Most families of children with Duchenne muscular dystrophy (DMD) first receive professional information about the disease at the time of their child’s diagnosis. Generally, as the families begin to build a supportive care system for their children, the parents will research DMD on their own or gather formal knowledge about the disease from professionals. However, gaining access to care is a major challenge because they often do not know how to ask the right questions. In particular, vulnerable populations may experience difficulties in assisting their child’s emotional adjustment to the disability and use of available services. The purpose of this study was to test the possible effects of psychosocial mediators of parental health, family hardiness and family support on family function in relation to the age at diagnosis of children with DMD. One hundred and twenty-six Taiwanese parents of children with DMD who are members of the Taiwan Muscular Dystrophy Association filled out questionnaires. Subjects received a phone call before and again within the first week after we mailed them a questionnaire, a stamped return-addressed envelope, and a consent form. The questionnaires included the Family Hardiness Index, Family Assessment Device, Family APGAR, Duke Health Profile and demographic questions. Hierarchical multiple regressions were conducted to test whether family hardiness, parental health, or family support mediated or moderated the association between age at diagnosis and family function. Family hardiness positively mediated the association between age at diagnosis and family function. These findings may help the design of interventions to develop family hardiness in families of children with DMD.

**Key Words:** Duchenne muscular dystrophy, family functioning, family hardiness, family support, parental health

children may help to prepare the families to participate in treatment decisions related to their children. Mediation hypotheses posit how or by what means a causal effect occurs, and how one variable predicts or causes an outcome variable. In contrast, moderators address when or for whom a factor is more strongly related to an outcome. More specifically, a mediator is defined as a potential intervening variable (M) that explains the relation between a predictor (X: independent variable) and an outcome (Y: dependent variable) [4,5]. In other words, a hypothesis that a predictor is related to or causes a mediator should have a theoretical rationale [6]. The predictor causes the mediator which, in turn, causes the outcome; the mediator should be something that can be changed. The main purpose of mediator analysis is to examine why an association between a predictor and outcome exists. Understanding such complex relationships among variables can also provide a theory-base for intervention.

CONSTRUCTION OF AN ANALYTICAL MODEL

We developed a Model for Stressors, Resources, and Functioning (Figure 1) [3] based on the Resiliency Model of Family Stress, Adjustment, and Adaptation [7]; using correlations among variables, confirmatory factors analyses, and hierarchical multiple regression analysis from predictor variables with dichotomous responses (two categories, usually coded 0 and 1); family income, employment, parent health, family support, family hardiness, child disability, and age at diagnosis were classified into two domains: family resources and family stressors [3]. These seven variables were termed components of family resources; the other variables (child disability and age at diagnosis) were termed components of family stressors.

*Family stressors* include a child’s disability and age at diagnosis, as measured by scales of individual complete dependence (Barthel Index) [8], and reported age when diagnosed with DMD. *Family resources* included five potential mediating variables that are all measured with a demographic sheet, the individual Duke Health Profile (Duke) [9], Family APGAR [10], and Family Hardiness Index [11]. These variables are defined below.

Parents’ employment and annual income were the most important variables regarding family characteristics of the family resources in our model. The Duke measures parental health as physical, psychological, social and emotional health of parents. *Family support* represents the parents’ satisfaction and examines how parents perceived support in the five attributes of adaptation, partnership, growth, affection, and resolve [12]. *Family hardiness* is conceptualized as the energy resource that includes commitment, challenge, and control [7]. Family function represents families’ abilities, including problem solving, communication, role, affective responsiveness, affective involvement, and behavioral control. It is used to evaluate family adaptation and uses a scale based on the individual Family Assessment Device (FAD) [12].

The objective of this study was to explore the influence of selected family resource variables on family function. The study hypotheses were as follows: family resources, including parental health, family hardiness and family support, might be mediated by or moderate the association between age at diagnosis and family functioning. If this assumption is true, the final effect of family stressors on the reduction of family function will depend on the effect of family characteristics, healthy parent status, family support, and family hardiness on family resources, in the intermediate process.

METHODS

**Study design and procedures**

This cross-sectional study used hierarchical multiple regression analysis to explore the mediating effects of parental health, family hardiness and family support
on the effects of the variables “age at diagnosis” and “family function” for parents of children with DMD.

The study was approved by the institutional review board of Kaohsiung Medical University, and participating parents with DMD children. A convenience sample (203 parents) was recruited from the Taiwan Muscular Dystrophy Association (TMDA) and from patient rolls at Kaohsiung Medical University Hospital. Members of the TMDA received a letter from the association inviting their participation in the study. Subjects who agreed to participate were mailed a packet consisting of two sets of questionnaires and informed consent forms. Follow-up telephone calls were made at 1 and 2 weeks after the questionnaires were mailed to promote the completion and return of the questionnaires by each parent. Subjects reported an average time of 40 minutes to complete the questions at home.

**Sample**

A “family” was defined as a child with DMD living with one or both biological parents. The parents, including eight single fathers, 26 single mothers, and 46 couples, returned completed questionnaires (62%). Their mean age was 43±6.1 years (range, 28–61 years). Most of the respondents were female (57%). Respondents’ religion was primarily Buddhist (50%) or Taoist (36%), and ethnicity was Taiwanese (76.2%), Hakka (11.9%), or Chinese (10.3%); over half had at least 12 years of education (57.7%) and were employed (56%); most were married (90.5%), and some worked as a laborer or farmer (26%). The families reported an annual income of less than US$10,000 (44%) to over US$30,000 (10%). Forty-eight (60%) families lived in urban areas.

The DMD children ranged in age from 3 to 25 years (mean, 14.3±4.6 years). Most were teenagers (41%) and needed wheelchair assistance (73%) or could not raise a hand to their mouth (46%). The mean age when diagnosed with DMD was 6.2±2.8 years (range, 1–15 years). The DMD children rated a score of 21–60 or 0–20 on the Barthel index, indicating severe dependency (47.5%) and complete dependency (35%), respectively. About 73% of the DMD children still received education at home.

**Data management and analysis**

Steps involved in analyzing the data included creating or transforming predictor and moderator variables (coding categorical variables, centering continuous variables), creating product terms, and structuring the equation. Mean-centered predictor variables (main effect variables before entering them in the model, which center a variable by subtracting the group’s mean from all observations) were used throughout the analysis to eliminate multicollinearity when variables and interaction terms entered the regression equation model. Hierarchical multiple regression analyses were conducted to test whether family hardiness, parental health, and family support mediated or moderated the association between age at diagnosis and family function.

An analysis resulted in an α of 0.05, and power of 0.86 was reached for the sample size of 126 subjects with a medium (0.15) effect on the regression [13]. There was high internal consistency and reliability using Cronbach’s α coefficients that ranged from 0.81 to 0.92 (Table 1) for the instruments in the present study. The construct validity of the FAD was appropriate [14]. FAD provided adequate evidence for the concurrent validity.

**Results**

**Relationships among predictor variables and dependent variables**

Based on the correlations, age at diagnosis of DMD showed a significant trend in its association with family

<table>
<thead>
<tr>
<th>Table 1. Normative data of the instruments in the current study</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Instruments</strong></td>
</tr>
<tr>
<td>-----------------</td>
</tr>
<tr>
<td>Barthel Index*</td>
</tr>
<tr>
<td>Duke Health Profile</td>
</tr>
<tr>
<td>Family Hardiness Index</td>
</tr>
<tr>
<td>Family APGAR</td>
</tr>
<tr>
<td>Family Function</td>
</tr>
</tbody>
</table>

*Total raw score ≤ 60 indicates severe dependency and total raw score > 60 indicates mild dependency. SD = standard deviation.
Mediation test

The most commonly used hierarchical multiple regression test for mediator effects involves three criteria for determining mediation, as shown in Figure 2 [15,16]. The first step in the process is to determine whether there is a significant association between a predictor and an outcome variable. A mediating relationship is one in which the path relating X to Y is mediated by a third variable (M). In this study, the correlation between age at diagnosis and family hardiness was −0.209, which was significant at *p* = 0.019. But the author hypothesized that this relationship was mediated by family hardiness, such that, if the child with DMD was younger at age of diagnosis, the family would have greater family hardiness, which would, in turn, lead to improved family function.

The author developed three equations (Figure 2) to test the hypothesis. The results are shown in Figure 3. First, testing showed that, for the mediation hypothesis to be true, the regression for each step had to be significant. Family function was regressed on age at diagnosis and family hardiness. Second, family hardiness was regressed on age at diagnosis to establish the mediator chain. In the third equation, family function was regressed on age at diagnosis and family hardiness. This provided a test of whether family hardiness was related to family functioning (β) as well as an estimate of the association between age at diagnosis and family function, controlling for family hardiness (τ) [16,17]. Results indicated that age at diagnosis was the best predictor for family function (β = 0.202, *p* = 0.023, Equation 1: Family function = 1.975 + 0.202 × age at diagnosis + 0.009) (Figure 3A).

Adding family hardiness to the equation did not increase the explained variance (R² = 0.547, F(2, 123) = 76.576, *p* = 0.00). The mediator function of family hardiness was substantiated first by the reduction of the path coefficient from 0.202 to 0.049 (Table 2), but age at diagnosis did not significantly decrease in the equation [β = 0.049, *p* = 0.425 (Figure 3A)]. Equation 2: Family function = 3.192 + 0.049 × age at diagnosis − 0.733 × family hardiness + 0.110; Equation 3: Family hardiness = 44.82 − 0.209 × age at diagnosis + 1.651) (Figure 3A). Third, the interaction between age at diagnosis and family hardiness (R² = 0.549, F(3, 122) = 51.670, *p* = 0.00; F change = 1.382, *p* = 0.242) was entered to assess its significance (Table 2).

A mediator model is supported if the model with a direct path between age at diagnosis and family functioning does not provide better fit to the data. With exclusion of age at diagnosis in the third step, the explained variance in family function significantly decreased for age at diagnosis (β = 0.066, *p* = 0.242) (Table 2). Keeping in mind that lower family function scores reflect better function, diagnosis at a later age was associated with lower family function. In addition, given the inverse scoring of FAD for family function (with lower scores indicating better function), these findings indicate that, as family hardiness scores increased, family function also increased. The causal model did not engage moderation because the path from the interaction of age at diagnosis and family hardiness to family functioning was not significant [18].

![Figure 2. Three-variable mediation model.](image-url)
(A) Mediator model

<table>
<thead>
<tr>
<th>Independent variable</th>
<th>Dependent variable</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at diagnosis</td>
<td>Family function</td>
</tr>
<tr>
<td>$\beta = 0.202, p = 0.023$</td>
<td></td>
</tr>
</tbody>
</table>

$\beta = -0.209, p = 0.019$

$\beta = -0.733, p = 0.000$

Step 2: Age at diagnosis

$\beta = 0.049, p = 0.425$

(B) Non-mediator model

<table>
<thead>
<tr>
<th>Parental health</th>
<th>Family function</th>
</tr>
</thead>
<tbody>
<tr>
<td>$\beta = 0.014, p = 0.879$</td>
<td></td>
</tr>
</tbody>
</table>

$\beta = -0.601, p = 0.000$

Step 2: Age at diagnosis

$\beta = 0.21, p = 0.003$

(C) Non-mediator model

<table>
<thead>
<tr>
<th>Family support</th>
<th>Family function</th>
</tr>
</thead>
<tbody>
<tr>
<td>$\beta = -0.007, p = 0.941$</td>
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</tbody>
</table>

$\beta = -0.659, p = 0.000$ 

Step 2: Age at diagnosis

$\beta = 0.198, p = 0.003$

**Figure 3.** Mediator model: age at diagnosis influences family function through family hardiness.

**Table 2.** Family function regressed on child’s age at diagnosis, family hardiness, parental health, and family support

<table>
<thead>
<tr>
<th></th>
<th>Step 1</th>
<th>Step 2</th>
<th>Step 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at diagnosis</td>
<td>0.202*</td>
<td>0.049</td>
<td>0.066</td>
</tr>
<tr>
<td>Family hardiness</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>$\beta = -0.733^t$</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>$\beta = -0.884^t$</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age at diagnosis $\times$ family hardiness</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>$R^2$</td>
<td>0.041</td>
<td>0.547</td>
<td>0.549</td>
</tr>
<tr>
<td>$F$</td>
<td>5.286*</td>
<td>76.576†</td>
<td>51.670†</td>
</tr>
<tr>
<td>$F$ change</td>
<td>5.286*</td>
<td>141.862†</td>
<td>1.382</td>
</tr>
<tr>
<td>Age at diagnosis $\times$ parental health</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>$R^2$</td>
<td>0.041</td>
<td>0.402</td>
<td>0.416</td>
</tr>
<tr>
<td>$F$</td>
<td>5.286*</td>
<td>41.368†</td>
<td>28.996†</td>
</tr>
<tr>
<td>$F$ change</td>
<td>5.286*</td>
<td>74.326†</td>
<td>2.945</td>
</tr>
<tr>
<td>Age at diagnosis $\times$ family support</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>$R^2$</td>
<td>0.041</td>
<td>0.475</td>
<td>0.483</td>
</tr>
<tr>
<td>$F$</td>
<td>5.286*</td>
<td>55.578†</td>
<td>37.962†</td>
</tr>
<tr>
<td>$F$ change</td>
<td>5.286*</td>
<td>101.584†</td>
<td>1.909</td>
</tr>
</tbody>
</table>

* $p < 0.05$; † $p < 0.01$. 
We found that age at diagnosis had an indirect effect on family function through the mediator family hardiness. The size of the indirect effect was the product of the age at diagnosis and family hardiness, and family hardiness and family function effects, that is \(-0.209 \times -0.733 \) (0.153). The direct effect was 0.049 and the total effect of age at diagnosis on family function was the sum of the indirect effect and the direct effect, which was 0.202 (Figure 3A). The strength of the mediation effect was estimated using the ratio of the indirect effect to the total effect, which equaled 75.5% for the proportion of mediated effect. The true indirect effect was estimated by the Sobel Test \( z \text{ value} = a \times b / \text{SQRT}(b^2 \times s_a^2 + a^2 \times s_b^2), \ z = 2.278, \ p = 0.022 \). This CI\(_{95} \) [0.023, 0.283] did not contain 0, so the null hypothesis of no indirect effect of family hardiness was rejected at \( \alpha = 0.05 \) [19].

**Moderation of parental health, family support and family function by age at diagnosis**

Moderator analyses were performed to examine the interaction effects between family resources and age at diagnosis on family function, after controlling the path from age at diagnosis to family hardiness, parental health or family support (Table 2) [18]. Solving the model to determine a moderating relationship formed a new variable, which was the product of the two predictors. For example, predicting family function (Y) from age at diagnosis (X) and family hardiness (M), created a variable \((XM) = X \times M\), then \(Y = b1X + b2M + b3X \times M + b0\). If the XM term was significant, there was a moderating relationship. Age at diagnosis was revealed as a mediator function. We tested the moderator effect from the interaction of age at diagnosis and family hardiness to family function (\(R^2 = 0.549, F(3, 122) = 51.670, p = 0.00\)), assessing their significance. After excluding age at diagnosis in the third step, the explained variance in family function significantly decreased with age at diagnosis (\(\beta = 0.066, p = 0.242\), SE = 0.006). The causal model did not engage moderation because the path from the interaction of age at diagnosis and family hardiness to family function was not significant (F change = 1.382, \(p = 0.242\)) (Table 2).

Age at diagnosis revealed that the non-mediator function and non-moderator function of parental health was substantiated first by increasing the path coefficient from 0.202 to 0.210. The second change was also significant in the path from age at diagnosis and parental health to family function. In the second step, age at diagnosis and parental health were also entered into the equation and contributed to explaining the variance in the measurement of family functioning (\(R^2 = 0.402, F(2, 123) = 41.368, p = 0.00\)) (Table 2). Age at diagnosis showed a small but significant addition in the equation (\(\beta = 0.21, p = 0.003\)), and the path from age at diagnosis to parental health was not significant (\(\beta = 0.014, p = 0.879\)) (Figure 3B). After statistically controlling the path from age at diagnosis to parental health and the path from parental health to family function, the previous relationship between age at diagnosis and family function became more significant (\(\beta = 0.21, p = 0.003\)) (Figure 3B). In the third step, the interaction between age at diagnosis and parental health was entered to assess its interaction significance (\(R^2 = 0.416, F(3, 122) = 28.996, p = 0.00, F\text{ change} = 2.945, p = 0.089\)) (Table 2). When excluding age at diagnosis in this third step, the explained variance in family function for age at diagnosis did not significantly decrease (\(\beta = -0.275, p = 0.347\)) (Table 2). When the regression coefficient for the interaction term of the age at diagnosis and parental health was entered into the third step of the hierarchical regression, there was no significant contribution to explaining the variance of family functioning (\(R^2\text{ change} = 0.014\)). The path from the interaction of parental health and age at diagnosis to family functioning was non-significant (Table 2).

Age at diagnosis revealed that the non-mediator function and non-moderator function of family support was substantiated first by decreasing the path coefficient from 0.202 to 0.198. The second change was also significant in the path from age at diagnosis and family support to family function. Age at diagnosis and family support were also entered into the equation and contributed to explaining the variance in the measurement of family function (\(R^2 = 0.475, F(2, 123) = 55.578, p = 0.00\)) (Table 2). Figure 3C shows that age at diagnosis was significantly suppressed in the equation (\(\beta = 0.198, p = 0.003\)), but the path from age at diagnosis to family support was not significant (\(\beta = -0.007, p = 0.941\)). After statistically controlling the path from age at diagnosis to family support and the path from family support to family function, the previous association between age at diagnosis and family function became more significant (\(\beta = 0.198, p = 0.003\)). In the third step, the interaction between age at diagnosis and family support was entered to assess their significance (\(R^2 = 0.483, F(3, 122) = 37.962, p = 0.00\)).
The finding that the children’s level of disability was not associated with family function or with any of the other predictor variables was surprising. The main supposition to explain this is that the parents participated in support groups that helped them adapt to the progressive course of the disease. These findings raise several important issues for future research. With respect to family resources, parents in this study reported poorer health status and more anxiety, depression, pain, and disability than members of the general population [21], a finding consistent with previous research on families of children with serious or disabling conditions. Tamplin and Goodyer [22] reported that mental health was highly correlated with all FAD scales for mothers of high-risk children, with only one or two scales for low- and high-risk fathers, respectively. Studies have revealed evidence that depressed adolescents were significantly more likely to be in families that were reported as dysfunctional on the FAD [22,23].

Mean family hardiness scores in this study were also lower than those for parents of children with asthma, cardiac conditions, or diabetes, as described in previous studies [7]. The reasons for these differences between parents of children with DMD and other conditions should be explored in future research. The positive association between hardiness and healthy family functioning indicates that parents of children with DMD who had greater hardiness to endure stressors also had greater health. In addition, from a health promotion perspective, the findings support the need to develop family hardiness through family support services that could be incorporated into health promotion programs in the long term [24].

This study was limited due to the small convenience sample and self-report measures. In using self-reports, the parents could have over-reported hardiness, support and strength, and under-reported their health condition. Addressing only parents in the TMDA, the present study produced findings that may be specific to one association and not generalizable to others. Furthermore, the cross-sectional design of this study limits the exploration of causal relationships among the predictors/mediators and outcome variables. A longitudinal study with a multilevel modeling technique design would allow further investigation of the multiple factors that influence the causal effects. Finally, this study did not account for economic differences between family hardiness and family function. Thus,
future studies might demonstrate other factors that mediate or moderate the associations between predictor and outcome variables. Owing to the dynamic nature of the phenomena examined in this study, qualitative interviews and other more in-depth, socially grounded research should be conducted in this area in the future.

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The author would like to thank Drs. Susan Instone, Allen Orsi, and Diane Hatton for their contributions to the completion of this research.

REFERENCES

中介因子影響杜顯型肌失養症兒童家庭之
家庭功能

陳季員
高雄醫學大學 護理學系

杜顯型肌失養症兒童在第一次被診斷時，大多數家庭才開始接受有關疾病的專業信
息。通常確立兒童診斷後父母將從專業人員收集和鑽研疾病的資訊且開始接受支持性
的照護系統。然而因他們不知如何問正確的問題，獲得可近性的照護是項嚴重的挑
戰。尤其此弱勢群體面對障礙過程須支持他們孩子的情緒調適和使用有效的服務系
統。本研究目的，是測試杜顯型肌失養症兒童之父母其健康、家庭耐力、和家庭支持
之變項的介入對預測變項“兒童障礙”和“診斷時的年齡”在正向家庭功能之影響。
加入台灣肌肉萎縮協會之 126 位杜顯型肌失養症兒童之父母填寫問卷，每位對象在問
卷寄出前及寄後一星期內會接到電話聯繫、並給予回郵信封和簽署同意書。問卷內容
包含家庭耐力量表、家庭評估指數、家庭支持量表、杜克健康量表和人口學資料。多
層模式及多層階序迴歸分析用於檢測 家庭耐力、父母健康、和家庭支持之變項介入或
調整“診斷時的年齡”及“家庭功能”之間的關係。家庭耐力介入功能在診斷時的年
齡和家庭功能關係間呈現正向增加的影響。研究發現有助於設計介入措施目標增強發
展杜顯型肌失養症兒童之家庭的優勢。

關鍵詞：杜顯型肌失養症，家庭功能，家庭耐力，家庭支持，父母健康
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通訊作者：陳季員醫師
高雄醫學大學護理學系
高雄市三民區十全一路 100 號