

# Modeling Family Outcomes: From the Family Ecosystem to the Iceberg Model

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**Abstract.** Today Rare Diseases are becoming in Europe a public health priority. In one hand, patients and their family have the wonderful hope of intensive research activities focusing on genetics, proof of concepts and drug development and on the other hand story telling and family biographies show minor progress in the clinical management and day to day care. Metaphorically the Rare Diseases history is like an Iceberg with a 10% visible part and 90% of unanswered solutions for a better quality of life or at least solutions to support the burden of the disease.

Family Portraits could be classified in a matrix with a vertical axis from behavior to emotion for the impact and from the family unit to the environment for the source of the problem. The model of Singer and Irvin derived from the pattern of coping of stress; called ABCX should be the blueprint for a real policy of care. The stressors here are the "family crisis". An adjustment balancing between external and internal family resources will target a positive adaptation. Applying the ABCX model and considering the ecosystem of the family centric approach as described by Urie Bronfenbrenner, the family outcomes could be defined as criteria allowing positive adaptations. Models provide pathways for a better understanding of patients' and families' needs.

**Keywords:** Family Outcomes, Patient-Driven Research, Health Models, Disease Burden, Health Behaviours

## 1. Introduction

The European Project for Rare Diseases National Plans Development (EUROPLAN) has addressed one unanswered question to date: "what is the scope of patient-driven research?" Identification of needs and priorities for translational and social research, modes of fostering them and promotion of interdisciplinary cooperative approaches has been largely recommended.

Beside the success stories of "orphan drug development" and genetic research during the last decades, family story telling and family sketches analysis show minor progress in the clinical management and day to day care beside tremendous unmet needs. Quality of Life and measurements of the burden of the disease have been neglected. A call to action for active research in the field of human and social sciences is urgent.

## 2. Modelling Family Outcomes

We reviewed and discussed different existing health models for a better understanding of the family outcomes and behaviours in the field of rare diseases and child disability.

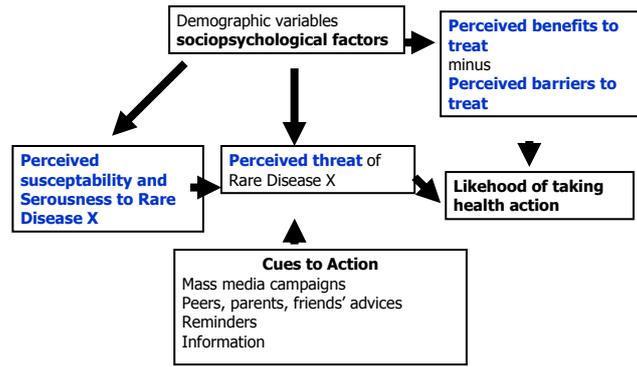
### 2.1. The Health Belief Model

The **health belief model** (HBM) was one of the first, and remains one of the best known social cognition models. It is a health behavior change and psychological model developed by Irwin M. Rosenstock in 1966 for studying and promoting the uptake of health services. The model was furthered by Becker and colleagues in the 1970s and 1980s [1] (figure 1). Originally, the model was designed to predict behavioral response to the treatment received by acutely or chronically ill patients, but in more recent years the model has been used to predict more general health behaviors. The HBM suggests that your belief in a personal threat together with your belief in the effectiveness of the proposed behavior will predict the likelihood of that behavior.

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This model fails to explicit behavior in hopeless situations like having a rare disease or a disability while a cure therapy is lacking.



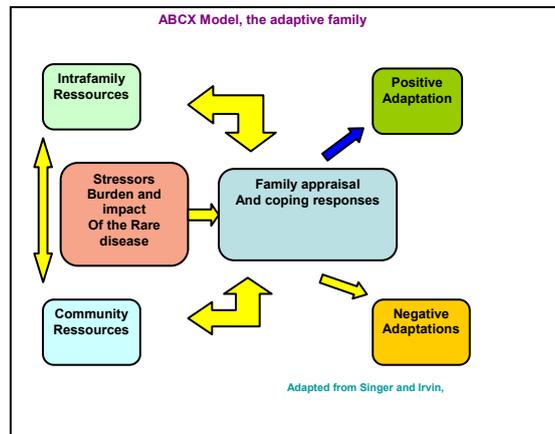
Adanted from MH Becker

Fig 1: The Health Belief Model

## 2.2. The Adaptive Model

Day to day life with a Rare Disease could be seen as an adaptive strategy using a stress coping model. The stressors here are the family crisis due to an unbalance between external and internal resources.

Efforts to understand and explain family response to stressful situations traditionally have been concentrated on the relations among three basic phenomena: the stressful event(s) and its associated hardships; the outcome of stress (crisis and adaptation); and the intervening factors between the two. The central question is, how much and what kinds of stressors, mediated by what family resources and processes, shape the course and ease of family adaptation [2]?



Adapted from Singer and Irvin,

Fig 2: The Adaptive Strategy (ABCX model, adapted from Singer and Irvin)

It appears, however, that the external environment, as the family experiences it, is more influential than the family's internal resources. This ability to perceive the overall situation as coherent, as one that "makes sense," and to perceive a "fit" between the family and the circumstances, in turn, is of great value to the family in facilitating its adaptation. Thus, the ABCX model fails to take into account the experience and the patient-expert phenomenon.

## 2.3. The Family Portraits

Portraits could be classified in a matrix with a vertical axis from behaviour to emotion for the impact and from the family unit to the environment for the source of the problem [3].

Reactions to disability, allow a construction of family typologies (Figure 3) having children with disabilities or rare diseases. This approach has benefited from progress and evolution of human and social sciences. The creation by Philipp Ferguson of a mapping matrix has provided 4 portraits of the reactions of families to disability based on 2 questions:

What is the nature of the parental response to having a child with a disability? (Vertical axis)

What is the source of this reaction? (Horizontal axis)

The answers to the first question were often and across scientific fields in emotional or behavioral categories. For the second question, it is tempting to respond by also two categories. Some reactions are inherent or normative impact of disability on the family or contextual influences within and outside the family. Here the horizontal axis is our Social Self. By combining the two to two categories, it is possible to conceptualize an approach with the answers to two questions about the nature and source of disability and the orientation of a support. Thus there are four approaches: psychodynamic, behavioral, psychosocial and socio-political with the characteristics of each parent.

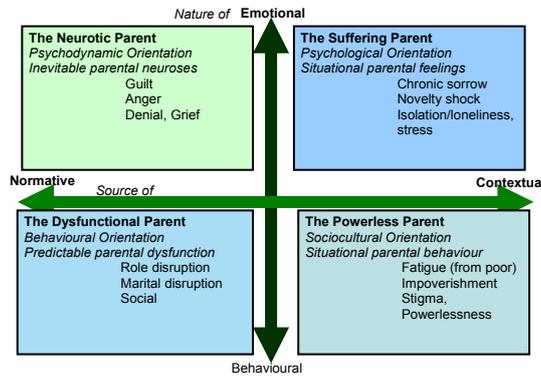


Fig 3: The Family Portraits (adapted from Ferguson)

## 2.4. The Iceberg Model

We have defined the Iceberg Model which seems responding to the family outcomes modelling. Metaphorically the Rare Diseases history is like an Iceberg with a 10% visible part and 90% of unanswered solutions for a better quality of life or at least solutions to support the burden of the disease.

Speaking of the Iceberg in disability issues could cast a chill! Icebergs as penguins and polar bears are the icons and symbols and an invitation to travel as a journey of initiation...

The Iceberg as the symbol is a truncated language. Is visible only the tip! The symbol from the Greek "symbolon" is an object in two parts, each owned by one person. The missing part is to seek to reform a coherent whole.

The metaphor of the iceberg can be applied to a parent of a child with a disability. He or she is like an Iceberg, a block of freshwater ice drifting on the sea, standing out the front of the polar glaciers or floating ice barrier as a detached belonging to a "nowhere". The metaphor is so evocative and speaks to us.

Icebergs formation challenges our educational and psychoanalytic approaches to understanding the life course.

The physical basis of formation of Icebergs invite to this journey of body "symbolic" in two parts, one visible and the other submerged.

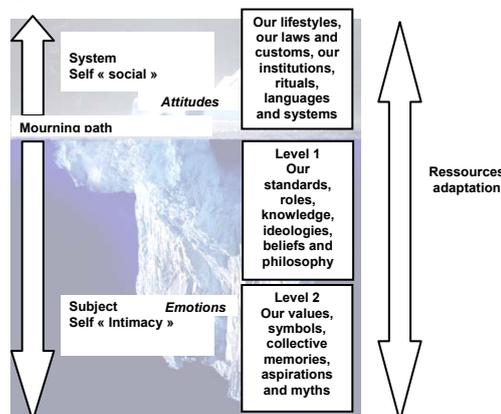


Figure 4: The Iceberg Model

The Iceberg model provides a systemic approach of the burden of a Rare Disease or any disease with disability, therefore an overview of patient outcomes and a model of resource adaptation.

Iceberg theory was embodied in the literature by Ernest Hemingway through his works like *The Snows of Kilimanjaro*, *Death in the afternoon*, or *the Old Man and the sea*. From his experience as a journalist in violence of the Spanish Civil War, he removed from his writing all unnecessary words, simplifying the structure of the sentence and focusing on objects and actions: "What we need is write one true sentence. Write the truest sentence that you know. "Wanting to" write like Cezanne painted ", that is to say, by removing all the" stuff "... In his novels, he emphasizes the courage to face adversity; his heroes are strong men, silent like his entire fathers child with a disability.

The lesson from the work of Hemingway, an intense creative energy, through the hollow of the story and an issue of will to live: "Make people feel something more than what they understand". ... Metaphorically, it represents the visibility and invisibility of disability, the social self and inner self separate from the waterline that is the journey of grief. In this sense it models a diagnosis of suffering and helplessness models called reinsurance plan of care ... A load on a floating model defies physical law promises further recommendations make sense!

Mourning in the sayings is "like an onion," you peel a little each day in his career, and it makes you cry! The grieving process takes time and is personal. It represents the waterline delineating between the visible and the invisible "Iceberg" individual. The steps are summarized in Figure 4. People experiencing repetitive shocks of varying intensity, state of amazement from the first word received as a diagnosis, at first glance social rejection, awkwardness, stigma, multifaceted pain and exhaustion to an injustice. This is the waterline, because suppressed feelings, the emotion, the masked depression, symptoms of unlabeled, hostility domesticated, pseudo-static polite acceptance resurface and become available. We must constantly fight to change the look when the disability is so visible? When the eye turns away from each other in passing of what one wants to see. See the reality of a different life, can be unbearable. Opening windows of nearby beings with multiple disabilities eyes diverted to the other, the vision of multiple disabilities may change.

That this look has changed is not limited to one of compassion or empathy! Being multiple disabilities is perhaps not standing by physical disability, but is not kneeling.

The vision is dependent on a complex physiology. We can not see because of an ophthalmologic problem, neurological or nervous. But look appeals to the desire to see and well discern in what experts called visual accommodation. The look first requires people to have their eyes open. Have a look that changes when you have open eyes that work, forces him to say that blindness somewhere in the head does not exist. One can look at whatever the state of the view.

This reminds us just be the singularity that exists. This singularity called a little more attention: attention so unique and so devoted to these professionals. Between the eyes and that kills the look of compassion, which changes the look is not enough, if the will to change is not there. Laws, records, year of disability associated with the lack of real resources commitment, and drive belts can not change the vision of everyday life, or the eyes of others. Blindness of the Society is still so common. Fighting for the next exchange, required energy and courage, the courage to be seen but not heard from parents, relatives and associations.

Multiple Disabilities reminds us of our individuality, our own body or the wooden puppet that we are still always manipulated. We can see, act or think for others instead of another? All these looks, all this shared vision of those who defend the status of being with multiple disabilities are only the blindness of a therapeutic group and individual. These looks reach the foundation of a new culture, the culture of disability. This culture requires above all the rapid changes in education reform by integrating disability issues at school and by including training in disability issues in the curriculum of legal studies, architecture, medical, economic and educational.

Contemplating this Visible requires political courage to build an inclusive society, open a little more "looking" toward the most vulnerable and ultimately to all.

The disadvantage of the symbol of the Iceberg in disability is often just 1/9th or 1/10th of things. It suits to discourse on the visible part.

Immersed in a world of silence and swarming with life forms in the variations of light and coldness of the ocean, the underwater part of iceberg contains all the "why, why me, how I'll do ...".

In the birth of a child with disability or a rare disease is described as a Parental painful journey to fulfill an impossible mourning "their identity is sprayed, self-esteem damage. The arms fall. The faces are frozen. Depression shade sails of lead ..." Finally death can be seen as the only issue to complete the course of mourning as seen waterline by changing the weight of pain that immerses all. This is perhaps the purest pain ever known living being.

Empathy and compassion are by no means the only illumination of the submerged part. The most complete description or the fairest of the Intimate Self engulfed not only purpose for a psychoanalytic treatment. She calls, she asks about individual responses to disability, the policies put in place and the fight against the labels and false beliefs.

Thumbnails image, social contexts and political support have changed in 100 years and were heavily tinted and colored political trends and scientific crossed moments in history.

Our Iceberg model seems to respond to the global human ecosystem, a social developmental family centric approach described by Urie Bronfenbrenner [4].

### **3. Conclusion**

Models provide pathways for a better understanding of needs and also a spectrum of Patient-Driven Research perspectives.

Research including Socio-psychological projects will provide cues to responses to so many day to day unmet needs and suffer beside the need to have a speculative curative therapy. Patient and family outcomes in the field of Rare Diseases and disability should be systematically assessed to respond to Public Health Priority.

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