contrary, we did not find consistency between our factor solution and Haggerty’s one, probably because of the differences in perspective (parents vs. adult patients) and healthcare organizational context.

Moreover, our data indicated that continuity of care was unrelated to the clinical characteristics of the child and the intensity of healthcare services received, indicating that parents perceive a high level of continuity of care regardless of the severity of the child’s condition.

The analysis of item responses underscores some issues that could be taken into account in order to improve continuity of care for children with special healthcare needs. We found that the main area of improvement concerned the role of the care coordinator. In fact, about 40% of parents stated that the main coordinator had poor or no attention in contacting other clinicians about healthcare received by the child and that often he/she did not keep in contact with parents when the child received healthcare by others professionals. Furthermore, management/informational continuity seemed to be a weakness in the continuity of care perceived by our families, because almost 75% of parents indicated that they have to provide, often or always, the report of a specialist’s visit to the person who was seeing their child, that they had to repeat information about the child’s health which should have been in his/her medical record and that the people who took care of the their child told different things about his/her health. To our knowledge, at present there are no other available quantitative studies investigating areas of discontinuity of care for children with special healthcare needs from parents’ perspective. Therefore, these areas should be taken into account by the healthcare providers to improve continuity of care in this specific setting and population.

The main strength of this study is that SpeNK-Q is the first instrument measuring continuity of care provided to children with special healthcare needs from the parents’ perspectives. Moreover, our study includes parents with different duration of the experience of care, thus increasing the sample variability as regards a core element of continuity of care, i.e. patient’s experience of care over time (Reid et al., 2002).

The study has some **limitations** to address. The first is the generalizability and utility of our instrument to assess continuity of care of children with chronic conditions or special healthcare needs other than preterm birth. The second limitation relates to the lack of information about test-retest reliability. We decided not to administer the questionnaire to the same participant at two different times to avoid burdening families who were living in difficult situations. The third limitation concerns the inability to assess the concurrent validity with other existing instruments because no validated instruments assessing continuity of care in children are available in Italian language. The fourth is the limited sample size that did not allow to use confirmatory factor analysis or item response theory analysis to examine the performance of items in deeper detail (Babyak & Green, 2010; Reeve, 2005).

3.1.5. Conclusion

The SpeNK-Q proved to be a promising instrument encompassing multifaceted components of continuity of care, which could be integrated in routine practice to assess the users’ experience of different healthcare models and procedures. Thus, SpeNK-Q may be used to identify areas of improvement from users’ perspective to be integrated with professionals’ and systems’ viewpoints (McDonald et al., 2014). This could represent a first step towards an experience based design approach in a public health perspective, by making the user integral to the process of redesigning services (Carr et al., 2011).

Further studies are needed to evaluate the test-retest reliability of the instrument, to analyze the item response in large samples, to confirm the factor structure and extend the psychometric properties of SpeNK-Q in children with other special healthcare needs recruited from other national and international settings.

3.2. Administration of SpeNK-Q to families recruited in SpeNK Project

Abstract

Introduction. Care of children with special health care needs requires coordination among multiple services and professionals and continuity of care in integrated health care pathways. The study aims to test the utility of SpeNK-Q Questionnaire as a measure of continuity of care in children with special health care needs and to assess continuity of care for these children from parents' perspective, within the Special Needs Kids (SpeNK) Project, which investigates sheltered discharge procedures for this population in Bologna province. **Methods**. A validated 20-item questionnaire (SpeNK-Q) with a 5-point response option was administered by phone to parents of 67 children with special health care needs enrolled in SpeNK Project at 8-15 months after hospital discharge from hospital facilities of St. Orsola Malpighi University Hospital of Bologna and of Local Health Authorities of Bologna and Imola. Percentages of item responses were calculated to identify differences in perceived continuity of care and the content analysis of answers to open-ended questions was carried out to identify key figures related to care and coordination of care. **Results**. Item responses indicate high levels of continuity of care (mean=4.3), with lower rates about the informational continuity and the maintenance of contacts of main care coordinator with parents about care received by the child from others. Responses to open-ended questions indicate a health care network as taking care of the most of child's care in over 40% of cases, with a central role of the FPs, whereas the main coordinator is mainly represented by the hospital (65.7%). **Conclusions**. This study confirms the utility of SpeNK-Q as a measure of continuity of care in children with special healthcare needs. Parents of children enrolled in SpeNK Project report high levels of continuity of care and the involvement of an array of services and professionals with the provision of health care to their children, with some improvement potential concerning the informational continuity and the maintenance of contacts of main care coordinator with parents.

Keywords: *special healthcare needs, children with special health care needs, continuity of care, family support, parents experience*.

3.2.1. Background

Children with special health care needs can be defined as those who "*have, or are at an increased risk for, a chronic physical, developmental, behavioral, or emotional conditions and who also require health and related services of a type or amount beyond that generally required by children*" (McPherson et al., 1998) (p.138). According to The National Survey of Children with Special Health Care Needs Chart Book 2009–2010 (US Dept of Health and Human Services, 2013), about 15% of children under 18 years of age in the United States are estimated to have special health care needs, and 23% of households with children have at least one child with special health care needs. This prevalence is growing in past decades, thanks to the advances in medical treatments that increase survival of children with serious congenital or acquired diseases. Similar to adults with chronic or complex health conditions, these children requires the implementation and coordination of an array of health care services and providers over time (van der Lee et al., 2007). In this context, the continuity of care has been widely recognized as an important factor for a high-quality healthcare provision to pediatric patients and their families (American Academy of Pediatrics, 2002). According to Haggerty et al. (2003), continuity of care can be defined as "*the degree to which a series of discrete healthcare events is experienced as coherent and connected and consistent with the patient's medical needs and personal context*". Up to now, poor attention has been paid to children and their families in assessing and measuring continuity of care, compared to adult and elderly patients with chronic or complex conditions. In children with special health care needs, the patient's needs, experiences, and perceptions are inevitably mediated by parents (Miller et al., 2009), whose involvement in the evaluation of services provided to their children is therefore indispensable. To our knowledge, most available instruments on continuity of care address specific adult populations and settings (Uijen et al., 2012), with the exception of a questionnaire measuring family's perspective about continuity of care in mental health care for pediatric population (Tobon et al., 2014).

Recently, we conducted a study aiming to develop and validate a quantitative measure of continuity of care in children with special health care needs from family's perspective, the SpeNK-Q Questionnaire (Rucci et al., 2015), on the basis of the findings of our qualitative study (Zanello et al., 2015). These studies are part of the Special Needs Kids (SpeNK) Project, aiming to examine existing sheltered hospital discharge procedures and integrated clinical pathways for children special healthcare needs in Bologna Province, and assess family's perspective and family pediatrician's role.

The aims of this study were: (1) to test the utility of our instrument to assess continuity of care in children with special healthcare needs other than preterm birth, on which it was validated, and (2) to evaluate continuity of care for children enrolled in SpeNK Project from parents' perspective.

3.2.2. Materials and Methods

3.2.2.1. Study design

Children and families were recruited to SpeNK Project at hospital discharge of the child from the participating units of hospital facilities at St. Orsola Malpighi University Hospital of Bologna and Local Health Authorities of Bologna and Imola.

The recruitment of children was conducted from October 1st 2012 to September 30th 2014 on incident cases meeting the following inclusion criteria: age from 0 to 16 years, residence in Bologna province, and the presence of at least one of the following conditions:

- Birth weight <1000 g;
  - Complex and/or chronic health conditions defined as: Need for technological assistance, Acute neurological deficit, Severe endocrinopathy, Complex genetic malformative pathology;
- Children with oncological diseases who need palliative care or particular community care;
- Newborns with mothers in contact with mental health services or drug addiction

Only first ever hospitalizations for the condition of interest were included. Written informed consent was obtained at recruitment from each parent to collect clinical data on children and to contact them during follow-up period (9 months from hospital discharge). The Ethics Committees of the Bologna University Hospital and of Bologna and Imola Local Health Authorities approved the study procedures.

3.2.2.2. Participants

At the end of follow-up (at least 8 months after hospital discharge), we contacted by phone the families recruited in SpeNK Project to invite them to answer to a telephone questionnaire about their experiences and perceptions on the healthcare provided to their children during the past months, after discharge. The inclusion criteria for SpeNK-Q administration were an adequate level of knowledge of Italian language and the telephone availability of recruited persons. The fluency in Italian language was appraised at recruitment by the personnel in charge. Of families of 82 recruited children, parents of 3 children (3.7%) were *a priori* excluded because they were not sufficiently fluent in the Italian language. Parents of 11 children (13.4%) were later excluded because it was not possible to contact them by phone (i.e. no answer, number off, etc.), after at least 5 attempts. In 1 case (1.2%), parents were not contacted because their child died during follow-up period.

In total, parents of 67 children (81.7% of SpeNK sample) were contacted at 8-15 months after hospital discharge of their children, during a 24-month period (June 2013 – June 2015), and completed the questionnaire by phone interview.

3.2.2.3. Instrument

The process of development and validation of the instrument is described in detail in the paper by Rucci et al. (2015). The SpeNK-Q questionnaire was derived from the results of the SpeNK-I qualitative study (Zanello et al., 2015) and was based on Haggerty's constructs of informational, management and relational continuity (Haggerty et al., 2003; Haggerty et al., 2012). SpeNK-Q was validated on a sample of parents of preterm birth children who completed 101 questionnaires at the second or subsequent planned follow-up visit after the child's hospital discharge.

SpeNK-Q consists of 20 items with a 5-point response option, to measure agreement or frequency, and two open-ended questions. The item statements explore parents' perspective about their relationship and interactions with: (1) the family pediatrician (knowledge of the child's medical history, partnership and confidence); (2) the main coordinator (knowledge of the child's health needs, continuity with other providers, services and clinicians); (3) the network of healthcare providers and services involving child care such as care provision, coherence and availability of information, parents' involvement and engagement, knowledge of the child, experiences of receiving advice, and healthcare systems. The two open questions ask respondents to identify: (1) the person who is in charge of most of the child's health; (2) the person who coordinates the child's health care (i.e. the main coordinator: for example, PIFC physician, Family Pediatrician, nurse, etc.). The structure of the instrument entails 5 factors explaining 60.2% of item variance: informational continuity; coordination of care; continuity of family-pediatrician relationship; family support; information on care plan. Appendix - SpeNK-Q includes the whole questionnaire in its original form (Italian language).

3.2.2.4. Analyses

IBM SPSS Statistics (version 20, Chicago, USA) was used for the descriptive analyses of the item responses to identify differences in levels of continuity of care. Content analysis was used to categorize the answers to open-ended questions.

3.2.3. Results

3.2.3.1. Participants

SpeNK-Q was administered to 64 families of 67 children. As regards the parental citizenship, families included Italian parents in 58.2% of cases (n=39), one Italian parent in 14.9% (n=10) of cases, and no Italian parents in 26.9% of cases (n=18). Families completed the questionnaire in average at 10.2 months (SD 1.2) after hospital discharge, ranging from 8 to 15 months, and their children had a mean age of 16.6 months (SD 15), ranging from 9 months to 7 years. The children were male in 53.7% (n=36) of cases. Table 6 summarizes characteristics of the children and number of children recruited at the participating units.

Table 6 – SpeNK-Q Administration: Characteristics of the Sample (n=67)

Gender, Male; n (%)	36 (53.7%)
Age, months; mean (±SD, range)	16.6 (±15, range 9.5-89.1)
Time from hospital Discharge, months; mean (±SD, range)	10.2 (±1.2, range 8.7-14.95)
Cause of inclusion; n (%)	
Encephalopathy/Neuropathy	8 (11.9)
Complex genetic/malformative pathologies	10 (14.5)
Prematurity < 1000 gr	31 (46.3)
Prematurity with other conditions	6 (9.0)
Other conditions	8 (11.9)
Social Problems	4 (6.0)
Discharge Procedure, Sheltered; n (%)	45 (67.2)
Care services activation*; n (%)	
- Community care services	61 (91.0)
- Individualized Care Plan (i.e. PAI)	41 (61.2)
- Home Care services (i.e. ADI)	43 (64.2)
- Hospital Follow-up	65 (97.0)
- Home Health Devices	13 (19.4)
Hospital Unit of recruitment; n (%)	
S.Orsola Malpighi University Hospital of Bologna	*41 (61.2)*
Pediatric Surgery Unit UH Bologna	4 (6.0)
Neonatology Unit UH Bologna	32 (47.8)
Child Neuropsychiatric Unit UH Bologna	1 (1.5)
Pediatric Unit UH Bologna	1 (1.5)
Pediatric Emergency Room UH Bologna	3 (4.5)
Maggiore Hospital LHA Bologna	*19 (28.4)*
Neonatology and NICU LHA Bologna	16 (23.9)
Pediatric Unit LHA Bologna	3 (4.5)
Hospital LHA Imola	*7 (10.4)*

* Percentages calculated on a total of 67 children for each item

3.2.3.2. Item responses and levels of continuity of care

The percentages of the item responses were calculated to identify areas with different levels of continuity of care according to parents' perspective (Table 7). The item endorsed with the lowest frequency was "the main coordinator keeps in contact with me even when my child receives healthcare by others" (59.7%). Furthermore, over 65% of the parents reported that they had to provide the results of a specialist's visit to the person who was seeing their child, and about a third of parents indicated that the people who took care of their child told them different things about the child's health. On the other hand, the majority of parents endorsed items on the knowledge of the main coordinator about the child's health needs (93.7%) and knowledge of the pediatrician about the child's medical history (91%). Moreover, about 90% of parents reported that they were given adequate information to take care of their child at home.

Table 7 – SpeNK-Q Administration: Responses to the item statements

	N	%	N	%	N	%
Factor 1: Informational continuity	**Never or Rarely**		**Sometimes**		**Always or Often**	
8. The people who took care of my child told me different things about his/her health [a]	44	66.7%	13	19.7%	9	13.6%
12. The person who was seeing my child ignored his/her recent medical history [a]	53	80.3%	8	12.1%	5	7.6%
13. The person who was seeing my child hadn't the results of last check, visit or test [a]	51	77.3%	8	12.1%	7	10.6%
14. I had to repeat information about my child's health which should be in his/her medical record	49	73.1%	10	14.9%	8	11.9%
15. I had to provide the results of a specialist's visit to the person who was seeing my child	22	32.8%	21	31.3%	24	35.8%
18. I felt my child was "well known" by the people who took care of him/her (pediatrician, specialists, nurses, physiotherapists, etc.)	3	4.5%	12	17.9%	52	77.6%
20. I felt abandoned by the healthcare system about the management of my child's healthcare	52	77.6%	9	13.4%	6	9.0%
Factor 2: Care coordination	**Strongly disagree or Disagree**		**Uncertain**		**Strongly agree or Agree**	
4. The main coordinator knows all my child's health needs [b]	1	1.6%	3	4.8%	59	93.7%
5. The main coordinator is always up-to-date about healthcare given by others [b]	0	0.0%	11	17.5%	52	82.5%
6. The main coordinator contacts other clinicians about healthcare received by my child [c]	2	3.2%	15	24.2%	45	72.6%
7. The main coordinator keeps in contact with me even when my child receives healthcare by others [c]	19	30.6%	6	9.7%	37	59.7%
Factor 3: Continuity of family-pediatrician relationship	**Strongly disagree or Disagree**		**Uncertain**		**Strongly agree or Agree**	
1. The pediatrician knows about the medical history of my child	3	4.5%	3	4.5%	61	91.0%

	N	%	N	%	N	%
2. The pediatrician takes into account what worries me most about my child's health	6	9.0%	8	11.9%	53	79.1%
3. I feel comfortable discussing with the pediatrician all the problems related to my child's health condition	7	10.4%	3	4.5%	57	85.1%
Factor 4: Family support	**Never or Rarely**		**Sometimes**		**Always or Often**	
9. Someone explained to me the consequences of my child's clinical conditions on his/her health [d]	9	13.8%	9	13.8%	47	72.3%
16. People who took care of my child gave me adequate information to take care of him/her at home	2	3.0%	5	7.5%	60	89.6%
17. People who took care of my child gave me adequate information to cope with minor problems or complications	11	16.4%	2	3.0%	54	80.6%
19. When things changed or went wrong, I could get answers or advice~~s~~ quickly [a]	4	6.1%	9	13.6%	53	80.3%
Factor 5: Information on care plan	**Never or Rarely**		**Sometimes**		**Always or Often**	
10. Someone explained to me which treatments were made for my child and why [d]	3	4.6%	9	13.8%	53	81.5%
11. Someone explained to me the plan of tests, visits and checks that my child should do	4	6.0%	9	13.4%	54	80.6%

[a] Missing data n=1 [b] Missing data n=4 [c] Missing data n=5 [d] Missing data n=2

Moreover, we calculated the mean score for each factor (see Table 8).

Values indicated that parents positively assessed continuity of care in average, with a minimal difference concerning the factor of "informational continuity".

Table 8 - SpeNK-Q Administration: Factors statistics

SpeNK-Q Factors (items)	**Mean**	**SD**	**Range**
Informational continuity (items 18, 20*, 13*, 14*, 12*, 15*, 8*)	4.1	0.7	2-5
Care coordination (items 4, 5, 6, 7)	4.2	0.7	2.2-5
Relationship with the pediatrician (items 3, 2, 1)	4.4	0.9	1.7-5
Family support (items 19, 16, 17, 9)	4.2	0.8	2-5
Care plan (items 10-11)	4.4	0.9	1-5

* Reverse scored item, calculated by subtracting the item score from 6

3.2.3.3. Open-ended questions and actors of healthcare provision

SpeNK-Q included two open questions asking respondents to identify: (1) the person who takes care of most of the child's health; (2) the person who coordinates the child's health care. As a result of the content analysis, it was possible to classify answers according to the actor(s) identified by parents (see Table 9).

As for the first question, in 6 cases (9%) family could not identify a professional taking care of most of child's health care. Over 40% (n=29) of answers identified a collaborating network of two or more actors (i.e. Family Pediatrician (FP), Hospital services, Community services), including the FP in the majority of cases (26/29). FP was mentioned as the only professional taking care of child's most health care in 22.4% of cases.

As for the second question, the majority of parents indicated an Hospital service as unique care coordinator (65.7%), identifying a single specific professional in most cases (n=39). In about 20% of cases the FP was mentioned as care coordinator, alone (n=4, 6.0%) or as part of a network (n=9, 13.4%). The community services and community professional were less frequently indicated as care coordinators.

Table 9 - SpeNK-Q Administration: Responses to open-ended questions

1. Thinking about health care received by your child, who takes care of most of his/her health care?	**N**	**%**
Family only	6	9.0%
Community service(s) only	2	3.0%
Hospital service(s) only	14	20.9%
Family Pediatrician only	15	22.4%
Network (FPs, Community, Hospital)	29	43.3%
- FP/Hospital	*14*	*20,9%*
- FP/Hospital/Community	*9*	*13,4%*
- FP/Community	*3*	*4,5%*
- Hospital/Community	*3*	*4,5%*
Others (i.e. outside Bologna province)	1	1.5%

2. Thinking about all the people who took care of your child's health, is there anyone which coordinates the health care? Who?	**N**	**%**
Nobody	1	1.5%
Family only	3	4.5%
Hospital service(s) only	44	65.7%
- Hospital service and team	*5*	*7.5%*
- Hospital professional (e.g. physician, nurse)	*39*	*58.2%*
Community service(s) only	3	4.5%
Family Pediatrician only	4	6.0%
Network (FPs, Community, Hospital)	10	14.9%
- FP/Hospital	*8*	*11,9%*
- FP/Community	*1*	*1,5%*
- Hospital/Community	*1*	*1,5%*
Others (i.e. outside Bologna Province)	2	3%

3.2.4. Discussion

The study aimed to assess continuity of care for newborns and children with special healthcare needs in Bologna province by administering the SpeNK-Q Questionnaire to parents of children enrolled in SpeNK Project: SpeNK-Q was easy to administer to parents and allowed to identify some improvement areas and some positive aspects into care for these children.

First of all, parents reported lower levels of "informational continuity" compared to the mean high evaluation of the item statements. In particular, according to the item responses, most of them had to provide (sometimes, often or always) the results of a specialist's visit to the person who was seeing their child, and about a third of them were told (sometimes, often or always) different things about the child's health by people who took care of their child. These results indicate that continuity of information on events and conditions of the patient could be improved. This concerns the use of information on prior events to provide care that is appropriate to the patient's current circumstances (Reid et al., 2002). The second improvement area concerns the "care coordination", since in 40% of case parents expressed a contrary or uncertain opinion about the main coordinator keeping in contact with them when the child receives healthcare by others. A qualitative meta-summary about experienced continuity of care for patients multiple clinicians (Haggerty, Roberge, Freeman, & Beaulieu, 2013), indicates that coordination and information transfer between professionals are "assumed until proven otherwise". A qualitative study by Miller et al. (2009) on parents' perceptions about continuity of care for children with special health care needs found that "*compartmentalization of services and information led to parents assuming a necessary, though at times, uncomfortable, coordinating role*". The lower levels of perceived continuity about information transfer and care coordination may represent potential barriers to a seamless management and provision of care.

On the other hand, over 90% of parents expressed a positive judgment about the knowledge of the main coordinator about the child's health needs and of the pediatrician about the child's medical history. This finding indicate that the main coordinator and the pediatrician represent key reference figures for the clinical management of the child's health condition, in accordance with the recommendations and guidelines at regulatory level.

Moreover, about 90% of parents reported that they were given adequate information to take care of their child at home. This finding is in accordance with the results of our qualitative study (SpeNK-I) (Zanello et al., 2015). As accounted by interviews and focus group, parents perceive the empowerment as a process starting from hospital setting which is essential to help them cope with the transition to the home care of their child.

Finally, the results of open-ended questions suggest some considerations about the identity and the level of involvement of actors participating in health care and care coordination for children with special health care needs. In some cases parents could not identify a professional, outside the family, as in charge of the case. On the other side, in over 40% of cases parents mentioned a collaborating network of two or more actors as taking care of most of child's health care, including hospital and community services and/or the FP. In almost two thirds of cases the FP was cited in parents' answers, alone or as part of the collaborating network. As regard the care coordination, in most cases parents were able to indicate a professional (or service) who coordinates the care of their child. According to their responses, the majority of parents recognize the Hospital as main coordinator and in most cases a professional in person. According to Haggerty et al. (2013), "*having a single trusted clinician who helps navigate the system and sees the patient as a partner*" undergirds the experience of continuity between clinicians. Our results show that most parents enrolled in SpeNK Project could identify a professional (mainly the Family Pediatrician as responsible clinician, and a Clinician from the hospital as care coordinator) as their reference figure in the health care pathway of their child.

To our knowledge, this study is the first investigating continuity of care for children with special health care needs, with a variety of conditions, using a validated instrument. Its main limitation is the limited sample size, which did not allow to use item response theory analysis to examine the performance of items in deeper detail or confirmatory factor analysis to verify the structure of the instrument (Babyak & Green, 2010; Reeve, 2005).

3.2.5. Conclusion

This study confirms the utility of SpeNK-Q as a measure of continuity of care in children with special healthcare needs. Results indicate that parents of children with special health care needs enrolled in SpeNK Project perceive high levels of continuity of care and the involvement of an array of services and professionals in the provision of health care to their children. Our findings also show some improvement areass in organization and coordination of care, concerning the informational continuity and the maintenance of contacts of main care coordinator with parents.

Chapter 4. The role of Family Pediatrician in care coordination

This chapter is based on the preliminary draft of a paper in preparation for publishing, as part of SpeNK Project, with permission of the authors.

Abstract

Introduction. Care coordination is widely recognized as an essential element of care for patients with chronic and complex medical conditions and their families. In care for children with special health care needs the Family Pediatrician (FP) plays a central role as care coordinator. This study aims to evaluate the FPs' activities of care coordination for children with special health care needs in the pediatric primary care setting, using an on-line measurement tool. **Methods**. Within the cohort prospective study SpeNK (Special Needs Kids), newborns and children with special health care needs were recruited at discharge from three hospital facilities in Bologna province, from October 1st 2012 to September 30th 2014. Their FPs were invited to participate in the study by recording with SpeNK-FP each encounter for the patient during a 9-month period after hospital discharge. SpeNK-FP was developed by adapting the CCMT© (Antonelli et al., 2008) to the Italian organizational context. **Results**. 40 FPs completed 382 questionnaires for 49 patients. The majority of patients (71.4%) were patients with special health care needs, without complicating familiar or social issues. The focus of encounter included in the majority of cases clinical issues. FPs reported "no need for care coordination" in more than 40% of records about patient's needs requiring care coordination. Activities implemented to meet the patient's needs included contacts with contacts with healthcare professionals and services in more than half of cases. According to FPs' subjective appraisal, 79.9% of encounters prevented an inappropriate services use. **Conclusions**. The study shows some difficulty for FPs to record their activities and the improvement potential as care coordinators for children with special health care needs.

Keywords: *children with special healthcare needs, pediatric primary care, family pediatrician, care coordination, assessment*.

4.1. Background

The coordination of care has been widely recognized as an important process of organization of patient care activities to facilitate the appropriate delivery of healthcare services and to achieve a high-quality, high-value, patient-centered healthcare system (Schultz & McDonald, 2014). The goal of care coordination is to support patients and their families requiring healthcare in their interaction with an increasingly complex healthcare system.

In the context of pediatric health care, care coordination has been defined as "a process that links children and youth with special health care needs and their families with appropriate services and resources in a coordinated effort to achieve good health" (American Academy of Pediatrics, 2005). Children with special health care needs can be defined as those who "*have, or are at an increased risk for, a chronic physical, developmental, behavioral, or emotional conditions and who also require health and related services of a type or amount beyond that generally required by children*" (McPherson et al., 1998). This definition refers to a "heterogeneous population" with a variety of "diagnoses and functional limitations", sharing in common a "high need for services" (Strickland et al., 2011). Care coordination for this population is associated with lower odds of unmet specialty care needs (Boudreau et al., 2014).

A key role in care coordination for children with special health care needs should be played by the primary care provider (Stille & Antonelli, 2004; Stille et al., 2005). However, some studies underlined the scant involvement of primary care providers in managing care of children with chronic conditions (Palfrey et al., 1980; Rowland, 1989).

In Italy, family pediatricians are trained specialists providing primary care for children up to 16 years of age in ambulatory and home settings and coordination of care for patients with chronic conditions (van Esso et al., 2010). Family pediatricians are in charge of assessing patients' needs, ordering diagnostic procedures, prescribing drugs, and referring patients to specialists and hospitals (Luciano et al., 2014).

The Italian Collective Agreement (July 2010) governs the relationship of the National Health Service with Family Pediatricians and includes, among the general objectives, the construction of an integrated network of services for taking in charge of children with special health care needs and for the government of healthcare and social pathways.

Nevertheless, research about the implementation of care coordination for children with special health care needs and the role of family pediatrician as care coordinator is limited.

In Emilia-Romagna Region the SpeNK (Special Needs Kids) Project has been designed to describe the implementation of existing sheltered hospital discharge procedures and integrated clinical pathways for children with complex or chronic health conditions and special healthcare needs and to assess the family's perspective on continuity of care and the role of family pediatrician.

The aim of this study is to assess the care coordination activities performed by the Family Pediatrician (FP) for children and newborns with special health care needs in the pediatric primary care setting, using an on-line measurement tool.

4.2. Materials and Methods

4.2.1. Study design

The study involved Family Pediatricians (FPs) who were in charge of children with special health care needs recruited to SpeNK Project, in order to assess the activities they performed for the coordination of care.

The involvement of FPs of children recruited to the SpeNK Project was arranged by the SpeNK Team in coordination with the Departments of Primary Care of the Health Authorities of Bologna and Imola, aiming to share the research objectives and instruments, initially with the Unions of Pediatricians and later with the family pediatricians, in person. FPs received an incentive for their participation in the study, as part of a contractual agreement between the Primary Care Departments and the Unions of Pediatricians.

4.2.2. Measurement Instrument

The SpeNK-FP was developed adapting the CCMT© by Antonelli et al. (2008) to the Italian organizational context. It was prepared whether online or in paper form and should be completed by the FP at each encounter regarding the child.

SpeNK-FP included an "identity record" and an "encounter record". The identity record collected the personal data of the patient (name, gender, birth date, etc.). The encounter record included the date of the encounter and 8 items aiming to collect information about the activity performed by the FP. The first item concerns the patient's complexity. We considered our inclusion criteria for the patients and decided to use a scale on 3 levels: children with mainly social needs (Level 1), children with mainly health needs (Level 2), children with both health and social needs (Level 3). The second item inquires the request(s) or problem(s) of the patient for which the encounter took place (e.g. make appointment with the FP, referral to a subspecialist). Item 3 investigates the need(s) for care coordination that emerged in the encounter (e.g. make appointments with other specialists). Item 4 investigates which activities were carried out by the FP (e.g. contacts with family or with hospital) and how (e.g. telephone contact, face-to-face meeting, etc.) to fulfill patient's needs emerged in the encounter. Item 5 examines the involvement of any other professional(s) in the care coordination. Item 6 inquires the time spent for care coordination. The two final items require the FP's appraisal about the outcomes that were occurred and prevented with the care coordination activity.

Appendix - SpeNK-FP includes the instrument in its original form (Italian language).

4.2.3. Training and Data Collection

The SpeNK Team planned and provided a 1-day training program about the study design and data collection procedures in two separated sessions for the pediatricians of Imola and Bologna respectively. During the training session the SpeNK Team presented to participants the study design, objectives and procedures and the measurement tool. FPs were asked to record each encounter during the follow-up period and the "Encounter" was defined as "any activity performed by the Family Pediatrician for the patient", involving the child or the family and including visit at the clinic, phone contact, etc.

A member of the SpeNK Team contacted the Family Pediatrician to alert the beginning of follow-up for each subject, to remind aims and procedures of the study, and to provide the login credentials and the instructions to use the SpeNK-FP on-line window. During follow-up, the same Team member maintained telephone contacts with FPs to provide technical support when needed. Ultimately, he alerted the end of the follow-up period for each subject to the FP.

4.3. Results

A total of 61 Family Pediatricians (FPs) who were in charge of 82 children with special health care needs recruited to SpeNK study were contacted and invited to participate in the study. Of these, 40 FPs (65.6%) completed the SpeNK-FP for a total of 49 (59.7%) subjects.

The total number of encounters entered was 382. A mean of 12.7 (SD=6.9) encounters were entered for single subject (median=12.5, range 1-28). Overall, the 40 FPs entered a mean of 17.0 (SD=10.3, median=15, range=1-36) encounters.

Table 10 shows characteristics of 40 FPs and Table 11 demographic characteristics and medical conditions of 49 children included in the analysis.

Table 10 – SpeNK-FP Study: Characteristics of Family Pediatricians (n=40)

Gender, Female, n (%)		31 (87.5%)
Age, years; mean (±SD)		55.3 (± 5.6); range: 37-66
No. of Practice Patients; mean (±SD)		855.8 (± 143.1); range: 425-1050
Form of Association, n (%)	Solo	8 (20%)
	Group	16 (40%)
	Network	14 (35%)
	Missing data	2 (5%)
Practice Location: Urbanization*, n (%)	Low	4 (10%)
	Middle	16 (40%)
	High	18 (45%)
	Missing data	2 (5%)

* Atlante Statistico dei Comuni 2014 Italia

Table 11 – SpeNK-FP Study: Characteristics of infants (n=49)

Gender, Male, n (%)		26 (53.1%)
Age, months; mean (±SD)		5.8 (±11.8); range: 0-76
Medical conditions, n (%)	Prematurity < 1000 gr	21 (42.9%)
	Complex genetic / malformative pathologies	7 (14.3%)
	Prematurity with other conditions	5 (10.2%)
	Encephalopathy / Neuropathy	5 (10.2%)
	Other conditions	7 (14.3%)
	Social Problems	4 (8.2%)

According to Levels of complexity, 3 subjects (6.1%, with a total of 19 encounters) were assessed at Level 1, 35 subjects (71.4%, with a total of 290 encounters) at Level 2, 11 subjects (22.5%, with a total of 73 encounters) at Level 3.

Table 12 shows the time spent for encounter at patient's complexity level. Time spent per encounter was less than 5 minutes in 5.3% of encounters of children at Level 1, 13.8% at Level 2 and 25% at Level 3. On the other hand, time spent per encounter was greater than 30 minutes in 5.3% of encounters of children at Level 1, 29.2% at Level 2 and 19.4% at Level 3.

Table 12 – SpeNK-FP Study: Patient Complexity Level and Time spent per encounter. N (%)

Time spent per Encounter	Level 1 Patients N (%)	Level 2 Patients N (%)	Level 3 Patients N (%)
Less than 5 minutes	1 (5.3%)	27 (13.8%)	18 (25.0%)
5-9 minutes	5 (26.3%)	40 (20.5%)	3 (4.2%)
10-19 minutes	4 (21.0%)	46 (23.6%)	12 (16.7%)
20-29 minutes	8 (42.1%)	25 (12.8%)	25 (34.7%)
30 minutes and greater	1 (5.3%)	57 (29.2%)	14 (19.4%)
N.o of Encounters with Time Spent recorded	19/19 (100%)	195/290 (67.2%)	72/73 (98.6%)

The focus of encounter is presented in Table 13. There were 850 records entered by the FPs for all encounters. The majority of records included growth and nutrition (29.6%), request of a visit of Family Pediatrician (24.2%) and developmental and behavioral issues (21.1%). All other focuses were indicated in less than 5% of records, except for medicines prescriptions (9.3%).

Table 13 – SpeNK-FP Study: Focus of encounters

Focus of encounters	**No. Recorded, n (%)***
Request of a visit of Family Pediatrician	206 (24.2%)
Medicines prescriptions	79 (9.3%)
Prescriptions of laboratory examinations	29 (3.4%)
Prescriptions of laboratory tests	3 (0.4%)
Need for prosthesis/devices	3 (0.4%)
Growth and nutrition	252 (29.6%)
Referral management	32 (3.8%)
Developmental and behavioral	179 (21.1%)
Educational and school	3 (0.4%)
Mental health	0
Social services (i.e. housing, food, clothing. …)	14 (1.6%)
Integrated Home Care (i.e. *ADI*)	21 (2.5%)
Legal and Judicial	1 (0.1%)
Other	28 (3.3%)

*Total 850 recorded focuses for 376 encounters

There were 364 records entered by the FPs about patient’s needs requiring care coordination. More than 40% (n=150) of records reported “no need for care coordination”, whereas “follow-up referrals” were indicated in 27.1% of records. All other needs were entered less frequently, from 9.6% (order laboratory examinations) to 1.1% (coordination services) of records (Table 14).

Table 14 – SpeNK-FP Study: Patient’s needs requiring Care Coordination

Patient’s needs requiring Care Coordination	**No. Recorded, n (%)***
Follow-Up Referrals	99 (27.1%)
Order Prescriptions	29 (7.9%)
Order Supplies	7 (1.9%)
Order Services	15 (4.1%)
Order Laboratory Examinations	35 (9.6%)
Order Laboratory Tests	16 (4.4%)
Coordination Services (schools, agencies, payers, etc.)	4 (1.1%)
Reconcile Discrepancies	9 (2.5%)
None	150 (41.1%)

*Total 364 recorded needs for 295 encounters

FPs recorded 468 activities to fulfill patient’s needs (Table 15). Among activities implemented to meet the patient’s needs, more than half included contacts with healthcare professionals and services and only 33.6% with parent/family. Referring to the contact methods, 34% (n=128) of encounters reported at least one face-to-face meeting and 30% (n=113) at least one telephone contact. Few contacts took place by e-mail (1.3%, n=5).

Table 15 – SpeNK-FP Study: Activity to fulfill patient's needs

Activity to fulfill patient's needs	**No. Recorded, n (%)***
Contact with Parent/Family	158 (33.6%)
Contact with School	0
Contact with Health-Social Services	33 (7.0%)
Contact with Hospital/Clinic	72 (15.3%)
Contact with Payers	1 (0.2%)
Contact with Professional Educator	9 (1.9%)
Contact with Pharmacy	3 (0.6%)
Contact with Agencies	1 (0.2%)
Contact with Home Care Personnel	30 (6.4%)
Contact with Consultants	49 (10.4%)
Contact with Other	1 (0.2%)
Written Communication	23 (4.9%)
Written Report to Health Authorities	0
Chart Review	4 (0.9%)
Clinical Audit	8 (1.7%)
Develop/Modify Written Care Plan	5 (1.1%)
Meeting/Case Conference	71 (15.1%)

*Total 468 recorded activities for 376 encounters

There were 422 records entered by FPs about staff involved in Care Coordination. More than 50% of records reported that no one was involved, other than FP. In less than 25% of records other physicians were involved in care coordination (Table 16).

Table 16 – SpeNK-FP Study: Staff involved in Care Coordination

Staff involved in Care Coordination	**No. Recorded, n (%)***
No one	219 (51.8%)
Other Physician(s)	93 (22.0%)
Nurse(s)	36 (8.5%)
Social Worker(s)	38 (9.0%)
Administrative Staff	13 (3.1%)
Other	23 (5.4%)

*Total 422 recorded staff for 376 encounters

Tables 17 and 18 show the subjective assessment of FPs about the outcomes prevented and occurred as a result of their care coordination activity. Near 80% of records reported that encounters prevented an inappropriate use of services (i.e. 24.4% visit to the pediatric office/clinic, 23.4% subspecialist visit, 18.3% visit to the emergency room, 13.8% hospitalization). Moreover, records about outcomes occurred mainly concerned the family (i.e. 31.2% advice on home management, 24.1% meeting of immediate needs, questions and concerns, 14.5% advocacy).

Table 17 – SpeNK-FP Study: Prevented Outcomes

Prevented Outcomes	**No. Recorded, n (%)***
Emergency Room visit	113 (18.3%)
Subspecialist visit	144 (23.4%)
Hospitalization	85 (13.8%)
Visit to Pediatric Office/Clinic	150 (24.4%)
Lab / X-ray	63 (10.2%)
Drugs	52 (8.4%)
Other	9 (1.5%)

*Total 616 recorded outcomes for 286 encounters

Table 18 – SpeNK-FP Study: Occurred Outcomes

Occurred Outcomes	**No. Recorded, n (%)***
Advised family /patient on home management	282 (31.2%)
Referral to Emergency Room	4 (0.4%)
Referral to subspecialist	22 (2.4%)
Referral for hospitalization	6 (0.7%)
Referral for pediatric sick office visit	11 (1.2%)
Referral to lab/X-ray	22 (2.4%)
Use of drugs	66 (7.3%)
Ordered equipment, diapers, transports	7 (0.8%)
Reconciled discrepancies	21 (2.3%)
Reviewed labs, specialist reports, *PAI*, etc.	109 (12.0%)
Advocacy for family/patient	131 (14.5%)
Met family's immediate needs, questions, concerns	218 (24.1%)
Unmet needs	0
Other	6 (0.7%)

* Total 905 recorded outcomes for 363 encounters

4.4. Discussion

This study aimed to assess the care coordination activities performed by the FPs for children and newborns with special health care needs in the pediatric primary care setting. To achieve this aim, we contacted FPs of children recruited to SpeNK Project and asked them to complete the SpeNK-FP at each counter regarding the child during 9 months after hospital discharge.

Our first finding is about the scant participation of FPs. In fact, only 65.6% of FPs recorded at least one contact, with 2 FPs recording only one encounter, regarding the 60% of subjects (49/82).

In Italian healthcare system, FPs are not required to record their activities and are remunerated on a capitation base/formula. On the contrary, in other countries (e.g. U.S.A.), they have to code and bill their activities to get the reimbursement by the government. This could partially explain the low compliance of our FPs. However, a study by Agrawal et al. (2012) found that the issues of time, reimbursement, billing and coding are perceived as significant barriers to care children and youth with special health care needs in a large sample of FPs in Illinois. This suggests that, in general, making FPs' care coordination activities documentable is complex and far away from being "measurable, auditable, and amenable to continuous quality improvement" (Antonelli et al., 2008). Moreover, the low compliance in the use of the online window could be explained by the age and gender of FPs involved. In fact, older people (55-74 years, representing 67.5% of our sample) as well as women (87.5%) tend to have lower digital competence (https://ec.europa.eu/digital-agenda/sites/digital-agenda/files/KKAH12001ENN-chap5-PDFWEB-5.pdf). Lastly, our study involves only FPs directly in charge of proving primary care to the children, whereas the study by Antonelli et al. (2008) involved the office staff of the practice. This could have implied a higher workload for FPs in their daily routine activities representing a barrier to the compliance.

Encounters were recorded by FPs across 3 patient complexity levels, with patients with mainly health needs (Level 2) representing the majority of the sample and receiving the majority of the encounters and time spent. Patients with both health and social needs (Level 3) received only 19% of all encounters but the time spent was less than 5 minutes in a quarter of cases.

This result is in contrast with the findings of the study by Antonelli et al. (2008), who argue that the presentation of an acute, family-based social stressor demands the provision of significant care coordination services. We wonder if, in our study population, the presence of social and familial problems adversely affects the contact with appropriate services (i.e. the family pediatrician as primary care provider), as suggested by the literature about the barriers to health services use and access in vulnerable groups (Ensor & Cooper, 2004; Jacobs, Ir, Bigdeli, Annear, & Van Damme, 2012; Scheppers, van Dongen, Dekker, Geertzen, & Dekker, 2006).

Focus of encounters indicates that the FP's role is mainly clinical (i.e. visits, developmental/ behavioral, growth/nutrition) and to a lesser extent of coordination (i.e. referral management, integrated home care, social services). In more than 40% of encounters FPs reported that there was no need for care coordination.

This result indicates that the role in coordination of care for children with special health care needs is played by FPs less than suggested by the recent literature on the topic (American Academy of Pediatrics, 2005; Antonelli et al., 2008; Boudreau et al., 2014; Gupta, O'Connor, & Quezada-Gomez, 2004). This finding is congruent with our recent qualitative study about the continuity of care for children with special health care needs, in which parents reported mixed perceptions and experiences about FPs, related to their centrality vs. marginality in the activation and coordination of the healthcare network (Zanello et al., 2015). The low level of care coordination activities as focus of the encounters could be explained by the lack of specific training and preparation about care coordination tasks (Stille & Antonelli, 2004), care of different conditions (Agrawal et al., 2013), education related to chronic illness and information about community resources (Liptak & Revell, 1989).

In most cases, the FP is involved in care coordination alone (over 50% of cases), accordingly to the recent literature about care coordination which identifies the primary care physician as principal provider of care coordination (American Academy of Pediatrics, 2005).

Among activities implemented to meet the patient’s needs, more than half included contacts with healthcare professionals and services. This result fits with the working definition developed by McDonald et al. (2014) stating that “organizing care involves the marshalling of personnel and other resources needed to carry out all required patient care activities and is often managed by the exchange of information among participants responsible for different aspects of care”.

According to the FPs’ subjective appraisals on prevented vs. occurred outcomes, 75% of recorded encounters prevented inappropriate service use, while most occurred outcomes concerned the family (i.e. advices on home management, advocacy, meeting family’s immediate needs, questions, concerns). Similarly to the study by Antonelli et al. (2008), the activities performed by FPs in care for children with special health care needs may prevent the higher cost of resources.

Our study has two main limitations. First, the small sample size and the low compliance of FPs do not allow to generalize our findings to the pediatric primary health care system. Second, the poor quality of data recorded about time spent does not permit to calculate the amount of time spent for specific care coordination activities, neither to estimate the costs related to the care of children with special health care needs.

Nevertheless, our study is the first in Italy examining and measuring with a standardized instrument the activities performed by FPs to care for children with special health care needs and to coordinate the care provided by multiple services and professionals. However, the enhancement of the assessment method could help in identifying areas of improvement in care coordination for children with special health care needs as concerns the role of FP.

4.5. Conclusion

The improvement of measurement, assessment and definition of FP's role in care coordination is a challenge for healthcare system and deserves attention to provide support and meet needs of children with special health care and their families. Nowadays, the activities performed by FPs in clinical practice with children with special health care needs showed that their role in care coordination is less central than advised by the recommendations for health care organization for this population.

Chapter 5. Utilization of healthcare resources

Abstract

Introduction. Although children with special health care needs represent a category with a low prevalence, they are major contributor to inpatient utilization, requiring integrated health care programs that involve primary, community, hospital and tertiary care services. The SpeNK (Special Needs Kids) Project aimed to examine resource utilization for newborns and children with special health care needs living in Bologna province. **Methods**. Newborns and children with special health care needs were recruited to SpeNK Project at discharge from 3 hospital facilities in Bologna province, from October 1st 2012 to September 30th 2014. Data collected for each subject were linked to administrative data extracted from Regional Healthcare System databases, referring to a 6-9 months period after hospital discharge, including both inpatient and outpatient healthcare services. Data were analyzed to assess resource utilization and used to develop an algorithm (SpeNK-A) to identify children "with special healthcare needs" on the basis of hospital discharge records. **Results**. The analyses showed a higher level of utilization of health services in infants enrolled to SpeNK Project compared with pediatric population, including a higher number of hospitalizations and hospital days, a more frequent use of specialty outpatient services for diagnostics, therapy and visits and almost no difference in the number of emergency room contacts. The SpeNK-A algorithm allowed to classify 16.5% of newborns discharged from the regional hospital facilities in the interval of interest as "having special health care needs", referring to 6 groups based on diagnoses in hospital discharge records: (1) Newborns with birth weight lower than 1000 gr., (2) Conditions associated with the extreme prematurity of newborn, (3) Malformations and other congenital diseases, (4) Irreversible diseases, (5) Neurological and metabolic degenerative diseases, (6) Neoplasms. **Conclusions.** Our findings confirm a higher level of utilization of inpatient services by children with special health care needs with no significant differences in Emergency Room visits, and some differences in the use of outpatient specialty services.

Keywords: *children with special healthcare needs, integrated health care pathway, care coordination, healthcare resources utilization*.

5.1. Background

Although newborns and children with complex or chronic health conditions represent a category with a low prevalence, a small cohort of recurrently readmitted pediatric patients is a major contributor to inpatient utilization, using a substantial amount of healthcare resources (Berry et al., 2011). Children with special health care needs are medically fragile and require frequent hospitalizations and Emergency Department visits even with improved primary care (Berman et al., 2005). Preventable and potentially preventable hospitalizations may be avoided with effective clinical diagnosis, treatment and patient education, with the active participation of patients in their care and adoption of healthy lifestyle behaviors (Kruzikas, 2004). Moreover, careful preparation of discharge and good follow-up after discharge may reduce risks of multiple hospital readmissions and the exposure to the risks of hospital acquired morbidity (American Academy of Pediatrics, 2008). Appropriate discharge planning and effective communication of hospital providers with patients, their families, and outpatient providers may be an important protective factor to minimize readmission (Kripalani, Jackson, Schnipper, & Coleman, 2007). To manage the transition of children with special health care needs from hospital to home, communication, proactive care planning with the involvement of the primary care provider and follow-up appointments with multiple outpatient specialty providers, equipment specialists, and home care nurses are required. These activities must take into account important differences in the situation and needs of child population compared with adults, concerning their developmental status and change during childhood and teen years, the critical mediating role played by parents between the children and their needs and the health care services, and the impact of the school context on shaping children’s social and health needs (Miller et al., 2009).

In Italy, the quality of services for children with special health care needs varies widely across regions. High-quality models foster integration across a continuum of care, including primary care providers (i.e. family pediatricians), hospitals and referral centers, with social and educational support guaranteed by specific agreements between health authorities, municipalities, and school authorities. To ensure the delivery of care at home as much as possible, the pediatric home care service (i.e. *Assistenza Domiciliare Pediatrica*), guaranteed by Local Health Authorities, connect specialist centers, community services and family pediatricians, in particular for patients requiring parenteral feeding, oxygen therapy, physiotherapy, etc. (Wolfe et al., 2013).

In Emilia-Romagna Region the Special Needs Kids (SpeNK) Project has been designed to describe the implementation of existing sheltered hospital discharge procedures and integrated clinical pathways for children with complex or chronic health conditions and special healthcare needs in Bologna province, to assess their utilization of healthcare resources, the family's perspective on continuity of care and the role of family pediatrician.

The primary aim of this study was to examine the utilization of both inpatient and outpatient healthcare services by children recruited to SpeNK Project during 6-9 months after the hospital discharge, through the data linkage with Regional Health Service databases. Secondary aims were (1) to compare SpeNK Children with pediatric population with respect to the use of healthcare services, and (2) to test an algorithm to categorize children with special health care needs on the basis of administrative data included in the hospital discharge record.

5.2. Material and Methods

5.2.1. Setting

Emilia-Romagna is a region in Northern Italy with around 4.4 million inhabitants, who receive health care by the Regional Health Service (RHS), which includes 8 Local Health Trusts (LHTs), 4 University Hospitals (UHs), 1 Hospital Trust and 4 Research Hospitals (IRCCS). Within each LHA, Family Pediatricians (FPs) are responsible for providing primary care in outpatient and home settings to children 0-14 years (16 years for specific conditions). They are responsible for assessing patients' needs, ordering diagnostic procedures, prescribing drugs, and referring patients to specialists and hospitals (Luciano et al., 2014). In accordance with the National Collective Agreement (2010) and the Regional Integrative Agreement (2011), FPs should participate in an integrated network of services, to improve the care of children with chronic conditions and the government of healthcare pathways, as "process owners" as regards the management of the child's health condition, especially with special needs. For children with complex social and/or healthcare needs, hospital facilities in Bologna Province adopt the "sheltered discharge" procedure, in order to promote the integration of healthcare services (hospital, specialist, primary and community care providers) across the transition from hospital to home setting.

5.2.2. SpeNK Sample and study population

SpeNK Sample consisted of children and families recruited at hospital discharge of the child from the participating units of the hospital facilities at the University Hospital of Bologna (St. Orsola Malpighi) and the two local Health Authorities of Bologna and Imola. The recruitment of children was conducted from October 1st 2012 to September 30th 2014 on incident cases meeting the following inclusion criteria: age from 0 to 16 years, residence in Bologna province, and the presence of at least one of the following conditions:

- Birth weight <1000 g;
- Complex and/or chronic health conditions defined as:
    - Need for technological assistance,
    - Acute neurological deficit,
    - Severe endocrinopathy,
    - Complex genetic malformative pathology;
- Children with oncological diseases who need palliative care or particular community care;
- Newborns with mothers in contact with mental health services or drug addiction

Only first ever hospitalizations for the condition of interest were included. Written informed consent was obtained at recruitment from each parent to collect clinical data on children.

The study population consisted in newborns and children aged 0-17 years resident in Emilia Romagna who were discharged between 1st October 2012 and 30th September 2014 from a hospital facility of the Regional Healthcare Service (RHS).

5.2.3. Data source and procedures

5.2.3.1. Utilization of healthcare resources

Administrative data about study population, including SpeNK sample, were extracted from RHS databases, referring to newborns and children aged 0-17 years, resident in Emilia Romagna Region, who were discharged from regional hospital facilities from 1st October 2012 and 30th September 2014. Databases included: hospital discharge records (SDO), specialty outpatient services (ASA), emergency room contacts (PS), home care services (ADI), certificates of birth attendance (CeDAP), death certificates (REM).

For subjects from SpeNK sample, administrative data were linked to data collected at recruitment. Through the data linkage with RHS administrative databases, subjects enrolled in SpeNK Sample were tracked to examine their utilization of healthcare resources within 6-9 months after hospital discharge.

To compare the utilization of healthcare resources between children with special health care needs and pediatric population, we selected two subgroups of SpeNK Sample and our study population. The comparison focused on SpeNK newborns with birth weight lower than 1000 gr.

(i.e. Extremely Low Birth Weight - ELBW), representing the majority of our sample, vs. newborns admitted at birth in the three hospital facilities of Bologna University Hospital and Bologna and Imola LHAs in the period of interest. All infants admitted at birth in the interval of interest in the three hospital facilities were selected and divided in three group referring to their health condition as recorded in the hospital discharge records (SDO). In accordance with an evaluation of a Pediatrician with the SpeNK Project Team, Group 0 included "healthy" newborns who were discharged to home with a record for Diagnosis Related Group (DRG) 391 (i.e. healthy newborn) and 391 (i.e. newborn with other significant disorders); Group 1 included ELBW newborns enrolled in SpeNK sample; Group 2 included newborns with intermediate conditions, excluded from other groups.

5.2.3.2. Development of SpeNK-A Algorithm

The SpeNK-A algorithm was based on the diagnostic codes and other variables recorded in the hospital discharge records (SDO), in order to categorize children with special health care needs in different groups. It was developed on the basis of an preliminary study by Abate et al. (2014) (unpublished) on palliative pediatric care in Emilia Romagna Region, which used a system to classify conditions requiring palliative care into four categories (Himelstein, Hilden, Boldt, & Weissman, 2004). In order to identify children with serious illness through administrative data, they associated specific ICD9CM diagnostic codes to 4 categories: (1) Conditions for which curative treatment is possible but may fail (e.g. advanced or progressive cancer or cancer with a poor prognosis, complex and severe congenital or acquired heart disease); (2) Conditions requiring intensive long-term treatment aimed at maintaining the quality of life (e.g. cystic fibrosis, muscular dystrophy), (3) Progressive conditions in which treatment is exclusively palliative after diagnosis (e.g. progressive metabolic disorders), (4) Conditions involving severe, non-progressive disability, causing extreme vulnerability to health complications (e.g. extreme prematurity, severe cerebral palsy with recurrent infection or difficult-to-control symptoms).

The SpeNK-A Algorithm derived from this study and was further developed to extend its applicability to children with other special health care needs, not limited to palliative care. The integration, further selection and categorization of diagnostic codes and variables into groups was performed by a Pediatrician with the SpeNK Project Team. The SpeNK-A algorithm classifies children with special health care needs into 6 groups: (1) Neoplasms, (2) Malformations and other congenital diseases (e.g. heart disease, renal abnormalities with organ failure), (3) Irreversible diseases (e.g. cerebral palsy, disability for brain damage and/or bone marrow), (4) Neurological and metabolic degenerative diseases, (6) Extremely low birth weight newborns, (5) Conditions associated with the extreme prematurity of newborn (e.g. encephalopathy, NEC). The Appendix - SpeNK-A Algorithm lists the diagnostic codes ICD9CM and variables referred to each group. To test the SpeNK-A algorithm we chose to consider newborns admitted at birth (as incident case) and discharged from regional hospital facilities in the interval of interest.

5.2.3.3. Ethical considerations

Parents recruited in SpeNK sample gave their written informed consent to the collection of clinical data of their children for SpeNK Project. The study protocol of SpeNK Project was approved by the Ethics Committees of the Bologna University Hospital and of the Bologna and Imola Local Health Authorities (LHAs).

All activities concerning the use of administrative data about the study population were conducted in accordance with Italian law on privacy (articles 20-21, D.Lgs. 196/2003) and in compliance with the principles of confidentiality and anonymity. The Regional Statistical Offices encrypted data and assigned to each subject unique identifier, which does not allow tracing the patient’s identity and other sensitive data. When encrypted administrative data are used to inform health care planning activities, studies are exempt from notification to the Ethics Committee, and no specific written consent is needed to use patient information stored in the hospital databases.

5.2.4. Statistical Analyses

IBM SPSS Statistics (version 20, Chicago, USA) was used for the statistical analyses. T-test for independent samples with pair-wise comparison and ANOVA were used to compare groups.

5.3. Results

5.3.1. SpeNK sample and study population

At the end of a 24-months period, a sample of 82 children was recruited (see Table 19), including a majority of preterm newborns with birth weight lower than 1000 gr. (42.7%) or children with other complicating conditions (12.2%). Children were male in 53.7% (n=44) of cases, had a mean age of 6.2 (±13.6, range 0-78) months at the time of recruitment, and in 78% of cases (n=64) were recruited within 28 days of age for an hospital admission related to birth, and after 28 days from birth in other 28% of cases (22%). Fifty-one (62.2%) cases were recruited at St. Orsola Malpighi University Hospital, 19 (23.2%) at Maggiore Hospital and 12 (14.6%) at Imola Hospital. A sheltered hospital discharge was attended to for about 65% of children. According to data collected at recruitment, the home care services were activated for 54 children (65.9%).

Considering 3 couples of twins, the sample comprised 79 families, including Italian parents in 40 cases (50.6%), one Italian parent in 14 cases (17.7%), and no Italian parents in 25 cases (31.6%). Thirty-six (45.6%) mothers and 29 fathers (36.7%) had foreign citizenship.

Table 19 – SpeNK Sample: Characteristics of Children (n=82)

Gender, Female; n (%)	38 (46.3%)
Age, months; mean (±SD, range)	6.2 (±13.6, range 0-78)
Cause of inclusion; n (%)	
Prematurity < 1000 gr	35 (42.7%)
Complex genetic / malformative pathologies	14 (17.1%)
Prematurity with other conditions	7 (8.5%)
Encephalopathy / Neuropathy	8 (9.8%)
Other complicating conditions	10 (12.2%)
Social Problems	8 (9.8%)
Discharge procedure; n (%)	
Standard	29 (35.4%)
Sheltered	53 (64.6%)
Care services activation	
Community care services; n (%)	70 (85.4%)
Home Health Devices; n (%)	18 (22.0%)
Individualized Care Plan; n (%)	48 (58.5%)
Home Care Services; n (%)	54 (65.9%)
Follow-up; n (%)	78 (95.1%)
Hospital Unit of recruitment; n (%)	
St. Orsola Malpighi UH Bologna	*51 (62.2%)*
Pediatric Surgery Unit UH Bologna	7 (8.5%)
Neonatology Unit UH Bologna	36 (43.9%)
Child Neuropsychiatric Unit UH Bologna	1 (1.2%)
Pediatric Unit UH Bologna	2 (2.4%)
Pediatric Emergency Room UH Bologna	5 (6.1%)
Maggiore Hospital LHA Bologna	*19 (23.2%)*
Neonatology and NICU LHA Bologna	16 (19.5%)
Pediatric Unit LHA Bologna	3 (3.7%)
Hospital LHA Imola	*12 (14.6%)*

As for the population of interest, from 1st October 2012 and 30th September 2014, 194,885 children aged 0-17 years resident in Emilia Romagna were discharged from the hospital facilities of the Region (SDO). In 21.2% of cases (n=41,355) the hospital facility was in Bologna Province, with 4,254 (10.3%) discharges from Imola LHA, 12,978 (31.4%) from Bologna LHA, and 24,123 (58.3%) from Bologna UH (St.Orsola Malpighi). Excluding newborns born before the interval of interest (with a subsequent discharge reported in that period), the total number of children discharged from regional hospital facilities was 194,450, of which 41,239 (21.2%) were discharged from the hospital facilities of Imola and Bologna LHAs and of Bologna UH. In these three hospital facilities 15,455 children were admitted at birth in the interval of interest.

5.3.2. Utilization of healthcare resources

Through the data linkage with administrative databases of the RHS, it was possible to track subjects enrolled in SpeNK Sample and examine their utilization of healthcare resources within 9 months after hospital discharge (see Table 20). Mean values were calculated by using the number of cases (i.e. subjects who used at least once the healthcare service considered) as denominator. Children had 74 days of hospitalization at birth on average, with a difference between subjects enrolled within vs. after 28 days from birth (69.1 vs. 92.7). All children used inpatient hospital services within 9 months of discharge (with missing data for one subject). Within 9 months from hospital discharge, subjects enrolled after vs. within 28 days from birth had more hospital readmissions (2.2 vs. 1.2) (mean=1.4) and more days of hospitalization (28.4 vs. 18.3) (mean=20.4). Less than half of children had emergency room visits (39/82). As for outpatient specialty services, children enrolled within 28 days from birth received on average three times more laboratory services (12.2 vs. 3.0), less services for therapy (4.3. vs. 7.7) and more services for diagnostics, rehabilitation, specialist visits, compared to others. Within 6 months after discharge, the home care service (i.e. ADI) was activated for 22% of children (18/82), with a majority of children who were enrolled after 28 days from birth.

Table 20 – SpeNK Sample: Utilization of healthcare resources within 9 months from hospital discharge

	Subjects enrolled ≤ 28 days from birth		Subjects enrolled > 28 days from birth		Total SpeNK Sample	
Utilization of healthcare resources	Mean	Cases/Total (%)	Mean	Cases/Total (%)	Mean	Cases/Total (%)
Days of hospitalization at birth, including birth event and transfers	69.1	64/64 (100%)	92.7	17/18 (94.4%)*	74.0	81/82 (98.8%)*
Days of hospitalization during hospital readmissions within 9 months from discharge	24.9	47/64 (73.4%)	28.4	17/18 (94.4%)*	20.4	64/82 (78.0%)*
Number of Emergency Room visits within 9 months from discharge	2.3	37/64 (57.8%)	4.0	2/18 (11.1%)	2.4	39/82 (47.6%)
Number of specialty outpatient services for Diagnostics within 9 months from discharge	5.6	60/64 (93.8%)	4.4	14/18 (77.8%)	5.4	74/82 (90.2%)
Number of specialty outpatient services for Laboratory within 9 months from discharge	12.5	42/64 (65.6%)	3.0	1/18 (5.6%)	12.2	43/82 (52.4%)
Number of specialty outpatient services for Rehabilitation within 9 months from discharge	2.6	29/64 (45.3%)	-	0/18 (0.0%)	2.6	29/82 (35.4%)
Number of specialty outpatient services for Therapy within 9 months from discharge	4.3	25/64 (39.1%)	7.7	3/18 (16.7%)	4.6	28/82 (34.1%)
Number of specialty outpatient services for Visits within 9 months from discharge	6.1	61/64 (95.3%)	5.3	15/18 (83.3%)	5.9	76/82 (92.7%)
Subjects with ADI Home Care service activated within 6 months from discharge		8/64 (12.5%)		10/18 (55.6%)		18/82 (22.0%)

*Missing n=1 (one subject recruited for social and familial issues, who had no hospital discharge record at birth and no hospital readmissions recorded in the hospital facilities considered)

To compare the use of healthcare resources between children with special health care needs and pediatric population we considered ELBW infants enrolled in SpeNK Sample and newborns admitted at birth and discharged in the three selected hospital facilities from 1st October 2012 to 30th September 2014. The categorization of children (n=15,454, excluding one child who died in the interval) in 3 groups referring to the information in the hospital discharge record resulted in 13,370 healthy newborns included in "Group 0", 37 ELBW newborns of SpeNK Sample included in "Group 1", and 2,047 newborns with intermediate conditions included in "Group 2".

Table 21 shows the comparison between groups with respect to the utilization of inpatient and outpatient healthcare services. Mean values were calculated using as denominator the number of cases (i.e. subjects who used at least once the healthcare service considered). The analysis of data showed a higher level of utilization of health services for Group 1 and 2 compared with Group 0. This entailed a significant higher number of hospital days in readmissions, a more frequent use of specialty outpatient service for diagnostics, therapy and visits than for laboratory and rehabilitation and almost no differences in the number of emergency room contacts.

Table 21 - Utilization of healthcare resources within 9 months from discharge: Comparison between groups

Utilization of healthcare resources	**GROUP 0** **Subjects with DRG 390-391 and discharge to home; Mean (cases/total; %)**	**GROUP 1** **ELBW newborns enrolled in SpeNK Sample; Mean (cases/total; %)**	**GROUP 2** **Other Subjects with intermediate conditions; Mean (cases/total; %)**
Days of hospitalization at birth, including birth event and transfers	2.5 (13,370/13,370; 100.0%)	88.9 (37/37; 100.0%)	11.3 (2,047/2,047; 100.0%)
Days of hospitalization in subsequent hospitalizations within 9 months from discharge	5.0* (1726/13,370; 12.9)	11.5* (27/37; 73.0%)	10.6* (532/2,047; 26.0%)
Number of Emergency Room visits within 9 months from discharge	1.5 (6,436/13,370; 48.1%)	1.9 (21/37; 56.8%)	1.6 (1,007/2,047; 49.2%)
Number of specialty outpatient services for Diagnostics within 9 months from discharge	1.8 (9,188/13,370; 68.7%)	5.8 (37/37; 100%)	2.7 (1,545/2,047; 75.5%)
Number of specialty outpatient services for Laboratory within 9 months from discharge	7.9 (1,341/13,370; 10.0%)	9.4 (29/37; 78.4%)	10.1 (374/2,047; 18.3%)
Number of specialty outpatient services for Rehabilitation within 9 months from discharge	1.6 (99/13,370; 0.7%)	2.3 (19/37; 51.4%)	2.4 (116/2,047; 5.7%)
Number of specialty outpatient services for Therapy within 9 months from discharge	1.7 (373/13,370; 2.8%)	3.5 (19/37; 51.4%)	2.9 (54/2,047; 2.6%)
Number of specialty outpatient services for Visits within 9 months from discharge	1.8 (3,111/13,370; 23.3%)	6.0 (37/37; 100.0%)	2.6 (924/2,047; 45.1%)

*Only comparison between Group 0 and Group 1, and between Group 0 and Group 2 were significant ($p<0.05$)

5.3.3. Application of SpeNK-A algorithm

Table 22 provides the results of SpeNK-A application on newborns born and discharged from regional hospital facilities (n=194,450) and from the hospital facilities of Imola and Bologna LHAs and of Bologna UH (n=41,239, 21.2%) from 1st October 2012 to 30th September 2014. Results indicate an estimate of 16.5% of children classified overall as "children with special needs", with a majority of malformation and other congenital diseases (Group 2).

Table 22 - SpeNK-A: Application of SpeNK-A Algorithm to regional population of newborns

SpeNK-A Group	Region, N (%)	3 Hospital Facilities, N (%)
None	162,389 (83.5)	34,453 (83.5)
Group 1: Neoplasms	6,458 (3.3)	1,820 (4.4)
Group 2: Malformations and other congenital diseases	18,342 (9.4)	3,669 (8.9)
Group 3: Neurologic and Metabolic Degenerative Diseases	1,958 (1.0)	585 (1.4)
Group 4: Irreversible Pathologies	4,741 (2.4)	579 (1.4)
Group 5: Extremely low birth weight	318 (0.2)	76 (0.2)
Group 6: Pathologies connected to newborn extreme prematurity	244 (0.1)	57 (0.1)
Total	194,450 (100.0)	41,239 (100.0)

5.3. Discussion

This study examined the utilization of healthcare services by children recruited to SpeNK Project within 9 months after their hospital discharge, through the analyses of administrative data extracted from the Regional Health Service databases. A focus on infants born in three hospital facilities of University Hospital of Bologna and of Local Health Authorities of Bologna and Imola during recruitment period allowed to compare the healthcare services used by a subgroup of SpeNK Children vs. pediatric population. An algorithm (SpeNK-A) was developed and tested on pediatric population to categorize children with special health care needs on the basis of administrative data included in the hospital discharge record.

The recruitment to SpeNK Project, from 1st October 2012 and 30th September 2014, in the hospital facilities of University Hospital of Bologna and Local Health Authorities of Bologna and Imola, resulted in a sample of 82 children with special health care needs. Our sample included a majority of preterm newborns with birth weight < 1000 gr. (42.7%), confirming that low birth weight infants can be considered a specific subgroup of children with special health care needs (McPherson et al., 1998). Low birth weight newborns (<2500 gr.) constitute about 6% of all newborns (Kowlessar et al., 2006) and are at increased risk of chronic conditions and of poor neurodevelopment (Gibertoni et al., 2015; Saigal & Doyle, 2008).

On the basis of information collected at recruitment, children recruited to our sample had a sheltered discharge procedure activated in almost 65% of cases, an hospital follow-up planned in majority of cases (over 90%) and community service activated in approximately 85% of cases, indicating a strong activation of a network of healthcare providers for these patients. Conversely, it is noteworthy that at recruitment the activation of home care service (i.e. ADI) was reported for 54/82 children, whereas the administrative data showed that, within 6 months from hospital discharge, the effective activation of the service occurred only for 18/82 children. This result may indicate a weakness in the health care network concerning the effective implementation of a service aiming to help parents and families to take care of their child at home as much as possible.

According to the analyses of administrative data, children recruited to our sample had 74 days of hospitalization at birth on average, with subjects enrolled after 28 days from birth hospitalized for about 23 days more. All children used inpatient services within 9 months of discharge, which is in accordance with what is known in literature about this population and its needs (Berman et al., 2005; Berry et al., 2011; Kripalani et al., 2007). Within 9 months from hospital discharge, subjects enrolled after 28 days from birth had more hospital readmissions and more related days of hospitalization, compared with those enrolled within 28 days. The recruitment to SpeNK Project involved the incident cases, meaning subjects with their first hospitalization for the condition meeting the inclusion criteria. Therefore, children enrolled within vs. after 28 days from birth have been identified as having “special health care needs” in different moments, apparently at birth or later. The slight differences in the use of outpatient specialty services between children with special health care needs diagnosed at birth vs. later, with first receiving more services for diagnostics, rehabilitation, specialist visits and many more laboratory services, and second receiving more services for therapy, may indicate different needs. This finding could be explained by different trajectories, as discussed by Quach, Jansen, Mensah, and Wake (2015), who identified four distinct “special health care needs” trajectories (i.e. persistent, emerging, transient, none), in a longitudinal cohort study on Australian children.

Moreover, the emergency room service pertained to less than half of children recruited to SpeNK sample, and entailed no significant differences between extremely low birth weight newborns of SpeNK sample and other newborns. This finding is in contrast with international literature, which indicates that children with special health care needs require frequent Emergency Department visits (Berman et al., 2005). This could be explained referring to the activation of a proactive healthcare network, helping families in care of their children with appropriate support of community services and primary care providers. A study by Ralston, Harrison, Wasserman, and Goodman (2015) about the hospital variation in health care utilization by children with medical complexity found that office and emergency visits varied to a lesser extent than inpatient and intensive care days. These results could be reflective of a high quality level in care for children with special health care needs provided by the Regional Healthcare Service, which includes specific procedures to manage the transition from hospital to home and the presence of a family pediatrician providing primary care with a specific role as “process owner” for care of these children. Further studies could explore if and in what extent the activation of a sheltered discharge procedure by the hospital impact on the utilization of emergency healthcare services.

Finally, the application of SpeNK-A algorithm resulted in an estimate of 16.5% of children “with special health care needs” among infants born and discharged from the regional hospital facilities from 1st October 2012 to 30th September 2014. This estimate is in accordance with international literature on the topic, despite the poorness of data at national level. Newacheck and Kim (2005) cite data from the 2000 Medical Expenditure Panel Survey (MEPS) in United States, according which 15.6% of children were identified as children with special health care needs. An estimate of the National Survey of Children with Special Health Care Needs Chartbook 2009–2010 in United States indicated that about 15% of children under 18 years of age have special health care needs and 23% of households with children have at least one child with special health care needs (US Dept of Health and Human Services, 2013). However, the selection of our study population implies the criterion of having been admitted to the regional hospital facilities, which excludes any infants born elsewhere (e.g. child born at home, or in facilities outside the region).

Our study is the first study on national territory to describe the utilization of healthcare resources by children with special health care needs on the basis of administrative data. Anyway, our study has one main limitation, that is the limited size of the SpeNK Sample. Further studies are needed to better understand the trajectories of special health care needs in pediatric population and the related use of healthcare services. Moreover, the further development of the SpeNK-A algorithm, in order to identify children with special health care needs on the basis of hospital administrative data, may represent an opportunity for targeting health care plans, improving quality and reducing health care costs.

5.4. Conclusion

This study confirms a higher level of utilization of health services of children with special health care needs compared with pediatric population, with significant differences in inpatient service, no significant differences in Emergency Room visits, and different frequencies in the use of outpatient specialty services according to the moment of recruitment, maybe due to different trajectories in children’s special health care needs, which deserve further investigation.

Conclusions

The broad focus of the SpeNK Project allowed to address different issues concerning children with special health care needs, including the current procedures and practices for their healthcare, the perspective of families, the role of the family pediatricians and the utilization of healthcare resources and services by this population.

The review of organizational procedures emphasized some variability among the practices and procedures in the healthcare of children with special needs in Bologna province. Despite a common interest in the implementation, organization and coordination of care for children with special health care needs, especially in the transition from hospital to community setting, procedures and practices need to be further validated and standardized.

The study of family perspectives with two different approach consented to explore first their experiences and perceptions with qualitative methods (Zanello et al., 2015) and then to develop a quantitative measure of continuity of care (Rucci et al., 2015). Parents’ narratives indicate that continuity of care and parent empowerment are important, with different issues from hospital setting to home care. The use of a quantitative measure allowed to identify some improvement areas related to the information continuity and care coordination and an array of professionals and services variously involved in care of their children. The care coordination seems to be controlled more by the hospital professionals, who provide follow-up care, than by the primary care provider. Overall, the study of parents’ perspectives demonstrated that families could be valuable partners in assessing the health care services provided to their children and could be consulted, by using validated and reliable instruments, to identify areas of care improvement and to outline suggestions for redesigning services, within an “Experience Based Design” approach (Cleveland, 2008).

The focus on family pediatricians highlighted their variable attitude in coordinating care of children with special healthcare needs, consistently with families’ perceptions, and a difficulty in recording their activities. It seems that their role as care coordinators for children with special health care needs, already formally defined at regulatory level (e.g. 2010 Collective National Agreement), needs to be more practically defined and implemented, beyond the individual attitude of the single professional to perform care coordination.

Lastly, the analyses of administrative data showed a higher level of utilization of health services of infants with special health care needs compared with pediatric population, mainly for inpatient service use. The low use of Emergency Room service could reflect the presence of a proactive healthcare network, which involves hospital services from the organization and management of sheltered discharge procedure to follow-up care, community services to support care at home and the family pediatrician as primary care provider. The differences in the use of outpatient specialty services according to the moment of the first identification of a special need (at birth or later) may be explained with different trajectories in children's special health care needs (Quach et al., 2015).

Overall, the results of the SpeNK Projects highlight that there is still some variability in continuity and coordination of care for children with special health care needs in Bologna province, although the integration and collaboration within the healthcare network are major goals according to regional and national recommendations.

In summary, the SpeNK Project represents a first step in describing and assessing the health care provided to children with special health care needs, based on a multi-method approach and with the involvement of different stakeholders (i.e. patients and their families, professionals, organizations). It suggests that the integration of multiple perspectives is useful to describe and evaluate healthcare pathways. The connection of viewpoints of users, professionals and systems (McDonald et al., 2014) could facilitate the identification of improvement potential into care for these children and help reduce the risk of fragmentation and discontinuity within the healthcare pathway.

References

Abate, M. E., Corvaglia, L. T., Di Gennaro, G., Melotti, R. M., Rosa, S., & Valentini, O. (2014). *La Rete delle cure palliative pediatriche.* (Master di 2° livello in Funzioni Direttive e Gestione dei Servizi Sanitari), Alma Mater Studiorum University of Bologna. (8546)

Agrawal, R., Shah, P., Zebracki, K., Sanabria, K., Kohrman, C., & Kohrman, A. F. (2012). Barriers to care for children and youth with special health care needs: perceptions of Illinois pediatricians. *Clin Pediatr (Phila), 51*(1), 39-45. doi: 10.1177/0009922811417288

Agrawal, R., Shah, P., Zebracki, K., Sanabria, K., Kohrman, C., & Kohrman, A. F. (2013). The capacity of primary care pediatricians to care for children with special health care needs. *Clin Pediatr (Phila), 52*(4), 310-314. doi: 10.1177/0009922813476572

Aller, M. B., Vargas, I., Garcia-Subirats, I., Coderch, J., Colomes, L., Llopart, J. R., . . . Vazquez, M. L. (2013). A tool for assessing continuity of care across care levels: an extended psychometric validation of the CCAENA questionnaire. *Int J Integr Care, 13*, e050.

American Academy of Pediatrics. (2002). The Medical Home. *Pediatrics, 110*(1), 3.

American Academy of Pediatrics. (2005). Care coordination in the medical home: integrating health and related systems of care for children with special health care needs. *Pediatrics, 116*(5), 1238-1244. doi: 10.1542/peds.2005-2070

American Academy of Pediatrics. (2008). Hospital discharge of the high-risk neonate. *Pediatrics, 122*(5), 1119-1126. doi: 10.1542/peds.2008-2174

Ames, K. E., Rennick, J. E., & Baillargeon, S. (2011). A qualitative interpretive study exploring parents' perception of the parental role in the paediatric intensive care unit. *Intensive Crit Care Nurs, 27*(3), 143-150. doi: 10.1016/j.iccn.2011.03.004

Antonelli, R. C., Stille, C. J., & Antonelli, D. M. (2008). Care coordination for children and youth with special health care needs: a descriptive, multisite study of activities, personnel costs, and outcomes. *Pediatrics, 122*(1), e209-216. doi: 10.1542/peds.2007-2254

Aujoulat, I., d'Hoore, W., & Deccache, A. (2007). Patient empowerment in theory and practice: polysemy or cacophony? *Patient Educ Couns, 66*(1), 13-20. doi: 10.1016/j.pec.2006.09.008

Babyak, M. A., & Green, S. B. (2010). Confirmatory factor analysis: an introduction for psychosomatic medicine researchers. *Psychosom Med, 72*(6), 587-597. doi: 10.1097/PSY.0b013e3181de3f8a

Bellin, M. H., Osteen, P., Heffernan, C., Levy, J. M., & Snyder-Vogel, M. E. (2011). Parent and health care professional perspectives on family-centered care for children with special health care needs: are we on the same page? *Health Soc Work, 36*(4), 281-290.

Berman, S., Rannie, M., Moore, L., Elias, E., Dryer, L. J., & Jones, M. D., Jr. (2005). Utilization and costs for children who have special health care needs and are enrolled in a hospital-based comprehensive primary care clinic. *Pediatrics, 115*(6), e637-642. doi: 10.1542/peds.2004-2084

Berry, J. G., Hall, D. E., Kuo, D. Z., Cohen, E., Agrawal, R., Feudtner, C., . . . Neff, J. (2011). Hospital utilization and characteristics of patients experiencing recurrent readmissions within children's hospitals. *JAMA, 305*(7), 682-690. doi: 10.1001/jama.2011.122

Boudreau, A. A., Perrin, J. M., Goodman, E., Kurowski, D., Cooley, W. C., & Kuhlthau, K. (2014). Care coordination and unmet specialty care among children with special health care needs. *Pediatrics, 133*(6), 1046-1053. doi: 10.1542/peds.2013-2174

Carr, V. L., Sangiorgi, D., Buscher, M., Junginger, S., & Cooper, R. (2011). Integrating evidence-based design and experience-based approaches in healthcare service design. *HERD, 4*(4), 12-33.

Cleveland, L. M. (2008). Parenting in the neonatal intensive care unit. *J Obstet Gynecol Neonatal Nurs, 37*(6), 666-691. doi: 10.1111/j.1552-6909.2008.00288.x

Colville, G., Darkins, J., Hesketh, J., Bennett, V., Alcock, J., & Noyes, J. (2009). The impact on parents of a child's admission to intensive care: integration of qualitative findings from a cross-sectional study. *Intensive Crit Care Nurs, 25*(2), 72-79. doi: 10.1016/j.iccn.2008.10.002

Davidoff, A. J. (2004). Identifying children with special health care needs in the National Health Interview Survey: a new resource for policy analysis. *Health Serv Res, 39*(1), 53-71. doi: 10.1111/j.1475-6773.2004.00215.x

Davidson, J. R., Brundage, M. D., & Feldman-Stewart, D. (1999). Lung cancer treatment decisions: patients' desires for participation and information. *Psychooncology, 8*(6), 511-520.

Diaz-Caneja, A., Gledhill, J., Weaver, T., Nadel, S., & Garralda, E. (2005). A child's admission to hospital: a qualitative study examining the experiences of parents. *Intensive Care Med, 31*(9), 1248-1254. doi: 10.1007/s00134-005-2728-8

Ensor, T., & Cooper, S. (2004). Overcoming barriers to health service access: influencing the demand side. *Health Policy Plan, 19*(2), 69-79.

Garland, R. (1991). The Mid-Point on a Rating Scale: Is it Desirable? *Marketing Bulletin, 2*, 66-70.

Gibertoni, D., Corvaglia, L., Vandini, S., Rucci, P., Savini, S., Alessandroni, R., . . . Faldella, G. (2015). Positive effect of human milk feeding during NICU hospitalization on 24 month neurodevelopment of very low birth weight infants: an Italian cohort study. *PLoS One, 10*(1), e0116552. doi: 10.1371/journal.pone.0116552

Gorusch, R. L. (1983). *Factor Analysis* (2nd ed.). Hillsdale, NJ: Lawrence Erlbaum Associates.

Gulliford, M., Cowie, L., & Morgan, M. (2011). Relational and management continuity survey in patients with multiple long-term conditions. *J Health Serv Res Policy, 16*(2), 67-74. doi: 10.1258/jhsrp.2010.010015

Gupta, V. B., O'Connor, K. G., & Quezada-Gomez, C. (2004). Care coordination services in pediatric practices. *Pediatrics, 113*(5 Suppl), 1517-1521.

Haggerty, J. L., Reid, R. J., Freeman, G. K., Starfield, B. H., Adair, C. E., & McKendry, R. (2003). Continuity of care: a multidisciplinary review. *BMJ, 327*(7425), 1219-1221. doi: 10.1136/bmj.327.7425.1219

Haggerty, J. L., Roberge, D., Freeman, G. K., & Beaulieu, C. (2013). Experienced continuity of care when patients see multiple clinicians: a qualitative metasummary. *Ann Fam Med, 11*(3), 262-271. doi: 10.1370/afm.1499

Haggerty, J. L., Roberge, D., Freeman, G. K., Beaulieu, C., & Breton, M. (2012). Validation of a generic measure of continuity of care: when patients encounter several clinicians. *Ann Fam Med, 10*(5), 443-451. doi: 10.1370/afm.1378

Harman, B. H. (1976). *Modern factor analysis* (3rd (revised) ed.). Chicago: University of Chicago Press.

Himelstein, B. P., Hilden, J. M., Boldt, A. M., & Weissman, D. (2004). Pediatric palliative care. *N Engl J Med, 350*(17), 1752-1762. doi: 10.1056/NEJMra030334

Hsieh, H. F., & Shannon, S. E. (2005). Three approaches to qualitative content analysis. *Qual Health Res, 15*(9), 1277-1288. doi: 10.1177/1049732305276687

Jacobs, B., Ir, P., Bigdeli, M., Annear, P. L., & Van Damme, W. (2012). Addressing access barriers to health services: an analytical framework for selecting appropriate interventions in low-income Asian countries. *Health Policy Plan, 27*(4), 288-300. doi: 10.1093/heapol/czr038

Knapp, C., Madden, V., Sloyer, P., & Shenkman, E. (2012). Effects of an Integrated Care System on quality of care and satisfaction for children with special health care needs. *Matern Child Health J, 16*(3), 579-586. doi: 10.1007/s10995-011-0778-9

Kowlessar, N. M., Jiang, H. J., & Steiner, C. (2006). Hospital Stays for Newborns, 2011: Statistical Brief #163 *Healthcare Cost and Utilization Project (HCUP) Statistical Briefs*. Rockville (MD).

Kripalani, S., Jackson, A. T., Schnipper, J. L., & Coleman, E. A. (2007). Promoting effective transitions of care at hospital discharge: a review of key issues for hospitalists. *J Hosp Med, 2*(5), 314-323. doi: 10.1002/jhm.228

Kruzikas, D. T. (2004). *Preventable hospitalizations : a window into primary and preventive care, 2000*. Rockville, MD: U.S. Department of Health and Human Services, Agency for Hearthcare Research and Quality.

Latour, J. M., van Goudoever, J. B., Schuurman, B. E., Albers, M. J., van Dam, N. A., Dullaart, E., . . . Hazelzet, J. A. (2011). A qualitative study exploring the experiences of parents of children admitted to seven Dutch pediatric intensive care units. *Intensive Care Med, 37*(2), 319-325. doi: 10.1007/s00134-010-2074-3

Liptak, G. S., & Revell, G. M. (1989). Community physician's role in case management of children with chronic illnesses. *Pediatrics, 84*(3), 465-471.

Luciano, L., Lenzi, J., McDonald, K. M., Rosa, S., Damiani, G., Corsello, G., & Fantini, M. P. (2014). Empirical validation of the "Pediatric Asthma Hospitalization Rate" indicator. *Italian Journal of Pediatrics, 40*. doi: Artn 7 Doi 10.1186/1824-7288-40-7

Mastroiacovo, P., & Costantino, A. (2007). Proposta di un modello assistenziale per bambini e adolescenti con malattie genetiche e/o disabilità ad elevata complessità assistenziale e per le loro famiglie (pp. 28): SIMGEPED.

McDonald, K. M., Schultz, E., Albin, L., Pineda, N., Lonhart, J., Sundaram, V., . . . Davies, S. (2014). *Care Coordination Measures Atlas* Rockville, MD: Agency for Healthcare Research and Quality.

McDonald, K. M., Sundaram, V., Bravata, D. M., Lewis, R., Lin, N., Kraft, S. A., . . . Owens, D. K. (2007) *Closing the Quality Gap: A Critical Analysis of Quality Improvement Strategies (Vol. 7: Care Coordination)*. Rockville (MD).

McPherson, M., Arango, P., Fox, H., Lauver, C., McManus, M., Newacheck, P. W., . . . Strickland, B. (1998). A new definition of children with special health care needs. *Pediatrics, 102*(1 Pt 1), 137-140.

Miller, A. R., Condin, C. J., McKellin, W. H., Shaw, N., Klassen, A. F., & Sheps, S. (2009). Continuity of care for children with complex chronic health conditions: parents' perspectives. *BMC Health Serv Res, 9*, 242. doi: 10.1186/1472-6963-9-242

Ministero della Salute. (2008). *Documento Tecnico sulle Cure Palliative Pediatriche*. Roma: Retrieved from http://www.salute.gov.it/imgs/C_17_pubblicazioni_797_allegato.pdf.

Newacheck, P. W., & Kim, S. E. (2005). A national profile of health care utilization and expenditures for children with special health care needs. *Arch Pediatr Adolesc Med, 159*(1), 10-17. doi: 10.1001/archpedi.159.1.10

Newacheck, P. W., Kim, S. E., Blumberg, S. J., & Rising, J. P. (2008). Who is at risk for special health care needs: findings from the National Survey of Children's Health. *Pediatrics, 122*(2), 347-359. doi: 10.1542/peds.2007-1406

Newacheck, P. W., Strickland, B., Shonkoff, J. P., Perrin, J. M., McPherson, M., McManus, M., . . . Arango, P. (1998). An epidemiologic profile of children with special health care needs. *Pediatrics, 102*(1 Pt 1), 117-123.

Palfrey, J. S., Levy, J. C., & Gilbert, K. L. (1980). Use of primary care facilities by patients attending specialty clinics. *Pediatrics, 65*(3), 567-572.

Perrin, E. C., Newacheck, P., Pless, I. B., Drotar, D., Gortmaker, S. L., Leventhal, J., . . . Weitzman, M. (1993). Issues involved in the definition and classification of chronic health conditions. *Pediatrics, 91*(4), 787-793.

Pless, I. B., & Douglas, J. W. (1971). Chronic illness in childhood. I. Epidemiological and clinicl characteristics. *Pediatrics, 47*(2), 405-414.

Quach, J., Jansen, P. W., Mensah, F. K., & Wake, M. (2015). Trajectories and outcomes among children with special health care needs. *Pediatrics, 135*(4), e842-850. doi: 10.1542/peds.2014-2431

Ralston, S. L., Harrison, W., Wasserman, J., & Goodman, D. C. (2015). Hospital Variation in Health Care Utilization by Children With Medical Complexity. *Pediatrics, 136*(5), 860-867. doi: 10.1542/peds.2014-3920

Reeve, B. B., & Fayers, P. . (2005). Applying item response theory modeling for evaluating questionnaire item and scale properties. In O. U. Press (Ed.), *Assessing quality of life in clinical trials: Methods of practice (2nd ed.)* (pp. 55-73). New York.

Reid, R. J., Haggerty, J., & McKendry, R. (2002). *Defusing the confusion: Concepts and measures of continuity of health care.* University of British Columbia, Vancouver: Centre for Health Services and Policy Research.

Rowland, D. (1989). Financing of care: a critical component. In S. REK (Ed.), *Caring for Children with Chronic Illness: Issues and Strategies* (Vol. 23). New York, NY: Springer.

Rucci, P., Latour, J., Zanello, E., Calugi, S., Vandini, S., Faldella, G., & Fantini, M. P. (2015). Measuring parents' perspective on continuity of care in children with special health care needs. *International Journal of Integrated Care, 15*(oct-dec), 1-17.

Saigal, S., & Doyle, L. W. (2008). An overview of mortality and sequelae of preterm birth from infancy to adulthood. *Lancet, 371*(9608), 261-269. doi: 10.1016/S0140-6736(08)60136-1

Scheppers, E., van Dongen, E., Dekker, J., Geertzen, J., & Dekker, J. (2006). Potential barriers to the use of health services among ethnic minorities: a review. *Fam Pract, 23*(3), 325-348. doi: 10.1093/fampra/cmi113

Schultz, E. M., & McDonald, K. M. (2014). What is Care Coordination? *International Journal of Care Coordination, 17*(1-2), 5-24. doi: 10.1177/2053435414540615

Starfield, B. (2003). Primary care and specialty care: a role reversal? *Med Educ, 37*(9), 756-757.

Stein, R. E., Bauman, L. J., Westbrook, L. E., Coupey, S. M., & Ireys, H. T. (1993). Framework for identifying children who have chronic conditions: the case for a new definition. *J Pediatr, 122*(3), 342-347.

Stein, R. E., & Silver, E. J. (1999). Operationalizing a conceptually based noncategorical definition: a first look at US children with chronic conditions. *Arch Pediatr Adolesc Med, 153*(1), 68-74.

Stille, C. J., & Antonelli, R. C. (2004). Coordination of care for children with special health care needs. *Curr Opin Pediatr, 16*(6), 700-705.

Stille, C. J., Fischer, S. H., La Pelle, N., Dworetzky, B., Mazor, K. M., & Cooley, W. C. (2013). Parent partnerships in communication and decision making about subspecialty referrals for children with special needs. *Acad Pediatr, 13*(2), 122-132. doi: 10.1016/j.acap.2012.12.003

Stille, C. J., Jerant, A., Bell, D., Meltzer, D., & Elmore, J. G. (2005). Coordinating care across diseases, settings, and clinicians: a key role for the generalist in practice. *Ann Intern Med, 142*(8), 700-708.

Strickland, B. B., van Dyck, P. C., Kogan, M. D., Lauver, C., Blumberg, S. J., Bethell, C. D., & Newacheck, P. W. (2011). Assessing and ensuring a comprehensive system of services for children with special health care needs: a public health approach. *Am J Public Health, 101*(2), 224-231. doi: 10.2105/AJPH.2009.177915

Tabachnick, B. G., & Fidell, L. S. (2013). Principal components and factor analysis *Using multivariate statistics*. Boston: Pearson.

Taylor, A., Lizzi, M., Marx, A., Chilkatowsky, M., Trachtenberg, S. W., & Ogle, S. (2013). Implementing a care coordination program for children with special healthcare needs:

partnering with families and providers. *J Healthc Qual, 35*(5), 70-77. doi: 10.1111/j.1945-1474.2012.00215.x

Terwee, C. B., Bot, S. D., de Boer, M. R., van der Windt, D. A., Knol, D. L., Dekker, J., . . . de Vet, H. C. (2007). Quality criteria were proposed for measurement properties of health status questionnaires. *J Clin Epidemiol, 60*(1), 34-42. doi: 10.1016/j.jclinepi.2006.03.012

Tobon, J. I., Reid, G. J., & Goffin, R. D. (2014). Continuity of Care in Children's Mental Health: development of a measure. *Adm Policy Ment Health, 41*(5), 668-686. doi: 10.1007/s10488-013-0518-0

Uijen, A. A., Heinst, C. W., Schellevis, F. G., van den Bosch, W. J., van de Laar, F. A., Terwee, C. B., & Schers, H. J. (2012). Measurement properties of questionnaires measuring continuity of care: a systematic review. *PLoS One, 7*(7), e42256. doi: 10.1371/journal.pone.0042256

Uijen, A. A., Schellevis, F. G., van den Bosch, W. J., Mokkink, H. G., van Weel, C., & Schers, H. J. (2011). Nijmegen Continuity Questionnaire: development and testing of a questionnaire that measures continuity of care. *J Clin Epidemiol, 64*(12), 1391-1399. doi: 10.1016/j.jclinepi.2011.03.006

US Dept of Health and Human Services, H. R. a. S. A., Maternal and Child Health Bureau. (2013). *The National Survey of Children With Special Health Care Needs. Chartbook 2009-2010.* . Rockville, MD: US Dept of Health and Human Services.

van der Lee, J. H., Mokkink, L. B., Grootenhuis, M. A., Heymans, H. S., & Offringa, M. (2007). Definitions and measurement of chronic health conditions in childhood: a systematic review. *JAMA, 297*(24), 2741-2751. doi: 10.1001/jama.297.24.2741

van Esso, D., del Torso, S., Hadjipanayis, A., Biver, A., Jaeger-Roman, E., Wettergren, B., . . . Primary-Secondary Working Group of European Academy of, P. (2010). Paediatric primary care in Europe: variation between countries. *Arch Dis Child, 95*(10), 791-795. doi: 10.1136/adc.2009.178459

Whelan P.J., R. L., Andrews T. (2011). Patient satisfaction rating scales v. patient-related outcome and experience measures. *The Psychiatrist, 35*(1), 32-33. doi: 10.1192/pb.35.1.32

Wolfe, I., Thompson, M., Gill, P., Tamburlini, G., Blair, M., van den Bruel, A., . . . McKee, M. (2013). Health services for children in western Europe. *Lancet, 381*(9873), 1224-1234. doi: 10.1016/S0140-6736(12)62085-6

Zampino, G. S., A. (2010). Pediatria della disabilità. *Prospettive in pediatria, 40*(159-160), 12.

Zanello, E., Calugi, S., Rucci, P., Pieri, G., Vandini, S., Faldella, G., & Fantini, M. P. (2015). Continuity of care in children with special healthcare needs: a qualitative study of family's perspectives. *Italian Journal of Pediatrics, 41*(1), 7. doi: 10.1186/s13052-015-0114-x

Appendix - SpeNK Recruitment Data Collection Tool

SCHEDA DI RACCOLTA DATI DI DIMISSIONE

Pz N.: |___|___|___|

ANAGRAFICA

Iniziali Pz. *(nome, cognome)*: |___| |___| Sesso: ☐ maschio ☐ femmina

Comune di nascita: __________________________   Data di nascita: |___|___|/|___|___|/|___|___|___|___|

Comune di residenza: ________________________

Cod. Fiscale: |___|___|___| |___|___|___| |___|___|___|___|___| |___|___|___|___|___|

RIFERIMENTO PER CONTATTI SUCCESSIVI DI FOLLOW-UP *(compilare almeno una delle due sezioni: madre o padre)*

MADRE	**PADRE**
Cognome: __________________________________	Cognome: __________________________________
Nome: _____________________________________	Nome: _____________________________________
Indirizzo: __________________________________ __________________________________	Indirizzo: __________________________________ __________________________________
Telefono: __________________________________	Telefono: __________________________________
Cittadinanza (madre):	Cittadinanza (padre):
☐ Italiana	☐ Italiana
☐ Straniera	☐ Straniera

MODULO DI CONSENSO INFORMATO FIRMATO: ☐ SI ☐ NO

PLS di riferimento:

Dott./Dott.ssa *(nome, cognome)*

____

Tel.: ____   Sede Ambulatorio: _________________________________ ____

e-mail: ____

RICOVERO INDICE

N. SDO: |___|___|___|___| |___|___|___|___|___|___|

Cod. Degente: |___|___|___| |___|___|___| |___|___|___| Corrisponde al campo "COD_BARRE" della SDO

Data di **ammissione** (ricovero): |___|___|/|___|___|/|___|___|___|___|

U.O. di ammissione:

☐ U.O. Neonatologia - Faldella	☐ U.O. Pediatria d'Urgenza, [...] - Bernardi
☐ U.O. Anestesiologia e Rianimazione - Baroncini	☐ U.O. Neonatologia e unità terapia intensiva neonatale (AUSL Bologna)
☐ U.O. Chirurgia Pediatrica di Bologna - Lima	☐ U.O. Pediatria (AUSL Bologna)
☐ U.O. Neuropsichiatria Infantile - Franzoni	☐ U.O. Pediatria e Nido (AUSL Imola)
☐ U.O. Pediatria – Pession	

Data di **dimissione**: |___|___|/|___|___|/|___|___|___|___|

U.O. di dimissione:

☐ U.O. Neonatologia - Faldella	☐ U.O. Pediatria d'Urgenza, [...]- Bernardi
☐ U.O. Anestesiologia e Rianimazione - Baroncini	☐ U.O. Neonatologia e unità terapia intensiva neonatale (AUSL Bologna)
☐ U.O. Chirurgia Pediatrica di Bologna - Lima	☐ U.O. Pediatria (AUSL Bologna)
☐ U.O. Neuropsichiatria Infantile - Franzoni	☐ U.O. Pediatria e Nido (AUSL Imola)
☐ U.O. Pediatria - Pession	

Motivo di inclusione nello studio: ____

Diagnosi principale di dimissione:
Cod. di diagnosi ICD9-CM: |___|___|___|___|___| Descrizione: ____________________________________

Diagnosi secondarie:

Secondaria 1: Cod. di diagnosi ICD9-CM: |___|___|___|___|___| Descrizione: ________________________________
Secondaria 2: Cod. di diagnosi ICD9-CM: |___|___|___|___|___| Descrizione: ________________________________
Secondaria 3: Cod. di diagnosi ICD9-CM: |___|___|___|___|___| Descrizione: ________________________________
Secondaria 4: Cod. di diagnosi ICD9-CM: |___|___|___|___|___| Descrizione: ________________________________
Secondaria 5: Cod. di diagnosi ICD9-CM: |___|___|___|___|___| Descrizione: ________________________________

Consulenze attivate in corso di ricovero:
☐ Servizio sociale
☐ NPEE *(neuropsichiatria dell'età evolutiva)*
☐ Fisiatria
☐ Altro *(specificare:* ______________________________
____*)*

Modalità di dimissione: ☐ dimissione ordinaria ☐ dimissione protetta

Attivazione dei servizi territoriali: ☐ SI ☐ NO
Se"SI", quali: ____
____

Necessità di fornitura di **presidi sanitari complessi a domicilio**: ☐ SI ☐ NO

Predisposizione di un **PAI** (Piano Assistenza Individuale): ☐ SI ☐ NO
Se"SI", quali servizi sono stati coinvolti/contattati? ____
____

Verifica del PAI: ☐ al bisogno ☐ programmata ☐ non prevista
Se "programmata": ☐ mensile ☐ trimestrale ☐ semestrale ☐ altro

Cure domiciliari:
☐ SID *(assistenza infermieristica e/o riabilitativa)*
☐ ADP *(assistenza domiciliare programmata)*
☐ Intensive e/o pallative
☐ ADI 1
☐ ADI 2
☐ ADI 3

E' previsto un **follow-up** presso la struttura ospedaliera di dimissione? ☐ SI ☐ NO

Trasferimento del paziente: ☐ SI ☐ NO
Se "Sì", indicare presso quale struttura: ____

Ulteriori dettagli (*procedure di dimissione, professionisti coinvolti,ecc.*):
____
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Appendix - SpeNK-I Interview Guide

AREA	TEMA	PROMPTS
PRESENTAZIONE	PSICOLOGA	Buongiorno(sera), Mi chiamo …. Sono uno/a psicologo/a e collaboro con il gruppo di ricerca …
	RICERCA E SCOPO INTERVISTA	In questa ricerca vogliamo dedicare una parte del lavoro al punto di vista di voi genitori sul percorso di cura del vostro bambino. Oggi vorrei farvi/le un’intervista per capire come avete vissuto questo percorso dall’inizio fino ad oggi. Mi interessa proprio il vostro/suo personale punto di vista, quindi sentitevi/si senta liberi/o/a di parlare sinceramente. Staremo insieme per un’ora circa. Siete/È d’accordo se registro l’intervista? (I dati rimangono anonimi e vengono usati solo a fini scientifici – per la ricerca)
INTRODUZIONE	DATI SOCIO-DEMOGRAFICI	(età, livello di istruzione, occupazione attuale, composizione del nucleo famigliare, nucleo abitativo)
	ESPERIENZA IN GENERALE	So che suo figlio è stato da poco dimesso. Com’è stato quel momento per voi? Mi può raccontare la sua esperienza?
CONDIZIONE CLINICA	CONOSCENZA	Quando informati, Cosa comporta, Che impatto ha sulla vita famigliare
	COMUNICAZIONE	Come, chi, quando, dove? Spiegata in termini comprensibili? Possibilità di chiedere chiarimenti? Risposte? Ascoltate le idee/aspettative? Ascoltate le paure/preoccupazioni? Si è sentita ascoltata/o?
	CONDIVISIONE INFO / SCELTE	Spiegate le conseguenze sulla salute? Presentate possibilità di scelta per le cure? È stato interpellato/a? Mostrato rispetto per quello che aveva da dire?
PIANO DELLE CURE	COMUNICAZIONE	Spiegato il percorso di cure previsto dopo la dimissione? In modo comprensibile? Lei si è fatto un’idea di cosa accadrà nel futuro/ora? Spiegato quali professionisti/servizi saranno coinvolti nel piano delle cure? Se aveva domande o dubbi le è stato possibile esprimerli? Le è stato permesso di esprimere le sue opinioni sulle cure proposte? Si è sentita ascoltata/o?
	CONDIVISIONE INFO / SCELTE	Presentate possibilità di scelta per il piano? È stato/a interpellato/a? Hanno mostrato rispetto per quello che aveva da dire?
	PROFESSIONISTI E SERVIZI COINVOLTI chi, come, perché disponibilità	Quali professionisti hanno condiviso con lei la dimissione? Si sono dichiarati disponibili ad essere contattati in futuro in caso di necessità? Le hanno indicato a chi rivolgersi o come fare per approfondimenti? Contatti con Pediatra? Altri Psicologi nelle UO?
POST DIMISSIONE	AUTONOMIA NELLA GESTIONE	Le sono state date indicazioni adeguate … - per gestire le cure quotidiane? (es. ausili o presidi, conoscenza) - per sapere come fare per farlo stare bene o meglio? - per gestire eventuali emergenze o imprevisti?
	RETE DI PROFESSIONISTI E SERVIZI Case manager, condivisione info e cure CONTINUITÀ DELLE CURE	C’è una persona / servizio più coinvolto di altri nel percorso di cura? Come mai mi ha indicato proprio lui/lei/quel servizio? Ritiene che i professionisti/servizi coinvolti condividano tra loro le informazioni cliniche su vostro figlio? Ritiene che siano d’accordo tra loro sulle piano delle cure? Ci sono professionisti o servizi più facili da raggiungere rispetto ad altri? E più difficili? Qual è attualmente la sua opinione sulla continuità delle cure che avete ricevuto da questa rete di servizi e professionisti? (…)
DOMANDE APERTE FINALI	ALTRE COSE IMPORTANTI	C’è qualcosa di importante per lei che non le ho chiesto e vorrebbe dire? (evento, episodio, incontro o altro)
CONCLUSIONE	RINGRAZIAMENTI CHIARIMENTI COMMENTI	La ringrazio per il tempo che mi ha dedicato. Ha qualche domanda da farmi? C’è qualche commento che vorrebbe fare sul nostro colloquio? (…) La ringrazio ancora una volta e la saluto.

Appendix - SpeNK-Q Questionnaire

AMBULATORIO NEONATO PRETERMINE
IL PUNTO DI VISTA DELLE FAMIGLIE

Caro Genitore,

Vorremmo sapere qual è stata la sua esperienza con i servizi sanitari (territorio, ospedale, pediatra di famiglia) che si sono occupati delle cure e dell'assistenza per suo/a figlio/a negli ultimi mesi, dopo la dimissione. Il suo contributo potrà servire a migliorare l'organizzazione dell'assistenza per altri genitori e bambini che vivono situazioni simili alla vostra.
Le chiediamo di compilare il questionario il più sinceramente possibile, in base alla sua esperienza. Può compilare il questionario da solo/a ma se vuole la posso aiutare.
Le informazioni ricavate dal questionario saranno trattate nel rispetto delle vigenti normative sulla privacy e utilizzate in forma anonima a fini scientifici e di ricerca.

DATA ☐ **RISPONDE** ☐ **Madre** ☐ **Padre** ☐ **Altro: ____________** **I.** ☐ **N.** ☐

Ripensando all'assistenza sanitaria ricevuta da suo/a figlio/a, Chi è che si prende cura di tutti o quasi i problemi di salute di suo/a figlio/a? Dove? Se più persone: Se dovesse indicarne uno, chi sarebbe?____
____

Ripensando al vostro Pediatra di famiglia…
Per ogni frase da 1 a 7 faccia un segno sul punteggio che esprime meglio il suo grado di accordo con la frase: i punteggi vanno da 1 a 5, dove 1 = Completamente contrario 2 = Contrario 3 = Incerto 4 = D'accordo 5 = Completamente d'accordo.

1. Il/la pediatra conosce la storia di salute di mio/a figlio/a.

☹		😐		☺
Completamente contrario	Contrario	Incerto	D'accordo	Completamente d'accordo
1	2	3	4	5

2. Il/la pediatra tiene conto di ciò che mi preoccupa di più per la salute di mio/a figlio/a.

☹		😐		☺
Completamente contrario	Contrario	Incerto	D'accordo	Completamente d'accordo
1	2	3	4	5

3. Mi sento a mio agio a discutere con il/la pediatra di tutti i problemi legati alla salute di mio/a figlio/a.

☹		😐		☺
Completamente contrario	Contrario	Incerto	D'accordo	Completamente d'accordo
1	2	3	4	5

Pensando a tutte le persone che si sono occupate della salute di suo/a figlio/a, c'è qualcuno che coordina l'assistenza sanitaria? Chi? Dove?
____
____

4. Questa persona conosce tutti i bisogni di salute di mio/a figlio/a.

☹		😐		☺
Completamente contrario	Contrario	Incerto	D'accordo	Completamente d'accordo
1	2	3	4	5

5. Questa persona è sempre aggiornata sull'assistenza sanitaria fornita da altri.

☹		😐		☺
Completamente contrario	Contrario	Incerto	D'accordo	Completamente d'accordo
1	2	3	4	5

6. Questa persona contatta altri specialisti sull'assistenza sanitaria ricevuta da mio/a figlio/a.

☹		😐		☺
Completamente contrario	Contrario	Incerto	D'accordo	Completamente d'accordo
1	2	3	4	5

7. Questa persona si tiene in contatto con me quando mio/a figlio/a riceve assistenza sanitaria da altri.

☹		😐		☺
Completamente contrario	Contrario	Incerto	D'accordo	Completamente d'accordo
1	2	3	4	5

Ripensando agli ultimi mesi, dopo la dimissione, e a quello che è stato fatto per la salute di suo/a figlio/a in questo periodo da tutte le persone coinvolte, dell'ospedale e del territorio …

Per ogni frase da 8 a 20 faccia un segno sul punteggio che indica quanto spesso le è capitato ogni episodio su una scala da 1 a 5, dove 1= Mai o quasi mai 2 = Raramente 3 = Qualche volta 4 = Spesso 5 = Sempre o quasi sempre.

8. Le persone che si sono prese cura di mio/a figlio/a mi hanno detto cose diverse sulla sua salute.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

9. Qualcuno mi ha spiegato che conseguenze hanno le condizioni cliniche di mio/a figlio/a sul suo stato di salute.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

10. Qualcuno mi ha spiegato quali trattamenti stava facendo a mio/a figlio/a e perché. (per es. esami, visite, medicinali, ecc.)

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

11. Qualcuno mi ha spiegato il piano di esami, visite e controlli da fare.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

12. La persona che stava visitando mio/a figlio/a ignorava la sua storia di salute recente.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

13. Alla persona che stava visitando mio/a figlio/a mancavano i referti dell'ultimo controllo, visita o esame.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

14. Ho dovuto ripetere informazioni sulla salute di mio/a figlio/a che avrebbero dovuto essere presenti nella cartella clinica.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

15. Ho dovuto dare i risultati di una visita specialistica alla persona che stava visitando mio/a figlio/a.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

16. Le persone che si sono prese cura di mio/a figlio/a mi hanno dato tutte le informazioni che mi servono per dargli/le le cure in casa.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

17. Le persone che si sono prese cura di mio/a figlio/a mi hanno dato tutte le informazioni che mi servono per affrontare eventuali piccoli problemi o complicanze.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

18. Mi è sembrato che le persone che si sono prese cura di mio/a figlio/a lo/la conoscessero bene. (per es. pediatra, specialisti, infermieri, fisioterapisti, ecc.)

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

19. Quando le cose sono cambiate o sono andate male, ho ottenuto subito risposte o consigli.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

20. Mi sono sentito/a abbandonato/a dal sistema sanitario per l'organizzazione dell'assistenza di mio/a figlio/a.

1	2	3	4	5
Mai o quasi mai	Raramente	Qualche volta	Spesso	Sempre o quasi sempre

Ripensando all'assistenza sanitaria ricevuta da suo/a figlio/a negli ultimi mesi, dopo la dimissione…
Quali sono gli aspetti che le sono piaciuti di più?

____
____
____

E di meno?

____
____
____

Grazie per la collaborazione!

Appendix - SpeNK-FP Data Collection Tool

SCHEDA PER L'ATTIVITÀ DI COORDINAMENTO DEL PLS

(Adattamento del *Medical Home Care Coordination Measurement Tool*© sviluppato da Antonelli et al., 2008)

SCHEDA ANAGRAFICA

Cognome: ____ *(campo obbligatorio)*

Nome: ____ *(campo obbligatorio)*

Data di nascita: |___|___|/|___|___|/|___|___|___|___| *(campo obbligatorio)*

Cod. Fiscale: |___|___|___| |___|___|___| |___|___|___|___|___| |___|___|___|___|___| *(campo obbligatorio)*

Cod. Esenzione: |___|___|___| |___|___|___|___|___|
(Cod. ICD-9-CM)

SCHEDA CONTATTO (<u>tutti</u> i campi sono obbligatori)

Data contatto: |___|___|/|___|___|/|___|___|___|___| *(campo obbligatorio)*

Livello complessità del paziente

Definire, sulla base dei dati clinici in proprio possesso, il livello di complessità assistenziale del proprio assistito (indicane SOLO UNO):

☐ Bambini senza particolari bisogni di assistenza sanitaria, <u>con</u> questioni familiari o problemi sociali complicanti
☐ Bambini <u>con</u> particolari bisogni di assistenza sanitaria, senza questioni familiari o problemi sociali complicanti
☐ Bambini <u>con</u> particolari bisogni di assistenza sanitaria, <u>con</u> questioni familiari o problemi sociali complicanti

Motivo del contatto

Specificare per quale delle seguenti problematiche/richieste del paziente è avvenuto il contatto (possibilità di scelta multipla):

☐ Richiesta visita al PLS
☐ Prescrizione farmaci
☐ Prescrizione esami strumentali
☐ Prescrizione esami di laboratorio
☐ Necessità protesi/ausili
☐ Crescita/nutrizione
☐ Invio ad uno specialista
☐ Sviluppo psicomotorio/comportamento
☐ Educazione/scolarizzazione
☐ Ritardo mentale
☐ Servizi sociali *(alloggio, alimentazione, abbigliamento, assicurazione, trasporti)*
☐ Richiesta ADI
☐ Questioni legali/giudiziarie
☐ Altro

Bisogni che richiedono attività di coordinamento

Indicare quali dei seguenti bisogni del paziente, emersi durante il contatto, richiedono attività di assistenza coordinata (possibilità di scelta multipla):

☐ Valutare ulteriori rinvii specialistici
☐ Richiedere farmaci
☐ Richiedere attrezzature sanitarie
☐ Richiedere servizi
☐ Richiedere esami strumentali
☐ Richiedere esami di laboratorio

☐ Servizi di coordinazione *(scuole, associazioni, assicurazione)*
☐ Risoluzione incomprensioni e mancate comunicazioni
☐ Nessuno

Attività messe in pratica

Specificare le attività da mettere in pratica per soddisfare i bisogni del paziente emersi durante il contatto (possibilità di scelta multipla):

☐ Contatti con genitori/famiglia (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con scuola (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con Servizi socio-sanitari (Pediatria di Comunità, Servizi Sociali, …) (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con ospedale/clinica (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con assicurazione (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con educatore professionale (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con farmacia (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con associazioni (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con personale dell'ADI (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con specialisti (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Contatti con altra figura (Se "SI", marcare una modalità)
    ☐ Telefono ☐ Incontri ☐ Lettera ☐ E-mail ☐ Fax
☐ Comunicazioni scritte (Se "SI", marcare una modalità)
    ☐ Rilascio certificati/moduli per scuole e tempo libero
    ☐ Rilascio referti/risultati indagini di laboratorio e strumentali
    ☐ Nessuna
☐ Notifiche/relazioni alle Autorità sanitarie (notifica malattie infettive, …)
☐ Revisione/aggiornamento documentazione paziente
☐ Audit clinico
☐ Sviluppare/modificare il piano di assistenza individuale (PAI)
☐ Incontri/conferenze sul caso

Staff coinvolto nell'attività di coordinamento

Nella programmazione delle attività di coordinamento specificare quali figure professionali del tuo staff sono coinvolte(possibilità di scelta multipla):

☐ Nessuno
☐ Altri medici
☐ Infermieri professionali
☐ Operatori sociali
☐ Personale amministrativo
☐ Altro

Tempo dedicato alle attività di coordinamento

Quantifica il tempo dedicato alla risoluzione del problema e alle conseguenti attività di coordinamento necessarie(indicane SOLO UNO):

☐ Meno di 5 minuti
☐ Da 5 a 9 minuti
☐ Da 10 a 19 minuti
☐ Da 20 a 29 minuti
☐ Da 30 a 39 minuti
☐ Da 40 a 49 minuti

☐ = o >50 minuti (prego annotare i minuti effettivi, se il tempo è >50') |___|___|___|minuti

Esiti dell'attività di coordinamento

Grazie alla presente valutazione, secondo la tua opinione, non è stato necessario per il paziente il ricorso a: (possibilità di scelta multipla):

☐ Visita al Pronto Soccorso
☐ Ulteriore visita specialistica
☐ Ospedalizzazione
☐ Visita alla clinica pediatrica
☐ Indagini di laboratorio/indagini strumentali
☐ Farmaci
☐ Altro, *specificare:*
____

Come risultato del presente contatto, si è verificato il seguente esito (possibilità di scelta multipla):

☐ Aver consigliato la famiglia/il paziente in merito alla gestione domiciliare
☐ Invio al Pronto Soccorso
☐ Invio ad ulteriore specialista
☐ Invio per ricovero
☐ Invio per visita alla clinica pediatrica
☐ Invio per indagini di laboratorio/indagini strumentali
☐ Utilizzo farmaci
☐ Aver richiesto attrezzature, pannolini, trasporti
☐ Aver risolto questioni legate a dati mancanti, mancate comunicazioni, problemi burocratici
☐ Aver riesaminato dati laboratorio, resoconti degli specialisti, programmi di educazione individuale,ecc.
☐ Attività di sostegno per la famiglia/paziente
☐ Aver provveduto ai bisogni immediati, alle domande e alle preoccupazioni della famiglia
☐ Problemi non risolti
☐ Altro

Note

____

____

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Appendix - SpeNK-A Algorithm

SpeNK-A Group	Diagnostic Codes (ICD9CM) and Variables indicated in Hospital Discharge Record
Group 1: Neoplasms	140.x - 208.91 Malignant neoplasms 225.x Benign neoplasm of brain and other parts of nervous system 235.x - 238.x Neoplasms of uncertain behavior 239.x Neoplasms of unspecified nature 212.7 Benign neoplasm of heart 213.0 Benign neoplasm of bones of skull and face 213.2 Benign neoplasm of vertebral column, excluding sacrum and coccyx 213.6 Benign neoplasm of pelvic bones, sacrum, and coccyx 215.0 Other benign neoplasm of connective and other soft tissue of head, face, and neck 224.1 Benign neoplasm of orbit 224.6 Benign neoplasm of choroid 224.8 Benign neoplasm of other specified parts of eye 224.9 Benign neoplasm of eye, part unspecified 227.3 Benign neoplasm of pituitary gland and craniopharyngeal duct 227.4 Benign neoplasm of pineal gland 227.5 Benign neoplasm of carotid body 227.6 Benign neoplasm of aortic body and other paraganglia 228.02 Hemangioma of intracranial structures 228.03 Hemangioma of retina V10.x Personal anamnesis of malignant neoplasm
Group 2: Malformations and other congenital diseases	740.x - 759.x Congenital anomalies 277.0x Cystic fibrosis 425.x Cardiomyopathy 271.1 Galactosemia 275.1 Disorders of copper metabolism 279.11 Digeorge's syndrome 330.1 Cerebral lipidoses 352.2 Other disorders of glossopharyngeal [9th] nerve 416.0 Primary pulmonary hypertension 585.6 End stage renal disease 429.89 Other ill-defined heart diseases 429.9 Heart disease, unspecified 723.0 Spinal stenosis in cervical region Gastrostomy (codes V55.1, V44.1) + 536.40 Gastrostomy complication, unspecified V15.1 Personal anamnesis of surgery of the heart and great vessels, with risk of disease
Group 3: Neurologic and Metabolic Degenerative Diseases	330.x Cerebral degenerations usually manifested in childhood 331.3 Communicating hydrocephalus 331.4 Obstructive hydrocephalus 331.5 Idiopathic normal pressure hydrocephalus (INPH) 331.6 Corticobasal degeneration 331.7 Cerebral degeneration in diseases classified elsewhere 334.x Spinocerebellar disease 335.x Anterior horn cell disease 336.x Other diseases of spinal cord 358.x Myoneural disorders 359.x Muscular dystrophies and other myopathies 333.71 Athetoid cerebral palsy 639.4 Metabolic disorders following abortion or ectopic and molar pregnancies 756.51 Osteogenesis imperfect 756.52 Osteopetrosis 243 Congenital hypothyroidism 272.7 Lipidoses

SpeNK-A Group	Diagnostic Codes (ICD9CM) and Variables indicated in Hospital Discharge Record
	277.5 Mucopolysaccharidosis 277.87 Disorders of mitochondrial metabolism 331.89 Other cerebral degeneration 331.9 Cerebral degeneration, unspecified 333.0 Other degenerative diseases of the basal ganglia 333.2 Myoclonus 333.6 Genetic torsion dystonia 340 Multiple sclerosis 341.2x Acute myelitis (transverse myelitis) 341.8 Other demyelinating diseases of central nervous system 341.9 Demyelinating disease of central nervous system, unspecified 360.21 Progressive high (degenerative) myopia 779.0 Convulsions in newborn
Group 4: Irreversible Pathologies	253.x Disorders of the pituitary gland and its hypothalamic control 320.x - 326.x Inflammatory diseases of the central nervous system 342.x Hemiplegia 343.x Infantile cerebral palsy 344.x Other paralytic syndromes 347.x Cataplexy and narcolepsy 348.x Other conditions of brain 427.4x Ventricular fibrillation, Ventricular flutter 430.x – 433.9 Cerebrovascular diseases 740.x Anencephalus and similar anomalies 741.x Spina bifida 742.x Other congenital anomalies of nervous system 756.1x Anomalies of spine 349.82 Toxic encephalopathy 349.89 Other specified disorders of nervous system 349.9 Unspecified disorders of nervous system 352.6 Multiple cranial nerve palsies 352.9 Unspecified disorder of cranial nerves 779.7 Periventricular leukomalacia
Group 5: Extremely low birth weight	Birth weight < 1000 gr
Group 6: Pathologies connected to newborn extreme prematurity	768.5 Severe birth asphyxia 768.7x Hypoxic-ischemic encephalopathy 777.5x Necrotizing enterocolitis in newborn 765.01 Extreme immaturity, less than 500 grams 765.02 Extreme immaturity, 500-749 grams 765.03 Extreme immaturity, 750-999 grams