ORIGINAL RESEARCH

A Case Control Study on Family Functionality and Coping Mechanisms Among Healthy Siblings of Pediatric Patients with Leukemia in Kythe-affiliated Hospitals in the Philippines

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Background: Leukemia is a leading cause of morbidity and mortality among pediatric patients in the Philippines, but it also significantly affects family dynamics and behaviors. Aside from the parents, siblings are also vulnerable to the effects of a chronic disease within the family but are not usually the topic of family studies.

Objective: To determine if having control-oriented or escape-oriented coping mechanisms is associated with the different levels and dimensions of family functioning among children ages 13 to 18 years old who have siblings diagnosed with leukemia.

Methodology: This case control study was conducted in two Kythe-affiliated hospitals in Luzon, Philippines. Using McMaster Family Assessment Device, adolescent siblings of leukemia patients who belong to dysfunctional families (n=8) were considered as the cases, while siblings who belong to functional families (n=5) were the controls. KidCOPE was used to identify coping mechanisms. Odds ratio for escape- and control-oriented coping mechanisms were computed using Fisher's exact test through MedCalc, while association of coping mechanisms with demographics was determined using Chi square test through SAS 9.4.

Results: Siblings of leukemia patients who belong to dysfunctional families are more likely to use escape-oriented coping mechanism (OR = 2.2; 95% CI = 0.0746 to 64.9082; p = 0.648) but also more likely to feel that it is not helpful to them (OR = 0.6471; 95% CI = 0.0111 to 37.6665; p = 0.8337). However, an association between coping mechanisms and family functionality cannot be established because the computed odds ratios are not significant. A significant association was found between the number of extended family members living with the sibling and efficiency of control-oriented coping mechanisms (p = 0.024).

Conclusion: Association between coping mechanisms of adolescent siblings of leukemia patients and family functionality was not established, but further studies with larger sample sizes are needed to confirm this.

Keywords: Case control, coping mechanisms, family functionality, siblings, pediatric patients, leukemia, KidCOPE, McMaster Family Assessment Device, Philippines

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Introduction

In the Philippines, leukemia is identified as one of the top causes of mortality among pediatric patients[1] and accounts for 50% of childhood cancer.[2] Due to its course, leukemia poses numerous challenges to Filipino families such as change in roles, logistical adjustments, and financial burdens.[3] These factors have the potential of influencing family functioning as well as their different coping mechanisms.

Cancer is a life-changing event that does not only affect the person who is sick but also the other members of the family. As such, siblings of the chronically-ill usually experience instability and disruption in their lives. Most of the family's energy may be focused on the ill child that well siblings are left-behind, forgotten, and neglected, making the family a crucial component on how siblings of chronically-ill patients cope.[4-7]

Changes in the general functioning of the family and the relationship between its members can influence the functioning of the healthy child.[8] Functional families are those that are able to maintain good relations and can foster adaptive coping strategies, whereas dysfunctional families lead to maladaptive coping, school problems and high propensity to engage in risky behaviors.[8-11] When a child experiences chronic pain, there are consequences both for the child and their family.[12] Thus, alterations and adjustments to the dynamics of the family, whether it be behaviorally, emotionally and/or cognitively, are often evident.[13] These adjustments, therefore, may significantly impact the healthy sibling especially in terms of one's coping mechanisms.

This study serves to bridge the gap by identifying the effects of family functionality on sibling's coping mechanism, although, there is very limited information regarding siblings of chronically-ill patients in the Philippine setting. Neglected children are said to perform more poorly compared to their non-maltreated counterparts by having lower grades, more suspensions, more disciplinary referrals, and more grade repetitions. Furthermore, neglect may also result to the child's resentment, jealousy, feeling of loneliness, as well as, adjustment problems and aggression in obtaining attention from their parents.[14-17] However, studies have shown that towards late adolescent, other children can also develop positive, protective coping mechanisms influenced by several family variables, thus, preventing maladaptive behaviors from progressing.[12,18,19] In addition to this, coping mechanisms are fairly stable and constant across situations. Therefore, it would be important to identify children who use maladaptive coping styles in dealing with a chronically ill sibling as this may affect their well-being and development.[4,20]

Therefore, this study aims to determine the association of having control-oriented or escape-oriented coping mechanisms in different levels of family functioning among children ages 13 to 18 years old who have siblings diagnosed with leukemia. It is hypothesized that belonging to a dysfunctional family would lead to greater odds of developing escape-oriented coping mechanisms, especially if the communication dimension is weak within the family.

Patients and Methods

Study Design, Setting, Population, and Sampling Methods

Participants partook in a case control study and were selected through convenience sampling among adolescent siblings ages 13-18 years old of patients diagnosed with leukemia affiliated with Kythe Foundation Inc., a non-government organization that caters to the psychosocial and emotional needs of hospitalized, chronically ill pediatric patients. Coping mechanisms were identified through self-administration of the KidCOPE questionnaire and family functioning level measured through the McMaster Family Assessment Device (FAD). Cases are those whose family general functioning is unfavorable, and controls are those with good general family functioning. To be included in the study, the participants must be healthy siblings (no chronic or progressive diseases that requires regular treatment or consultation to the doctor such as, but not limited to, mental illness, diabetes, cardiovascular, respiratory, urogenital and renal diseases, congenital diseases, and physical

disabilities) of diagnosed leukemia patients (ALL, CLL, AML, or CML) for at least six months already but not in relapse, and parents who are also willing to participate. McMaster FAD was also administered to a parent or guardian of the leukemia patient aside from the sibling. The average of their scores was computed to determine family functionality. When multiple siblings were eligible for the study in a single family, the participant was selected through simple randomization.

A total of 103 families were screened but only 39 were eligible to participate and were subsequently invited. Three weekend schedules were designated for the study, with the participants having the liberty of choosing based on their availability. However, because of logistical and schedule concerns of some participants, only 13 families were able to participate in the study. The study was approved by the ASMPH Institutional Review Board as well as the partner organization (Kythe), and was subsequently conducted in the designated Kythe Foundation Inc. provincial offices or in the Kythe Foundation, Inc. main office to ensure the confidentiality, comfort, and ease of the participants.

Tools

The McMaster FAD is a 60-item, self-administered screening tool for adolescents and adults 12 years old and above, designed to assess family dynamics within specific areas of functioning. The six dimensions assessed are

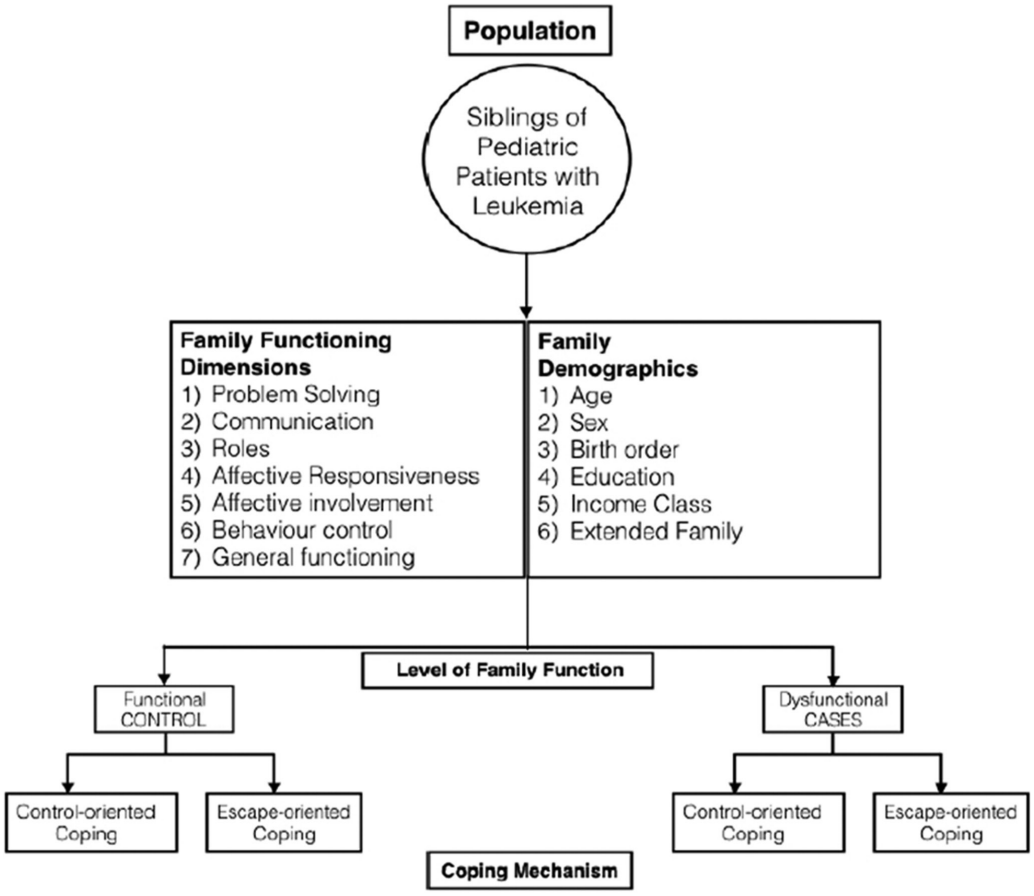

Figure 1. Conceptual framework of the study.

problem solving, communication, functions/positions of individuals within the family, affective responses, affective involvement, and behavior control.[21] These six dimensions are postulated to have the most impact on the emotional and physical health of family members.[22] Multiple studies have shown the tool's reliability having an internal consistency within 0.50 to 0.80. It also has a low correlation with social desirability, meaning it cannot be easily influenced by the biases of the participants.[21]

KidCOPE is a brief, clinical checklist that screens for cognitive and behavioral coping in children and adolescents. The version used in this study is an 11-item 4-point response scale that caters to children between 13-18 years old. It is used to assess the perceived frequency and efficacy of helpful and not-so-helpful coping strategies. Included in these strategies are distraction, social withdrawal, cognitive restructuring, self-criticism, blaming, problem solving, emotional regulation, wishful thinking, social support, and resignation.[21] According to Hernandez[4], the conciseness of KidCOPE may act as a significant limitation, but it is still a frequently endorsed coping scale due to ease of use and brevity, which can both be considered as assets and liabilities.[23] Another limitation is that specific cut-off scores and population norms are not available for this instrument.[21]

Methods and Procedures

For reliability and validity of the tools in the Philippine setting, the McMaster FAD and KidCOPE were forward- and back-translated through two independent resource persons and were subjected to a pre-test for validation by administering them to five participants of the study population. Comments and suggestions were integrated to the revised questionnaires.

Recruitment of eligible participants was done after profiling of leukemia patients in two Kythe-affiliated hospitals. Complete informed consent forms, detailing the objectives and methodology of the study, were given and explained to the parents/guardians at the start of the session. Assent forms containing the aforementioned details were also provided to the actual adolescent participants. To address the possible risks such as psychological discomforts when answering the questionnaires, a Child Life Coordinator (CLC) from the Kythe Foundation, Inc. was on standby.

Each participant and his parent/guardian answered a basic information questionnaire to elicit demographics information. After reading and explaining the instructions, the two questionnaires (KidCOPE and McMaster FAD) were given to each individual to be answered for a maximum of 30 minutes. A debriefing session was organized after answering the questionnaires.

Data Analysis

Statistical analysis was conducted using SAS 9.4. Reliability analysis of the questionnaires was done by measuring Cronbach's alpha. For family functioning, the study used the scoring system of the McMaster FAD. Scores for the negative items were transformed and computed accordingly. Scores of each individual were computed by getting the sum of all the values of the items for each scale and dividing them by the number of items for that dimension. Family score for each scale was the average of the participants' and parents' scores.[24] A General Functionality score of 2.00 or above indicated problematic family functioning. Responses with missing values for more than 40% of the items were not included. For coping mechanisms, the study used the scoring of KidCOPE where each item was assigned to a specific coping mechanism in terms of frequency and efficacy of use. The different coping mechanisms were classified into two general groups: control-oriented and escape-oriented.[25,26] The average score for each general group was obtained for each participant. A participant was deemed to have a predominantly control-oriented coping mechanism when his/her average score of items under control-oriented coping mechanism was higher than his/her average score of items under escape-oriented coping mechanism, and vice versa.

Chi square test with SAS 9.4 was used to determine the strength of association between family functioning, patient demographics, and coping mechanisms (Tables 3 & 4).

Paired t-test was used to determine differences in the two general groups of coping mechanisms. Fisher's exact test was used to compute for the odds ratios through MedCalc (Table 5).

Ethical Considerations

To ensure confidentiality, codes were assigned to each participant. All gathered data were stored in a password-protected Google drive accessible only to the researchers and the preceptor for the purpose of the study. Participants were compensated with rice meals and transport allowance after completing the questionnaire. It was also emphasized that their participation in the study would not affect the treatment and service they are receiving from the foundation.

Training with the Child Life Coordinators (CLCs) of Kythe Foundation, Inc. was conducted for the interviewers regarding proper debriefing. This was conducted after each session to assess their emotional and psychological status after the test taking. Throughout the data collection period, no participant showed adverse behavior due to the research conducted. Completion was subject to approval of the coordinators. Precaution for maladaptive behavior and coping during test-taking was referred to the CLCs assigned for intervention.

If the participant is willing, the result of the study can be shared to them through their assigned CLCs for appropriate disclosure and discussion of possible interventions.

Results

Demographics

A total of 13 participant families took part in the study. Table 1 shows the summary of the participants' demographics. Siblings' ages ranged from 13-18 years old with a mean age of 14.85. Families had 4-11 members. Most of the siblings spend at least 2 hours a day with their families (92.30%). Most families' incomes fall under Class 2 to 4, with income of more than PhP40,000 (53.85%). Majority of the participant families' religion is Roman Catholic (92.31%).

Table 1. Summary of participants' family demographics.

	Mean	SD	Median
Age of participant	14.85	1.68	15
Age of sibling with leukemia	8.77	5.59	6
Family Size	6.46	1.98	6
Household Size	7.38	2.84	7
Extended relatives in household	2.38	3.04	1
Annual income class (FIES 2012) 1 - Under Php40,000 2 - Php40,000-59,999 3 - Php60,000-99,999 4 - Php100,000-249,999 5 - Php250,000	2.00	1.03	1
Hours spent with immediate family	6.76	4.38	8
Hours spent with extended family	4.25	3.49	3.5

Family Functionality

Based on the results of the McMaster Family Assessment Device, majority of the families were dysfunctional in terms of general functioning (61.5%) and in the dimensions of communication (84.6%), affective responsiveness (76.9%) and affective involvement (76.9%). Problem solving was the only dimension wherein majority of the families were functional (69.2%). It is important to note that all families were dysfunctional in the dimensions of roles and behavior control. Based on general functioning, a total of 8 cases and 5 controls were found.

Table 2. Family functionality based on McMaster Family Assessment Device.

	Functional	Dysfunctional
General Functioning	5	8
Problem Solving	9	4
Communication	2	11
Roles	0	13
Affective Responsiveness	3	10
Affective Involvement	3	10
Behavior Control	0	13

Siblings' Coping Mechanisms

Of the participants, only 1 (7.7%) used escape-oriented coping mechanisms more frequently than control-oriented ones. In terms of efficacy, all participants find it more helpful to use control-oriented coping mechanisms.

Chi square Analysis, Paired t-test, Odds ratio

The efficiency of control-oriented coping is correlated with the number of extended family living with the siblings ($p < 0.024$). However, there was no significant relationship found between any of the variables among different coping mechanisms and family functioning dimensions ($p > 0.05$).

Table 3. Association of demographics and coping mechanism.

Coping	Demographics	Chi square value
Escape- Oriented (Frequency)	Sex	8.775 (p = 0.187)
	Age	35.2083 (p = 0.506)
	Birth Order	10.4464 (p = 0.577)
	Extended Family	3.8458 (p = 0.698)
	Education	4.0379 p = (0.672)
	Income	19.5000 (p = 0.362)
Escape- Oriented (Efficiency)	Sex	5.9583 (p = 0.545)
	Age	50.375 (p = 0.176)
	Birth Order	18.7649 (p = 0.174)
	Extended Family	9.8313 (p = 0.198)
	Education	13.0000 (p = 0.072)
	Income	19.5000 (p = 0.553)
Control- Oriented (Frequency)	Sex	3.1417 (p = 0.678)
	Age	30.3333 (p = 0.449)
	Birth Order	11.9167 (p = 0.291)
	Extended Family	7.3667 (p = 0.195)
	Education	7.8788 (p = 0.163)
	Income	15.1667 (p = 0.439)
Control- Oriented (Efficiency)	Sex	8.0708 (p = 0.233)
	Age	30.8750 (p = 0.711)
	Birth Order	12.6905 (p = 0.392)
	Extended Family	14.5507 (p = 0.024)
	Education	2.5756 (p = 0.839)
	Income	16.9722 (p = 0.525)

An association between family functionality and sibling coping mechanism cannot be proven because the odds ratio values were not significant. Among the participants, siblings in dysfunctional families were 2.2 times more likely (OR = 2.2; 95% CI = 0.0746 to 64.9082; z statistic = 0.457; p = 0.648) to use escape-oriented coping mechanisms, compared to those who have functional families. In terms of efficacy, siblings in a dysfunctional family are 1.54 times more likely (OR = 0.6471; 95% CI = 0.0111 to 37.6665; p = 0.8337) not to feel escape-oriented coping mechanisms as helpful to them.

Table 4. Contingency table for coping mechanisms and family functionality. Where zeros cause problems with computation of the odds ratio or its standard error, 0.5 is added to all cells (a, b, c, d)[27,28]

		Coping Mechanism (Frequency)		Total
		Escape-Oriented	Control- Oriented	
Family Functionality	Dysfunctional	1	7	8
	Functional	0	5	5
	Total	1	12	
Odds Ratio			2.20	
		Coping Mechanism (Efficacy)		Total
		Escape-Oriented	Control- Oriented	
Family Functionality	Dysfunctional	0	8	8
	Functional	0	5	5
	Total	1	13	
Odds Ratio			0.65	

Discussion

Findings of the study showing an association between the presence of extended family members and efficiency of control-oriented coping mechanisms add to the conflicting results from previous studies. In the context of the Filipino culture, the findings of this study on preferred coping mechanisms demonstrate how crucial social and family relationships are in the Filipino family especially adolescents.

Association of Demographics and Coping Mechanisms

Demographics and coping mechanisms in this study only show significance between efficiency of control-oriented coping and number of extended family, but current literature has different findings. Studies are conflicting regarding present age and sex of the sibling as associated with coping mechanisms. One study shows that adjustment problems were more common among siblings of older age and of the female sex.[29] Another study reported however that females show more coping efforts than males as they are more affected in terms of interpersonal problems than the opposite sex. Although, the study also mentions that coping is not related to sex in terms of the type of coping, be it approach or avoidance.[30] As for birth order, extended family, education, and income, there have been no studies that demonstrate their association with coping mechanisms. As such, there is a need to delve deeper into this facet of adolescent studies. It is however known that Filipino families, in general, view family as a unit for support and closeness as compared to just being a traditional family having a father, mother, and siblings.[31] Lapena, et al.[32] stated that Filipino youth generally cope by sharing their problems with someone. It showed that in rural areas, the mother is the most commonly approached by Filipino adolescents followed by their friends. While in urban areas, female friends were the first approached followed by their mothers. Only 3% of rural and 2% of urban youth in the study mentioned keeping their problems to themselves. Consistent with literature, although statistically insignificant, participants of the study used social support as the most frequently coping mechanism with a score of 2.1 out of 4, tying with emotional regulation (calming) and resignation. However, it has a score of 2.7 out of 4 among respondents, third highest following emotional regulation (calming) as the most efficiently used coping mechanism among the participants with a score of 3.5 out

of 4 and wishful thinking being the second most efficiently used with a score of 2.8 out of 4.

Family Functioning and Coping Mechanism Association

Based on the results of this study, the computed odds of employing escape-oriented coping and finding it more helpful than control-based mechanisms by belonging to a dysfunctional family is not significant. But, the odds ratio on the frequency of use of escape-oriented coping mechanisms among dysfunctional families was consistent with previous studies stating that subgroups from this population generally have maladaptive coping mechanism.[14-17] Nonetheless, the results of this study are not statistically significant to generalize to the study population due to the limitations encountered. In addition, the definition and tool used for coping mechanism in other studies may differ from that used in this study, which may yield different results given that certain factors will fall in different categories.

Filipino Families' Context in Relation to Coping Mechanisms and Family Functionality

Chronic illnesses in the family have been noted in a systematic review of Van Schoors, et al.[33] to reorganize family roles and priorities, leading to disruption from normalcy. In another study, many Filipino families experience severe caregiver strain. Many attribute strains to lack of resources but the study mentioned faith, religion, and support from within the family as a source of help.[34] One main factor that drives the formation and development of coping mechanisms in adolescents is the cultural background of individuals.[35] In the context of Filipino teenagers, it was found that most of the time they resort to social interaction when dealing with difficult and stressful situations.[36] As mentioned earlier, the youth cope by seeking support from family, friends or other people in the environment[32], towards people whom they believe can enlighten them and give them proper advice in a given situation.[37] This is seen in the study, as social coping mechanism is one of the most used by the adolescents. As seen in the results, problem solving deemed to be the highest dimension for both functional and dysfunctional families. According to Lapena[32], problem-solving skills of the youth are influenced by different internal factors such as self-confidence, inherent talents, skills and intelligence, and belief in God. The younger ones are said to have *lakas ng loob* that helps them deal with their problems while the older youths are *madiskarte sa buhay*. In that, both exemplified confidence and strong faith in oneself, which helps them cope with difficult situations. In the study, both parents and siblings also reflected similar factors during the debriefing session; they exemplified optimism in overcoming hurdles through *pagpapaubaya, pagbibigay and pasasalamat sa mga grasya*. As a matter of fact, as stated in De Leon and Balila's[36] study, adolescents who have good family problem solving skills are less likely to engage in maladaptive behaviors. Their findings show that Filipino adolescents demonstrate a problem-focused approach in coping; they often choose the best strategy in dealing with difficult situations.[36]

Limitations and Strengths

This paper has several limitations. As the sample size is only composed of 13 participants, margin of error increases thereby limiting the level of precision. In addition to this, the ability to find statistically significant differences between groups is compromised. Therefore, the results of the study cannot be generalized to the population of interest. Another issue that arises is the low Cronbach reliability index of KidCOPE and McMaster FAD (<0.7) from the sample population. Thus, it would be advisable to increase the population to garner higher levels of precision, power, Cronbach reliability and decreasing margin of error. Furthermore, the inclusion and exclusion criteria may be refined so as to not limit the number of participants that may qualify for the study.

The study however also has its strengths. As discussed earlier, this study aimed to be the first study in the country which delves into the coping mechanism and family functionality among healthy siblings of pediatric children

with leukemia making it a pilot study that can be utilized by future researchers. In addition to this, strict abidance to the inclusion and exclusion criteria was conducted, making the results reflective of a careful selection process. The use of a wide landscape of participants proved to be of key importance in finding participants, as there are a limited number of prospective participants fitting the inclusion and exclusion criteria if it were limited to only a single location. It is important though to ensure that selected locations have the same level of environmental factors such as development in the city, access to media, transportation, and the like.

Recommendations

To our knowledge, this is the first study to tackle on the possible association between family functionality, with its different dimensions, and coping mechanisms among siblings of chronically ill patients in the Philippines. Further studies are needed in order to have a better understanding of the dynamics between the two, in light of the difference in family dynamics and culture as compared with Western families, where most literature are from. Improvements can be made in the sample size and exploring the association of the presence or absence of extended family members further through possible inclusion of other malignancies with similar consequences such as leukemia. The association between number of extended family members and efficiency of control-oriented coping mechanisms could be explored in the future as a means of enforcing the importance of having external family support during times of chronic illnesses. It is also interesting to look into the roles, behavior control, and problem-solving dimensions of Filipino families, given the data gathered in this study. Information regarding this field of study can help in the development of support programs and recommendations for psychosocial and emotional care not only of chronically ill patients, but their siblings as well. This highlights the need for a family physician and/or family psychiatrist/ counsellor who should be made available to the families of patients with chronic illness.

Conclusion

Although odds ratios show that siblings in dysfunctional families were 2.2 times more likely to have an escape-oriented coping mechanism and 1.54 times more likely to not see escape-oriented coping mechanisms as helpful to them, these are not statistically significant. A significant association was found between efficiency of control-oriented coping and extended family; however, no other significance was found in the other coping mechanisms and demographics variable. There was no significant association among variables in coping mechanisms and family functioning dimensions. Further studies are needed in this topic in order to implement programs and projects for the well-development of siblings of chronically-ill patients. An increase in the sample size is need and refining the inclusion and exclusion criteria can be considered. Looking into roles, behavior control, and problem-solving family dimensions of Filipinos in relation to coping mechanisms is also recommended in future studies.

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