

Strategic analysis of malformations congenital care: proposal of approach and development of care pathways

Luciane Binsfeld (<https://orcid.org/0000-0001-7513-5266>)¹

Maria Auxiliadora de Souza Mendes Gomes (<https://orcid.org/0000-0001-5908-1763>)¹

Rosana Kuschnir (<https://orcid.org/0000-0002-0971-4590>)²

Abstract *This paper aims at presenting a proposal for grouping cases for the organization of health services and care pathways. This is an exploratory study in the field of health services planning and management, which used, as its methodology, documentary and bibliographic research as well as interviews with specialists by using nominal group technique. From the strategic analysis, four groups were identified: smaller CM; CM with late surgical approach; CM with immediate surgical approach; and CM incompatible with life. The proposition started from the articulation of clinical, epidemiological and health planning knowledge to assist in the management and organization of congenital malformations care. The strategic analysis proved to be adequate and allowed us to identify case groups that demand a homogeneous set of care strategies and care in health services with a similar profile. This proposal can also contribute to regional planning and management of care for other complex health problems and conditions, which demand the articulation of specialized services and high technological density.*

Key words *Congenital malformations, Health planning, Health management, National health networks, Maternal child health services*

¹ Instituto Nacional de Saúde da Mulher, da Criança e do Adolescente Fernandes Figueira, Fundação Oswaldo Cruz. Av. Rui Barbosa 716, Flamengo. 22250-020 Rio de Janeiro RJ Brasil. lucianebinsfeld@gmail.com

² Escola Nacional de Saúde Pública Sergio Arouca, Fundação Oswaldo Cruz. Rio de Janeiro RJ Brasil.

Introduction

Difficulties in organizing the set of elements involved in healthcare process regarding attention level, care practices and services have been demanding the development of new strategies concerning integrations and care coordination^{1,2}.

In this context, the proposal of structuring care pathways rises aiming at promoting the organization of steps to be followed by users in the most logical, systemic and humanized ways in order to ensure that the necessary resources, due to diagnosis and designed therapy will be properly available^{1,2}.

In order to qualify the design of such care pathways it is necessary to trigger clinical/ epidemiological knowledge – from the identification of the best care practices – to the planning/ management to decide the most effective health service organization³.

In a view to do so, it is important to improve analyses tools to incorporate knowledge and medical rationality, epidemiology and planning so as to identify the best approach strategies regarding health issues. It is also important to consider approaches which can ensure some safe and qualified care, based on the best available evidence, as well as the optimization of resources in healthcare organization.

For child and teenager care, changes in the profile of sickening due to higher survival of extremely premature birth, children who suffer from chronic conditions and birth defects, have occupied a significant spot both in national and international setting for health policies⁴⁻⁶. In this context, congenital malformations are characterized as complex chronic condition and rare disease, and it is the second cause of neonatal deaths in Brazil and it is one of the main causes of pediatric hospital assistance^{7,8}.

To qualify congenital malformation care it is important to guarantee a consistent framework related to care pathways to ensure the integration of different care processes, care units and attention at regional levels. However, in the group called congenital malformations there are multiple pathologies, consisting of different typologies and care needs, thus leading to the question: which would be the best way to approach this health problem considering the organization of care process as well as care sequence?

Taking this question into account, a strategic analysis concerning attention to congenital malformation was applied to identify which elements guide the care sequence and health services at a

regional perspective. For such analysis, the proactive posture of a state/region manager was considered, observing if this professional should seek to manage care demands, of a problem or health condition, with planning and management elements to organize qualification and supply when it comes to care profile in health service.

The aim is to present the result of this strategic analysis and to design a proposal for grouping congenital malformations in order to prepare care pathways, considering care needs and elements to contribute to better organizing health services.

Methods

It is an exploratory study in the field of planning and organization of health services, setting an even bigger research called Planning and Programming of Health Care: Organization of Health Care to Congenital Malformation⁹. This project had four moments, and the results shown in this paper refer to the first one: strategic analysis of congenital malformations. The subsequent moments were: definition of recommendations as well as the strategic prerequisites to the organization of specialized services: situational diagnosis in the State of Rio de Janeiro (RJ); development of a proposal to organize congenital malformation care in the state of RJ⁹.

Bibliographic research and expert consultation – through Nominal Group techniques (NG) were the methodological strategies used in this study. This technique is indicated in the case of small group works when searching for a common ground among experts and it includes presential meetings – individual and collective ones – from the contextualization of a problem as well as questions that can be responded and debated by the group in search of a set of solutions or recommendations to the subject^{10,11}.

These were the following steps when applying the technique with the group: contextualization of the problem; introduction of questions that would guide the ideas; global registration and consolidation of generated information; presentation of material; discussion aiming at clarification and selection of the most significant proposals.

The participants of the Nominal Group are experts who work with congenital malformation care in addition to managers in mother-child health areas in Rio de Janeiro. The first ones constituted the Nominal group called “NG experts” and included a representative of each of the areas:

fetal/obstetric medicine, neonatology, pediatric surgery, pediatric and clinical genetics.

These experts work at a reference unit consisting of the highest numbers in congenital malformation births in the State of RJ and they coordinate a structured working model to design national clinical guidelines to the main congenital malformations of immediate surgical approach nature: congenital diaphragmatic hernia, spina bifida and gastroschisis¹². Such guidelines have been under analyses by Ministry of Health and the evidences supported the experts' discussion presented in the nominal group.

The second group called "Amplified NG" is constituted by the above experts and the representatives of mother-child care, as follows: one from de reference unit; one from the City Health department of RJ; the other one from the State health department of RJ.

Bibliographical research and experts consultation in NG were guided by the following questions: *How are congenital malformations classified? What are the characteristics of processes/activities and the technologies required for congenital malformation care? Taking the above question into account, what enables congenital malformations differentiation and/or aggregation?*

The subjects were invited at different times during the project whether collectively or individually in order to discuss the core questions, to elaborate and to analyze proposals and results. The main researcher coordinated the work with the Nominal Group. Also, she has drawn up a set of information and synthesis from the bibliographical review that were presented as guiding elements to the debate and, also, she has consolidated the results in summaries for analyses and group debates.

It is also important to highlight that in this study technological resources and procedures which are already attached at health public services were also taken into account here, as the focus of the project is to organize care as well as available services to draw a care process and also a design regarding attention to congenital malformation diagnosed at prenatal time.

To accomplish strategic analysis framework and the consolidation of results, the principles of *démarche stratégique* methodology were applied, mainly the strategic segmentation one.

Démarche stratégique is an approach that proposes to logically define the mission of a hospital, placing it at an ideal perspective in a coordinated health care. The method comprehends the following stages: pre-existing analysis; strategic

segmentation; analysis of values and competitive position; elaboration of a portfolio of activities; action plan.

At segmentation phase, the goal is the multi-criteria analysis in order to identify homogenous groups concerning activities, information description concerning the population, the pathologies and technologies, and attention involved in a specific health problem care. From this description on, the goal is to reduce discrepancies among segments and/or to identify similarities and promote grouping depending on one or more variables^{13,14}.

These elements supported the organization of the process of strategic analysis thereby presented in this study. However, it is not the project goal to apply the method at all phases or instruments.

Results

Congenital malformations constitute the group of birth defects where these ones are defined as morphological defects in an organ or at some part of the body, being the result of a process of intrinsic abnormal development present at the birth moment^{15,16}.

Regarding the first question, referring to the classification of congenital malformation, there are two very common typologies. The first, from morbidity and/or clinical repercussion that classifies congenital malformation as minor- the ones that do not provoke significant issues to the subject – and major ones – when they bring clinical consequences or even serious aesthetic problems. The second typology classifies malformation from its lethality as compatible or incompatible with life¹⁷.

International Classification of Diseases (CID 10)¹⁸ introduces a categorization based on systems and/or body segments, as congenital malformation are part of Chapter XVII – congenital malformation, defects and chromossomic anomalies. In this typology the groups related to nervous system are included: eye, ear, face and neck; circulatory system; respiratory system; lip and palate fissures; digestive system; genital organs; and osteomuscular system.

Regarding the second question, from the records and systematization of information provided by Nominal Group Experts, technology characteristics were identified and also the necessary process of attention aiming at a proper care.

The set of information topics has elements that correspond to criteria which are initially

shown by *démarche stratégique* to the multicriteria analysis of a problem – population, pathology, technologies and processes – and two more added on: medical specialization and the activities involved. Some other relevant questions also emerged concerning care when it comes to the referred pathologies, health policies and strategies related to the attention process and congenital malformation care, which were included as cross-cutting themes. Information provided by the group was consolidated in Figure 1.

The consolidation of the third question occurred from the result systematization of both previous questions and the creation of some key questions related to the ways of classification, the characteristics of service care, technologies and competences that are required to congenital malformation care. The questions were placed in the NG Experts and enabled the identification of presence of differences and similarities among the set of pathologies composing the big group of congenital malformations (Chart 1).

Every malformation has in common the fact that they encompass care process to the pregnant, the newborn and the child, but not only the strategic moment of intervention but also care process changes according to the classification paths. The possibility of diagnosis and the ways of treatment, too.

The two first questions inserted in Chart 1 remind the ways of classification: major and minor, compatible or incompatible with life, which inform the severity and the impact of these conditions and the care sequence design and, the distinct therapeutical processes for pregnancy and neonatal care.

The third question refers to diagnosis and considers the possibility of congenital malformation being detected – the majority of cases – through obstetric ultrasound and if its detection would change the course of prenatal follow-up as much as the delivery planning and neonatal care. Thus, it enables to highlight cases in which the follow-up by fetal medicine and genetic service is recommended in order to mother-child monitoring, besides the assessment of in- utero necessity and/or identification of essential prognostic markers to plan the birth and neonatal care^{19,20}.

The following question, involving surgical intervention, was included because it is the main procedure to differ the treatment of congenital malformation after birth as well as the moment of its performance. In some cases, the guarantee of surgical care soon after delivery (24-48h) to fix and/or maintain life impacts directly on

prognosis for reducing neonatal morbidity²¹. Concerning the reference identification of surgical medical specialization, it guides the analysis of care profile of attention and the reference staff who need to be available and able at the location to provide all care required and necessary at the time.

For transportation, in this case, it was considered as that which would happen between health units to promote surgical intervention if the unit would not have its own service. Transportation needs analysis was included because at times it is not recommended in some cases, as in major congenital malformations, as it can worsen the injury and/or newborn condition, it also can increase infection and neonatal morbidity conditions. The segment is related to follow-up after hospital discharge and the identification of cases that demand some closer connection to a specialized outpatient clinic so as to provide care shared with primary attention.

Applying the key questions to NG experts plus the consolidation of results have enabled the grouping of congenital malformation and the making of typologies that can guide the design of care pathways and the organization of attention services. The process of grouping has resulted in the definition of four groups of congenital malformation, derived from the main classification ways, as major and minor; compatible and incompatible with life and also some assistance segments, identified according to the main medical specialization involved in care (Chart 2).

However, it was not the intention to classify every malformation in some case or even line of care. For this reason, examples were placed in results description. Some malformation groups vary a lot according to its presentation, as neurologic and cardiac malformations and can embark, from cases that are classified as incompatible with life to others that are diagnosed only in neonatal period. Therefore, clinical evaluation and the making of therapeutical projects that will define the programming of care for each case and the care process to be followed.

Discussion

This study follows the premise that health planning processes if intended to be effective and able to promote concrete and consistent changes in the paths for the organization of services and attention network, need to solidify interlocution with the clinic²³.

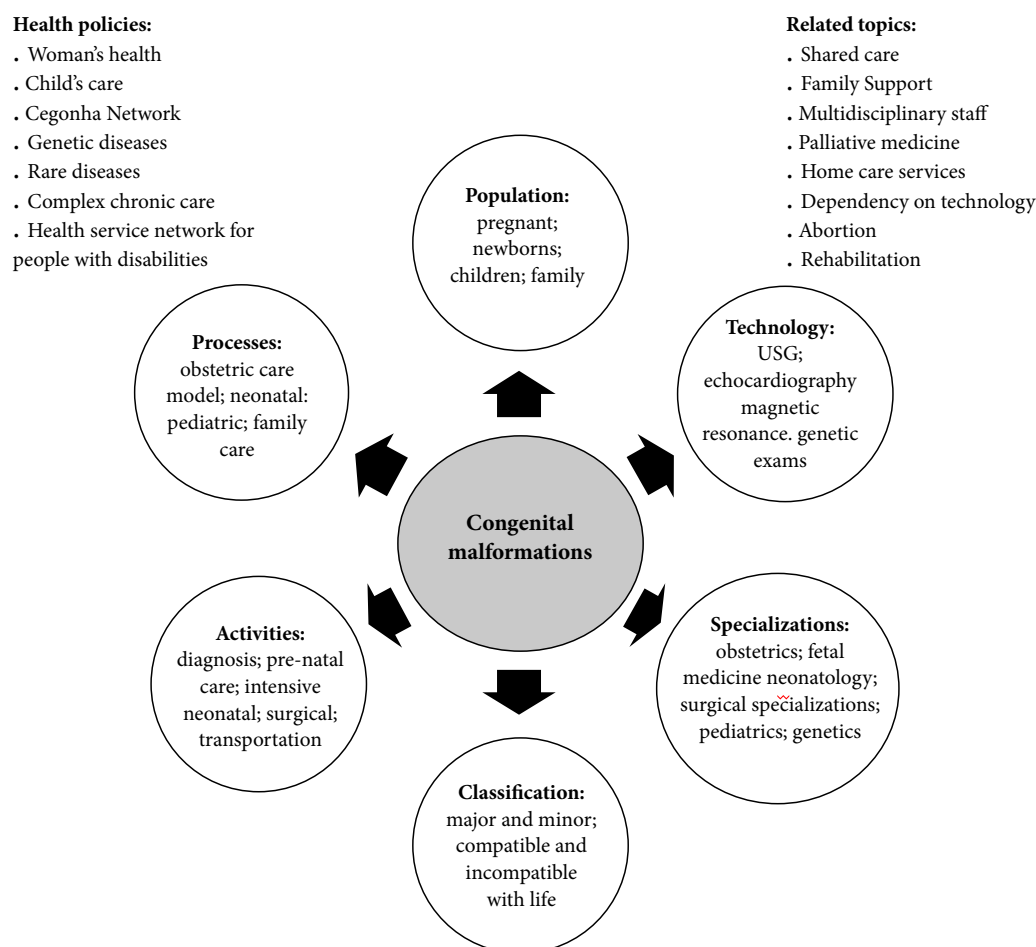


Figure 1. Summary of information obtained through strategic analysis.

Source: Author's, adapted from Crémadez M¹⁹.

To make such interlocution easier, it is necessary to develop articulation strategies of the different rationalities of clinical area, epidemiology and planning to identify new paths to approach health problems.

In *démarche stratégique* perspective, the strategic analysis stage was structured to help health organizations to identify, in its inward environment, homogenous groupings of activities or focus of activities, that are characterized by approaching the same strategic problem. The premise is that the construction of segments/ groups considers several criteria and not only biomedical ones, or organizational divisions as those established by institutional charts for departments, hospital wards and health services^{13,14}.

These questions were embedded to the present study, that stemmed from clinical elements as pathology characteristics, the assistance process and the assistance needs, but focusing on the organization of a set of specialized and hospital services. Thus, the study adopted meso management perspective to analyze health problems, understanding it is the space for management and organization of health services at regional/ state levels.

Considering so, the strategic analysis process of congenital malformation resulted in the identification of cases grouping that demand therapeutical resources, care processes and the design of similar care process to health attention network.

Chart 1. Strategic analysis according to the care profile of congenital malformation.

Primary characteristic of care	Keywords regarding pathology, technology and care							
	Is the MC classified as major or minor?	Is the Pathology compatible with life?	Does the diagnosis in prenatal alter the management of pregnancy?	Is there any demand of neonatal surgery?	Is the surgical intervention immediate 24/48hs)?	What is the main surgical specialization involved?	Is there any interference on the prognosis due to the Newborn transportation?	Is the outpatient follow-up specialized?
MC which demands neonatal and outpatient approach	Minor	Yes	No	No	No	Not applied	No	No
MC which demands neonatal and pediatrics surgical care	Major	Yes	Yes	Yes	No	Pediatric, plastic and orthopedic surgery	No	Yes
MC which demands early neonatal surgical care	Major	Yes	Yes	Yes	Yes	Pediatric and cardiac surgery and neurosurgery	Yes	Yes
MC which demands pre-natal and neonatal approach	Major	No	Yes	No	No	Not applied	Not applied	Not applied

Source: Author's.

The first group, called **Minor congenital malformation**, comprises several malformations concerning eyes, ear, face and limbs- as polydactyly, lip pits, pre-auricular appendages, bowed legs, pigmented nevus, and others – that are relatively frequent in populations and do not cause major problems of clinical repercussion to the bearer¹⁷.

Despite the fact they develop during the fetal period, they are not necessarily diagnosed in prenatal or they can even be not visible at the moment of birth, and they can be asymptomatic and/or have late manifestation. Newborn's physical examination is the main strategy to early detection¹⁷.

In these cases, pediatric follow-up happens mainly at basic attention level. From the first evaluation therapeutic plan is defined and, when necessary, an articulation with specialized service for treatment is also performed, and also surgical correction and/ or rehabilitation.

Major congenital malformations were separated in three groups. The first, **Congenital malformations of late surgical approach** that are compatible with life. As they do not represent any immediate life risk, at early neonatal period and also as they do not constitute any surgical emergency, they are so-called late ones. The main feature of these malformations is that there is an aesthetic impact, be it functional and/ or psychological, requiring specialized surgery, specialized outpatient follow-up and multidisciplinary health team. The main problems include facial segments and extremities, cleft lip and palate and genitals (ambiguous genitalia)¹⁷.

Diagnosis can happen at prenatal or at morphological and physical examination of the newborn. Utterly important for the surgical care is that hospital units are identified for the reference of cases and conformity of experienced staff to evaluate and manage these malformations, both at neonatal and pediatric time.

Chart 2. Groups of congenital malformations identified in order to organize care pathways and care segments (CS).

Classification	Groups of congenital malformations	Examples of pathologies that are part of the group
Minor	Group 1 – Minor congenital malformations	
	SA 1.1 Neonatal and pediatric approach	Talipes calcaneus; dysmorphic ears
Major	Group 2 – Congenital malformations of late surgical approach	
	SA 2.1 Pediatric surgical approach	Palate fissure; ambiguous genitalia
	SA 2.2 Orthopedic surgical approach	Congenital hip dislocation; Congenital club foot
	Group 3 – Congenital malformations of immediate surgical approach	
	SA 3.1 Pediatric surgical approach	Gastroschisis; congenital diaphragmatic hernia
	SA 3.2 Neurosurgery approach	Spina Bifida; mielomeningocele
	SA 3.3 Cardiac surgical approach	Aorta coactation; pulmonary atresia
	Group 4 – Congenital malformations incompatible with life	
SA 4.1 Neonatal and obstetric approach	Anencephaly; renal agenesis	

Source: Author's.

Another important aspect is to define a set of strategies of proper support to the family who need to be guided by a multidisciplinary health team about the impacts of the diagnosis to the newborn, options of treatment and follow-up^{24,25}. Such cases can require long-term outpatient care with a multidisciplinary staff, plastic surgeries for correction, home care, rehabilitation and psychological specialized care. The goal is to avoid functional complications (visual, hearing loss or ambulation) and esthetic and psychological problems (as in ambiguous genitalia case) in childhood/ teenagerhood years having impact also in adulthood^{24,25}.

Congenital malformations of immediate surgical approach are also compatible with life, but they bring serious clinical consequences to the newborn and they are classified as surgical emergency, representing the main cause of early morbidity and mortality. Among the malformations in this group, there are spina bifida, congenital heart defects, congenital diaphragmatic hernia, omphalocele, gastroschisis, and others²⁶⁻²⁸.

The majority of these malformations can be diagnosed at prenatal time – from the obstetric and/or morphological ultrasound – early diagnosis enables the definition of strategies to be followed during prenatal, the fetal treatment and the proper programming for the birth at reference units regarding high-risk pregnancy care. Due to its severity, delivery and neonatal care need to be performed at specialized centers, places that have real conditions to provide intensive neonatal surgical care, avoiding the transportation of the newborn between health units^{9,22}.

These cases are characterized as complex and chronic conditions and the units of reference need to be prepared for childcare who can become dependent on this technology, or who can depend on outpatient follow-up, also health care and, in some cases, palliative care, as well²⁹⁻³¹.

The third group, **Malformations incompatible with life**, includes the cases in which the severity of the congenital defects make impracticable extra-utero life maintenance. Such malformations can be diagnosed at prenatal, but there is no intervention that can change this diagnosis leading to fetal death during pregnancy, at the parturition soon after birth³².

The main incompatible with life malformation is anencephaly, but rarest cases can also be included, as bilateral renal agenesis, or the pancreas, acrania, and the complex and severe malformations as well as malformation syndromes³². In Brazil, the possibility of stopping a pregnancy is included in the law only in cases of anencephaly; for the other malformations incompatible with life there are different views and debates under bioethical, legislative and law perspectives^{32,33}.

However, units of reference for this group need to have multidisciplinary staff who are ready and well prepared for approaches of different situations, as the ones that implicate in the right of therapeutical anticipation of the delivery or, also, for cases that opt for going on with the pregnancy developing and improving support strategies for the mother and her family in all the stages and care processes, and the genetic counseling considering future pregnancies³²⁻³⁴.

In this work, strategic joint analysis of congenital malformations enabled a different view of the characteristic features of pathologies and medical specializations, bringing the care process to a more systemic perspective. At the same time, it broadened the dialogue of different areas and cores of knowledge, allowing a more collective way of explaining and approaching congenital malformations to the organization of a service network.

To this purpose, typologies were created, from the analysis of ways of classification and criteria used by clinic and epidemiology but that not necessarily reproduced them to the design of care pathways³⁵.

The discussion related to the need of identification of new typologies to guide the organization of health systems is present in the model of attention to chronic conditions framework aiming at using the health conditions concept³⁵. As shown by a Mendes, *classical typology classifies diseases as communicable and in chronic non-communicable diseases, having as criterion the etiopathogenesis. Such typology works well, mainly in the field of epidemiological studies, but it is not enough to support the organization of health attention systems*³⁵.

Another tool that has been used by health systems for patients grouping in agreement with resources use is Diagnosis Related Groups (DRG), in its various ways. One of its goal is to fit resources available to the needs of the patients groups assisted by hospital units, qualifying both care process and care profile. This methodology has been progressively embedded by countries to redefine their payment systems and as a tool of clinical governance, to cost management and assistance quality assessment^{36,37}.

The two references cited above seek new approaches for health problems considering the organization of service and attention care, where the first typology – health conditions – focus on a model of attention and organization in health services system while the second – DRG – produces new typologies from patients' grouping aiming at reorganizing in-house processes concerning hospital services.

However, reports and health strategic analysis proposals are still very little frequent for the identification of new ways of approach, or health problems grouping to subsidize the organization, programming and planning processes of specialized services and care pathways through a state or regional perspective.

As observed by Castellanos, description and explanation of an issue or health situation are not

a part of who describes it or even his/her position at the very moment of such description³⁸. As planning and programming processes take place at different system management levels or even at an organization, there will also be different ways of approaching a health problem, whether by clinical professionals or by unit health managers and the different levels of management.

Thus, the use and completion of health problems analysis tools – as the strategic analysis methodology presented in this work – can expand the articulation of different knowledge of clinic, epidemiology and health management and contribute to care pathways organization at regional levels.

The possibility to include health problems approach to the regional planning processes from case grouping can contribute to the definition of reference units, to the articulation of services and professionals, and also to the previous pact of responsibilities as well as the definition of care coordination mechanisms that are suitable for each line of care.

In the case of congenital malformation, an approach in wider perspective- without identifying the groups of case to the design of care pathways – would not contribute to a proper definition of care profile and identification of reference centers. On the other hand, excessive decomposition (as pathologies) could make care sequences structure so specific that they would lose sight of optimization and integration resources of care that concentration and grouping of case could offer. Without the definition of case groups, possibly, there would not be the minimum assistance number in each attention spot, and it impacts positively in the quality of care for some congenital malformations, as the ones of surgical approach³⁹.

Thus, it is believed that planning and programming processes at the level of care, if intended to reach higher effectiveness, need to consider the way as health problems are defined, described and approached at every attention level and at management of the health system space.

Strategic analysis, however, when decomposing a complex reality in subsets can lead researchers to lose sight of interfaces and connections of this problem with other areas of attention and sectional policies. For this reason, two elements were added once they emerged during the work of strategic analysis: public policies and related subjects.

In the case of congenital malformations, this interface occurs from initiatives that are at the level of health policies considering chronic com-

plex conditions in children and teenagers, rare diseases – and at the level of organization and broadening of care strategies and practices – such as home care service, palliative care and family support^{40,41}.

Therefore, it is important to highlight that the elaboration of a proposal aiming at organizing services and care pathways, no matter the health problem/ condition or, even the level of system it is designed to, needs to identify interfaces and open a dialogue with different areas and subjects, widening the articulation between care and management.

Finally, there should be highlighted how suitable the strategic analysis proved itself to be and allowed the grouping identification of cases

which demand homogenous set of care strategies and health services with similar care profile, so as to contribute to organizing congenital malformation attention. Such analysis can contribute, precisely, to think of regional planning processes and in the case of health problems that are characterized as chronic complex conditions and require the articulation of specialized services of high technologic density.

Thus, the use of strategic analysis and the grouping of cases in order to elaborate care pathways can also be understood as an effort to organize different rationalities (clinical, epidemiology and planning/ management) to ensure the completeness on the answer to a health problem of some population.

Collaborations

L Binsfeld, M Gomes and R Kuschnir participated in the manuscript's conception, writing and review.

References

- Kalichman AO, Ayres JRCM. Integralidade e tecnologias de atenção à saúde: uma narrativa sobre contribuições conceituais à construção do princípio da integralidade no SUS. *Cad Saude Publica* 2016; 32(8):1699-1712.
- Silva NEK, Sancho LG, Figueiredo WS. Entre fluxos e projetos terapêuticos: revisitando as noções de linha do cuidado em saúde e itinerários terapêuticos. *Cien Saude Colet* 2016; 21(3):843-851.
- Organizacion Pan-Americana de la Salud (OPAS). *Redes integradas de servicios de salud: conceptos, opciones políticas y hoja de ruta para su implementacion en las Americas*. Washington DC: OPAS; 2010. [acceso 2022 maio 25]. Disponible en: https://www.paho.org/hq/dmdocuments/2010/APS-Redes_Integradas_Servicios_Salud-Conceptos.pdf
- Bernardino FBS, Gonçalves TM, Pereira TID, Xavier JS, Freitas BIBM, Gaíva MAM. Tendência da mortalidade neonatal no Brasil de 2007 a 2017. *Cien Saude Colet* 2022; 27(2):567-578.
- Souza CDF, Magalhães MAFM. Novo século, velho problema: tendência da mortalidade infantil e seus componentes no Nordeste brasileiro. *Cad Saude Colet* 2021; 29(1):133-142.
- SzwarcwaldCL, Leal MC, Almeida WS, Barreto ML, Frias PG, Miranda M, Theme Filha MM, Soares RM, DomingueM, Franca EB, Gama S, Coccolini CS, Victora C. Child health in Latin America. In: *Oxford Research Encyclopedia of Global Public Health*. US: Oxford University Press; 2019. <http://dx.doi.org/10.1093/acrefore/9780190632366.013.37>
- Pinto M, Gomes R, Tanabe RF, Costa ACC, Moreira MCN. Análise de custo da assistência de crianças e adolescentes com condições crônicas complexas. *Cien Saude Colet* 2019; 24(11):4043-4052.
- França EB, Lansky S, Rego MAS, Malta DC, França JS, Teixeira R. Principais causas da mortalidade na infância no Brasil, em 1990 e 2015: estimativas do estudo de carga global de doença. *Rev Bras Epidemiol* 2017; 20(Supl. 1):46-60.
- Binsfeld L. *Planejamento e programação nas redes de atenção à saúde: organização da atenção às malformações congênitas* [tese]. Rio de Janeiro: Instituto Nacional de Saúde da Mulher, da Criança e do Adolescente Fernandes Figueira/Fiocruz; 2020.
- Centers for Disease Control and Prevention. gaining consensus among stakeholders through the nominal group technique. *Evaluations Briefs* 2018. [cited 2022 maio 25]. Available from: <https://www.cdc.gov/healthyyouth/evaluation/pdf/brief7.pdf>
- Carvalho ICBM, Rosendo TMSS, Freitas MR, Silva EMM, Medeiros WR, Moutinho NF, Pimenta IDSE, Gama ZAS. Adaptation and validation of the World Health Organization's on Safe Childbirth Checklist for the Brazilian context. *Rev Bras Saude Mater Infant* 2018; 18(2):401-418.
- Guerra FAR, Llerena JC Junior, Gama SGN, Cunha CB, Theme Filha MM. Defeitos congênitos no município do Rio de Janeiro, Brasil: uma avaliação através do SINASC (2000-2004). *Cad Saude Publica* 2008; 24(1):140-149.
- Crémadez M. *Le management stratégique hospitalier*. Paris: Elsevier Masson; 1997.
- Artmann E, Llerena Junior JC, Pereira LT, Binsfeld L, Rivera FJU. Análise estratégica de um centro de genética médica em um instituto de pesquisa nacional em saúde no Brasil: desafios para o SUS. *Cien Saude Colet* 2021; 26(Supl. 2):3481-3492.
- Vieira T, Giugliani R, organizadores. *Manual de genética médica para atenção primária à saúde*. Porto Alegre: Editora Artmed; 2013.
- Horovitz DDG, Cardoso MHCA, Llerena JC Junior, Mattos RA. Atenção aos defeitos congênitos no Brasil: características do atendimento e propostas para formulação de políticas públicas em genética clínica. *Cad Saude Publica* 2006; 22(12):2599-2609.
- Leite JCL, Dewes LO, Giugliani R. *Manual de defeitos congênitos*. Porto Alegre: Editora Livres; 2007.
- World Health Organization (WHO), Centers for Disease Control and Prevention, International Clearinghouse for Birth Defects Monitoring Systems. *Birth defects surveillance: atlas of selected congenital anomalies* [Internet]. 2014. [cited 2022 maio 25]. Available from: <https://apps.who.int/iris/handle/10665/127941>
- Bruns RF, Araujo Júnior E, Nardoza LMM, Moron AF. Ultrassonografia obstétrica no Brasil: um apelo à padronização. *Rev Bras Ginecol Obstetr* 2012; 34(5):191-195.
- Telles J. Protocolo mínimo da ultrassonografia morfológica do segundo trimestre. *Rev Soc Ultrassonografia* 2013; 14(18):21-24.
- Moreira MEL, Lopes JMA, Carvalho M. *Recém-nascido de alto risco: teoria e prática do cuidar*. Rio de Janeiro: Editora Fiocruz; 2004.
- Barreiros CFC, Gomes MASM, Gomes Junior SCS. Mortalidade por gastrosquise no estado do Rio de Janeiro: uma série de 10 anos. *Rev Saude Publica* 2020; 54:63.
- Onocko R. *O planejamento no labirinto: uma viagem hermenêutica*. São Paulo: Hucitec; 2003.
- Bolla BA, Fulconi SN, Baltor MRR, Dupas G. Cuidado da criança com anomalia congênita: o olhar da família. *Rev Esc Anna Nery* 2013; 17(2):284-290.
- Diseth TH, Emblem R. Long-term psychosocial consequences of surgical congenital malformations. *Semin Pediatr Surg* 2017; 26(5):286-294.
- Rajiah P, Mak C, Dubinsky TJ, Dighe M. Ultrasound of fetal cardiac anomalies. *AJR Am J Roentgenol* 2011; 197(4): W747-W760.
- Haddock C, Skarsgard ED. Understanding gastroschisis and its clinical management: where are we? *Expert Rev Gastroenterol Hepatol* 2018; 12(4):405-415.
- Binsfeld L, Gomes MASM, Kuschner R. Malformações congênitas de abordagem cirúrgica imediata no estado do Rio de Janeiro, Brasil: análise para a organização do cuidado em rede. *Cad Saude Publica* 2022; 38(2):e00109521.
- Rabello CAFG, Rodrigues PHA. Saúde da família e cuidados paliativos infantis: ouvindo os familiares de crianças dependentes de tecnologia. *Cien Saude Colet* 2010; 15(2):379-388.

30. Costa MTF, Gomes MA, Pinto M. Dependência crônica de ventilação pulmonar mecânica na assistência pediátrica: um debate necessário para o SUS. *Cien Saude Colet* 2011; 16(10):4147-4159.
31. Drucker LP. Rede de suporte tecnológico domiciliar à criança dependente de tecnologia egressa de um hospital de saúde pública. *Cien Saude Colet* 2007; 12(5):1285-1294.
32. Löwy I. Indicação de aborto por anomalia fetal: como abordar um assunto difícil. *Cad Saude Publica* 2020; 36(Supl. 1):e00188618.
33. Fonseca SC, Domingues RMSM, Leal MC, Aquino EML, Menezes GMS. Aborto legal no Brasil: revisão sistemática da produção científica, 2008-2018. *Cad Saude Publica*.2020; 36(Supl. 1):e00189718.
34. Domingues RMSM, Fonseca SC, Leal MC, Aquino EML, Menezes GMS. Aborto inseguro no Brasil: revisão sistemática da produção científica, 2008-2018. *Cad Saude Publica* 2020; 36(Supl. 1):e00190418.
35. Mendes EV. Entrevista: A abordagem das condições crônicas pelo Sistema Único de Saúde. *Cien Saude Colet* 2018; 23(2):431-436.
36. Mihailovic N, Kocic S, Jakovljevic M. Review of diagnosis-related group-based financing of hospital care. *Health Serv Res Manag Epidemiol* 2016; 3. DOI: <https://doi.org/10.1177/2333392816664789>
37. Meng Z, Hui W, Cai Y, Liu J, Wu H. The effects of DRGs-based payment compared with cost-based payment on inpatient healthcare utilization: a systematic review and meta-analysis. *Health Policy* 2020; 124(4):359-367.
38. Castellanos PL. Sobre el concepto de salud enfermedad. *Bol Epidem OPS* 1990; 10(4):1-7.
39. Sacks GD, Ulloa JG, Shew SB. Is there a relationship between hospital volume and patient outcomes in gastroschisis repair? *J Pediatr Surg* 2016; 51(10):1650-1654.
40. Moreira MCN, Albernaz LV, Sá MRC, Correia RF, Tanabe RF. Recomendações para uma linha de cuidados para crianças e adolescentes com condições crônicas complexas de saúde. *Cad Saude Publica* 2017; 33(11):e00189516.
41. Pinto M, Madureira A, Barros LBP, Nascimento M, Costa ACC, Oliveira NV, Albernaz L, Campos DS, Horovitz DDG, Martins AJ, Moreira MCN. Cuidado complexo, custo elevado e perda de renda: o que não é raro para as famílias de crianças e adolescentes com condições de saúde raras. *Cad Saude Publica* 2019; 35(9):e00180218.

Article submitted 20/01/2022

Approved in 03/10/2022

Final version submitted 05/10/2022

Chief editors: Romeu Gomes, Antônio Augusto Moura da Silva

