Functional defecation disorders in children
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AWARENESS AND IMPLEMENTATION OF THE 2014 ESPGHAN/NASPGHAN GUIDELINE FOR CHILDHOOD FUNCTIONAL CONSTIPATION

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ABSTRACT

Objective: To assess if physicians approach children with functional constipation according to the 2014 ESPGHAN/NASPGHAN guideline.

Methods: We invited pediatricians and pediatric gastroenterologists in the Netherlands and the U.S. to participate in this anonymous survey using a self-developed questionnaire containing 19 multiple-choice questions concerning evaluation and treatment of children with constipation.

Results: We included 328 physicians (67% from the U.S., 34% from the Netherlands). The majority of U.S. responders (53%) worked in primary care whereas all Dutch responders worked in a hospital. In total, 31% of responders were not familiar with the guideline, (38% U.S. responders vs. 16% Dutch responders, \( P < .001 \)). Perianal inspection was frequently (‘often’ or ‘always’ on a 5-point Likert scale) conducted by 78% of responders. Digital rectal examination was frequently done by 42%. Inquiry about sexual abuse was made by 18%. Commonly reported reasons for omitting these items were perceived patient or parental discomfort. Most implemented initial non-pharmacological interventions included: a toilet training program (89%), optimizing fluid and fiber intake (86% and 81%), a defecation diary (62%) and a reward system (60%). Polyethylene glycol was the most prescribed medication for disimpaction (68%) and maintenance treatment (57% for infants, 97% for children ≥1 year).

Conclusions: Many responders were not familiar with the ESPGHAN/NASPGHAN guideline for functional constipation. Nonetheless, therapeutic decisions correlated fairly well with recommendations from the guideline, especially for children ≥1 year. Guideline awareness and adherence remain to be improved. Future studies should focus on exploring strategies to improve guideline implementation through the development of digital learning tools.
INTRODUCTION

Functional constipation (FC) is a common problem in childhood, with a worldwide reported prevalence up to 29.6%. The diagnosis is based on the pediatric Rome IV criteria. Symptoms of FC include infrequent, painful defecation, hard stools, fecal incontinence and abdominal pain. These bothersome symptoms negatively affect quality of life in patients and their families and contribute to increased health care utilization and associated costs.

In 2014, the European Society for Pediatric Gastroenterology, Hepatology, and Nutrition (ESPGHAN) and the North American Society for Pediatric Gastroenterology, Hepatology, and Nutrition (NASPGHAN) published the ESPGHAN/NASPGHAN guideline for FC in children. This guideline provides evidence-based recommendations for the evaluation, treatment and follow-up of children with FC. Key recommendations include that the diagnosis is based on history and physical examination and that the Rome criteria should be used to define FC. If only one of the Rome criteria is present and the diagnosis of FC is uncertain, a digital rectal examination (DRE) is recommended. A normal fiber intake, fluid intake and physical activity level are recommended and the routine use of pre- or probiotics is not recommended in the treatment of childhood constipation. Non-pharmacological interventions consist of demystification, explanation, and guidance for toilet training (in children with a developmental age of at least 4 years). For disimpaction, the guideline recommends polyethylene glycol (PEG) orally 1-1.5g/kg/day for 3 to 6 days or an enema once per day for 3 to 6 days if PEG is not available. PEG with or without electrolytes is also recommended as the first-line maintenance treatment.

In order to improve medical care, it is important that physicians are aware of guidelines and that these guidelines are adhered to in clinical practice. Previous studies on guideline adherence in the Netherlands and the U.S. have suggested that approximately 30-40% of patients do not receive care according to current evidence. Focht et al. and Yang et al. have shown poor familiarity with and low adherence to the NASPGHAN guidelines for childhood FC from 1999 and 2006 respectively.

These studies addressed the need for educational strategies to improve guideline adherence in order to achieve effective management of FC in children. Unfortunately, educational interventions have shown low levels of improvement regarding guideline implementation. Numerous potential barriers may impede guideline awareness, these barriers can occur at the level of the individual professional, the healthcare team, the healthcare organization or the wider environment. Therefore, it is important to understand the specific barriers to change certain practices and to tailor strategies to improve guideline adherence to these specific barriers.
Our primary aim of this study was to investigate physicians’ familiarity with the 2014 ESPGHAN/NASPGHAN guideline. Our secondary aim was to assess if physician’s approaches are consistent with this guideline. Moreover, we hypothesized that physicians would avoid items of the medical history and physical examination that could be perceived as uncomfortable or embarrassing, such as inquiring about sexual abuse and performing a perianal inspection or DRE. Therefore, our tertiary aim was to assess how frequently physicians inquire about sexual abuse and perform perianal inspection and DRE in the evaluation of FC.

METHODS

Study population
We included physicians working in pediatrics and pediatric gastroenterology in either the Netherlands or the U.S. We invited all participants attending the following meetings to partake in this survey:

1. Pediatric gastroenterology review course organized by the European Postgraduate Gastrointestinal School (EPGS) held in Amsterdam, the Netherlands in September 2016
3. Regional pediatric gastroenterology symposium held in Amsterdam, the Netherlands in December 2016
4. NASPGHAN Board Review Course held in Scottsdale, AZ, U.S. in February 2017

For this study, only surveys answered by physicians who reported that they treated children with FC were included.

Furthermore, we invited general pediatricians from a secondary and a tertiary hospital in Amsterdam to partake in this survey study and we sent an email to all registered pediatric gastroenterologists in the Netherlands inviting them to participate.

Questionnaire
We developed a questionnaire containing 19 multiple-choice questions. The questions assessed factors related to the work setting, familiarity with the 2014 ESPGHAN/NASPGHAN guideline and the diagnostic and therapeutic approach towards children with FC. The questionnaire is provided as Supplemental Digital Content (http://links.lww.com/MPG/B148).
The self-reported frequency with which items from the medical history and physical examination were put into practice was scored on a 5-point Likert scale (never-rarely-sometimes-often-always). For the interpretation of the results, we considered ‘often’ or ‘always’ on the 5-point Likert scale as ‘frequent’ performance of an item. For each question, responders had the option to an open answer in case they considered the multiple choice answers to be insufficient.

**Statistical analysis**

Analyses were conducted using IBM SPSS Statistics v23.0 (IBM, Amsterdam, the Netherlands). For questions where the responder had to choose one of the provided answers (questions 1-7, 9, 11, 13, 16-19, see Supplemental Digital Content, http://links.lww.com/MPG/B148), only cases with available data were analyzed; cases with missing data were excluded from these analyses. For questions with multiple possible answers, only affirmative answers are reported as proportion of the total group (questions 8, 10, 12, 14, 15, see Supplemental Digital Content, http://links.lww.com/MPG/B148). Confidence intervals were calculated according to the Clopper-Pearson method. For the comparison of dichotomous categorical variables between two groups, Fisher’s exact tests were performed. *P*-values of < .05 were considered to be statistically significant.

This study was approved by the Medical Ethics Committee of the Academic Medical Center.

**RESULTS**

**Participants**

In total, 418 questionnaires were returned. Ninety questionnaires were excluded (Figure 1).

**Responder characteristics and familiarity with the guideline**

Responder characteristics are shown in Table 1. Of the 328 included questionnaires, 67% were filled out by physicians from the U.S. and 34% by physicians from the Netherlands. The majority of responders (92%) were attending physicians and 58% of them worked in the field of general pediatrics. Significantly more responders from the U.S. (50%) worked in pediatric gastroenterology compared to responders from the Netherlands (26%, *P* < .001). Just over half of U.S. responders (53%) worked in primary care or private practice whereas all responders from the Netherlands worked in a hospital setting. A total of 31% (CI 26-36%) reported that they were not familiar with the 2014 ESPGHAN/NASPGHAN guideline, unfamiliarity with the guideline was more common in responders from the U.S. compared to the Netherlands (Table 2). Moreover, unfamiliarity with the guideline was significantly more common in pediatricians compared to pediatric gastroenterologists and in physicians
with more than 15 years of experience compared to physicians with less experience (Table 2). Most frequently reported reasons to deviate from the guideline were: patient’s or parent’s preference for a different laxative (29% [CI 24-35%]), unfamiliarity with the guideline (27% [CI 23-33%]), insufficient clinical response to the maximum recommended dosage (26% [CI 21-31%]), patient or parental preference for a different treatment than laxatives (19% [CI 15-24%]).

FIGURE 1. Flowchart inclusion completed questionnaires. 
Abbreviations: FC, functional constipation; FI, fecal incontinence; NL, Netherlands; U.S., United States.

TABLE 1. Responder characteristics and familiarity with the guideline for childhood functional constipation.

<table>
<thead>
<tr>
<th></th>
<th>Total (n=328)</th>
<th>Dutch (n=110)</th>
<th>U.S. (n=218)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n (%)</td>
<td>n (%)</td>
<td>n (%)</td>
</tr>
<tr>
<td>Field of practice</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pediatrics</td>
<td>191 (58)</td>
<td>81 (74)</td>
<td>110 (51)</td>
</tr>
<tr>
<td>Pediatric gastroenterology</td>
<td>137 (42)</td>
<td>29 (26)</td>
<td>108 (50)</td>
</tr>
<tr>
<td>Title/role</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Attending</td>
<td>300 (92)</td>
<td>100 (91)</td>
<td>200 (92)</td>
</tr>
<tr>
<td>Resident</td>
<td>28 (9)</td>
<td>10 (9)</td>
<td>18 (8)</td>
</tr>
<tr>
<td>Practice setting</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tertiary hospital</td>
<td>87 (27)</td>
<td>28 (26)</td>
<td>59 (27)</td>
</tr>
<tr>
<td>Secondary hospital</td>
<td>126 (38)</td>
<td>82 (75)</td>
<td>44 (20)</td>
</tr>
<tr>
<td>Primary care/private practice</td>
<td>115 (35)</td>
<td>-</td>
<td>115 (53)</td>
</tr>
<tr>
<td>Years of experience</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>0-5 years</td>
<td>75 (23)</td>
<td>24 (22)</td>
<td>51 (23)</td>
</tr>
<tr>
<td>5-10 years</td>
<td>47 (14)</td>
<td>22 (20)</td>
<td>25 (12)</td>
</tr>
<tr>
<td>10-15 years</td>
<td>44 (13)</td>
<td>17 (16)</td>
<td>27 (12)</td>
</tr>
<tr>
<td>&gt;15 years</td>
<td>162 (49)</td>
<td>47 (43)</td>
<td>115 (53)</td>
</tr>
</tbody>
</table>
Diagnostics

Perianal inspection was frequently (often or always on the 5-point Likert scale) conducted by 78% [CI 73-82%] of responders in the evaluation of children with FC. DRE was frequently performed by 42% [CI 36-47%] and inquiry about sexual abuse was frequently made by 18% [CI 14-22%]. Comparisons between different groups of physicians are shown in Table 2.

Frequently reported reasons for omitting these items in the diagnostic workup for FC were discomfort for the child, parent or physician (see Figure 2). Most physicians (65% [CI 60-70%]) reported no suspicion of sexual abuse as the primary reason not to elicit a history of sexual abuse. An important reason not to perform perianal inspection was the perception that this would not provide useful information (11% [CI 8-14%]). The most commonly reported reason for not performing a DRE was if a child fulfilled sufficient criteria for the diagnosis of FC based on the history (40% [CI 35-46%]). Moreover, 17% [CI 13-21%] of responders reported to avoid a DRE because they felt that this would not provide useful information. Significantly more U.S. responders compared to Dutch responders reported both child and parental discomfort as reasons not to perform a DRE (43% [CI 36-50%] vs. 27% [CI 20-37%], P = .010 and 16% [CI 11-22%] vs. 5% [CI 2-10%], P = .003 respectively).

Treatment

Most commonly implemented non-pharmacological interventions included instating a toilet training program (89% [CI 85-92%]), optimizing fluid and fiber intake (86% [CI 81-89%] and 81% [CI 77-85%]), keeping a defecation diary (62% [CI 56-67%]), instating a reward system (60% [CI 54-65%]) and prescribing pre- or probiotics (23% [CI 19-28%]). Comparisons between different groups of physicians are shown in Table 2.

PEG was the most prescribed medication for disimpaction (68% [CI 64-74%]), followed by enemas (25% [CI 21-30%]). The preference of 6% [CI 4-9%] of responders was unclear since they checked both boxes (PEG and enemas). The primary reasons for choosing PEG over enemas for disimpaction were patient comfort (69% [63-75%]), the drug being well-tolerated by patients (53% [CI 46-60%]) and its easiness to use (47% [CI 40-54%]). The primary reasons for choosing enemas over PEG were a more rapid effect (63% [CI 52-74%]) and perceived higher effectiveness (56% [CI 45-67%]).

For infants, PEG was the preferred medication for maintenance treatment in 57% [CI 51-62%], followed by lactulose (31% [CI 26-36%]), either PEG or lactulose (7% [CI 4-10%]) and milk of magnesia/magnesium hydroxide (5% [CI 3-8%]). For children ≥1 year of age, 97% [CI 94-99%] of responders preferred PEG as laxative for maintenance treatment.
Table 2: Guideline awareness and implementation of items of the medical history and physical examination and non-pharmacological interventions, compared between responders from the Netherlands and the U.S., pediatricians and pediatric gastroenterologists, physicians with more than 15 years of experience and physicians with less experience. Fisher's exact test, bold if \( P < .05 \).

<table>
<thead>
<tr>
<th>Country</th>
<th>U.S. (n=218)</th>
<th>Pediatric GI (n=137)</th>
<th>Pediatric GI (n=137)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n (%, CI)</td>
<td>n (%, CI)</td>
<td>n (%, CI)</td>
</tr>
<tr>
<td>NL (n=110)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Familiarity with the guideline</td>
<td>82/110 (75% [69-81])</td>
<td>98/110 (89% [85-93])</td>
<td>98/110 (89% [85-93])</td>
</tr>
<tr>
<td>Unfamiliar with the guideline</td>
<td>5/110 (4% [3-6])</td>
<td>12/110 (11% [9-13])</td>
<td>12/110 (11% [9-13])</td>
</tr>
<tr>
<td>Frequent implementation of items of the medical history and physical examination</td>
<td>8/110 (7% [5-9])</td>
<td>69/110 (62% [56-68])</td>
<td>69/110 (62% [56-68])</td>
</tr>
<tr>
<td>Inquirry sexual abuse</td>
<td>34/110 (31% [23-40])</td>
<td>24/217 (11% [8-16])</td>
<td>24/217 (11% [8-16])</td>
</tr>
<tr>
<td>Perianal inspection</td>
<td>98/110 (89% [82-94])</td>
<td>157/218 (72% [66-78])</td>
<td>157/218 (72% [66-78])</td>
</tr>
<tr>
<td>Digital rectal examination</td>
<td>37/108 (34% [26-44])</td>
<td>99/217 (46% [39-52])</td>
<td>99/217 (46% [39-52])</td>
</tr>
<tr>
<td>Non-pharmacological interventions</td>
<td>96/108 (89% [82-94])</td>
<td>185/217 (85% [80-89])</td>
<td>185/217 (85% [80-89])</td>
</tr>
<tr>
<td>Toilet training</td>
<td>95/108 (88% [81-90])</td>
<td>130/136 (95% [90-99])</td>
<td>130/136 (95% [90-99])</td>
</tr>
<tr>
<td>Optimizing fluid intake</td>
<td>87/108 (81% [73-89])</td>
<td>169/189 (89% [83-93])</td>
<td>169/189 (89% [83-93])</td>
</tr>
<tr>
<td>Optimizing fiber intake</td>
<td>54/108 (50% [42-62])</td>
<td>117/136 (86% [78-93])</td>
<td>117/136 (86% [78-93])</td>
</tr>
<tr>
<td>Keeping a defecation diary</td>
<td>54/108 (50% [42-62])</td>
<td>117/136 (86% [78-93])</td>
<td>117/136 (86% [78-93])</td>
</tr>
<tr>
<td>Implementing a reward system</td>
<td>67/108 (62% [54-74])</td>
<td>100/136 (74% [66-80])</td>
<td>100/136 (74% [66-80])</td>
</tr>
<tr>
<td>Prescribing pre-or probiotics</td>
<td>8/108 (7% [4-14])</td>
<td>69/136 (51% [43-60])</td>
<td>69/136 (51% [43-60])</td>
</tr>
</tbody>
</table>

* Self-reported frequency with which items from the medical history and physical examination were put into practice was scored on a 5-point Likert scale (never-rarely-sometimes-often-always), the answers 'often' or 'always' on the 5-point Likert scale were considered as frequent performance of an item.
DISCUSSION

In our sample of Dutch and U.S. physicians working in pediatrics and pediatric gastroenterology, 31% of responders were not familiar with the 2014 ESPGHAN/NASPGHAN guideline for childhood FC. Unfamiliarity with the guideline was more common among general pediatricians, physicians from the U.S. and physicians with >15 years of experience. Nonetheless, overall therapeutic decisions correlated fairly well with recommendations from this guideline, especially for children ≥1 year of age.

Two studies on the awareness and implementation of previous childhood FC guidelines have been published before. Focht et al. reported results from a cross-sectional survey study among 366 pediatricians across the U.S., assessing familiarity and implementation of the 1999 NASPGHAN guideline for childhood FC. They found that only 8% of pediatricians were familiar with that guideline. Later, Yang et al. performed a cross-sectional survey study among 967 physicians in the U.S. (771 trainees, 196 attendings) evaluating familiarity and implementation of the 2006 NASPGHAN guideline. In this study, 84% of responders reported being unfamiliar or only slightly familiar with the 2006 NASPGHAN guideline. In our current study, awareness of the 2014 ESPGHAN/NASPGHAN guideline was better than awareness of older guidelines reported in the two previous studies, although even in the current study still 31% of responders were unfamiliar with the 2014 ESPGHAN/NASPGHAN guideline.
In 2015, Ciullo et al. published an abstract on adherence to the 2014 ESPGHAN/NASPGHAN guideline. In their study among 147 Italian pediatricians, 63% of responders were unfamiliar or slightly familiar with the guideline. The awareness of the guideline was poor compared to our study, but it should be taken into account that this study was performed when the guideline was only recently published.

In our current study, guideline awareness was much higher among pediatric gastroenterologists than among general pediatricians. Although this result was to be expected, it does raise questions on how to improve guideline awareness among general pediatricians, who treat the majority of children with constipation. Interestingly, physicians with less experience reported better familiarity with the guideline, suggesting that junior physicians are more aware of recent guidelines potentially because they are confronted with these guidelines during their training. In an attempt to achieve better awareness and implementation of the 2014 ESPGHAN/NASPGHAN guideline, in 2015 ESPGHAN developed an online course on childhood FC. This course is accessible via United European Gastroenterology (UEG) after obtaining a log in that is available for free through https://www.ueg.eu/myueg/myueg. In a recently published short communication, Mallon et al. described promising results to improve FC guideline awareness and implementation using a “spaced-education” game. The concept of spaced-learning involves repetition of highly condensed information. In the study by Mallon et al., questions were sent to pediatric primary care providers on a weekly basis. Through an adaptive process, incorrectly answered questions were re-sent after one week and correctly answered questions after two weeks. Participants earned points based on their performance and could eliminate questions by answering them correctly twice. A competition element was added by assigning participants to teams. This intervention significantly improved the performance of participants, comparing final scores with baseline. Hopefully, such attempts to improve guideline awareness and implementation will lead to better guideline awareness and implementation in the future.

Previous studies have reported that DRE is often avoided in clinical practice. A study from Saudi Arabia showed that DRE was routinely performed in the evaluation of children with constipation by only 39% of pediatric providers versus 79% of pediatric gastroenterologists. Similarly, a study from Brazil demonstrated that only 43% of non-gastroenterology pediatricians performed a DRE. In the study by Focht et al., 43% of U.S. pediatricians reported to either never or only occasionally performing a DRE on new patients presenting with constipation. In the more recent study by Yang et al., DRE was never or rarely performed by 61%. In the current study, patient and parental discomfort were commonly reported as reasons not to perform a DRE and this was more frequently reported by U.S. physicians as compared to the Dutch physicians. Patient discomfort has been previously reported as a reason to avoid a DRE as part of the physical examination
in both children and adults.\textsuperscript{10,18,19} Moreover, examination of the anorectum seems to be a sensitive issue for parents and physicians. Perhaps the difference between Dutch and U.S. responders indicates that cultural differences exist regarding the opinions on this topic, even between two Western countries. The recommendations regarding DRE have also changed in guidelines over time. While the 1999 NASPGHAN guideline recommended to perform a DRE in all children with FC\textsuperscript{20}, the 2014 ESPGHAN/NASPGHAN guideline recommends that this is only necessary in children who fulfill only one of the Rome criteria and where a DRE may therefore help to establish a diagnosis of FC.\textsuperscript{7} Although a fecal mass in the rectum remains one of the diagnostic Rome IV criteria\textsuperscript{2}, the perceived value of a DRE seems to have decreased over the past two decades. However, a DRE can provide valuable information on the presence of a rectal fecal mass, anorectal sensation, and sphincter tone.\textsuperscript{7,21–24} Moreover, in a child with functional fecal incontinence it can be difficult to differentiate between FC and functional nonretentive fecal incontinence and in these children, detection of a fecal mass during DRE will help to establish a diagnosis of FC and thereby guide management.\textsuperscript{7,25}

In accordance with the guideline, the majority of responders in the current study reported to use PEG as treatment of first choice for disimpaction in children. The guideline also recommends PEG as the first-line maintenance treatment of FC in children of all ages.\textsuperscript{7} For the treatment of children ≥1 year of age, this recommendation was well adhered to in our study sample. For the treatment of children <1 year of age, most responders indeed reported to use PEG as treatment of first choice, but lactulose was also commonly used as treatment of first choice. The guideline recommends lactulose if PEG is not available.\textsuperscript{7}

In the present study, a remarkably high rate of physicians reported to prescribe pre- or probiotics to children with FC, this rate was significantly higher among U.S. physicians compared to Dutch physicians. These findings confirm results from a previous survey study among 74 pediatric surgeons and pediatric gastroenterologists, where 18% of responders reported to prescribe pre- or probiotics to children with FC.\textsuperscript{26} Currently, there is no sufficient evidence to support the use of pre-, pro- or synbiotics in the treatment of childhood FC.\textsuperscript{7,27} It is therefore unclear why physicians prescribe pre- or probiotics and which pre- or probiotics they actually prescribe. This should be sought out in future studies and it may be necessary to educate physicians about the benefits, risks and costs of prescribing pre- and probiotics.

Some limitations need to be taken into account when interpreting the results of this study. Because we primarily invited participants of selected meetings focused on pediatric gastroenterology, our results are at risk of selection bias. In order to include more general pediatricians who did not attend a pediatric gastroenterology focused meeting, the questionnaire was also distributed in two regional hospitals (one secondary care center, one tertiary care center). Furthermore, the questionnaire was distributed among all pediatric
gastroenterologists registered in the Netherlands (n=47) and returned by 29 (response rate 62%). Unfortunately, it is not possible to reliably calculate the response rate for the entire study. The questionnaire was distributed at conferences with heterogeneous participant compositions which cannot all be retrieved accurately. When interpreting the results of the comparisons between the Netherlands and the U.S., it should also be taken into account that the majority of U.S. physicians worked in primary care or private practice, which is highly uncommon in the Netherlands. Furthermore, a larger proportion of U.S. responders were pediatric gastroenterologists rather than general pediatricians.

In conclusion, in our sample of physicians working in pediatrics and pediatric gastroenterology, almost a third of responders were not familiar with the ESPGHAN/NASPGHAN guideline on FC. Nevertheless, therapeutic decisions correlated fairly well with recommendations from the guideline, especially for the treatment of children ≥1 year of age. Future research should assess guideline adherence in other countries and aim to develop strategies to improve guideline awareness and adherence. Digital learning tools carry low costs and can be distributed easily. Therefore, it appears valuable to further explore these options and to optimize the distribution and use of such tools. Optimally, the effect of such tools should also be studied in research settings. Furthermore, specific barriers that impair guideline awareness and adherence, especially in general pediatricians, need to be further investigated in order to tailor interventions appropriately.

ACKNOWLEDGEMENTS

The authors sincerely thank all responders who participated in this survey.
REFERENCES


