

Validation and preliminary results of a survey on decision of treatment in neonatal critical infants administered to neonatologists in Buenos Aires, Argentina

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Introduction

Therapeutic decision in critically ill infants has raised, and continues to raise a profound ethical debate. The birth of premature or disabled infants with neurological damage and with poor future quality of life is an important challenge in decision making by neonatologists.¹

There are numerous publications on the bioethical criteria followed by specialists on the treatment of critically ill infants within the frame of a clinical and a specific sociocultural context.² However, publications on the bioethical behavior of neonatologists in underdeveloped countries are very few.³ In this study we have develo-

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1 VERGAHEN E, SAUER P. *End-of-life decisions in newborns: an approach from the Netherlands*. Pediatrics 2005; 116: 736-739; ID. *The Groningen Protocol. Euthanasia in severely ill newborns*. N England J Med. 2005; 352: 959-962.

2 NUFFIELD COUNCIL ON BIOETHICS. *Critical care decisions in fetal and neonatal medicine: ethical issues*. Plymouth: Latimer Trend&Company; 2006; PELLEGRINO E. *Decisions to withdraw life-sustaining treatment: a moral algorithm*. JAMA 2000; 283: 1065-1067; DE SARASQUETA P. *Mortalidad infantil por malformaciones congénitas y prematuridad en la Argentina: análisis de los criterios de reducibilidad*. Archivos Argentinos de Pediatría 2006; 106 (2): 153-158; COUNCIL ON BIOETHICS. *Human dignity and bioethics: essays commissioned by the President's Council on Bioethics*. Washington D.C.: Government printing office; 2008: 505-512.

3 FAJARDO CA, GONZALEZ S, ZAMBOSCO G ET AL. *End of life, death and dying in neonatal intensive care units in Latin America*. Acta Paediatr 2012; 101 (6): 609-613; SILBERBERG A,

ped an anonymous survey to determine those bioethical decisions by neonatologists in Buenos Aires Capital City and in the province of Buenos Aires (Argentina).

Validation of surveys has become a useful tool to strengthen results obtained with them and is an optimal procedure to analyze this bioethical debate on treatment decision in critically ill infants. The main goal of this paper is to show the process of validation of the survey. Additionally, we show some preliminary correlating data and the behavior of neonatologists on euthanasia, treatment withdraw and future quality of life.

MATERIAL AND METHODS

1. Preparation of the survey

Based on a previous study on neonatologists in the province of Cordoba (Argentina)⁴, we designed a new survey to explore decision making in neonatologists in Buenos Aires Capital City and two other sanitary regions in the province of Buenos Aires.⁵

This new survey introduced new data such as: age, sex, School of Medicine where medical degree was obtained, public or private affiliation, legal context and religious beliefs. The survey had two sections, the first one, which consisted of 23 questions on a multiple-choice format. Six of them required a justification for the choice made. The second part of the survey consisted of two clinical cases each one with 8 questions to be answered with a scoring of 1 to 5 (totally agree to totally disagree) based on a Likert scale.

GALLO E. *Managing end-of-life decisions in critical infants: a survey of neonatologists in Cordoba, Argentina*. Acta paediatr. 2013; 102: e475-e477.

4 SILBERBERG A. *Valoración moral de las decisiones de los neonatólogos de Córdoba (Argentina) en el tratamiento de neonatos prematuros extremos*. Roma: Pontificia Universidad de la Santa Cruz; 2010; SILBERBERG A, GALLO E. *Managing end-of-life decisions in critical infants: a survey of neonatologists in Cordoba, Argentina...*

5 For our Survey, of the numerous publications of the EURONIC study, we have particularly taken in consideration the paper by Samaan et Al.: SAAMAN MC, CUTTINI M, CASOTTO V ET AL. *Doctor's and nurses' attitudes towards neonatal ethical decision making in Ireland*. Arch Dis Child Fetal Neonatal. 2008; 93: F217-F221.

2. Design and Population studied

The survey was distributed among neonatologists from all maternities and private and public reference hospital located at Buenos Aires Capital City and sanitary regions V and VII in the province of Buenos Aires (Argentina). Estimated time to respond the survey was 12 min.

The study covered a population of 8.275.765 inhabitants.⁶ The total number of births in this population is 139.712, of which 11.675 are premature (37 weeks gestation).⁷ See Table 1. Other socio-economical data showed that 58% of the mothers in Buenos Aires Capital City and 78% in Buenos Aires province completed their secondary studies without, however reaching tertiary or university degrees. Six percent of homes in Buenos Aires Capital City and 26% in Buenos Aires province were under the line of poverty (families with unmet basic needs).

There are no official data on the number of neonatologists attending the population studied. However, the National Ministry of Health reports a total number of 1.500 specialists in Argentina.⁸ From our request to the 36 Clinical Units analyzed in this study a number of 580 specialists that met inclusion criteria has been established. All these neonatologists were invited to participate in the present study.

6 INDEC. *Censo Nacional de Población 2010*. (consulted on September 15, 2014 in: http://www.censo2010.indec.gov.ar/CuadrosDefinitivos/P1-P_Buenos_Aires.pdf); MINISTERIO DE SALUD. *Población, grupo etario y sexo. Provincia de Buenos Aires. Año 2010*. (consulted on September 15, 2014 in [http://www.ms.gba.gov.ar/estadisticas/Estadísticas demográficas](http://www.ms.gba.gov.ar/estadisticas/Estadísticas%20demográficas)).

7 SECRETARÍA DE POLÍTICAS, REGULACIÓN E INSTITUTOS. DIRECCIÓN DE ESTADÍSTICAS E INFORMACIÓN DE SALUD. *Sistema estadístico de salud*. (consulted on September 19, 2014 in: <http://www.deis.gov.ar/publicaciones/archivos/Boletin142.pdf>). There are no accounts on the number of premature newborns in sanitary regions V and VII in the province of Buenos Aires. However, since the total number of births for the whole province of Buenos Aires is known and regions V and VII represent a 47.8 %, figures for premature births may be estimated from this percentage. See Table 1.

8 Data provided by the National Ministry of Health (April 23, 2014).

3. Inclusion and Exclusion Criteria

All neonatologists working in sanitary centers and hospitals with the following profiles were included in the present study: 1. Specialists in neonatology with a official degree; 2. Physicians doing their clinical practice in hospitals with the following characteristics: *a.* Complexity level IIIA and IIIB, in those cases where the annual number of births was ≥ 1.500 ; *b.* Complexity level II, in those cases where the annual number of births was ≥ 2.000 ; *c.* Clinical centers of derivation for neonatal diagnostics and treatment of surgical pathology and high level of complexity; 3. Neonatologists who treat newborns in geographic areas of Buenos Aires Capital City and sanitary regions V and VII in Buenos Aires province (Argentina).

Thirty six hospitals met inclusion criteria. In Buenos Aires Capital City there were 20 hospitals, 10 public and 10 private. In sanitary regions V and VII of Buenos Aires province there were 16 hospitals of which 14 were public and 2 were private.

Medical doctors which worked as neonatologists with, however, no official degree of specialization were excluded of this study. Similarly, physicians working only on ambulatory care and no direct contact with critically ill infants at the moment of birth were excluded as well.

4. Data Collection Method

Once the surveys were completed data was analyzed following descriptive statistics. Parametric data was presented in percentages and shown in tables to facilitate analysis.

The first part of the survey was transformed to a Likert scale based in the 5 options of the multiple-choice. In those six questions where justification was requested, answers were grouped according to frequency and converted to a Likert scale as well. In this way, both the first and second parts of the survey were analyzed following the Likert scale.

Results obtained were introduced into an Excel page and a dynamic table was used for analysis following several statistic procedures.

5. *Research Fields*

The survey explores the influence of vitality and future quality of life in critically ill infants on decision making by neonatologists. To analyze both items 5 research fields were considered.

The first field corresponded to euthanasia. This was studied considering a neonate with a good neurological perspective and, however, a bad life prognosis (Case 1) and, conversely, a child with a bad neurological perspective and a good life prognosis (Case 2).

The second field corresponded to the decision of whether or not to initiate a treatment, either the decision of initiating a futile treatment (without objective benefits), the decision of not providing life sustaining treatment (LST) or cardiopulmonary resuscitation (CPR) in case of bad neurological perspective and good life prognosis.

The third field corresponded to treatment suspension. The decision to stop a futile treatment in case of a clinical worsening of the disease and a bad life prognosis was considered. Also the decision to stop administration drugs of LST and mechanical ventilation (MV) in a case with good neurological perspective and a bad life prognosis was requested. Finally, we explored the decision to withdraw nutrition, hydration and analgesia both in neonates with good neurological perspective and bad life prognosis and in case of a bad neurological perspective and a good life prognosis.

The fourth research field explored the influence of life quality in neonates with malformations in the choice treatment. The last field corresponded to elements influencing neonatologists behavior such as legal frame, transcendence perspective of life, religious beliefs and limited economical resources.

6. *Survey Validation*

For validation of the survey a pilot study was divided in two steps. Step one corresponded to evaluation of the survey by 20 pediatricians of private and public hospitals. These physicians considered if the timing to complete the survey was reasonable and within the 12 minutes planed. Also they had to consider the clarity and self-explai-

ning characteristics of the survey. The results of this step were positive for all these parameters.

The second step of the pilot study verified whether or not the survey specifically explored the research fields previously described. That is if the survey reached significant levels of reliability by means of internal consistency.

To evaluate internal consistency we applied Cronbach alfa, with values between 0 (no consistency) and 1 (perfect consistency). Cronbach alfa was applied to 138 surveys. Internal consistency was also analyzed through factor analysis (*varimax rotation*) with extraction of *eigenvalues* and the corresponding variance explained for each factor.

Additionally, a correlation between factors examined was made with the non parametric test of Spearman. In this way, euthanasia research field was correlated in both clinical cases and with the decision to deny CPR or to withdraw LST and nutrition.

Research field corresponding to treatment initiation was correlated with decision to apply futile treatment and to deny LST and CPR.

Research field corresponding to treatment withdraw was correlated to variables such as decision to apply futile treatment and with no additional therapeutic procedures. Pharmacological withdraw of LST was correlated to withdrawal of MV. Also, decision to apply minimal care was correlated to withdrawal of nutrition and hydration in both cases presented and also with analgesia administration.

Research field corresponding to influence of life quality was correlated with seven congenital malformations.

Finally, for the field corresponding to other elements influencing treatment choice, economical burden was correlated to limited resources, gestation age of the neonate with minimal weigh and ethic primacy over legal aspects with practice of euthanasia.

RESULTS

Results of the first step of validation of the survey showed that it may be completed in 12 minutes or less. Cronbach alfa was 0.94 in all 138 surveys analyzed (standardized alfa coefficient: 0.93; the

worst alfa coefficient when a question was eliminated was 0.93). Factorial analysis allowed extraction of two factors (roots) with explained variances, 48.9% and 33.4% that corresponded, respectively, to fields of euthanasia and treatment withdrawal.

Non parametric test of Spearman showed significant levels of correlation for decisions referring to euthanasia (Table 2), treatment withdrawal (Table 3) and quality of life (Table 4). Instead, there was no good correlation for decisions on treatment initiation and existence of other elements (economical burden, limited resources, gestation age of the neonate and minimal weigh at birth) that influenced decision making.

Preliminary results show that only 2% of neonatologists indicated euthanasia in both cases presented. Also in both cases, between 70 and 75% of neonatologists rejected withdrawal of LST. Less than 6% of physicians indicated nutrition withdrawal in both cases.

Preliminary results of the survey also showed that 81% of neonatologists considered that economical burden of treatments have a low influence on decision making with critically ill infants, even when resources are limited, as often occurs in Argentina. Eighty seven percent of neonatologists considered that life has a transcendence meaning and therefore there is a meaning that goes beyond our sensitive world.

DISCUSSION

Validation of survey applied to a wide number of specialists constitutes a valuable tool to participate in the ethical debate on decision making on critically ill infants. There are a few examples within the EURONIC study on decision making by neonatologists in several European countries.⁹ Our present study analyzed a significant population of neonatologists within Argentina in an area that corresponds

⁹ CUTTINI M, KAMINSKI M, SARACCI R ET AL. *The EURONIC Project: a European concerted action on information to parents and ethical decision-making in neonatal intensive care*. Paediatric and Perinatal Epidemiology. 1997; 11: 461-474; CUTTINI, NADAI, KAMINSKI. *End-of-life decisions in neonatal intensive care...*

to 20.6% of argentine inhabitants and 19% of newborns, both premature and full-term births.

Due to the wide extension of Argentina we decided to choose three representative areas with the higher number of births in the country in order to make the study feasible. Specifically, we studied the behavior of a group of neonatologists in Buenos Aires Capital City and two other sanitary regions of Buenos Aires province. However, it should be mentioned that we excluded sanitary region VI in Buenos Aires province, which is also very extensive and may be analyzed in the future.

The two steps of validation of the survey showed positive results. Specifically, Cronbach alfa reached a general score of 0.94, expressing an excellent internal consistency.

The non parametric Spearman test showed a good correlation in three of the five research fields analyzed, euthanasia, treatment withdrawal and quality of life in those neonates with different disabilities. These three fields allowed the consideration of the importance given by neonatologists to viability and future quality of life in neonates.

Our preliminary results show that a wide majority of specialists rejected euthanasia even in case of children with a low quality of life and bad life prognosis. With respect to treatment suspension, physicians show reluctance to withdraw a treatment that may lead to death of the neonate even if death is not intentionally pursued.

As has been reported in the EURONIC study, social and cultural factors were the most important elements in predicting attitudes and practices by neonatologists.¹⁰ With the collection of the complete data, covering all neonatologists in the geographical areas chosen for our study, we expect to establish the relevance of these social and cultural characteristics, since they could explain the rejection to euthanasia by argentine neonatologists. Also, a more complete study

10 SAAMAN, CUTTINI, CASOTTO ET AL. *Doctor's and nurses' attitudes...*; REBAGLIATO M, CUTTINI M, BROGGIN L ET AL. *Neonatal end of life decisions making: Physicians' attitudes and relationship with self-reported practices in 10 European countries*. JAMA 2000; 284: 2451-2459.

will help to establish a concordance of our present preliminary data with data obtained from the larger population of neonatologists.

In coincidence with Saaman et Al.,¹¹ in our study sanitary resources are not considered as major factors in decision making by neonatologists. However, while there is no shortage of those resources in European countries, in Argentina, and particularly in the geographical regions chosen for our study, this may be a rather common situation. It appears that decisions by the neonatologists tested in this study are instead conditioned by a transcendence sense of meaning that they give to life.

Conclusion

Our survey is viable since it was always completed within the stipulated time and, also because all physicians tested expressed it was clear and self-explaining. Internal consistency was considered excellent, established according to Cronbach alfa coefficient.

Correlations chosen to explain the validity of the survey showed a significant association between euthanasia and LST withdrawal and between quality of life and resuscitation, also with the existence of congenital malformations.

Finally, our preliminary results show that argentine neonatologists reject euthanasia, give a low priority to the shortage of economical resources and give life a transcendence meaning.

11 CUTTINI, NADAI, KAMINSKI. *End-of-life decisions in neonatal intensive care...*

Table 1.
Population and number of births including premature
in the geographical regions chosen for the study

	Argentina	Buenos Aires Capital City	Sanitary Region V Buenos Aires	Sanitary Region VII Buenos Aires	Population
Inhabitants	40.117.096	2.890.151	3.131.892	2.253.722	8.275.765
Births	738.318	43.733	55.302	40.677	139.712
Premature	61.393	3.487	8.188		11.675
Between					
32-36 weeks	No data	2.974	7.182		10.156
Between					
28-31 weeks	No data	322	653		975
≤ 27 weeks	No data	191	352		543

Table 2.
Non Parametric Spearman test on euthanasia

	r	p
Euthanasia, Case 1 vs Euthanasia, Case 2	0.75	0.0001
Euthanasia, Case 1 vs deny LST, Case 2	0.59	0.0001
Euthanasia, Case 1 vs LST even if needed, Case 2	0.60	0.0001
Euthanasia, Case 2 vs No CPR even if needed, Case 2	0.71	0.0001
Euthanasia, Case 1 vs Withdrawal of LST drugs, Case 2	0.60	0.0001
Euthanasia, Case 2 vs Withdrawal of LST drugs, Case 2	0.72	0.0001
Euthanasia, Case 1 vs Nutrition withdrawal, Case 2	0.62	0.0001
Euthanasia, Case 2 vs Nutrition withdrawal, Case 2	0.76	0.0001

Table 3.
Non Parametric Spearman test on treatment withdrawal

	r	p
Administration of futile treatment vs No treatments added even after worsening of the disease , Case 1	Non significant	
Withdrawal of LST drugs, Case 1 vs Withdrawal MV, Case 1	0.79	0.0001
Withdrawal of LST drugs, Case 2 vs Withdrawal MV, Case 2	0.86	0.0001
Minimal care Administration vs R Withdrawal nutrition and hydration, Case 1	0.18	0.036
Minimal care Administration vs Withdrawal nutrition and hydration, Case 2	0.17	0.04
Minimal care Administration vs Analgesia administration, Case 1	Non significant	

Table 4.
Non Parametric Spearman test on quality of life

	r	p
Quality of life vs Burden that a disabled represents for the family	0.49	0.0001
Quality of life vs Resuscitation anencephalic child	Non significant	
Quality of life vs Resuscitation in renal agenesis	0.26	0.002
Quality of life vs Resuscitation in trisomy 13	Non significant	
Quality of life vs Resuscitation in trisomy 18	0.18	0.03
Quality of life vs Resuscitation in trisomy 21	Non significant	
Quality of life vs Resuscitation in pulmonary malformation	0.23	0.006

Parole chiave: processo decisionale in medicina, eutanasia, neonatologia.

Key words: medical decision making, euthanasia, neonatology.

RIASSUNTO

Le decisioni terapeutiche in bambini gravemente malati con danni neurologici e previsione di scarsa qualità di vita futura hanno sollevato una profonda discussione etica.

La validazione delle survey è uno strumento utile per rafforzare i risultati ottenuti e sembra essere una procedura ottimale per analizzare questo dibattito, in particolare se le survey si concentrano sul comportamento clinico dei neonatologi nei paesi non sviluppati.

L'obiettivo di questo lavoro è duplice: in primo luogo, si intende illustrare il processo di validazione di una survey da somministrare a 580 neonatologi argentini, che rappresentano un terzo del numero totale di specialisti in Argentina. In secondo luogo, si intende presentare alcuni risultati preliminari che mettono in correlazione l'eutanasia, la sospensione dei trattamenti e la qualità della vita attesa.

Per la validazione è stata condotta uno studio pilota su neonatologi, divisa in due fasi. La prima fase è corrisposta alla valutazione della survey da parte di 20 pediatri. Essi hanno valutato se i tempi per completare la survey fossero ragionevoli. I pediatri hanno dovuto, inoltre, valutare chiarezza e comprensibilità della survey. I risultati di questa fase sono stati positivi. Nella seconda fase dello studio pilota si è verificato se la survey esaminasse in modo specifico i campi di ricerca precedentemente descritti, cioè se essa avesse raggiunto livelli significativi di affidabilità sulla base della consistenza interna.

Il coefficiente alfa di Cronbach è risultato di 0,94 in tutte le 138 survey analizzate. L'analisi fattoriale ha permesso l'estrazione di due fattori (radici) con una varianza rilevata 48,9% e 33,4%, che corrispondevano rispettivamente ai campi relativi all'eutanasia e alla sospensione dei trattamenti. Il test non parametrico di Spearman ha mostrato livelli significativi di correlazione per le decisioni che si riferiscono all'eutanasia, alla sospensione del trattamento e alla qualità della vita.

I risultati preliminari della survey hanno evidenziato inoltre che il 98% dei neonatologi rifiutavano l'eutanasia e più del 70% non era d'accordo con la sospensione dei trattamenti di sostegno vitale sia nei neonati con buone prospettive neurologiche e una prognosi infausta, sia in quelli con una prospettiva neurologica negativa e prognosi di buona sopravvivenza.

La nostra survey è praticabile e ha un alto livello di consistenza interna secondo il coefficiente alfa di Cronbach. Le correlazioni hanno mostrato una significativa associazione tra l'eutanasia e la sospensione dei tratta-

menti di sostegno vitale e tra la qualità di vita e la rianimazione anche in presenza di malformazioni congenite. Infine, i nostri risultati preliminari mostrano che i neonatologi argentini rifiutano l'eutanasia.

SUMMARY

Validation and preliminary results of a survey on decision of treatment in neonatal critical infants administered to neonatologists in Buenos Aires, Argentina.

Therapeutic decision in critically ill infants with neurological damage and poor future quality of life has raised a profound ethical discussion.

Validation of surveys is a useful tool to strengthen the results obtained and appears to be an optimal procedure to analyze this debate, particularly if they focus on the clinical behavior in neonatologists in undeveloped countries.

The goals of this paper have been twofold. Firstly, to show the process of validation of a survey to be administered to 580 argentine neonatologists which represent one third of the total number of specialists in Argentina. Secondly, to show some preliminary results correlating euthanasia, treatment withdrawal and probable quality of life.

For validation a pilot study in neonatologists was divided in two steps. Step one corresponded to evaluation of the survey by 20 pediatricians. These physicians considered if the timing to complete the survey was reasonable. Also they had to consider the clarity and self-explaining characteristics of the survey. The results of this step were positive for all these parameters. The second step of the pilot study verified whether or not the survey specifically explored the research fields previously described. That is if the survey reached significant levels of reliability by means of internal consistency.

Cronbach alfa was 0.94 in all 138 surveys analyzed. Factorial analysis allowed extraction of two factors (roots) with explained variances, 48.9% and 33.4% that corresponded, respectively, to fields of euthanasia and treatment withdrawal. The non parametric test of Spearman showed significant levels of correlation for decisions referring to a: euthanasia, treatment withdrawal and quality of life.

Preliminary results of the survey also showed that 98% of Neonatologists rejected euthanasia and more than 70% disagree with suspension of life support treatment both in the neonate with good neurological perspective and bad life prognosis and when there was a bad neurological perspective and a good life prognosis.

Our survey is viable and has a high internal consistency established according to Cronbach alfa coefficient. Correlations showed a significant as-

sociation between euthanasia and life support treatment withdrawal and between quality of life and resuscitation also with the existence of congenital malformations. Finally, our preliminary results show that argentine neonatologists reject euthanasia.