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Understanding the Variation in Use of Screening DXA Scans in Pediatric Patients with Celiac Disease

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Abstract Celiac disease (CD) is an immune-mediated genetic disorder occurring secondary to gluten exposure and increasing the likelihood of low bone mineral density (BMD). As there are no published guidelines for dual-x-ray absorptiometry (DXA) scanning in pediatric CD patients, we characterized current practices of pediatric gastroenterologists in ordering screening DXA scans for pediatric CD patients. To accomplish this, A REDCap survey was distributed to the NASPGHAN listserv. There was a total of 231 (11%) responses. The majority (60%) of clinicians do not order screening DXA scans because they don't believe it is clinically necessary. Patient factors influenced ordering screening DXA scans with fracture history driving ordering and tissue transglutaminase (tTG) level not affecting practice. Physician factors such as practice type and experience were not associated with ordering screening DXA scans. Case scenarios, showed wide variation in management based on DXA results.

Keywords: *celiac disease, bone, survey, guidelines, pediatric*

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1. Introduction

Celiac disease (CD) is one of the most common causes of intestinal malabsorption in childhood, with an estimated prevalence of 0.7-1% in North America and Europe and has long term complications including malnutrition, development of autoimmune diseases, and small bowel cancer [1,2]. Malabsorption is secondary to loss of villous cells in the small intestine leading to impaired absorption of nutrients [1]. With this loss, alteration of calcium absorption negatively influences bone development and bone density [3,4]. Despite recent advances in testing, delay in diagnosis remains an issue and children may have longstanding mucosal damage and malabsorption prior to diagnosis [5]. Previous studies have shown that altered bone metabolism and decreased bone mineral content in children with CD leads to osteoporosis and potential increased risk of fracture [3,6,7,8,9].

The mainstay for treatment of CD is a strict gluten-free diet which typically leads to a rapid clinical response [10]. There is also evidence that adherence to a gluten-free diet can reverse intestinal mucosal damage as well as the resulting abnormalities in growth and bone mineral density [7,11,12,13].

Bone status and measurement of bone density is accomplished through the use of dual x-ray absorptiometry (DXA) [4]. To obtain accurate readings from DXA scans, children are required to be cooperative which can be difficult for the younger population. Currently, CD is not a reimbursed indication for DXA scan for all insurance carriers and can result in large charges to patients. DXA scanning is widely used for the assessment of bone health in patients with CD despite these barriers. Currently, there are no published guidelines for the use of DXA scanning to monitor bone density in pediatric patients with CD. This lack of standardization has the potential to result in underuse and under diagnosis of low BMD or overuse and excess burden and cost for families.

Alternatively, there are recommendations and guidelines in the adult literature for appropriate use and timing of DXA scans in patients with newly diagnosed CD [14,15,16]. Multiple papers attempting to validate these recommendations have found mixed results. The British Society of Gastroenterology found that screening DXA scans were not justified for all patients with CD due to the low yield of positive findings [14]. The Canadian Journal of Gastroenterology released a position statement only recommending DXA scans in patients who have been on a strict gluten-free diet for one year [17]. A review of these publications suggest that screening DXA at diagnosis is not justified in all patients with newly diagnosed CD and should only be performed in specific populations [17,18]. As the physiology of pediatric bone disease differs from adults, further research is necessary to develop similar guidelines for pediatrics.

In order to understand common practices across North America, we surveyed clinicians regarding their use of DXA scans in pediatric patients with CD. The aims of the present study were to describe variations in current physician practices in ordering scans and to

determine management patterns of patients with low BMD. We also analyzed patient clinical factors and physician factors contributing to ordering of screening DXA scans.

2. Materian and Methods

A 14-item survey on ordering patterns of screening DXA scans in pediatric patients with CD was designed in REDCap [19] and received IRB approval from the Children's Hospital of Philadelphia. The survey was distributed electronically to all pediatric gastroenterologists in North America who receive emails from the North American Society for Pediatric Gastroenterology, Hepatology, and Nutrition (NASPGHAN) listserv. A total of 2,051 members received the survey and 231 members (11%) completed the survey. The survey was sent out in March of 2017 followed by 3 reminder emails over a 3-month period of time.

The main variables of interest were frequency of ordering screening DXA scans, patient information influencing ordering of screening DXA scans (lab results, endoscopy results, body mass index [BMI], and history of fracture), and physician factors (location of practice, type of practice, experience, and volume of practice) associated with ordering of screening DXA scans. Multiple case scenarios were presented to describe variability in management based on DXA results. Management options included change in medications, change in diet, behavioral changes, repeating DXA scans, laboratory testing, referral to a dietician or no changes to management. The survey also prompted clinicians to rate what information they would like to see in national guidelines.

2.1. Statistical Analysis

STATA (StataCorp. 2015. Stata Statistical Software: Release 14. College Station, TX: StataCorp LP) software was used for statistical analysis. All data are presented as percentages. Covariates were analyzed using Chi-square testing with a binary outcome of ordering a DXA scan or never ordering a DXA scan.

3. Results

A total of 231 clinicians (11%) responded to the survey covering the Midwest, South, Northeast, and Western regions as well as Canada (Table 1). Most respondents reported that they did not order screening DXA scans for pediatric patients with CD. Of the clinicians surveyed, a total of 146 (63%) clinicians never order a screening DXA scan, 38 (17%) clinicians order between 1-25% of the time, and 47 (20%) clinicians order more than 25% of the time. Of those who never order a DXA scan, 127 (89%) did not feel it was clinically necessary. Patient factors did significantly influence the decision to order a screening DXA scan ($p<0.0001$). Providers were most influenced by history of fracture and least influenced by tTG levels. Alternatively, BMI and endoscopy results had conflicting influence in a provider ordering a screening DXA scan (Table 2). Ordering of a screening DXA scan differed significantly based on region ($p=0.03$) with those from the Northeast ordering scans more frequently than other regions. There were no other statistically significant associations between physician factors and ordering of a screening DXA scan.

Table 1. Physician Demographics (n=231)

Region*: n (%)	Midwest: 35 (21)
	Northeast: 48 (29)
	South: 36 (21.5)
	West: 37 (22)
	Canada: 11 (6.5)
Type of practice: n (%)	Hospital based: 155 (87)
	Non-hospital based: 23 (13)
Years practiced: n (%)	0-5 years: 61 (34)
	6-10 years: 32 (18)
	11-20 years: 30 (17)
	21-30 years: 40 (22.5)
	>30 years: 15 (8.5)

* Northeast: DE, MA, NH, NJ, NY, PA; Midwest: IL, IN, KS, MI, MO, NE, OH;
South: AL, DC, FL, GA, LA, MD, NC, OK, SC, TN, TX, VA;
West: AZ, CA, CO, NM, NV, UT, WA.

Table 2. Patient factors influencing ordering of screening DXA and Content for national guidelines; rating from 1-5

Patient Factors Influencing ordering screening DXA (n=81)					
	1 (not influential/ important)	2	3	4	5 (very influential/ important)
tTG level: n (%)	50 (62)	7 (9)	8 (10)	9 (11)	7 (8)
Endoscopy biopsy results: n (%)	32 (39.5)	12 (15)	12 (15)	14 (17)	11 (13.5)
BMI: n (%)	28 (35)	11 (13.5)	11 (13.5)	18 (22)	13 (16)
History of fracture: n (%)	9 (11)	6 (7)	5 (6)	12 (15)	49 (61)
Content for national guidelines (n=193)					
Surveillance guidance: n (%)	14 (7)	7 (4)	30 (16)	60 (31)	80 (42)
Interval for repeat scans: n (%)	11 (6)	10 (5)	21 (11)	68 (36)	81 (42)
Clinical scenarios: n (%)	10 (5)	6 (3)	30 (16)	70 (36)	76 (40)
Red flags: n (%)	9 (5)	6 (3)	19 (10)	76 (39)	83 (43)
Treatment guidance: n (%)	6 (3)	4 (2)	16 (8)	61 (32)	106 (55)

When clinicians were presented with case scenarios, there was no consensus with management plans. These case scenarios were similar except for the DXA result of either a z-score of -1.5 (normal/borderline) or -2.0 (abnormal). The severity of low bone density did not seem to alter management choices. For example, "change in diet", "referral to dietitian", and "change in medications" were selected at similar frequency for both case scenarios despite notable differences in DXA scan results (data not shown).

Overall, clinicians felt that guidelines regarding screening DXA scans for pediatric patients with CD would be extremely valuable. When evaluating all of the suggested content for guidelines, 79% of respondents rated all categories a 4 or 5 out of 5 in importance (Table 2).

4. Discussion

This is the first study that investigates DXA ordering practices and common practices for management of BMD in the United States and Canada for pediatric patients with CD. There is identified variation and currently, there are no guidelines regarding screening DXA scans, repeat DXA scans, or treatments based on DXA scan results for pediatric patients with CD. The most recent NASPGHAN guideline [20], published in 2005, for diagnosis and treatment of CD, discuss bone health in pediatric patients with CD but do not provide specific recommendations for screening and management of low BMD. The adult literature does suggest that screening DXA scans are unnecessary and only specific patients should undergo a DXA scan at diagnosis of CD.

In our sample of North American clinicians, the majority are not ordering screening DXA scans on pediatric patients with newly diagnosed CD. The majority of these clinicians did not feel that a scan was clinically necessary.

There was a significant difference in ordering screening DXA scans based on patient factors such as history of fracture, BMI, esophagogastroduodenoscopy (EGD) results, and tTG levels. Most notable, the majority of providers order screening DXA scans in patients who previously had a fracture. This is consistent with literature indicating increased fracture risk in patients with CD [9,15]. Clinicians were less influenced by tTG level, endoscopy results, and BMI although there is no literature to support this decision-making process.

When presented with case scenarios, clinicians had extreme variability in management choices. The primary difference in clinical scenarios were the DXA z-score result, one of which was extremely abnormal (z-score = -2.0) and one of which was borderline (z-score = -1.5). Interestingly, management options were chosen at similar rates in both scenarios. This is likely because there are no consensus guidelines regarding appropriate management in the setting of a low BMD on DXA scan.

Overall, clinicians felt that a guideline would be extremely valuable. Clinicians would like to see recommendations about which patients should have DXA scans, at what interval repeat scans should be performed, red flags which would be predictive of low BMD, and treatment guidance based on DXA scan results. This is consistent with content in adult guidelines [17].

This study provides evidence of variability in physician practices in the diagnosis and management of bone disease in pediatric patients with CD. Although the majority of clinicians are not ordering screening DXA scans on patients newly diagnosed with CD, about 40% of respondents do order DXA scans in certain scenarios. Those who are ordering scans routinely did not have consistent management plans based on case scenarios. This variability could result in poor quality of care in pediatric patients with CD.

Current research shows that low BMD improves approximately one year after treatment of CD with a strict gluten-free diet [11,12,13]. This suggests that we may not need to screen patients who are compliant with their diet. Further research evaluating incidence of low BMD in pediatric CD and risk factors predictive of low BMD is needed to develop evidence-based guidelines.

There were multiple limitations of this study. Although our sample has representation across the United States and Canada, there was a low response rate so the results may not reflect practices of the remainder of the clinicians in the country. This survey is based on self-reported practices and no validation was completed to confirm the responses.

5. Conclusions

Lack of standardization for use of DXA scans in children with CD has the potential to lead to under and over-ordering of DXA scans in this population. Because DXA scans are sometimes difficult for patients to perform and can be costly to families, it is important to order the scan only in patients with a clear need for monitoring of BMD. This study found variability in the use of DXA scans in pediatric patients with CD as well as variability in treatment of patients with low BMD. Clearly, there is a need and desire by clinicians for clinical guidance in this area.

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Statement of Competing Interests

The authors have no conflict of interest to declare.

Abbreviations

CD: Celiac disease; DXA: Dual x-ray absorptiometry; BMD: Bone mineral density; tTG: Tissue trans-glutaminase; BMI: Body mass index; EGD: Esophagogastroduodenoscopy; NASPGHAN: North American Society for Gastroenterology Hepatology and Nutrition

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