

Family intervention as a diagnostic complement in expanded pediatrics. The puzzle model

E. Eleonora De Petre, M.D.^a

ABSTRACT

The puzzle model is a brief intervention, an interdisciplinary diagnostic and therapeutic tool focused on the family. Its aim is to determine the impact of family dynamics on eliciting and developing the patient's symptom. The adult and the child joint participation, by means of discourse in the case of the adult, and cognitive and projective games, in the case of the child, enable, through mutual interaction, the disclosure of the family structure and dynamics. The therapist, based on the elements arising from the interaction of both parties during the interview, reframes or sets up the new perspective of the described problem.

The objective of this article is to describe the technique used. Two clinical cases and some results of this model are presented as an example.

Key words: family therapy, children, psychosomatic disorder, interaction

<http://dx.doi.org/10.5546/aap.2013.128>

INTRODUCTION

Consultation due to behavioral or psychosomatic symptoms is common in pediatrics, and they affect the child's health, growth and development.

Based on the concepts of expanded pediatrics,¹ since the family is preferably the core around which the earliest bonds and interactions are created and intertwined, it is important to look at it from the systemic family therapy approach to contribute to the pediatric diagnostic process.^{2,3} This multidisciplinary intervention model is provided as a diagnostic and therapeutic tool for dealing with this type of symptoms taking into account that the child's problem can be part of or arise from a dysfunctional family system and social environment.²

Unlike classic, systemic or psychoanalytic family therapies,³ this model of care turns the child into the main character. Its aim is to determine the impact of family dynamics on eliciting and developing the patient's symptom and enables to do a differential diagnosis.

The therapist constructs a hypothesis that discloses the likely reasons for family imbalance. This comes from the possibility of disclosing the child's perceptions evidenced through the interaction between the child's projective game and the joint participation of parents.^{6,7}

Based on the emotions elicited and shared among participating members and on the therapeutic intervention, the family group's own resources are evidenced and can be used to establish new modes of relationship among the family members as a solution to the problem that had led to consultation.^{8,9} The child's feelings and emotional needs, which were ignored by the adult up to that moment, become the center of attention.

The resolution of symptoms will therefore be the consequence of the possibility of reframing or setting a new perspective on the problem.

The objective of this article is to describe the model and provide the example of two clinical cases.

Description of the therapeutic technique

The puzzle model is based on three to five semi-structured, interdisciplinary, diagnostic and therapeutic interviews, each one having a specific objective (*Table 1*). The projective game played by the child in the presence of his/her father serves as the main vehicle for change in the family system (*Annex 2*). The team consists of a task force specialized in family approach in which the pediatrician that refers the child is included.

There is a thin line between organic and psychopathological states, so it is likely that on the first visit the pediatrician should look deeply for organic causes.

If necessary, the patient or family members that appear as the source of the problem will be referred to a more protracted therapy so as to consolidate the results achieved (*Annex 1*).

a. Pediatrician and Psychiatrist for Children and Adolescents in the Outpatient Facilities of Hospital de Niños R. Gutierrez and member of the Systemic Family Team of the Mental Health Center No. 1 of the City of Buenos Aires.

E-Mail:

E. Eleonora De Petre, M.D: eledepetre@hotmail.com.ar

Conflicto de intereses: Ninguno que declarar.

Recibido: 10-8-2012

Aceptado: 12-18-2012

Clinical cases

1. Nine-year old boy. Reason for consultation: referred by his school due to introversion, lack of eye contact and of social integration. He does not write nor participate in class. Lack of emotional contact: he does not let anyone kiss or touch him. He showed adequate intelligence and learning knowledge. Asperger's syndrome is suspected.

His family history included the separation of his parents due to violence. At two years old, his father would lock him up and prevent him from being in contact with his mother. Mother custody. No contact with his biological father for the last five years. The mother has been with a new partner for the past four years. She claims the child's behavior is due to the damage caused by his father.

The boy is collaborative and expressive during consultation. The projective game played by the boy in the presence of his mother allowed her to realize the child's need of having a father, how strong he was, and that he was not afraid of his biological father. The mother discloses before the boy the unclear reasons why she did not let him get close to his father nor his stepfather. Reframing the problem led to the family rearran-

gement. The boy started going for a walk with his stepfather and describes him as a spectacular magician.

Changes were observed in the third interview: he lets his mother kiss him, he stares at people in the eyes, he started relating with his schoolmates, he paints with colors and gets very good grades at school.

2. Two-and-a-half year old girl. Reason for consultation: lack of weight gain and retarded speech development (says only three words). The child is irritable and aggressive. Maturation delay is suspected because of paternal sexual abuse on a single occasion referred in a pediatric consultation by the mother. Family history: parents have been separated since the girl's birth. Paternal contact on only four occasions. The mother puts the girl's behavior down to this cause. General attitude: the girl responds to the therapist's instructions and seems calm and collaborative. From the moment projective games started, the mother sees that her daughter represents herself apart from the rest of the family group, she does not make a representation of her mother, and she places her baby sister as the main female figure. From the mother's

TABLE 1. Participation of the family, the therapist and the co-therapist during the interviews

Participation of the family	<ul style="list-style-type: none"> • Parent's discourse: reason for consultation, genogram, described history • Emotions detected in the child and parents upon arrival and during the interview • Child intervention through cognitive games • Moments of interference during the games • Child intervention through cognitive and projective games for the family evaluation • Parental emotion and reflection triggered by the game. • Shared emotional experience of affective findings with a therapeutic effect
Therapist intervention	<ul style="list-style-type: none"> • Interview directionality • Translation of the emotional experience into words based on the sensitive experience lived by the therapist • Comparison and overlapping of parent's and child's gazes • Indication of tasks
Co-therapist intervention	<ul style="list-style-type: none"> • Observation of the child throughout the interview: detection of moments of interference during games • Recording of projective games

discourse, it comes up that the girl is not a priority to her, that the girl makes her feel exhausted, and that she has to scold her all the time. In a moment of emotion, the mother perceives her daughter's sadness, who starts crying upon the therapist's intervention. This enables the reframing of the problem as mother-daughter indifference. The mother was advised to share moments with her daughter without scolding nor repressing her. In the second interview (15 days later), the following changes were observed: the girl gained 400 g, says phrases using a large number of words, and is peaceful at home.

DISCUSSION

This model helps to get a comprehensive examination of the patient that makes the differential diagnosis easier, by evaluating the involvement of family dysfunctionality as a determining factor of the occurrence, worsening or evolution of symptoms of uncertain organic cause. At the same

time, it allows to evaluate the patient's cognitive and behavioral aspects.

The innovation posed by this model lies in the creation of a dynamic structure mediated by projective games played by the child and the contrast with the adult's discourse. A traditional approach would allow the adult to preserve him/herself based on the possibilities granted by the verbal discourse. The child establishes an affective construction through the projective game and, in this way, she can make her voice heard.

The game, by acting as a facilitator of intra-family communication discloses the conflict that is probably causing the symptomatology. By acting as the main vehicle for change, it generates family learning and constructs the prognosis and treatment to follow.

Likewise, the model considers a paradigm shift by conducting a joint family interview taking the child as the main character.

The model framing is characterized by the pre-

TABLE 2. Interview description, objectives and contents

Interviews	Specific objectives	Contents
First	<ul style="list-style-type: none"> • Development of a hypothesis based on the interaction between projective games and the parent's discourse 	<ul style="list-style-type: none"> • Family dynamics triggered by the game
Second	<ul style="list-style-type: none"> • Detection of the underlying problem or hidden reason for consultation 	<ul style="list-style-type: none"> • Symptom evaluation • Accomplishment of the task assigned • Changes and difficulties observed: in the patient in particular and in other members of the family.
Third to fifth	<ul style="list-style-type: none"> • To investigate the efficacy of the therapy and define a new therapeutic approach for those who require it 	<ul style="list-style-type: none"> • Changes and difficulties observed: in the patient in particular and in other members of the family • Intervention on the family subsystem detected as the source of the problem • Reevaluation of the patient in the emotional and cognitive areas and also in relation to bonds • To work with the extended family group, if necessary, and to attach teachers' reports, if relevant

FIGURE 1. Representation of unique and particular family structure using cubes and figures



sence and subsequent follow-up by the referring pediatrician, who as a consequence of this intervention has found out unknown aspects about the patient. The involvement of the pediatrician as recipient of the patient's trust gives place to a strong therapeutic bond, thus enabling a deep and fast intervention in the system. Multiple emotions are freed that are shared by the family members during the intervention.⁹

This type of therapy causes changes in a short time because of the emotional involvement of the family during the interviews, turning therapy into the necessary means for the resolution of symptoms. These processes continue when the interviews are over, based on the maternal and paternal resources unveiled, and the supportive pediatric follow-up.

The model can be easily reached by a population with unmet needs, generally naive to psychological treatments, and works as an extension of a pediatric consultation.

The model was applied in hospital patients. Between July 2010 and October 2011, 87 patients

FIGURE 2. Representation of emotions grasped by the child. It allows the child to describe feelings of sadness, joy, pleasure or displeasure in relation to him/herself or his/her parents, and also to state his/her family values.



aged 2-12 years old were seen. Out of them, 11 were dropouts. Out of the 76 patients followed-up, an association between symptoms and a dysfunctional family dynamics was seen in 67, who showed a favorable evolution since the short-term intervention. The remaining 9 patients were referred for other treatments. The possibility of continuing a more prolonged therapy was offered to those requiring it.

As a conclusion, the puzzle model can be used as an effective therapeutic and diagnostic tool to tell the difference between symptoms of uncertain organic cause in the context of a pediatric consultation and it can be used to refer patients to other treatments in the case they required them. ■

REFERENCES

1. Pérez, Aurora. *Psicoanálisis; pediatría, familia y derecho*. 1era Ed. Buenos Aires: Carybe; 2001.
2. David M. Fergusson, Michael T. Lynskey. Adolescent Resiliency to Family Adversity. *J Child Psychol Psychiat* 1996;37(3):281-92.
3. Berenstein I. *Familia y enfermedad mental*. 2ª Reimpresión. Ed Paidós. Buenos Aires; 1984.

ANNEX 1

Description of the therapeutic model

The objective is that the therapist is able to find out the hidden dysfunctional relationship or transactional pattern resulting in symptoms (hypothesis) and which cannot be directly elicited from the adult's discourse.

The interview structure and the conversation directionality are guided by the therapist in terms of game symbolization.

Participating members are: the pediatric patient, from 2 to 12 years old, identified as the carrier of the problem, and accompanying adults, either parents or caregivers, who play such role. The child mainly participates through a cognitive and projective game (Annex 1), while adults participate through their discourse. Simultaneity of attention enables to generate interactions and create an environment with situations and exchanges similar to those at their home in the presence of a therapist.

The duration of each interview is determined by the time of development and outcome of an emotional process, experienced during the visit.

Steps of the therapeutic setting:

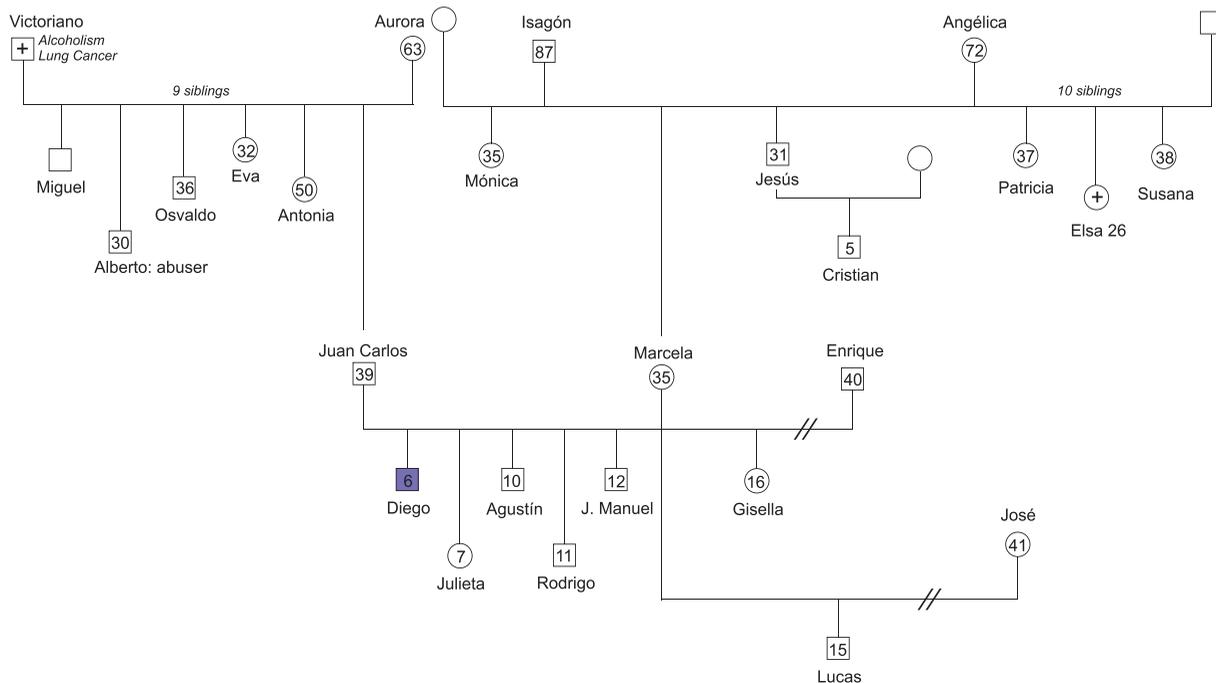
1) *Family genogram*: it includes three generations and reconstructs the unique history of the patient and his/her family (Figure 1).

The adult participates in two different ways: on the one side, he/she puts the glaring problem into words, provides information on fundamental landmarks and the historical context. This permits to give a special sense to the disease, situating it within the silent history that each person tells the others and the one that he/she tells him/herself.¹

In this stage of the interview the following is detected: a) significant discourses related to beliefs or emotional thoughts, attitude determinants, and b) memories, situations or people referred as emotional triggers.

Simultaneously, the child is active, playing the suggested games, therefore his/her spontaneous interventions and interruptions or discontinuities should be detected in relation to the mother's or father's story. The child's spontaneity is essential so as not to induce his/her replies and to detect foci of interest or concern.

FIGURE 1. Genogram



2) *Projective games*: to disclose the family structure and dynamics (Annex 1).

The set provided to the child consists of wooden cubes and figures that may be used freely or arranged in a puzzle with increasing difficulty. They represent images on which the child do projections at the request of the therapist: a) make up stories; b) personify animals to which the child ascribes certain capabilities and characteristics; and c) order figures or people by grouping them into subsystems to which the child assigns a meaning, by creating a determined spatial distribution. Each puzzle provides the therapist the possibility to assess different aspects of the patients and the family dynamics. This also allows evaluate cognitive skills.

The exclusive intervention of the child leads to a first moment of surprise, followed by the participation of all those who are present. The child is given the chance to become the center of attention during the consultation visit. The child's impressions are shared with the adult through the reflections and associations triggered by the game; these associations stem from shared experiences, enabling the interaction among the participating members.

The child expresses situations, perceptions and desires through games, using images that adults can explain. At the same time, feelings and intuitions can be alleviated when putting them in words. The cognitive activity maintained during the game buffers the emotions generated in the child when listening to the parent's discourse.

The game allows to assess children from two years old on, even if they are not capable of expressing themselves through drawing.

This moment in the consultation is therapeutic for the child because it allows him/her to feel recognized and to have his/her feelings unveiled.

From the parental standpoint, an impact occurs which directly involves them in the situation from the moment they pick up the family visual representation as perceived by the child. This modifies and enriches the discourse employed up to that moment, bringing to light what has been hidden explaining the reality of the situation based on the child's representation.

3) *Working with emotions*: emotions elicited are a core piece of treatment. Traumatic events, deeply rooted parental beliefs, fears or secrets lea-

ding to attitudes that give rise to symptoms, are reviewed. From the possibility of a regression, i.e., of returning to a less differentiated stage of development, so as to once again recreate what has been destroyed by trauma or disease, parents are able to become the architects for change.²

4) *Construction of a hypothesis*: based on the decoding of the meaning of interactions, it is possible to infer the family structure and functioning and, at the same time, to redefine the symptom that arose, in terms of a context that involves every member of the family.

With the contribution of the elements that emerge as pieces of a puzzle, a hypothesis can be constructed that enables the subsequent reframing or change of approach to the problem, that later can be confirmed by parents, thereby conferring veracity to it.³

5) *Recommendations to the family*: these are specific tasks indicated by the therapist to the family based on the hypothesis posed.

6) *Follow-up*: once the symptom has resolved, the patient is followed-up with the referring pediatrician. However, this model opens up the possibility of providing psychotherapy to those members of the family who thus require it based on the discovery of a problem different to the one they had consulted upon before.

Due to the high drop out rate among patients with chronic conditions, especially psychopathological ones,⁴ and with a higher dropout rate among low-income populations, telephone calls serve as a way of finding out the evolution of patients who are not coming back, in many cases because of their favorable course. ■

REFERENCES

1. Greenhalgh T, Hurwitz B. Narrative based medicine. Dialogue and discourse in clinical practice. London: BMJ Books; 1998.
2. Canevaro A. Quando volano i cormorani. Terapia individuale sistemica com il coinvolgimento dei familiari significativi. Roma, Italia: Edizioni Borla; 2009.
3. Bergman JS. Pescando barracudas. Pragmática de la terapia sistémica breve. 1a ed. España: Paidós, 1987.
4. Belmar NA, De La Parra Cieciva G, Guillón M, Valdés C. Deserción de pacientes de psicoterapia: exploración de factores pronósticos en base a la evaluación psicométrica de ingreso. *Rev Chil Neuro-Psiquiatr* 1995;33(3/4):273-8.

ANNEX 2

Projective games to evaluate the family

Games are offered following a specific order. The child is invited to put together the puzzles being offered. Then, based on the puzzle, the therapist prompts the child to play with his/her imagination, enabling him/her to use the same games in a projective fashion. Each puzzle will help to evaluate different aspects in relation to the child and the family dynamics.

The parent participates in the same games when being invited to think what part of the child's story calls his/her attention. In this way, the game is compared to the reality perceived by the adult. The differences found between both realities, or the reasons that might account for what the child is telling, help to shed a light on the key aspects of the family dynamics.

In the first two puzzles, the therapist participates only by observing. As of the third puzzle, the therapist gets directly involved by guiding the child's plot.

The interrelation between all the games and parental interventions provide the keys for the therapist to pose the hypothesis.

Figures:

1. The therapist observes if the puzzle is put together entirely or partially, and the degree of difficulty it entails.
2. The therapist observes whether the child includes the egg and the chick inside the chicken.
3. The patient is induced to choose the child figure he/she likes the most, and is asked to describe what the child figure might be doing. Intentionality is evaluated.
4. The child is invited to mix up the figures and rearrange them in a new order. Then the child has to find which figure he/she is and identify the rest of the figures by name. Relations are evaluated with the extended family.
5. The child is asked to think that he/she is standing in front of an imaginary house. He/she is asked whether he/she wishes to walk in. If the child agrees, the walls of the puzzle are removed and the different rooms are exposed for the child to walk around doing different activities and finding different people to join him/her. Examples: Who are at the table with him? Who is inside the house? What TV program did the child watch and what episode? The child is prompted to make a wish upon the star that can be seen from the room.
6. The child is situated in this scenery and is asked to image where he/she could be or who could the persons therein be. The child is asked to concentrate on the character disguised in front of the tree to whom he/she should confer a benevolent or intimidating attitude. In contrast, regarding the character of the girl dressed in white who is carrying some flowers, the child will assign her the capacity to give affection by means of the distribution the character will do of the flowers. The child will situate the pet as a member or not of the family, and with the car the child will indicate whether somebody is arriving or leaving (those who move around).
7. City: in this new context, the child will choose a place to live and who he/she wishes to live with. In the neighboring house, the child will place other people (known or unknown). The child is invited to take a ride with the cars; thereby recreational aspects can be evaluated. The different people who are significant to the child will be distributed in a particular way in the cars (setting up different subsystems). When arriving at the coast, these same people will be distributed for a boat ride. The child will indicate who travels on the tugboat and who on the ship. The executive capacity and the capacity for change will be evaluated.
8. Circus: the child is invited to be part of the circus staff together with his/her family, who will be assigned to the different characters: a tightrope walker, a juggler, a clown holding balloons, a clown holding balls, a tamer, a lion, and an elephant. Spatial and functional relationship among the members is evaluated.
9. Zoo: the child is asked to identify the animals and him/herself and his/her family by assigning each animal a specific characteristic. The caged animals will be invited to be released or not providing a reasoning behind it. The therapist will pay special attention to the spatial distribution (central or peripheral positions, closeness, habitat assigned to each animal: air, water or earth). Animals will be invited to walk around in the search for other animals' company.
10. Bears: represent emotions captured by the child. It enables to express feelings of sadness, joy, pleasure or displeasure in relation to him/herself or his/her parents, and also to reveal family values.
11. Cubes: the child represents his/her family structure, roles, hierarchies, alliances, closeness or distance using wooden cubes and figures.

The figures represent a father, a mother, a boy, a girl and a baby. The child is offered to add more figures, if he/she wishes to. After putting together the puzzle, the child will indicate who he/she is and then will tell who the rest are. It is important to know that in many cases the patient uses the father or mother figures to

represent him/herself or the baby to represent one of his/her parents. This choice allows to evaluate roles within the family. Likewise, the height to which each figure is placed is related to the hierarchy or significance within the family. Spatial distributions are useful to indicate emotional distances. ■

Sequence of games offered during therapy

1) Figure: identity



2) Chicken and chick: mother-child bond



3) Kids: intent



4) Family with grandparents: extended family



5) House: perception of her/himself at home and wishes



6) Girl with flowers: emotional capacities and fears



7) City: subsystems, executive capacity and flexibility



8) Circus: intrafamily relationships



9) Zoo: subsystems, identifications, attitudes, dangers and emotional blockages



10) Bears: moods



11) Cubes and figures: family structure



Administration of the National Screening Test (PRUNAPE) at home level in inner cities

Horacio Lejarraga, M.D.^a, María Cecilia Pascucci, M.D.^a, Celina Lejarraga, M.D.^a and Fernando Nunes, M.D.^a

ABSTRACT

The administration of the National Screening Test (*Prueba Nacional de Pesquisa*, PRUNAPE) at households with unmet basic needs (UBNs) can be associated with a high proportion of false positive results. In order to reduce such proportion, the adequacy of the household was assessed in a survey conducted in 2012 by the Environmental Health Division of ACUMAR in La Matanza and Florencio Varela districts, based on five factors: uneven floor, scarce space according to certain PRUNAPE guidelines, environmental noise, privacy, and lack of necessary furniture. If household conditions were inadequate, the PRUNAPE was administered in a trailer located nearby; if the child was “non collaborative” at home, the test was readministered in the trailer. Psychomotor development was evaluated in 2174 children younger than 6 years old. All surveyed households were adequate for the administration of the PRUNAPE. In households, 98 children (4.4%) were “non collaborative” with the PRUNAPE and were referred to the trailer; 58 (59%) of them attended the trailer and 28 (48%) passed the test. The PRUNAPE may be administered in the field, in households with unmet basic needs provided the described precautions are taken.

Key words: psychomotor development, screening for developmental disorder, development surveys, population with UBNs, PRUNAPE

<http://dx.doi.org/10.5546/aap.2013.132>

INTRODUCTION

The National Screening Test (*Prueba Nacional de Pesquisa*, PRUNAPE¹) has been validated² for the timely detection of developmental disorders in children younger than 6 years old.³ It was designed for its administration in a pediatric outpatient office^{1,4} and has also been used in prisons⁵ and in-the-field household surveys.⁶ In 2012, the Environmental Health

Division (*Dirección de Salud Ambiental*, DSA) of the Matanza-Riachuelo River Basin Authority (*Autoridad Cuenca Matanza-Riachuelo*, ACUMAR), a state agency established for the environmental sanitation of 14 districts on the borders of the City of Buenos Aires and the Riachuelo river, conducted a survey on nutrition, development and toxicological analysis (ENU DPAT II).⁷ The survey was conducted in areas with a high prevalence (30% or higher) of families with unmet basic needs (UBNs), in the districts of La Matanza (LM) and Florencio Varela (FV). Children were assessed in their homes, but the household conditions and surrounding area (environmental noise, lots of people watching, etc.) might interfere with the result of the test. One of the possible results of the PRUNAPE is that the child is “non collaborative.” While in a clinical setting such result would be recorded as “the child did not pass the test,”¹ when the survey is administered at home, such result calls into question whether it was because of psychomotor retardation or children did not pass the test due to environmental interferences.

The objectives of this study were:

- To find out the household adequacy for the administration of the PRUNAPE survey in a population with 30% or more basic unmet needs.
- To assess the possibility of administering the test in a trailer located near the house when the household conditions are deemed inadequate or when a “non collaborative” result is obtained in a household considered adequate.

POPULATION AND METHODS

After having determined census areas by radius, sampling points were randomly defined and approximately 30 children younger than 6 years old were located by point. Based on the spatial study, population characteristics and data processing, the final sample size was estimated at 1000 children younger than

a. Environmental Health Division, Matanza-Riachuelo River Basin Authority (*Autoridad Cuenca Matanza Riachuelo*, ACUMAR). Health and Environment Department of the National Ministry of Public Health. City of Buenos Aires..

E-mail:
Horacio Lejarraga M.D.: cursotesis07@gmail.com

Conflicto de intereses: None.

Received: 8-3-12
Accepted: 12-18-12

6 years old in each district. Households were visited by the staff responsible of making lists of the households and children younger than 6 years old and those in charge of visiting the selected houses to explain the survey and obtain the consent, with the help of neighborhood leaders and district authorities. All interviewed families accepted the invitation to participate and signed the informed consent. Twenty-seven professionals (pediatricians, psychologists, educational psychologists, general practitioners, a speech therapist, psychomotor specialists, and occupational therapists) were trained on how to administer the test using adapted versions of Hospital Garrahan's PRUNAPE education programs.⁸ The replicability of the PRUNAPE results (intra- and inter-observer) was studied before administering the survey in a blinded fashion to 25 children and showed a 92% and a

86.4% matching rate, respectively; and a Kendall's kappa coefficient \pm standard error of 0.75 ± 0.16 ($p = 0.0001$) and 0.77 ± 0.21 ($p = 0.003$) for intra- and inter-observers, respectively; these results indicate an "adequate match."⁹

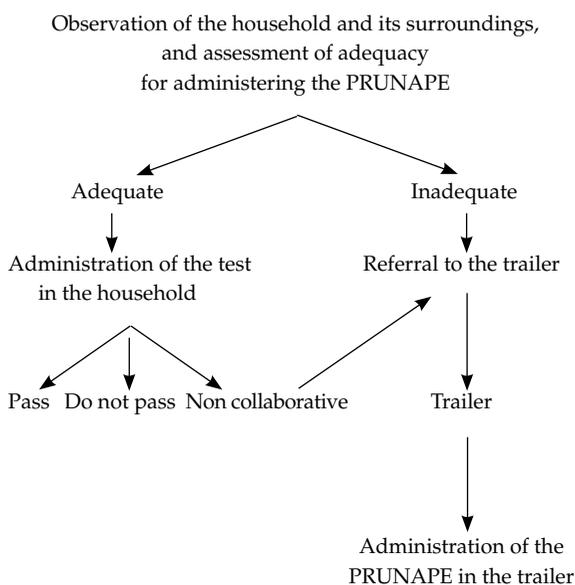
The guidelines indicated in the *Figure* were used to reduce the proportion of children who failed the PRUNAPE survey due to the "non collaborative" result in the population with UBNs.

Two steps were followed to reduce the number of "non collaborative" children: 1) to define the adequacy of household conditions; 2) if household conditions were not acceptable as per the criteria established, the child was referred to the trailer for the administration of the PRUNAPE. If the test was done at the household and the result was "non collaborative," the child was also referred to the trailer for a retest.

Five conditions, shown in *Table 1*, were identified to qualify the household adequacy.

The observer applied the criteria by combining all possible factors for interference with a qualitative judgment and, based on this assessment, followed the steps indicated in the *Figure*. A trailer was located nearby, and inside it there was a closed, private and silent room that was 3 meters wide and 5 meters long, adequate for drawing blood and performing other medical procedures, with a table and two chairs.

FIGURE 1. Guidelines for administering the PRUNAPE together with the ENUDPAD II



RESULTS

Table 2 shows the results of applying assessment guidelines for evaluating the household and referral to the trailer of non collaborative children during the administration of the PRUNAPE at home.

No households were considered inadequate, as a result, no child was initially referred to the trailer. There were 98 children who were "non collaborative" at home and were then referred to the trailer. Out of these 98 (4.5%) referred children, 58 (59%) attended the trailer, and out of these 58, 28 (48%) passed the test. There were significant differences between both districts

TABLE 1. Household conditions for the administration of the PRUNAPE

Item	Condition
Floor	Regular surface.
Space	Adequate for testing motor skills (jumping with both feet, etc.). A courtyard is also acceptable.
Pieces of furniture	A chair and a table (to assess the baby).
Environmental noise	Lack of environmental noise.
Privacy	No excess of people watching the test procedure

in the proportion of non collaborative children and in the number of children who attended the trailer the following day. These results show three relevant findings: 1) no household was considered inadequate for the administration of the PRUNAPE in the neighborhoods with a high population with UBNs; 2) in this context, the proportion of “non collaborative” children was very low (7% and 2%); and 3) out of the non collaborative children, more than half passed the test once it was readministered.

COMMENT

It is encouraging to confirm that the PRUNAPE can be administered at homes in populations with UBNs. In other regions, such as the Ecuadorian Amazonia, households do not meet these conditions, so some of the PRUNAPE’s guidelines were modified for its implementation.¹⁰

The fact that 41.8% of children had a “non collaborative” result at their household and then passed the test administered in the trailer allows us to deduce that the lack of collaboration at home may be due to factors not related to the child’s development. Giving the child a second “chance” of taking the test in the trailer enabled us to “recover” children that would have otherwise been negatively classified. Such differences in results for the same child may occur even if the test is administered in the same context because it is not 100% reproducible, as with any other tests. These guidelines are intended to eliminate disturbing factors, but differences in results continue when the test is administered in the trailer or at home, and these differences may also be found when the test is repeated in the pediatrician’s office, and this is more likely related to the child’s mood.

There is no information to account for the differences found between both districts, but we now know that a different proportion of “non collaboration” can be obtained in different studied areas.

There is scarce scientific data regarding the proportion of “non collaborative” children at household level. In Cardiff, results were “doubtful” for 3.9% of 208 children,¹¹ while Frankenburg found “doubtful” results in 9.8% of 2343 children from a subgroup of items of the Denver test.¹² In studies conducted in children from a low socioeconomic level in rural areas of the United States, the proportion of doubtful cases ranged from 7% to 9%.¹³ The PRUNAPE standard regarding the classification of “non collaborative” children as “not passing the test” results reasonable; for example, Glascoe found that these children had a more retarded psychomotor development than those that did pass the test.¹⁴

For this reason two subgroups of children can be found in the “non collaborative” category: one subgroup consists of children who do not collaborate with the test due to circumstantial reasons, but that do have a normal development; the other subgroup (detected by Glascoe in his study) is composed of children with an actual mild retardation. The guidelines prepared for our survey may be useful to reduce the proportion of non collaborative children due to environmental or circumstantial factors.

This study did not focus on the result of a second test administered at the household, which would have provided valuable information for comparing such results with those obtained at the trailer. No data on the total proportion of children who either passed or not the test are provided because they were not relevant to the core subject of our investigation; but we do know that populations with UBNs may include a very high proportion of children with developmental disorders.¹⁵

The fact that the PRUNAPE can be administered at the household following the algorithm herein used is relevant because it allows to assess children’s development in surveys administered in the field in socially

TABLE 2. Number and percentage of non collaborative children at home and results obtained in the trailer

Districts	Assessed in the household	Non collaborative	Attended the trailer	Passed the PRUNAPE in the trailer
La Matanza (LM)	975	72 (7.4%) *	41 (56.9%) **	22 (54%)
Florencio Varela (FV)	1199	26 (2.2%)*	17 (65.4%) **	6 (35%)
Total	2174	98 (4.5%)	58 (59%)	28 (48%)

* OR= 3.55, for 95% IC: $p < 0.01$ (between FV and LM).

** OR= 4.98, for 95% IC: $p < 0.004$ (between FV and LM).

disadvantaged groups, who are the recipients of the efforts made by the Environmental Health Division.

Acknowledgments

Thanks to the Fundación Garrahan for donating the PRUNAPE materials (Technical Manual, supplies, forms) to Acumar.

To the teams of the Observatory and the Methodology Group of Environmental Health Division of Acumar, who helped with the ENUDPAT II survey: Graciela Algorry, Julian Antman, Graciela Blatt, Daniel Currá, Alberto Derlindati, Paula Das Neves, Diana Di Pietri, Alberto Derlindati, Iván Insúa, Mariana Kesselman, Alicia Masautis, Estela Rugolo, María Teresa Teso, and Alicia Yanicelli. ■

REFERENCES

1. Lejarraga H, Kelmansky FD, Pascucci MC, Salamanco G. Prueba Nacional de Pesquisa. Publicación Fundación Hospital Garrahan. Buenos Aires. 2005.
2. Pascucci MC, Lejarraga H, Kelmansky D, Alvarez M, et al. Validación de la Prueba Nacional de Pesquisa PRUNAPE. *Arch Argent Pediatr* 2002;100(5):374-385.
3. Pascucci MC, Lejarraga H. Detección temprana de Trastornos de Desarrollo en niños menores de seis años Boletín Oficial del Ministerio de Salud. Servicio Nacional de Rehabilitación y Promoción de la Persona con Discapacidad 1998. Boletín N° 13, 3-5.
4. Lejarraga H. La detección oportuna de problemas de desarrollo. La Prueba Nacional de Pesquisa (PRUNAPE). pp 447-450. In: Comité Nacional de Pediatría Ambulatoria. Sociedad Argentina de Pediatría. Manual para la Supervisión de la Salud de Niños, Niñas y Adolescentes 2010. Coordinadores: Boggiano E, Breitman F, Andrade M. SAP – Fundasap. 2010. Pags. 447-50.
5. Lejarraga H, Berardi C, Ortale S, Contreras MM, et al. Crecimiento, desarrollo, integración social y prácticas de crianza en niños que bien con sus madres en prisión. *Arch Argent Pediatr* 2011;109(6):485-91.
6. Dirección de Salud Ambiental. Resultados de la Encuesta de Nutrición, Desarrollo Psicomotor y Análisis Toxicológico en el ámbito de la Cuenca Matanza Riachuelo (CMR). ENUDPAT I, 2010. ACUMAR. Autoridad Cuenca Matanza-Riachuelo. MSP. 2011
7. Dirección de Salud Ambiental. Encuesta de Nutrición, Desarrollo y Análisis Toxicológico en los Municipios de Florencio Varela y La Matanza. ENUDPAT II, 2011-2012. ACUMAR. Autoridad Cuenca Matanza-Riachuelo. MSP. 2012.
8. Servicio de Crecimiento y Desarrollo. 2012. Curso de Capacitación para la Administración de la Prueba Nacional de Pesquisa PRUNAPE. Dirección de Docencia e Investigación. Hospital Garrahan. www.garrahan.ewdu.ar/cursos/prunape
9. Altman DG. Practical statistics for medical research 1991. London, UK: Chapman and Hall.
10. Senplades. DEIN. Primera Encuesta de Desarrollo Infantil Integral. Secretaría nacional de Planificación y Desarrollo y INEC Instituto Nacional de Estadísticas y Censos. Gobierno de la República del Ecuador. Quito. 2011.
11. Bryant GM, Favies KJ, Newcombe RG. Standardisation of the Denver Developmental Screening Test for Cardiff Children. *Dev Med Child Neurol* 1979;(3):353-364.
12. Frankenburg WK, Ker CY, Engelke SA, Shaefer ES, et al. Validation of New Denver Developmental Screening test items: a preliminary study. *J Pediatr* 1988, 112, (4),560-66.
13. Harper DC, Waeker DP. The efficiency of the Denver Developmental Screening test with rural disadvantaged pre-school children. *J Pediatr Psychol* 1983;(8):273-83.
- 14) Glascoe FP. Are overreferrals on developmental screening tests really a problem? *Arch Pediatr Adolesc Med* 2001; 155(1):54-9.
- 15) Lejarraga, H; Menéndez, A; Menzano E, Fattore MJ; et al PRUNAPE: Pesquisa de trastornos de desarrollo en el primer nivel de atención. *Arch Argent Pediatr* 2008; 106(2):119-25.

Early detection of neonatal cholestasis using a stool color card screening

Margarita D. Ramonet, M.D.^a, Silvia Gómez, M.D.^a, Silvia Morise, M.D.^a, Lidia Parga, M.D.^b, Patricia Caglio, M.D.^a, Mirta De Micheli, M.D.^b and Armando Garsd, M.D.^c

- a. Department of Pediatrics.
- b. Neonatology Division.
Department of Mother and Child.
Hospital Nacional Prof. Alejandro Posadas.
- c. Spectrum Pharmaceuticals, Inc.,
157 Technology Dr, Irvine, CA 92618. USA.

Conflict of interest: None.

Financiamiento:
Graphic materials were and are currently financed by Laboratorio Roemmers, which does not have any products related to the treatment of neonatal cholestasis.

E-mail:
Margarita D. Ramonet, M.D.: margarita.ramonet@gmail.com

Received: 6-6-2012
Accepted: 12-21-2012

ABSTRACT

The objective of this study was to identify, by early screening, one-month old infants with acholic or hypocholic stools for the early detection of biliary atresia and other causes of neonatal cholestasis.

The study was essentially exploratory (observational, prospective, statistically underpowered and with no clinical intervention) and used a color card screening of all newborns attending the first month of life checkup from 1999 to 2002 in a public hospital in Argentina.

Of 12 484 newborn infants, 4239 were examined at their first month of life checkup visit with the stool color card. Eighteen infants were identified with hypocholic stools, of whom only four were proven to have cholestasis.

Although no case of biliary atresia was detected, screening by stool color cards proved useful for the detection of other causes of neonatal cholestasis.

Key words: color card screening, neonatal cholestasis, biliary atresia, Matsui's method.

<http://dx.doi.org/10.5546/aap.2013.136>

INTRODUCTION

Reduced biliary flow in the neonatal period causes jaundice, choluria and acholic or hypocholic stools,^{1,2} which are all typical signs of neonatal cholestasis (NC). Biliary atresia is the most common cause of obstructive jaundice during the first three months of life and accounts for 40-50% of all liver transplants in the world.^{3,4} There is a strong consensus in pediatric literature indicating that efforts to detect NC should not be delayed because its prognosis depends on early diagnosis and management.^{2,3}

The objective of this study was to test a method for the identification of one-month old infants with acholic or hypocholic stools by early screening in order to eventually rule out cholestasis and thus try to adapt the methodology developed by Matsui, et al. in Japan in the specific context of a public hospital in Argentina.⁵

POPULATION AND METHODS

The study was conducted between August 1st, 1999 and July 31st, 2002 in the Department of Mother and Child Health of a tertiary care hospital in Greater Buenos Aires. In this period, 12 484 infants were born.

The study was essentially exploratory (observational, prospective, with no statistical power and no experimental clinical intervention). Newborn infants (NBIs) admitted to the hospital received the institution's standard of care based on the patient's clinical condition and the consent provided by one of the parents.

With the authorization of Matsui, et al., a

Spanish version of the eight-color card was used: from normal (dark brown, colors 5 to 8) to abnormal (yellowish, colors 1 to 4) (Figure). Mothers were also given a card with additional explanatory information at the time of discharge. They were asked to compare the color of their infant's stools to the colors indicated in the card on a weekly basis and to assign the latest stools a number in the corresponding box and deliver it back to the pediatrician at the first month of life checkup.

Posters were posted in the Neonatology and Pediatric areas of the hospital as part of the educational program so that mothers would understand the relevance of using the stool color card.

The hospital's Medical Record Committee authorized the use of graphic materials (color cards) in the medical records of NBIs.

FIGURA 1. Spanish version of the color card used in the pilot program of Hospital Nacional Prof. A. Posadas.

	1	Please write the number that resembles the COLOR OF THE STOOLS of your child in the corresponding box a few days prior to the first month of life checkup. Remember to bring this card to that appointment. The color of my child's stool resembles number _____ Date _____ Information Child's first and last names: _____ MR _____ Date of Birth: _____ Hospital where the baby was born: _____ Mother's Name: _____ Date of Checkup: _____ To be completed by the doctor If the mother selected numbers 1-4, please observe if the child has: jaundice (No, Moderate or Severe). _____ _____ Color of stool, confirmed by doctor, number _____ Date _____ If color corresponds to numbers 1-4, please contact immediately: _____
	2	
	3	
	4	
	5	
	6	
	7	
	8	
Department of Pediatric Gastroenterology Hospital Nacional P.A Posadas – Villa Sarmiento, Haedo Province of Buenos Aires Telephone: (011) 4469-9300 Ext. 1511 Fax: (011) 4469-9220		

RESULTS

At the first month of life checkup, 4239 NBIs (33.9%) returned with their color card. Out of them, 18 (0.42%) had jaundice and stools corresponding to colors 1-4. The following procedures were done in this subgroup: medical history, physical exam, lab tests, additional tests, tests for establishing the probable etiology and a hepatobiliary ultrasound with four hours fasting.

All patients with NC detected during the assessment period were treated with fat-soluble vitamins and ursodeoxycholic acid at 20 mg/kg/day while cholestasis persisted.

The subsequent direct observation of stools was normal in 14 patients, with self-limiting jaundice during the assessment period, and normal color was confirmed later by physical exam and lab results. In the 4 remaining patients acholia was confirmed. Lab findings and other additional tests (abdominal ultrasound, eye exam, thoracic spine X-ray, heart exam, liver puncture biopsy) confirmed the following definitive diagnoses: Alagille's syndrome, luetic hepatitis, idiopathic or transient neonatal cholestasis and biliary lithiasis (Table). No case of biliary atresia was identified during the study period.

DISCUSSION

The etiological diagnosis of cholestatic syndrome improves prognosis because it allows to

manage diseases which can be clinically or surgically treated in a timely manner. A hepatoporoenterostomy (Kasai operation) is universally recognized as the primary treatment for biliary atresia and its prognosis depends on age at the time of the surgery and also on the expertise of the surgical team.⁶⁻⁹

In 1995, Matsui, et al. designed in Japan the color card system for identifying biliary atresia. The authors performed a screening using this method in a population of 17 641 NBIs at the first month of life checkup for a period of one year and identified two cases of biliary atresia in three patients with hypocholic or acholic stools. Thanks to the early detection program, it was possible to perform the Kasai operation within 60 days of age.⁵

A similar screening method used in a pilot regional study with a cohort of 119 973 NBIs from 49 Taiwanese hospitals showed a return rate of 65.2% out of a total of 78 184 NBIs. Abnormal stools were found in 94 cases (0.12%). Biliary atresia was confirmed in 29 (30.9%) NBIs, 8 (8.5%) cases were neonatal hepatitis and 3 NBIs had the diagnosis of other chronic, non-surgical, liver diseases. The remaining 54 cases with transient discoloration of stools normalized during the three-month follow-up.¹⁰

World incidence of biliary atresia is 1:8000-1:18 000 live births, so there was an extremely low

TABLE 1. Main findings and definitive diagnosis of four cases that had acholic stools

Case	Gender	Birth weight (kg)	Onset of acholia (days)	Liver enlargement	DB Mg (%)	AST/ALT (IU/L)	AP/GGT (IU/mg%)	Cholesterol (mg%)	Gallbladder (US)	Biopsy/puncture	Diagnosis (Age in days)
1	F	3.23	2	Firm	3.8	207/96	1901/513	299	Small	Poor bile ducts	Alagille's syndrome (42)
2	F	1.39	4	Firm	3	150/80	1660/310	151	Normal	Non-specific hepatitis	Luetic hepatitis (37)
3	M	2.75	4	Firm	12	340/225	771/56	250	Normal	Giant-cell transformation	Idiopathic neonatal cholestasis (39)
4	M	3.55	15	Elastic	2,6	167/97	2377/1466	308	Stones	Canalicular and hepatocytic-cholestasis	Biliary lithiasis (35)

DB: direct bilirubin; AST: aspartate-aminotransferase; ALT: alanine-aminotransferase; AP: alkaline phosphatase; GGT: gamma-glutamyl transpeptidase.

chance of finding cases of biliary atresia in this three-year study. However, the method detected other cholestatic disorders that also benefit from a timely diagnosis and treatment.^{11,12}

It is interesting to observe that the patient with Alagille's syndrome had a 4 year old sister with a prior diagnosis of peripheral pulmonary stenosis and pruritus, and she was diagnosed with the same syndrome during follow-up at our site.

After the end of the study in 2002, and based on this experience, color card screening was implemented as a routine practice in the hospital. Up to this date, the use of this strategy allowed to detect newborn infants with NC; two were diagnosed with biliary atresia at 38 and 42 days of age and underwent an early Kasai operation.

The low compliance observed regarding the rate of return can be attributed to the characteristics of our health care system and, especially, to the studied population. The population seeking care at our hospital is mainly vulnerable, with a 15.8% of unmet basic needs, high rates of incomplete education, adolescent pregnancies, unemployment and precarious housing conditions.¹³ Most mothers who receive obstetric care in their pregnancy and childbirth do not return for the first month of life checkup because they are not continuously linked to the hospital, cannot access transport facilities and seek care in primary care sites near their place of residence.

Future efforts aimed at increasing the level of awareness of health teams and mothers in relation to the significance of a routine early screening are needed. It is necessary to implement intensive health educational programs in the community and to develop guidelines for health care professionals aimed at achieving an effective management and a timely referral of all newborn infants younger than 30 days of age with jaundice and hypocholelic or acholic stools for an effective diagnosis and treatment.

The significance of this study lies in that this non-invasive methodology enables to detect patients with neonatal cholestasis and benefit many of them through an early treatment. For this reason, authors believe that the use of the color card screening should be added to neonatal screening programs.

CONCLUSION

The detection of acholic or hypocholelic stools by the color card screening in newborn infants is feasible, simple and of low cost.

Acknowledgments

Thanks to the pediatricians and neonatologists of the Department of Mother and Child Health of Hospital Nacional Prof. Alejandro Posadas. To Fernando Álvarez, M.D., for reviewing the manuscript; to Jorge Roca for his technical assistance, and to Laboratorio Roemmers for printing the color cards. v

BIBLIOGRAPHY

- Balistreri WF, Grand R, Hoofnagle JH, Suchy FJ, et al. Biliary atresia: current concepts and research directions. *Hepatology* 1996; 23:1682-92.
- Suchy FJ. Approach to the infant with cholestasis. In: Suchy FJ, Sokol RJ, Balistreri W, Eds. *Liver diseases in children*. Philadelphia, 2nd ed: Lippincott, 2001; Pags. 187-94.
- Sokol RJ, Mack C, Narkewicz MR, Karrer FM, et al. Pathogenesis and outcome of biliary atresia: current concepts. *J Pediatr Gastroenterol Nutr* 2003;37:4-21.
- Schreiber RA, Kleinman RE. Biliary atresia. *J Pediatr Gastroenterol Nutr* 2002; 35: S11- S16
- Matsui A, Dodoriki M. Screening for biliary atresia. *Lancet* 1995; 345:1181.
- Kasai M, Suzuki S. A new operation for "non-correctable" biliary atresia: hepatic portoenterostomy. *Shuyutsu* 1959; 13: 733-39.
- Chardot C, Carton M, Spire-Bendelac N, et al. Prognosis of biliary atresia in the era of liver transplantation: french national study from 1986 to 1996. *Hepatology* 1999;30:606-11.
- Hung PY, Chen CC, Chen WJ, Lai HS, et al. Long-term prognosis of patients with biliary atresia: a 25 year summary. *J Pediatr Gastroenterol Nutr* 2006;42:190-95.
- Serinet M, Wildhaber B, Broué P, et al. Impact of Age at Kasai Operation on Its Results in Late Childhood and Adolescence: A Rational Basis for Biliary Atresiam Screening. *Pediatrics* 2009; 123; 1280-86.
- Chen S-M, Chang M-H, Du J-Ch, et al. Screening for biliary atresia by infant stool color card in Taiwan. *Pediatrics* 2006;117: 1141-54.
- Bakshi B, Sutcliffe A, Akindolie M, Vadamalayan B et al. How reliable can paediatric professionals identify pale stool from cholestaic newborn? *Arch Dis Fetal Neonatal*. Ed 2011, Aug 17 [Epub ahead of print].
- Jacquemin E. Dépistage de l'atresie des voies biliaires et couleur des Selles : méthode de l'échelle colorimétrique. *Arch Pediatr* 2007; 14(3):303-5.
- Anuario Estadístico de la República Argentina 2004, *INDEC*, 1^a ed., Buenos Aires; 2004. Pags. 69-72.