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The relationship between neuroticism, coping styles and emotions in women with Mayer-Rokitansky-Küster-Hauser syndrome: A moderated mediation analysis¹

Abstract Objectives: Study participants are 46 women (age 23.48 ± 4.88 years) with Mayer-Rokitansky-Küster-Hauser (M-R-K-H) syndrome. Occurrence of the M-R-K-H syndrome is one in 4000-5000 female children. It was investigated (a) whether coping styles mediate the effect of neuroticism (N) on positive (PA) and negative (NA) affect, and (b) whether this mediation is moderated by the level of N as well as (c) whether this moderated mediation is moderated by length of awareness of illness. Methods: Neuroticism, coping style as well as positive and negative emotions were assessed using the Polish version of NOE-FFI, CISS and Scale of Emotional State. Results: Emotion coping style fully (for PA) or partly (for NA) mediated the relationship between neuroticism and emotions and these mediations were moderated by the level of N. Additionally, direct effect of neuroticism on NA was moderated by the time for which a patient has been aware of the disease. Conclusions: The level of neuroticism and the length of illness are important factors for the psychological functioning of women with M-R-K-H Syndrome. In women with medium and high neuroticism, (positive) effect of N on NA was indirect and also direct – but only in those who have been aware of the diagnosis for 6 years or more. Relations with positive emotions were different: the indirect effect (negative) was observed only in women with a moderate and low neuroticism.

Key words: Mayer-Rokitansky-Küster-Hauser Syndrome, neuroticism, coping style, affect, mediation, moderated mediation

Introduction

Mayer-Rokitansky-Küster-Hauser syndrome named M-R-K-H after the first letters of the names of researchers who described it, is a congenital absence of uterus and vagina, or their serious malformation (Ulrich et al., 2004). In most cases ovaries are developed normally and their usual functions are maintained. Thus no hormonal imbalance is observed and tertiary sex features may develop normally in girls with M-R-K-H syndrome i.e. nipples, pubic and axillary hair and female body shape (Fedele, Bianchi, & Berlanda, 2006; Ulrich et al., 2004). The occurrence rate of M-R-K-H syndrome is estimated differently. Some authors suggest that it happens once in 4000-5000 female births,

others claim that it occurs in 1-5% of female newborns (Panici, Bellati, Boni, Francescangeli, Frati & Marchese, 2007).

The etiology of M-R-K-H syndrome has not yet been fully explained. Inhibited growth of Müllerian ducts is most often claimed to be the cause of the syndrome, it usually occurs between the 8 and 10 week of foetal life and is caused by certain harmful factors e.g. ionizing rays, microbiological, chemical and hormonal factors, immunological or other disorders (Panici et al., 2007; Sołtysiak, 2008). Some authors also emphasize the role played by gene mutation. However it is known that patients with M-R-K-H syndrome have normal karyotype - 46 XX (Ulrich et al., 2004; Skałba, 1998).

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The most common first clinical symptom of M-R-K-H syndrome is the primary lack of menstruation in girls with correctly developed tertiary sex features. Therefore the syndrome is discovered quite late i.e. most often in patients aged around 16, who alarmed by the lack of menstruation, go to see a doctor. Other characteristic symptoms include abdominal pain of varying intensity and frequency. Pain in the chest, vomiting and even bleeding from the urinary bladder may also appear (Rzepka-Górska & Błogowska, 2000).

The treatment of patients with M-R-K-H syndrome consists of surgical creation or dilation of the vagina. Currently there are various methods of performing the surgery. Since the surgery requires a patient to co-operate with a doctor for several months, and be sexually active, the surgery is not performed in the puberty period. Presently M-R-K-H syndrome is ranked second in diagnoses preceded by a primary lack of menstruation (Rzepka-Górska & Błogowska, 2000).

The diagnosis of this syndrome is highly stressful for a teenage patient and her parents. Reaction of the family and environment is an additional emotional burden. Many mothers blame the syndrome on their behaviour during pregnancy, even though extensive research does not confirm it, and that significantly affects the atmosphere in which a girl grows up and impacts her relationship with mother (Bidzan, 2006). Female infertility itself, as well as the necessary long-term treatment, are not only a physical difficulty for a woman but most of all a huge emotional burden for the woman affected by this problem and for her partner and family as well. The information that a woman will not be able to bear children normally regardless of what she does and what treatment she undergoes - as is the case for women with M-R-K-H syndrome – may have several psychological consequences. Furthermore, taking into account the fact that in most cases this information is conveyed in the puberty period, it may have a considerable impact on psychosocial development.

In this paper the authors analyze the relation between the level of neuroticism, the coping style and positive and negative emotions in a group of women with M-R-K-H syndrome. The level of neuroticism seems to be important since this factor defines susceptibility to negative emotions such as fear, confusion, discontent, anger, guilt, or susceptibility to psychological stress. The elements that make up neuroticism are anger, aggressive hostility, depressive mood, impulsivity, oversensitivity and excessive self-criticism (Costa & McCrae, 1992a; Zawadzki, Strelau, Szczepaniak & Śliwińska, 2007). The results of research conducted so far show that infertile women score higher on all fear measuring scales, moreover they are more emotionally unsteady (Bidzan, 2006). In addition, research results show that blaming oneself and avoidance coping are the tools used most frequently by those women in difficult situations (Cwikel, Gidron & Sheiner, 2004; Jarmołowska, 2007).

The coping style is defined as "a set of coping strategies characteristic for a given individual" i.e. a set of cognitive and behavioral efforts, some of which may be made in a specific difficult situation (Heszen-Niejodek, 2000, p. 484). The research conducted by Bidzan and Józefiak (2006, after: Bidzan, 2006) showed that women who felt that they did not fulfill their role of a mother did indeed use escapism as a method of fighting stress. The research also showed that there is a significant difference in the avoidance subscale arising from the fact that women who found procreation difficult were indeed more eager to divert attention from the cause of stress than healthy women.

However, there are certain discrepancies in contemporary literature as far as the results of research in this field are concerned. Differences in research reports are visible mainly in conclusions on the social and psychological functioning of women facing the problem of infertility. The differences appear for example in conclusions concerning the dominance of one of the coping styles in women who are not able to have children. Some research shows that the avoidant coping style prevails in those women, other reports claim that the dominant style is the emotion-oriented (Bidzan, 2006; Cwikel, Gidron & Sheiner, 2004; Jarmołowska, 2007).

Some researchers focus mainly on the negative emotions experienced by women who are not able to have children (Klimek, 1986). They also underline that psychopathological symptoms, in particular depression, occur much more frequently in infertile women (29.7%) than women who do not have this problem (2.7%); it must be underlined that infertile women did not have any psychiatric disorders before (Bidzan, 2006). Results of research conducted among girls with M-R-K-H syndrome show a higher level of fear symptoms (1.2 times higher) and depression (1.4 times higher) as compared to results recorded in the control group of healthy girls (Laggari et al., 2009). It was also the girls with M-R-K-H syndrome who scored over 30 points in the selfdescriptive Beck Depression Inventory (BDI) which suggests deep depression as compared to the control group and girls with another gynecological condition e.g. polycystic ovary syndrome. Other research showed that infertile women had twice the high depression level compared with fertile women. The same research also showed that women, who have been coping with the problem of infertility for two or three years, display the most intense depressive behaviors as compared to women who have been aware of their infertility for one or over six years (Cwikel, Gidron & Sheiner, 2004). The authors explain this phenomenon with the fact that initially women, who found out that they are not able to have children, are more hopeful and optimistic and that protects them from symptoms of depression. However persisting experiences related to infertility as well as its treatment, significantly increase differences in the depth and frequency of depression symptoms in women, who experienced treatment failure, as compared to those women

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who have not experienced a failure yet. Moreover, authors claim that after six years women get used to the fact that they will not have children and it is the acceptance of this fact that protects them against depression. Therefore, the research referred to in this paper also took into account the time factor.

The aim of the study

The main aim of the study was to investigate relations between neuroticism, coping styles and positive and negative emotions in a group of women with M-R-K-H syndrome, and define the mechanisms behind relations existing between those variables. The first thing to be investigated was whether the coping style plays an intermediary function between neuroticism and positive and negative emotions in a group of women with M-R-K-H syndrome (see Fig. 1, line ab). The testing of such mediation requires a comment since both neuroticism (one of the personality traits described in the five-factor personality model by Costa and McCrae [1992a]), and the coping style (dispositional variable) are relatively constant subject variables having the status of personality variables. Still, neuroticism was given priority in this relation. The coping style is shaped based on inborn and personality factors in the course of a learning process (mainly related to identification of coping behaviors that turned out to be more or less effective; cf. Heszen-Niejodek, 2000).

Another target set was to check whether the mediation tested earlier is moderated by the level of neuroticism in women participating in the research (cf. line d on Fig.1) i.e. whether it is triggered by the level of neuroticism. In the light of data on the role of time factor in the dynamics of the emotional condition of women with M-R-K-H syndrome, the last step was to define whether the time elapsed since a patient was diagnosed with M-R-K-H syndrome is an additional moderator of mediation moderated by neuroticism (cf. line e Fig. 1.).

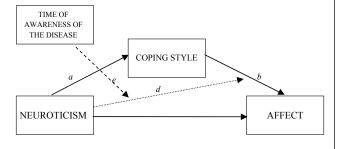


Figure 1. Conceptual moderated mediation model (solid line - direct effects; dotted line - moderated mediation by neuroticism; dashed line - moderation moderated mediation by time of awareness of M-R-K-H syndrome)

Method

Participants and procedure

The study covered 46 women diagnosed with M-R-K-H syndrome, aged 18-37 years (M = 23.48; SD = 4.88). The predominant majority of women participating in the research (91.3%) had secondary or higher education. 37% of women had a romantic relationship (no-data ratio was 43.5%). The average syndrome awareness period in the group covered by the research was 6.24 years (SD = 4.34) and ranged from 1 to 17 years.

The research was conducted individually. Women selected to participate in the research were in the age range 18-37 years as this is usually the period in which permanent romantic relationships are established and families started. In view of the specific character and rare occurrence of M-R-K-H syndrome, women to participate in the research were recruited through contact established with the population of sick women who also helped to look for other people to participate in the research.

Measures

Neuroticism was measured based on Personality inventory NEO-FFI by Costa and McCrae (1992b; Polish adaptation: Zawadzki, Strelau, Szczepaniak & Śliwińska, 2007). Out of 60 statements 12 refer to neuroticism (e.g. "I often feel tense and jittery" $\alpha = .81$). Respondents were asked to take a stance on each statement using the 5-point Likert scale.

Coping style. The coping style was assessed based on Endler and Parker's Coping Inventory for Stressful Situations (CISS) (1990; Polish adaptation: Strelau, Wrześniewski, Jaworowska & Szczepaniak, 2005), which makes it possible to assess the task-oriented (e.g) "Focus on the problem and see how I can solve it" $\alpha = .66$), emotions - oriented (e.g."Feel anxious about not being able to cope"; $\alpha = .65$) and avoidance coping style (e.g. "Take some time off and get away from the situation"; $\alpha = .40$). Respondents were asked to express their attitude towards statements concerning tendency to take remedial action in difficult situations, based on the 5-point Likert scale.

Positive and negative emotions. Positive emotions (e.g. "joy", "pleasure"; $\alpha = .90$) and negative emotions (e.g. "depression", "fear"; $\alpha = .94$) were studied based on the Emotional State Survey by Heszen-Niejodek, Januszek and Gruszczyńska (2001). Each scale is based on 10 adjectives. Respondents were asked to identify the level at which they currently experience a given emotion, based on a 7- point scale.

Statistical analysis

The analysis of mediation and moderated mediation was based on non-parametric bootstrapping procedure and computational macro for SPSS 19 (Heyes, 2012). The essence of the method is to generate a bigger number of samples of the same size as the initial sample through sampling with replacement (in analyses presented N=5000), and then for each sample created identify the range which the value of a given parameter in a population is probable to fall into (Preacher & Hayes, 2008). If the range does not include zero, the indirect effect is significant and mediation is shown. The possibility of parallel testing of several potential mediators and mediation moderators is the advantage of this method. It is also the most frequently used and recommended method for mediation analyses (ibidem).

Results

Descriptive statistics and intercorrelations between neuroticism and coping styles (task, emotions and avoidance-oriented), and positive and negative affect were presented in Table 1. Results obtained were in line with functional characteristics of the variables. Only the avoidance-oriented coping style was not significantly related to neuroticism or emotions, thus this variable was excluded from further analyses.

Table 1. Descriptive statistics and intercorrelations (N = 46).

Variables	M	SD	TASK	EMOT	AVOID	PA	NA
N	31.24	9.96	39**	.69**	.042	37*	.69**
TASK	43.11	4.34		32*	22	.35*	19
EMOT	55.22	8.37			.31*	46**	.66**
AVOID	32.22	4.74				27	.13
PA	31.63	10.32					39**
NA	22.80	12.00					

Note: N = neuroticism; TASK = task-oriented coping style; EMOT = emotion-oriented coping style; AVOID = avoidance-oriented coping style; PA = positive affect; NA = negative affect. * p < .05; ** p < .01.

Initial analyses excluded the relation between secondary controlled variables (such as age, education, romantic relations, time from diagnosis) and principal variables therefore secondary variables were not included in analyses of mediation and moderated mediation as co-variants.

Mediation

First the effect of neuroticism on positive affect through coping styles (task- and emotion-oriented) was studied. Bootstrapping results showed that this relation was fully mediated by the emotion-oriented coping style (direct effect: $\beta = -.04$; p = .853; indirect effect $\beta = -.26$; p < .01). The higher the neuroticism level, the stronger the tendency to cope based on the focus on negative emotions, and in consequence the lower the intensity of positive emotions (cf. Fig. 2).

Identical tests were performed for the relation of neuroticism and negative emotions, however this time only partial mediation was obtained (cf. Fig. 2). The relation between neuroticism and experiencing negative emotions

was direct ($\beta = .58$; p < .01) and indirect ($\beta = .30$; p < .01) through emotion-oriented coping style. In both cases a higher level of neuroticism was accompanied by a higher level of negative emotions.

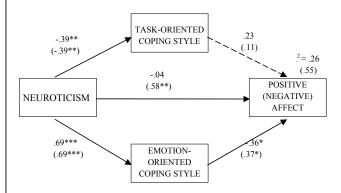


Figure 2. Results of mediation analysis for positive and negative affect (in parentheses). *Note*: Values presented are standardized coefficients (*p<.05; **p<.01; ***p<.001).

Moderated mediation

Analyses of moderated mediations were performed to check whether indirect effects between neuroticism and emotions depend on a specific level of neuroticism $(M, \pm 1 SD)$. Test results, separate for positive and negative emotions, are presented in Table 2.

Table 2. Bootstrapped indirect effects of neuroticism on positive and negative affect via emotion-oriented coping style at specific values of the moderator (neuroticism, N).

Positive affect						Negative affect				
N	β	SE	LL BCA	UL BCA		В	S	Е	LL BCA	UL BCA
- 1 SD	40**	.19	84	07		.20	.1	7	11	.57
Mean	28**	.14	56	02	.3	31**	.1	4	.05	.57
+ 1 SD	16	.23	68	.24	.4	13**	.1	9	.09	.81

Note: ** p < .01; 5000 bootstrapping resamples; LL BCA and UL BCA = Lower level and Upper level of the bias-corrected and accelerated confidence interval for $\alpha = .05$.

Results obtained show that the emotion-oriented coping style mediated the effect of neuroticism on positive affect at the mean and at low levels (-1 SD) of neuroticism, but not when neuroticism was high (+1 SD). What is more, the impact of the indirect effect was the biggest when the level of neuroticism was low. Thus in women with low and medium level of neuroticism this feature resulted in a weaker emotion-oriented coping style, and was in consequence linked to a higher level of positive emotions experienced.

Different results were obtained for negative emotions, namely the indirect effect of emotion-oriented coping style was growing together with the increase of neuroticism level and was significant when its level was medium or high The relationship between neuroticism, coping styles and emotions in women with Mayer-Rokitansky-Küster-Hauser syndrome 5

(+1 SD). Thus medium and, in particular, high level of neuroticism intensified negative mood indirectly through strengthening the emotion-oriented coping style.

The next step was to verify whether the aforementioned results depend on the time lapsed from the diagnosis of M-R-K-H syndrome (moderated mediation moderation). Thus models took into account the length of time for which a patient has been aware of the condition as a moderator of relations moderated by neuroticism. For positive emotions model, the interaction effect: neuroticism x neuroticism level x time from diagnosis, was not significant (direct effect, $\beta = .13$; p = .344). The indirect effect between neuroticism and positive emotions moderated by the level of neuroticism was not additionally moderated by the time from diagnosis ($\beta = .00$ with a 95%-confidence interval from -.05 to .14).

Those relations were different in case of negative emotions: moderation of moderated mediation was not observed $(\beta = .00 \text{ with a } 95\%\text{-confidence interval from -.01 to .06}),$ but the direct effect of neuroticism on negative emotions was moderated both by the level of neuroticism and the time lapsed from diagnosis (cf. Table 3; since the number of participants was small, the effect of low and medium and higher level of neuroticism was tested in subgroups). Direct positive relation of neuroticism and negative emotions occurred only in women with a medium and high level of neuroticism who have known the diagnosis for 6 or more years. However, the impact of this effect was the biggest in women who have known the diagnosis for over 10 years.

Table 3. Bootstrapped direct effects of neuroticism on negative affect at specific values of the level of neuroticism (1. moderator: N) and duration of M-R-K-H syndrome in years (2. moderator: TIME).

N	TIME	В	SE	N	TIME	В	SE
- 1 SD	1.90	.24	.50	M+1 SD	1.90	.56	.56
- 1 SD	6.24	.60	.36	M+1 SD	6.24	1.10**	.36
- 1 SD	10.58	.95	.48	M+1~SD	10.58	1.59***	.41

Note: ** p < .01; *** p < .001; 5000 bootstrapping resamples

Discussion

The aim of the study was to establish whether (1) a stress coping style is a mediator of the relation neuroticism - positive and negative emotions in a group of women with M-R-K-H syndrome, and (2) whether the mediation is moderated by the level of neuroticism in women and the time lapsed from diagnosis.

The results of bootstrapping yielded the fact that emotion-oriented coping style partly mediated the relationship between neuroticism and negative affect. Moreover, direct effect between those variables was moderated by the level of neuroticism as well as the time of awareness of M-R-K-H syndrome (it occurred only in women with a medium and high level of neuroticism who have known the diagnosis for 6 years or more). It might be an indication of the fact that as the time goes by neuroticism has ever bigger impact on the emotional state of women with M-R-K-H syndrome and thus makes them experience negative emotions such as uncertainty, disappointment, anger and depression. It is even possible that a negative spiral is created: experiencing negative emotions intensifies the feature of neuroticism which makes further experiencing of negative emotions even deeper. Results obtained are in opposition to studies conducted by Cwikel, Gidron and Sheiner (2004), who describe curvilinear dependence between the period for which a patient has been aware of their condition and emotional adaptation to infertility, where in the long-term horizon "time heals all wounds".

The relation between neuroticism and positive emotions was fully mediated by the emotion-oriented coping style and was independent of time that lapsed since from the diagnosis. However the level of neuroticism itself turned out to be an important moderator of this relation: indirect relation via emotion-oriented coping occurred de facto only in a group of women with this feature at a medium or low level. In those cases neuroticism led to a lower tendency to cope through orientation on negative emotions, and later to a higher level of positive emotions experienced.

Based on the above, it may be assumed that an attempt to modify the coping style e.g. through psychotherapeutic activities, could increase the intensity of positive emotions. The coping style is indeed a certain permanent but not rigid predisposition to cope, however it is rather about preferring certain precautionary measures with the main element of the coping style being flexibility (cf. Heszen-Niejodek, 2000). Attempts to modify this predisposition could be based not only on impacting the number and type of coping strategies available but also flexibility of using them. Thus the target should be to make it easier for women with M-R-K-H syndrome to focus on a task or on positive emotions. What seems to be important in this context is to reformulate the situation, change the way in which it is perceived, focus on resources available and not the element that cannot be achieved. Research results suggest that it is not the fact of not having children itself that has a negative impact on the psychophysical functioning but only the perception of this fact as a major problem in one's life. Research shows a clear difference between two types of relations – childless due to health or life problems, and those that do not have children because they decided not to. Extensive data show that partners who are not able to conceive despite a strong desire and an urge to have a child, either naturally or with the help of latest technologies and medications offered by modern medicine, feel unhappy and unfulfilled contrary to those couples for whom the decision not to have a child is a conscious choice. The vast majority of those couples are very happy (Dabrowska-Caban, 2001), and describe their relations as agreeable and tight (Kalus, 2001). Couples who

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deliberately choose not to have children usually have a different life perspective as their main aim is self-fulfillment (Dąbrowska, 2003). Results of the research also show that people who do not want to have children look for a partner who has the same attitude to this issue (Kalus, 2001). Furthermore, it turns out that in case of couples who make a conscious decision not to have children, the wives are more independent and more involved in professional career or other activity whereas in family environment they are dominant and have a tendency to compete. The husbands on the other hand are sensitive and submissive (Dąbrowska, 2003, 2005; Dabrowska-Caban, 2001). Additionally, it turns out that both women and men, who chose not to have children, value their own education highly, are more engaged in career development devoting more time to it than people their age who are parents (Kalus, 2001). Apart from that, those who consciously chose not to have children enjoy life, care for their relationship, talk to each other a lot and do not feel obligated to respect religious imperatives and prohibitions (Dąbrowska, 2003, 2005; Dąbrowska-Caban, 2001; Kalus, 2001). Other authors claim that couples who consciously decided not to have children show each other more love and intimacy, they spend more time together and care more for their intellectual development (Slany & Szczepaniak-Wiecha, 2003).

Folkman and Moskowitz (2000, 2004, 2006) identify several possible ways to tackle the problem and possibilities of experiencing positive emotions i.e.: positive reappraisal, adaptive goal processes, reordering priorities, benefit finding, infusion of ordinary events with positive meaning. Positive reframing of the situation of infertility in order to have a more constructive look at life plans may be difficult for the women with M-R-K-H syndrome because of the specific nature of their situation. People often tend to concentrate on aims that are impossible to achieve. This is connected to the principle of scarcity (Cialdini, 1988). This group of women knows they will not have children normally regardless of what they do and what treatment they undergo. For some part of this group (with more complicated picture of the illness) it can be also difficult to take the role of woman in terms of sexual life and to build intimate relationships.

Positive reappraisal of the situation can be also difficult because of the stereotypes relating to the role of women and social pressure. Bidzan (2006) refers to studies that showed that 83% of people, who are not able to have children, feel there is social pressure to have children. The pressure is usually exerted by a partner and parents, less frequently by friends and grandparents. This intensifies feelings related to not having a child and the sense of being stigmatized by the society. Moreover, some studies reveal that people who do not have children are perceived as egoistic, selfish and unhappy. Furthermore, what is very characteristic is the fact that people who do not have children are very often perceived as ill-adapted, lazy and not very responsible

(Dąbrowska, 2003, 2005; Dąbrowska-Caban, 2001; Kalus, 2001).

Based on the initial analyses of simple correlations, avoidance-oriented coping turned out not to be related to neuroticism or to positive or negative emotions in a group of women covered by the research, and was therefore excluded from the main analyses. The results of different studies of the relationship between avoidance-oriented coping and affect and neuroticism are usually different: significant correlations between those variables are observed (cf. Endler & Parker, 1990; Higgins & Endler, 1995; Murberg, 2009), but also their lack (cf. Endler & Parker, 1990; Roesch, Wee & Vaughn, 2006; Szczepaniak, Strealu & Wrześniewski, 1996). The results obtained seem to fit into non-conclusive data on the role of avoidant behaviour of women with M-R-K-H syndrome, who focus mainly on social contacts. There are studies confirming that a tendency to withdraw from people, which intensifies over time, can be observed in childless women; and this in turn has further negative consequences (Klimek, 1986; Jarmołowska, 2007). Other studies however, suggest that infertile women have numerous but superficial social contacts. There are also reports indicating that infertile women are very sensitive to other people's reactions, they blame them for their own problems, are mistrustful and very often feel threatened in close relations (Jarmołowska, 2007). Still it needs to be mentioned that in the presented reports the avoidance coping sub-scale was characterized by unsatisfactory psychometric ratios despite the fact that it has been identified through a factor analysis.

Other interesting result of the studies presented in this article is the fact of a negative correlation between positive and negative emotions. The links between them are usually not important (Folkman, 1997). It is worth analyzing whether it is connected with the specificity of the research group. Numerous research shows that infertile women focus to a large extent on the inability to have children and find it difficult to think about issues other than infertility (Klimek, 1986; Jarmołowska, 2007). Research covering childless couples shows that women more often than men consider the desire to fulfill themselves as mothers more important than the harmony of married life, moreover their evaluation of the marriage and their position in it is lower. It is much harder for them to accept the unintended childlessness and they do everything to give birth to their own child, and finally consider adoption as the last resort (Jarmołowska, 2007). Thus it may be presumed that such an intense focus on one aspect makes it difficult to parallely fulfill aims on different planes and get satisfaction from them. Thus the emotions experienced vary between positive and negative states, depending on the current perception of the problem of infertility, and the level of hope for solving it. Furthermore the prevalence of positive over negative emotions, declared by women with M-R-K-H syndrome, turned out to be statistically significant, which is similar to a series



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of studies on coping with a disease (t[45]=3.209, p<.001) (cf. Ben-Zur, 2002; Rabkin, McElhiney, Moran, Acree & Folkman, 2009). This result seems to fit in with a general trend for the so-called positive offset (positivity appears more frequently; Diener & Diener, 1996).

To conclude, based on the data obtained, it may be expected that in a group of women with M-R-K-H syndrome, the positive and negative affect are at least partly modulated by permanent personality traits such as neuroticism - directly and/or indirectly, based on a tendency to focus on one's own negative emotional state. The reformulation of the situation mentioned before and building the skill to generate positive emotions seems to be a significant aim of psychological activities in this research group. Fredrickson (1998, 2006) underlines that particularly in long-term stress situations positive emotions may play several adaptation functions: they make it possible to rebuild physical, intellectual and social resources, and they also increase the scope of attention and activity.

The reports presented are not however free from limitations. The number of women in a group covered by the research may be seen as a drawback of this research, and in consequence the reliability and accuracy of conclusions drawn on this account. M-R-K-H syndrome is however a relatively rare congenital disorder. It is estimated that in Poland approx. 0.2% of women suffer from this condition. Only 404 items related to this condition can be found in EBSCO database. And almost all of them refer only to the medical aspect of this condition (only three referred to the emotional state and the adaptation of women with M-R-K-H syndrome), and are a case study in substantial majority (empirical studies - available to the authors - conducted in a group where N = 5, cf. Laggari, 2009). In view of the specific character of this condition it is difficult to obtain a larger research group. This factor also influenced the number of women in a sample in the research presented in this paper.

A certain limitation on the research presented is also the result of its crosswise perspective. Longitudinal measurement would make it possible to define not only the emotion dynamics i.e. situational variables, but also possible changes regarding more permanent features such as coping styles and neuroticism. Only the longitudinal approach would make it possible to really explore the mechanisms of dependencies between personality traits, coping style and emotions and their dynamics in the relation of the time from learning the diagnosis. Furthermore, taking into account situational aspects of coping (preventive strategies) would most probably make it possible to better understand the range of remedial activities taken by women with M-R-K-H syndrome. Nevertheless, the results described make it possible to identify primary psychological and social problems related to M-R-K-H syndrome, and to formulate proposals related to psychological actions addressed to this group of women.

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