DEPRESSION AMONG PARENTS OF CHILDREN DIAGNOSED WITH RARE DISEASES

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Abstract

The **aim** of the study is to identify the level of depression among parents of children diagnosed with a rare disease. **Materials and methods**: a number of 29 parents were included in the research. Parents had to complete the items of Beck Depression Inventory. A number of twelve questions were address and socio-demographic data were also registered. Data were processed using *IBM SPSS Statistics*, version 23. **Results and discussion**: parents have a minimal depression (M=8.27 ± 11.70). Marital status and child's age were identified to relate with the level of depression. Parents who have children aged between 6 and 12 years old score higher on BDI compared to the groups 0-6 years old or of those older than 12. Parents who registered higher scores on depression were more frequently involved in psychological or psychiatric consultations. Parents who believe the mother is more involved have higher scores on BDI. **Conclusion**: The support for parents having a child diagnosed with a rare disease must be offered by a multidisciplinary team and the psychological care of both children and parents must be considered.

Keywords: *rare diseases, psychological support, parents, depression, phenylketonuria, cystic fibrosis and hypothyroidism*.

1.INTRODUCTION

Cystic fibrosis, phenylketonuria and congenital hypothyroidism are rare diseases, their incidence being less than 1: 2500 cases. Cystic fibrosis is the most common autosomal recessive disease in the Caucasian population that affects all organs except the brain. Phenylketonuria and congenital hypothyroidism have as a common element the mental retardation that may occur in the absence of an early diagnosis and the delay of treatment initiation.

Living with a chronic disease is difficult for both the patient and his family. Experiences such as anxiety, depression and stress are evident throughout the life of the patient with chronic illness, but the degree of this distress and psychosocial problems are influenced by the severity of health condition and by the support of family, friends and community.

Children with a rare disease present several psychological symptoms and social difficulties, usually identified by mothers who are usually the ones responsible (IORGA et al, 2017). Educational, social and family aspect must not be ignored, since they are part of the daily life and assure a high quality of life (GÖTZ&GÖTZ, 2000).

After the diagnostic of a rare disease, the most common reactions are: shock, denial, sadness or anger. The psychological support for both child and family members must be provided, along with the multidisciplinary medical care. A rare disease is not affecting only the patient but also the caregiver who must provide attention, helping the child in following the medical restriction (food, drinks, physical activity etc.) and be prepared to interfere whenever necessary in order to assist the child's medical needs. Maternal adjustment was proved to be correlated with lower rates of perceived stress in mothers and family functioning provided high levels of supportiveness. Also, regarding the coping mechanism, maternal adjustment was related to less use of palliative coping style (THOMPSON et al, 1992). The socio-economic aspect and the marital status proved to influence the level of stress. For example, single mothers seem to experience more stress-related symptoms in comparison to those who are married (MACPHERSON et al, 1998).

The care of a pediatric patient diagnosed with a rare disease must be done in a multi-disciplinary team, including a psychologist and a psychiatrist. The demanding period of adolescence is complicated both for the patient and the caregiver and that is why a close relationship with psychologists must be taken into consideration.

The aim of the present study is to identify the presence of depression among parents of children with rare diseases and to identify the factors related to it.

2.MATERIALS AND METHODS

A number of 35 parents were included in the research. The subjects are parents of children diagnosed with a rare disease: phenylketonuria, cystic fibrosis and hypothyroidism. All parents took part in the researcher during the hospitalization of their children, between February and May 2016. The papers were delivered to the parents by the pediatrician or psychologist. The questionnaires were returned on the same day. The documents which were not fully completed were excluded from the research. In the end, a number of 29 questionnaires were considered for the study.

Parents must complete three documents. Before filling in, mothers and fathers were informed about the goal of the study, were assured about confidentiality of data and they were told that they could withdraw from the study without any consequences. The study was approved by the Ethical Committee of "Sfanta Maria" Hospital for Children in Iasi, Romania.

First, they had to answer to the items of *Beck Depression Inventory* (*BDI* - BECK et al., 1961). The psychological instrument contains a number of 21 items. The subjects must choose the answer evaluated on a 4th-grade scale of intensity (from 0 to 3). The total score (from 0 to 63) represents the sum of subscores for each item and reveals the level of depression. The 21 symptoms refer to: mood, pessimism, sense of failure, lack of satisfaction, feelings of guilt, sense of punishment, self-hate, self-accusations, self-punitive wishes, crying spells, irritability, social withdrawal, indecisiveness, body image, work inhibition, sleep disturbance, fatigability, loss of appetite, weight loss, somatic preoccupations and loss of libido. Cronbach's alpha score for this inventory is 0.949 and proves a strong reliability.

The subjects had also to answer nine questions and to provide socio-demographic and medical data like age, gender (male/female), and environment (urban/rural), level of education (primary/secondary/high school/university), marital status (married/single), child's age or type of disease.

The data analysis was conducted using IBM SPSS Statistics (IBM, Tokyo, Japan), version 23. Means and standard deviations were used for descriptive analysis and for comparative and correlational analysis was performed either for independent samples t tests or One-way ANOVAs, depending on the number of available responses for each item (LABAR, 2008).

3.RESULTS

Socio-demographic and medical data: The sample consisted of 29 parents (23 mothers and 6 fathers). Their children are treated for one of the three following chronic disease: phenylketonuria (31% of children), hypothyroidism (31%), and cystic fibrosis (38%). 51.7% of them are living in rural areas. Data regarding the parents' level of education revealed that 3.4% of the parents have primary education, 26.9% secondary education, 53.8% attended high school, and 15.4% attended college. The age of the parents is ranged from 18 to 55 (M = 33.48 ± 8.44) and of the children with an M = 7.10 ± 6.32, from 1 to 22 years old. Regarding marital status, 85.2% are married and are 14.8% single parents. The register child age at the time of diagnostic ranged between 1 month and 11 years.

Depression. Parents with children suffering from a rare disease have a minimal level of depression (M=8.27 ± 11.70**)**. The total means and standard deviations and for both male and female parents are presented in Table 1.

Table 1. Means and standard deviations for BDI and items

	Items	**M ± St. dev. total**	**M ± St. dev. for mothers**	**M ± St. dev. for fathers**
1	mood	.41±.77	.47±.84	.16±.40
2	pessimism	.31±.71	.39±.78	.00±.00
3	sense of failure	.31±.84	.39±.94	.00±.00
4	lack of satisfaction	.55±.94	.65±1.02	.16±.40
5	guilty feelings	.27±.64	.34±.71	.00±.00
6	sense of punishment	.41±.98	.52±1.08	.00±.00
7	self-hate	.37±.77	.34±.64	.50±1.22
8	self-accusations	.34±.66	.39±±.72	.16±.40
9	self-punitive wishes	.10±.30	.13±.34	.00±.00
10	crying spells	.51±.87	.60±.94	.16±.40
11	irritability	.44±.82	.56±.89	.00±.00
12	social withdrawal	.27±.59	.30±.63	.16±.40
13	indecisiveness	.65±1.11	.78±1.20	.16±.40
14	body image	.27±.75	.34±.83	.00±.00
15	work inhibition	.55±.94	.69±1.01	.00±.00
16	sleep disturbance	.65±.97	.69±1.06	.50±.54
17	fatigability	.62±.82	.69±.87	.33±.51
18	loss of appetite	.24±.51	.30±.55	.00±.00
19	weight loss	.06±.25	.08±.28	.00±.00
20	somatic preoccupations	.41±.68	.47±.73	.16±.40
21	loss of libido	.44±.98	.52±1.08	.16±.40
	Total BDI	8.27 ± 11.70	9.73±12.68	2.66±3.38

The chi square tests showed an association between item 20 of the BDI and the type of disease: $\chi 2(4) = 12.576$, p=.014. We identified one negative and one positive adjusted residuals: in the case of cystic fibrosis, there were less participants in the category of aparents who are not worried about their health than would be expected by chance; also, there were more participants in the category of parents who are worried about physical problems than would be expected by chance.

There is also an association between item 2 of the BDI and marital status: $\chi 2(3) = 11.530$, p=.009. We identified three negative and three positive adjusted residuals: in the case of married individuals, there were less participants in the categories of people who are discouraged and who feel there is no hope than would be expected by chance; also, there were more participants in the category of people who are not discouraged than would be expected by chance. In the case of single individuals, there were less participants in the category of people who are not discouraged than would be expected by chance; also, there were more participants in the categories of people who feel discouraged and who feel there is no hope than would be expected by chance.

Chi square tests also show an association between item 3 of the BDI and marital status: $\chi 2(3) = 8.568$, p=.036. We identified two negative and two positive adjusted residuals: in the case of married individuals, there were less participants in the category of people who feel they fail more than others and more in the category of people who feel they didn`t fail than would be expected by chance. In the case of single individuals, there were more participants in the category of people who feel they fail more than others and less in the category of people who feel they didn`t fail than would be expected by chance.

Chi square tests show an association between item 18 of the BDI and marital status: $\chi 2(2) = 6.322$, p=.042. We identified one negative and one positive adjusted residual: in the case of married individuals, there were less participants in the category of people who have no appetite than would be expected by chance. In the case of single individuals, there were more participants in the category of people who have no appetite than would be expected by chance.

Items. A number of 12 questions were formulated in order to identify various aspects related to the consequences of the diagnostic. The questions and the distribution of answers are presented in Table 2.

Table 2. The items and the recorded results

	Items	**(%)**
1.	Time spent with the child diagnosed with a rare disease	6.9% - less than other children 44.8% - same as other children 34.5% - more than other children 6.9% - can`t tell
2.	I discussed the diagnostic with my family	96.6% - yes
3.	My relationship with my partner has changed	3.4% - yes 93.1% - no
4.	My family ….	58.6% - encouraged me 34.5% - helped me 3.4% - no modifications 3.4% did not answer
5.	I turn to ….	79.3% - partner 10.3% - friends 3.4% - doctor 3.4% - other parents who have children diagnosed with the same disease
6.	My relationship with my child has changed	3.4% - yes 72.4% - no
7.	Screening for the disease	48.3% - yes 48.3% - no
8.	I understand the difficulties of the disease	37.9% - yes 44.8% - no 6.9% - can`t tell
9.	I consulted another doctor	48.3% - yes 44.8% - no
10.	I went to a psychologist	44.8% - yes 51.7% - no
11.	I went to a psychiatrist	27.6% - yes 69% - no
12.	The mother is more involved	48.3% - yes 44.8% - no

We performed either independent samples t tests or the one-way analysis of variance (ANOVA), depending on the number of available responses on each item. The analysis using independent samples t tests showed no difference in depression among parents according to child's sex(t (27) = 1.028, p =.313), living environment (t (27) = .917, p =.369), or marital status (t (25) = -1.170, p =.253). Also,one-way ANOVA analyses showed no difference in depression among parents considering the child's disease (F (2.26) = 2.254, p = .125).

We identified that child's age influences parent's depression (F (2.25) = 32.423, p = .000). Parents who have children aged between 6 and 12 years old score higher on BDI compared to the groups 0-6 years old and over 12 years old.

Time lapsed from diagnosis influences depression: (F (2.25) = 15.567, p = .000). In the case of children diagnosed more than 5 years ago, parents score higher on BDI than in the case of children diagnosed less than 2 years ago or between 3 and 5 years ago.

Time spent with the child does not influence depression. No statistical significant differences between participants who spend less time / more time / same time / can't appreciate with the child diagnosed with a rare disease compared to their other children concerning depression. (F (3.23) = 2.946, p = .054).

All subjects declared that they discussed the child's diagnosis with family members and that their families encouraged or helped them after the medical diagnostic confirmation. Only one participant considered that the relationship with the partner changed. A number of 23 parents (79.3%) reported they turned to their partners and only one parent considered that the relationship with his child has changed after they were told about the disease. Mothers and fathers who believe the mother is more involved have higher scores on BDI:t (25) = 2.612, p =.019).

Concerning screening for the disease, the independent samples t test showed no statistical differences in depression between parents who advised family members to do a screening and those who did not give such advice (t (26) =.680, p =.503).

The results of one-way analysis of variance showed that whether the parents understand the difficulties of the disease does not influence depression (F (2.23) = 3.273, p = .056). More specifically, there were no statistically significant differences between participants who understand / do not understand / can't appreciate the difficulties of the disease concerning depression.

There was no difference in depression between parents who consulted another doctor and those who did not (t (25) = 1.336, p =.193). But we identified that parents who went to a psychologist have higher scores on BDI compared to those who did not(t (26) = 3.553, p =.004) and also for those having psychiatric consultations since the diagnostic of their child (26) = 3.549, p =.036).

4. DISCUSSION

This paper aimed at assessing the level of depression experienced by mother and fathers of children with rare diseases (phenylketonuria, cystic fibrosis and hypothyroidism).

The results of this research identified that 44% of parents were provided with a psychological care and a psychologist and more than one third had an appointment with a psychiatrist. Also, we showed that almost half of parents looked for a second medical opinion regarding the disease of their children. The level of minimal depression, obtained by analyzing this sample, is explained by these supporting interventions. We appreciate that the minimal score for depression in the case of parents of children with rare diseases is due to the psychological, psychiatric and medical support provided by the medical team for both parent and child. Our results are in line with the scientific literature. Many studies revealed that mother of children with a rare disease did not experience more psychological problems in comparison to those from other groups (FOSTER et al., 1998).

Regarding the impact of child's disease on the relationship between mother and father, our study revealed no major influence. Similar results were obtained by proving that no important impact was identified on marital satisfaction or depression of caregivers of children with rare diseases (QUITTNER et al., 1998)

Some other studies focusing on the quality of life of children and parents proved that family

therapy decreased the level of stress among parents with children with rare diseases (DALLVE et al, 2006) and social support predicts less parental emotional impact (HAYES & SAVAGE, 2008).The parent's level of stress influences the pediatric patient's quality of life (WONG & HERIOT, 2008). Some researchers identified that the mother is more psychologically vulnerable during the first few months of diagnostic, but the depression rate decreases over some time (GLASSCOE et al, 2007).

This study identified that the time lapsed from diagnosis influences depression: higher scores being identified in parents with more than 5 years of treatment for the disease of their children. This result is somehow contradictory with the data presented by the literature. One reason could be related to the fact that some of the considered rare diseases are diagnosed since birth (N = 24, 82.8%) and children are more demanding before and during the adolescence. As our result proved, that parents who have children aged between 6 and 12 years old score higher on BDI compared to the groups 0-6 years old and over 12 years old, pointing that psychological assistance during this critical period of life is important for both patient and caregiver.

5. CONCLUSION

It is very important to provide psychological support for parents having a child diagnosed with a rare disease. Since a rare disease is affecting child but also family members, both family centered therapy and client centered therapy must be provided. Especially during the preadolescence period, the patient must be carefully taken care of in order to diminish depression and distress and also to empower the caregivers to face these difficult psychological situations.

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