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Psychological Factors and Outcomes in the Surgical Treatment of Pediatric Patients With Median Arcuate Ligament Syndrome

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ABSTRACT

Objective: Median arcuate ligament syndrome (MALS) is a frequently overlooked cause of chronic abdominal pain (CAP), and results in many symptoms that mimic other gastrointestinal conditions that result in CAP. A small, but growing body of literature indicates that surgery improves quality of life (QOL) in patients with MALS. The purpose of the current study was to examine the psychological characteristics of pediatric patients with MALS to determine their prevalence and impact on surgical outcomes.

Methods: Thirty-two pediatric patients completed psychological assessments before surgery, and 6 months postsurgical intervention. Descriptive analyses and *t* tests were conducted to characterize the sample and compare psychosocial and QOL items. To explore possible associations between coping and ultimate changes in QOL, exploratory multiple regressions were conducted.

Results: Comorbid psychological conditions were common, occurring in about half the sample before and after surgery. Current pain significantly improved, as well as patient and parent-reported QOL constructs ($P_s < 0.05$). Parent-reported observations of patients using catastrophizing or helpless strategies to cope with pain before surgery was significantly associated with changes in patient and parent-reported QOL following surgery ($P_s = 0.04$).

Conclusions: Comorbid psychological conditions are common in pediatric patients with MALS, and are maintained following surgery. While surgery improved pain and QOL, the need for presurgical psychological interventions for MALS is implicated.

Key Words: chronic abdominal pain, coping, median arcuate ligament syndrome, pediatric, quality of life, surgical outcome

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Chronic abdominal pain (CAP) is common in pediatric populations and is associated with multiple negative and long-term psychological sequelae (1–3). Although there are many

What Is Known

- Median arcuate ligament syndrome is a frequently overlooked cause of chronic abdominal pain and is treated surgically.
- Pediatric median arcuate ligament syndrome surgery improves quality of life.

What Is New

- Comorbid psychological conditions are common in pediatric patients with median arcuate ligament syndrome and there is no evidence to support that surgical intervention impacts comorbid diagnoses or symptoms.
- Pediatric patient catastrophizing may serve as an intervention target to improve pre- and postsurgical quality of life outcomes.

possible etiologies for pediatric CAP, one rarely diagnosed cause is median arcuate ligament syndrome (MALS). MALS is a vascular compression syndrome resulting from compression of the celiac artery by diaphragmatic crus fibers (4). This compression may result in many symptoms that mimic other gastrointestinal (GI) conditions that result in CAP, including chronic GI syndromes, inflammatory bowel disease (IBD), and functional GI disorders (FGIDs) (5–7).

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Possibly due to the clinical overlap of symptoms and similarity of psychosocial profiles with other conditions causing CAP (6), MALS is a diagnosis that is frequently overlooked. Similar to FGIDs, MALS tends to occur in young females and produces symptoms of post-prandial, mid-epigastric pain, and other constitutional symptoms, such as weight loss, nausea, and vomiting (5–7). MALS is, however, considered to be a diagnosis of exclusion. Before exploring MALS as a diagnosis, a complete series of studies is conducted to rule out common causes of abdominal pain. Initial workup for a diagnosis of MALS begins with a noninvasive mesenteric duplex ultrasound; findings consist of elevated velocities of the celiac artery with normalization of the velocities during deep inspiration, suggesting celiac artery compression. The MALS diagnosis is confirmed with ultrasound findings, either through magnetic resonance angiography, computed tomography (CT) angiography, or standard angiography. Images consistent with MALS demonstrate a “J-hook” or web-like conformation at the origin that normalize during inspiration (5).

Decades after it was first described (4), MALS remains a controversial and vexing condition. In adult populations, 13% to 50% of healthy patients may exhibit radiographic features of celiac artery compression, yet a much smaller percentage of patients report symptoms consistent with MALS (8). The link between symptoms and the narrowing of the celiac axis, celiac nerve plexus, or both remains unknown. Our approach to patients with MALS is based on the assumption that compression of either the celiac artery or surrounding nerves result in pain. It is, however, carefully explained to the patient and his/her family that there is controversy surrounding the diagnosis, as the pathophysiologic mechanism of the pain is unclear. Furthermore, due to the unclear mechanisms, surgery as a treatment option is approached with great care and caution.

All patients with MALS exploring surgery as a treatment option undergo separate evaluations by our interdisciplinary team, including the following services: general pediatric surgery, vascular surgery, psychiatry, and pain. Input from each member of the team is critical, not only to the decision to proceed with surgery but also for the preoperative preparation, immediate postoperative care, and long-term follow-up. For example, if a pediatric patient presents with severe depression, psychiatric services may be required from the team before scheduling surgery. Also of critical importance is the appropriate management of patient and family expectations. There should be complete candor that while surgical release has been reported to be successful in improving abdominal pain in 70% to 80% patients, that there is a possibility of persistent postoperative pain (7).

Purpose

A growing number of studies have indicated that surgical intervention improves quality of life (QOL) in patients with MALS (5,6,9). Samples have, however, been small and more research is required to understand this population’s needs, and improve outcomes for pediatric patients with MALS. Therefore, the purpose of the current research was to characterize MALS in pediatric patients by prospectively examining patients’ psychological characteristics before and following surgical intervention for MALS. Given the high prevalence of presurgical comorbid psychological conditions in pediatric patients with MALS (6) and the recommended practice to screen and treat comorbid mood symptoms in the presence of pain (10), we hypothesized that surgery would not significantly affect psychological symptoms. Secondly, given previous results indicating improvements

in QOL following surgery for MALS (5,9), we hypothesized that QOL would improve following surgical intervention from both the patient and parent perspective. Finally, presurgical coping strategies were explored to identify possible targets for improving postoperative QOL in pediatric patients with MALS.

METHODS

Participants

Participants were eligible for inclusion in this study if they completed a pre- and postsurgical psychological assessment, completed other presurgical assessments and tests as part of the standardized presurgical MALS protocol (eg, duplex ultrasound, GI workup, etc) (5), underwent surgical release of the celiac artery for MALS, and were under 18 years of age throughout the assessment period.

Procedure

In compliance with the University of Chicago Institutional Review Board, informed patient assent and parental consent were obtained before the presurgical psychological assessment. The presurgical assessment occurred in conjunction with other medical and pain assessments associated with the presurgical protocol (5). The postsurgical assessments occurred at 6 months following surgery. The pre- and postsurgical psychological assessments both included a semistructured interview developed by the University of Chicago Median Arcuate Ligament Syndrome (UCMALS) team to assess current functioning, a structured psychological interview, as well as patient and parent questionnaires.

Measures

Demographic

Participants were asked to report demographic characteristics, including gender, race/ethnicity, age, and others, in the semistructured pre- and postsurgical interview developed by the MALS team (11,12).

Psychological

The Mini International Neuropsychiatric Interview for Children and Adolescents (MINI-Kid) is a brief, structured diagnostic interview for children and adolescents (13). Diagnoses are based on the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV)* and *International Statistical Classification of Diseases and Related Health Problems, Tenth Revision (ICD-10)* criteria (14,15).

Psychosocial Pediatric Patient Self-report

Pediatric patients were asked to report their current and highest experience of pain on a 0 to 10 scale before and following surgery, with higher ratings indicating greater experiences of pain.

The Child Depression Inventory (CDI) is a self-report scale measuring depressive symptoms in youth (16). The Multidimensional Anxiety Scale for Children (MASC) is a self-report scale measuring symptoms of anxiety in youth (17). Higher scores indicate greater symptomatology for both measures.

The Eating Disorder Examination-Questionnaire (EDE-Q) is a self-report scale of key eating disorder behaviors and cognitions (18). Higher scores indicate greater disordered eating symptoms. Given the frequent symptom of significant weight loss in pediatric

MALS, the EDE-Q is included in the assessment battery to provide a self-report assessment of possible symptoms of an eating disorder.

The Pediatric Quality of Life Inventory (PedsQL) measures health-related QOL; assessing physical, emotional, social, and school functioning (19). The Pediatric Pain Coping Inventory (PPCI), a questionnaire in the PedsQL series, measures the use of pediatric pain coping strategies, with subscales specifically querying: cognitive self-instruction, problem solving, distraction, seeking social support, and catastrophizing or helplessness (20). Higher scores on both measures indicate higher QOL and use of coping, respectively.

Psychosocial Parent Report

Parents were asked to complete the parent report versions of the EDE-Q, PedsQL, and PPCI (ie, the Parent Eating Disorder Examination-Questionnaire [PEDE-Q] (21), the Parent-Report of the PedsQL (19), and the Parent-Report of the PPCI (20)). Subscales and scoring on the measures are identical to the pediatric self-report versions (see above).

Medical Evaluation

A complete GI work up was performed, as previously described (5). All patients had a duplex ultrasound in the University of Chicago Medicine's accredited vascular laboratory to fully evaluate the visceral vessels. Confirmatory studies, including CT angiogram, magnetic resonance angiography, or conventional angiogram, were used to corroborate duplex findings (5).

Surgery

Surgical release of the celiac artery compression was performed via a laparoscopic approach. Guiding principles and goals of the procedure include division of the median arcuate ligament, including overlying lymphatics and soft tissue releasing the celiac artery with division of the celiac nerve plexus. Intra-operative duplex was used to verify adequate release of the celiac artery (5). Follow-up duplex was performed at 6 months.

Data Analysis

Descriptive analyses were conducted to identify demographic characteristics, pre- and postsurgery. The CDI and MASC were examined using standardized *T* scores; the EDE-Q, PEDE-Q, PedsQL, and PPCI were examined using total scores. *t* Tests were conducted to compare pre- and postsurgical psychosocial and QOL

characteristics. Exploratory multiple regressions with forward entry were used to examine possible relationships between presurgical coping strategies and changes in QOL following surgery.

RESULTS

Participants

Participants were 32 pediatric patients (10–17 years of age) diagnosed with MALS, who underwent surgical release of the celiac artery at the University of Chicago Medicine between 2011 and 2017. Table 1 displays the demographic and surgical characteristics of the sample. The sample was primarily female (93.8%) and non-Hispanic Caucasian (75.0%). About 19% of the sample (*n* = 6) also had a comorbid GI condition before surgery (ie, Crohn disease, gastroesophageal reflux disease, irritable bowel syndrome). Laparoscopic release of the median arcuate ligament was performed successfully in all 32 cases. Table 1 demonstrates the demographics of the cohort and shows that release of the ligament resulted in overall improvement in celiac artery blood flow, including, the celiac artery peak systolic velocity (PSV), the celiac to aortic PSV ratio, and the variation of PSV with inspiration (*P*s < 0.0001).

Representativeness of Study Patients With Median Arcuate Ligament Syndrome

To ensure the sample was representative of all pediatric patients presenting for surgical intervention for MALS at the university, all baseline characteristics included in the current study were compared between the study sample and 36 pediatric patients with MALS assessed and treated at the University of Chicago Medicine who did not complete their postsurgical follow-up assessments. There was no evidence to suggest significant differences in baseline variables between those included and not included in the present study (*P*s > 0.06).

Psychosocial Characteristics Following Surgery

Over half of the sample (*n* = 18) met criteria for at least 1 comorbid psychological condition before surgery. These conditions included mood disorders (ie, major depressive disorder), anxiety disorders (ie, panic disorder, agoraphobia, specific and social phobia, posttraumatic stress disorder, and agoraphobia), adjustment disorder, learning disorders and attention deficit/hyperactivity disorder, and rumination. About half of the sample (*n* = 17) continued to meet criteria for a comorbid psychological condition following surgery, with no evidence to suggest a significant difference in the

TABLE 1. Demographic and surgical characteristics pre- and postsurgical release

| Characteristic | Pre | Post | <i>P</i> |
|---|-----------------|---------------|----------|
| Age, <i>M</i> (SD) | 14.8 (1.8) | 15.6 (1.7) | — |
| Female, <i>n</i> (%) | 30 (93.8) | — | — |
| Non-Hispanic Caucasian, <i>n</i> (%) | 24 (75.0) | — | — |
| Intact family status, <i>n</i> (%) | 26 (81.3) | — | — |
| Comorbid GI condition, <i>n</i> (%) | 6 (18.8) | — | — |
| Psychological condition, <i>n</i> (%) | 18 (56.3) | 17 (48.6) | 0.8 |
| Peak systolic velocity, <i>M</i> (SD) | 410 cm/s (108)* | 266 cm/s (97) | <0.0001 |
| Celiac/aortic PSV ratio, <i>M</i> (SD) | 2.3 (0.7) | 1.5 (0.5) | <0.0001 |
| Change in PSV with Inspiration, <i>M</i> (SD) | 151 cm/s (116) | 59 cm/s (77) | <0.0001 |

GI = gastrointestinal; *M* = mean; PSV = peak systolic velocity; SD = standard Deviation.

*Includes 4 patients with occlusions at rest and flow upon deep inspiration. For these values, the PSV was taken at deep inspiration.

TABLE 2. Psychosocial characteristics pre- and postsurgical release, M (SD)

| Measure | Pre | Post | <i>P</i> |
|-----------------------------------|-------------|-------------|----------|
| Pediatric patient | | | |
| Current pain | 4.1 (2.6) | 2.7 (2.8) | 0.049 |
| Highest pain | 8.0 (2.7) | 7.0 (3.0) | 0.2 |
| CDI total <i>T</i> score | 45.9 (9.8) | 42.3 (7.4) | 0.1 |
| EDE-Q total score | 0.2 (0.3) | 0.3 (0.7) | 0.4 |
| MASC total <i>T</i> score | 47.5 (12.3) | 45.8 (13.6) | 0.6 |
| PPCI cognitive self-instruction | 0.9 (0.4) | 1.1 (0.4) | 0.1 |
| PPCI problem solving | 0.9 (0.4) | 1.0 (0.3) | 0.2 |
| PPCI distraction | 0.8 (0.3) | 1.0 (0.3) | 0.08 |
| PPCI seeks social support | 0.8 (0.4) | 0.9 (0.4) | 0.5 |
| PPCI catastrophizing/helplessness | 1.0 (0.4) | 0.9 (0.4) | 0.3 |
| PPCI total | 0.9 (0.3) | 1.0 (0.2) | 0.2 |
| PedsQL physical | 56.6 (21.3) | 72.3 (21.5) | 0.006 |
| PedsQL emotion | 64.2 (21.4) | 73.5 (21.3) | 0.09 |
| PedsQL social | 82.4 (17.8) | 84.7 (15.5) | 0.6 |
| PedsQL school | 56.8 (21.5) | 71.4 (22.9) | 0.01 |
| PedsQL psychosocial | 67.4 (17.8) | 75.6 (18.2) | 0.08 |
| PedsQL total | 63.5 (16.8) | 74.4 (18.1) | 0.02 |
| Parent | | | |
| PEDE-Q total score | 0.2 (0.4) | 0.2 (0.4) | 0.9 |
| PPCI cognitive self-instruction | 0.6 (0.4) | 0.8 (0.4) | 0.07 |
| PPCI problem solving | 1.0 (0.3) | 1.0 (0.3) | 0.9 |
| PPCI distraction | 0.8 (0.3) | 0.7 (0.3) | 0.6 |
| PPCI seeks social support | 1.0 (0.4) | 0.9 (0.4) | 0.6 |
| PPCI catastrophizing/helplessness | 1.1 (0.4) | 0.9 (0.4) | 0.05 |
| PPCI total | 0.9 (0.3) | 0.9 (0.3) | 0.5 |
| PedsQL physical | 54.8 (26.9) | 77.2 (18.7) | <0.001 |
| PedsQL emotion | 62.7 (22.9) | 75.2 (17.9) | 0.02 |
| PedsQL social | 81.7 (18.4) | 88.3 (15.0) | 0.1 |
| PedsQL school | 62.0 (26.5) | 63.0 (28.7) | 0.2 |
| PedsQL psychosocial | 59.5 (25.1) | 69.0 (25.3) | 0.04 |
| PedsQL total | 68.4 (19.3) | 78.2 (17.0) | 0.004 |

Current and highest pain ratings made on a 0–10 scale.

CDI = Child Depression Inventory; EDE-Q = Eating Disorder Examination-Questionnaire; MASC = Multidimensional Anxiety Scale for Children; PEDE-Q = Parent Eating Disorder Examination-Questionnaire; PedsQL = Pediatric Quality of Life; PPCI = Pediatric Pain Coping Inventory.

prevalence or types of diagnoses following surgical intervention (*P* = 0.8; see Table 1).

Table 2 displays psychosocial characteristics pre- and postsurgery. Patient-reported current pain levels (0–10 scale)

significantly decreased following surgery (*P* = 0.049). There was no evidence to suggest significant changes in the highest experience of pain following surgery (*P* = 0.2). As hypothesized, there was no evidence to suggest significant differences in psychological symptoms, as measured by the CDI (depression), EDE-Q (eating disorder behaviors or cognitions), MASC (anxiety), or PPCI (coping) following surgery (*P*s > 0.1). There was also no evidence to suggest significant changes in the parent-report of these characteristics following surgery (ie, P-EDEQ, PPCI; *P*s > 0.07).

Changes in Quality of Life Following Surgery

Consistent with the hypothesis, significant improvements in QOL were reported in terms of patient-reported physical (*P* = 0.006), school (*P* = 0.01), and overall (*P* = 0.02) functioning. There was no evidence to suggest significant improvements in patient-reported emotional, social, or psychosocial QOL following surgery (*P*s > 0.06). Parent-reported QOL showed significant improvement in physical (*P* < 0.001), emotional (*P* = 0.02), psychosocial (*P* = 0.04), and overall (*P* = 0.004) functioning on the PedsQL. For all other measures of QOL from the parent-perspective, there was no evidence to support significant change following surgery.

Coping Strategies and Changes in Quality of Life

Patient-reported changes in overall QOL ranged from –21 to +56 points on the PedsQL (mean = 9.82 ± 18.81); parent-reported changes in overall QOL ranged from –23 to +42 points (mean = 11.31 ± 18.79). Forward regression models indicated that the parent-report of the use of catastrophizing/helplessness by patients as a pain coping strategy was significantly associated with changes in patient (*P* = 0.04) and parent-reported (*P* = 0.04) QOL following surgery (Table 3). Parental report of this type of coping uniquely accounted for 40% of the variance in changes in patient- and parent-reported changes in QOL. There was no evidence to suggest significant associations with any other domains of pain coping.

DISCUSSION

More than half of the pediatric patients with MALS undergoing surgical release of the celiac artery initially presented with a comorbid psychological condition. Consistent with our hypotheses, self-reported psychosocial symptoms were not significantly impacted by surgery, with the exception that the patients' current

TABLE 3. Presurgical use of pain coping to predict changes in quality of life

| Variable | Pediatric patient-reported post-operative change in QOL | | | | |
|---|---|--------|------|----------|-----------------------------|
| | B | SE (B) | β | <i>t</i> | r _p ² |
| PPCI catastrophizing/helplessness subscale, parent report | 18.29 | 8.29 | 0.40 | 2.21* | 0.40 |
| Variable | Parent-reported post-operative change in QOL | | | | |
| | B | SE (B) | β | <i>t</i> | r _p ² |
| PPCI catastrophizing/helplessness subscale, parent report | 18.24 | 8.31 | 0.40 | 2.20* | 0.40 |

Variables excluded from the model include parent and pediatric patient-reported presurgical use of problem solving, distraction, seeking social support; and patient-reported use of catastrophizing/helplessness for coping. PPCI = Pediatric Pain Coping Inventory; QOL = quality of Life.

**P* ≤ 0.04.

experience of pain significantly improved following surgery. Patient- and parent-reported QOL significantly improved following surgery. Exploratory analyses indicated that parent observations of pediatric patients' use of catastrophizing as a coping strategy was significantly associated with changes in QOL over time.

Pediatric patients and their families seeking surgical release of the celiac artery tend to present with a particular focus on relieving the physical symptoms of MALS, particularly post-prandial pain. Comorbid psychological conditions were, however, common in the sample. Additionally, psychological symptoms were not impacted by surgical intervention. This is with the noted exception that surgery significantly improved subjective pain; however, we are hesitant to over interpret the findings of 1 self-report pain measure. These findings therefore suggest that, regardless of the etiology of these comorbid psychological symptoms in pediatric patients with MALS, such symptoms do not necessarily resolve with surgery and/or the subsequent reduction of pain.

Consistent with previous findings (5,6,9), patient and parent-reported QOL improved overall following surgery. Parental reports of the pediatric patients' use of specific coping strategies were related to changes in QOL. Items that comprise this subscale include pain responses such as: crying, anticipating the pain to worsen, wishing the pain away, irritability, thinking there is nothing to do to stop the pain, and so on (20). These analyses were exploratory and conducted with a small sample, and we are therefore hesitant to over interpret the findings. This association may, however, indicate that maladaptive coping strategies may be an important factor related to pre- and postsurgical QOL in pediatric patients with MALS. This association is also in line with previous findings, in that catastrophizing is a common coping style in pediatric patients with CAP (22) and targeting catastrophizing thoughts in pediatric patients mediated symptom reduction in an intervention targeting functional abdominal pain (23). Maladaptive pediatric pain coping, such as catastrophizing, may be an important target for pediatric patients with MALS.

Although we are hesitant to over interpret the possible relationship between poor pain coping strategies with QOL, it is likely that presurgical psychological interventions focusing on positive coping would benefit patients. Indeed, the current data demonstrate the presence and tenacity of comorbid psychological conditions and symptoms in patients with MALS. Furthermore, it is unclear whether any of these psychological symptoms preceded the physical symptoms of MALS, or perhaps had an exacerbating role in the experience of MALS symptoms. It is likely that psychological intervention would help to ameliorate psychological symptoms and possibly impact the physical symptoms of MALS, specifically the experience of pain. It is unclear whether these processes were to occur whether decisions to pursue surgical intervention may also change. Beyond this line of inquiry, it has previously been recommended to screen and treat comorbid mood symptoms in the presence of pediatric CAP (10). Additionally, strategies such as cognitive refocusing have been associated with lower patient- and parent-reported worst pain experiences and lower mood symptoms (20). Cognitive behavioral therapy therefore stands as a possible treatment intervention, with a focus on increasing positive coping strategies, and demonstrated efficacy in the treatment of pediatric pain management (24–29) and mood symptoms (30–32). Future research should explore the impact of a presurgical psychological intervention, such as cognitive behavioral therapy, on the pre- and postsurgical QOL and functioning of pediatric patients with MALS.

The present study is the largest of its kind to examine the presence and impact of psychological factors on the surgical treatment of a commonly overlooked condition causing CAP in a pediatric population. Limitations should be, however, noted in the interpretation of these findings. First, the sample was small. Nevertheless, a sample of 32 is large for a pediatric MALS sample (8), and significant

results were able to be detected. Furthermore, this sample represents a geographically varied population, with many patients traveling from out of state for specialty services from the interdisciplinary UCMALS team. Given the travel necessary to this specialty clinic, a possible placebo effect regarding postsurgical pain reduction, however, may also be considered. Second, the follow-up period was 6 months following surgery. It is unclear how the present results extend to longer-term follow-up for pediatric patients. Third, the number of analyses conducted in this study increases the risk for type I error. Future examinations of larger samples of pediatric patients with MALS will be needed, as well as the implementation and examination of psychological interventions to improve outcomes.

In summary, the present study builds upon previous findings that surgery improves self-reported pain and QOL for patients with MALS (5,6,9). Furthermore, comorbid psychological conditions are common in pediatric patients with MALS (6), and there is no evidence to suggest that the amelioration of pain due to surgical intervention improves psychological symptoms. Ineffective pain coping before surgery may also impact changes in QOL following surgical intervention. Therefore, the findings support the need to continue to better understand the needs of this unique population, and for psychological interventions targeted toward pediatric patients with MALS.

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