Multiple Hereditary Exostosis and Missed School Days

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INTRODUCTION:

Multiple Hereditary Exostosis (MHE) is a rare autosomal dominant disorder characterized by many benign bone tumors called osteochondromas or exostoses. There is a paucity in the current literature on how pediatric patients with MHE are impacted. This study aimed to evaluate the impact of pain, psychosocial factors and surgical intervention on missed school days in those with MHE. We hypothesized that patients with MHE will have higher generalized pain scores, and there will also be an association between generalized pain scores and number of missed school days.

METHODS:

Patients diagnosed with MHE ≤18 years old were prospectively enrolled and asked to complete surveys related to medical history, family history, generalized pain and missed school days, as well as PROMIS surveys on mobility, pain interference, physical activity, peer relationships and Neuro-QOL surveys on stigma, anxiety, and depression. Aside from the PROMIS Mobility survey, a lower score on all PROMIS/Neuro-QOL surveys indicated a more positive outcome. For patients < 8 years old, parents completed the surveys. Demographic and surgical data was collected from patients' EMR.

One-sample t-tests were used to compare patients' t-scores to normative survey scores. Negative binomial regression models were used to examine the association between the number of missed school days and survey scores. Spearman's correlation assessed the relationship between PROMIS scores and surgery count, as well as the associations between patient age, missed school days, and survey scores.

RESULTS:

Thirty patients (mean age 11.2 ±3.8 years; 15 females and 15 males) with MHE were included in the final analysis (Table 1). Patients' mean PROMIS/Neuro-QOL scores did not significantly differ from the normative mean (Table 2).

Patients reported missing a mean of 7.2 days (range 0-50 days) per 10-month school year because of pain. Missed school days were significantly correlated with patients' pain interference (r=0.41, p=0.041) and depression (r=0.45, p=0.026). The correlation between patients' missed school days and perceived stigma was trending towards significance (r=0.39, p=0.057). Stigma was, however, a significant predictor of missed school days (p = 0.049), with each standard deviation increase from the normative mean associated with a 15.5% increase in missed school days.

Patient age was positively correlated with number of missed school days (r=0.41, p=0.0391) and greater pain interference (r=0.38, p= 0.0488). Number of surgeries was significantly correlated with decreased mobility (r=-0.39, p=0.043) and increased pain interference (r=0.51, p=0.007). The relationship between number of surgeries and stigma was trending towards significance (r=0.36, p=0.065).

DISCUSSION AND CONCLUSION:

Perceived pain interference, depression, and social stigma among pediatric MHE patients impacts number of missed school days. Additionally, adolescent/teenage patients tend to experience greater pain interference and miss more school. By understanding these challenges, providers can better support and plan treatment for pediatric MHE patients and promote quality of life.

Table 1: Demographics

Variable	N=30
Age, mean (SD)	11.2 (3.8)
Sex, N (%)	
Female	15 (50.0%)
Male	15 (50.0%)
Race/ethnicity	
Hispanic or Latino/a/e	19 (63.3%)
Non-Hispanic or Latino/a/e White	8 (26.6%)
Non-Hispanic or Latino/a/e Black	1 (3.3%)
Middle Eastern or North African	1 (3.3%)
Other	1 (3.3%)
Preferred Language	
English	24 (80.0%)
Spanish	6 (20.0%)
Insurance status	
Medi-Cal	14 (46.7%)
PPO	11 (36.7%)
HMO (Commercial)	5 (16.7%)

Continuous variables are presented as mean (standard deviation)

and categorical variables are presented as N (%)

Table 2: Mean PROMIS/Neuro-QOL Scores compared to normative mean

Survey Basis	Mean T-Score (SD)	Range	p-value
PROMIS Pain Interference	43.25 (8.61)	34.0 – 59.2	0.9994
PROMIS Mobility	48.09 (8.11)	31.6 – 58.5	0.1087
PROMIS Physical Activity	47.37 (8.78)	28.8 – 66.7	0.1068
Neuro-QOL Stigma	44.94 (6.78)	37.1 – 58.4	0.9990
Neuro-QOL Anxiety	44.55 (6.64)	37.5 – 58.1	0.9990
Neuro-QOL Depression	42.70 (6.66)	36.4 – 61.0	0.9999
PROMIS Peer Relationships	50.9 (10.71)	32.3 – 64.4	0.5880

Normative Mean T-Score=50