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Translation and Cultural Adaptation of the ISTH-Bleeding Assessment Tool to European Portuguese

Tradução e Adaptação Cultural do Questionário ISTH-Bleeding Assessment Tool para Português Europeu

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ABSTRACT

Introduction: A distinctive bleeding history is the first diagnostic stage of congenital hemorrhagic diseases. Furthermore, since it is in fact an assessment of the patient's phenotype, it is not a (mere) complementary diagnostic method or test - it is an integral and intrinsic step to establish the patient's clinical history, which is a medical act. The International Society on Thrombosis and Haemostasis-Bleeding Assessment Tool (ISTH-BAT) is a standardized bleeding assessment tool used during the diagnostic workup of suspected bleeding disorders. This study aimed to translate and culturally adapt the ISTH-BAT questionnaire to the European Portuguese language.

Methods: The translation and cultural adaptation were based on the International Society for Pharmacoeconomics and Outcomes Research Guidelines and initiated after obtaining permission from the authors. The process involved three English to European Portuguese independent forward translations and a back translation. Discrepancies were gradually resolved. A harmonized version was presented to hemophilia patients and hemophilia carriers for cognitive debriefing.

Results: The forward and back translations did not produce important discrepancies. However, some issues were identified in the cognitive debriefing, which led to the cultural adaptation of medical terms for better understanding.

Conclusion: A formal translation and cultural adaptation process ensures that the new version maintains the same concepts as the original. After translation, several changes were necessary to ensure that the questionnaire was understandable by non-medical patients. We propose a European Portuguese version of the ISTH-BAT, which will require validation in further studies.

Keywords: Blood Coagulation Disorders; Patient Reported Outcome Measures; Portugal; Surveys and Questionnaires; Translation

RESUMO

Introdução: A identificação da história clínica completa da sintomatologia hemorrágica do doente é a primeira etapa no diagnóstico de qualquer coagulopatia congénita. Além disso, por se tratar de uma avaliação do fenótipo hemorrágico do doente, este não é (apenas) um método ou exame complementar de diagnóstico - é uma etapa essencial e intrínseca na definição da história clínica do doente, o que constitui um ato médico. O *International Society on Thrombosis and Haemostasis-Bleeding Assessment Tool* (ISTH-BAT) é um instrumento de avaliação da hemorragia padronizado, utilizado quando existe suspeita de doença hemorrágica. Este estudo teve como objetivo a tradução e adaptação cultural do questionário ISTH-BAT para o português europeu.

Métodos: A tradução e a adaptação cultural foram baseadas nas normas da International Society for Pharmacoeconomics and Outcomes Research e iniciadas após a obtenção da autorização dos autores da versão original. O processo envolveu três traduções independentes do inglês para o português europeu e uma retroversão. As discrepâncias foram gradualmente resolvidas. A versão traduzida conciliada foi apresentada a doentes com hemofilia e portadoras de hemofilia para identificarem problemas de compreensão do questionário.

Resultados: As traduções diretas e reversa não mostraram discrepâncias importantes. No entanto, foram identificadas algumas questões nas entrevistas cognitivas, que levaram à adaptação cultural de termos médicos para uma melhor compreensão do questionário pelos doentes.

Conclusão: Um processo formal de tradução e adaptação cultural garante que a nova versão mantém os mesmos conceitos que a versão original. Após a tradução, foi necessário realizar alterações para garantir que o questionário fosse compreensível para os doentes. Propomos uma versão em português europeu do ISTH-BAT, que deverá ser validada em estudos futuros.

Palavras-chave: Distúrbios da Coagulação Sanguínea; Inquéritos e Questionários; Medidas de Resultados Reportadas pelo Doente; Portugal; Tradução

KEY MESSAGES

- We propose a European Portuguese version of the International Society on Thrombosis and Haemostasis-Bleeding Assessment Tool (ISTH-BAT).
- The ISTH-BAT is recommended to evaluate bleeding phenotype in patients with suspected bleeding disorders.
- The European Portuguese version was translated and culturally adapted and must be collected by a physician like the original version.
- The absence of psychometric property analysis and its validation are the main study limitations.

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INTRODUCTION

Bleeding disorders include von Willebrand disease (VWD), platelet disorders, hemophilia, and other clotting factor deficiencies.[1] A bleeding history is important to identify those who may have an inherited bleeding disorder. A structured bleeding history should explore mucocutaneous bleeding symptoms (e.g., epistaxis, gum bleeding, prolonged bleeding from minor wounds, menorrhagia, gastrointestinal tract bleeding), bleeding in relation to previous hemostatic challenges (e.g., surgery, dental extractions, trauma) and bruising with minor trauma from an early age.[2]

A great deal of clinical information on a patient can be obtained using a Bleeding Assessment Tool (BAT) that can help prioritize those individuals who require laboratory testing. Multiple BATs have been published, and studies have shown them to be useful in the evaluation of patients with a possible bleeding disorder.[3] At present, the International Society on Thrombosis and Haemostasis (ISTH) endorses the BAT consensus, the ISTH-BAT, which reports a positive or abnormal bleeding score of ≥ 4 in adult men, ≥ 6 in adult women and ≥ 3 in children. Each domain is scored from zero (absence of bleeding symptoms) to four (symptoms requiring extensive medical intervention), and the overall bleeding score is determined by summing the scores for all domains.[4,5]

In 2019, an International Working Group was established by the European Hematology Association. It proposed a diagnostic approach for patients referred for investigation of bleeding symptoms, and decided that the first evaluation of bleeding phenotype must include the ISTH-BAT,[6] a tool that can help identify hemophilia patients. A study by Borhany *et al* showed that the bleeding score (BS) was significantly higher in hemophilia A and hemophilia B patients than in controls, with no significant difference between newly diagnosed and known hemophilia patients. Furthermore, there were no significant differences in the BS between hemophilia A and B patients. However, the BS was higher in severe hemophilia A patients than in mild hemophilia A patients and lower in pediatric patients than in adult patients.[7]

The use of a standardized bleeding score proved to be potentially useful to further dissect the association between von Willebrand factor function and bleeding, to establish an optimal diagnosis of type-1 VWD (VWD-1) and to evaluate the bleeding risk in VWD patients.[8] A 2019 Platelet Physiology Standardization Subcommittee (SSC) study tested the utility of the ISTH-BAT in a large cohort of patients with a well-defined diagnosis of inherited platelet disorder in comparison with two parallel cohorts, one of patients with VWD-1 and one of healthy controls (HC). They concluded that the ISTH-BAT discriminated inherited platelet function disorders (IPFD) from healthy controls efficiently, while having lower accuracy in distinguishing IPFD from VWD-1. Therefore, the ISTH-BAT appears to be useful for identifying subjects requiring laboratory evaluation for a suspected inherited platelet function disorders, once VWD is preliminarily excluded.[9] Although VWD is the most frequently inherited bleeding disorder, it remains heavily underdiagnosed. The identification and diagnosis of VWD patients will be improved through further improvements in the design and digital availability of BATs, as well as in ongoing and future outreach campaigns, including on social media. Some authors launched a website (www.vwdtest.com) that provides easily accessible information on VWD to individuals who may suffer from abnormal bleeding, as well as a simple online bleeding assessment tool and guidance on the next steps to diagnose a potential bleeding disorder.[10]

One of the largest studies on hemophilia carriers and the first to systematically document the use of the ISTH-BAT in this group showed that hemophilia carriers experience abnormal bleeding, including hemarthrosis. Overall, the bleeding score is higher in women with Type 1 VWD and in descending order in hemophilia carriers and Type 3 VWD obligate carriers.[11]

Based on these data, BATs have been recommended in a number of clinical settings by national and international guidelines.[6,12,13] BATs are not without limitations, including lack of sensitivity in those without hemostatic challenges (such as children and men), recall bias, score saturation with recurrent symptoms, and the inability of positive BATs to distinguish between different types of mild-to-moderate bleeding disorders.[14]

Given the current recommendations on the use of the ISTH-BAT to evaluate bleeding phenotype in patients with suspected bleeding disorders, we aimed to translate and culturally adapt the ISTH-BAT from the original English to European Portuguese.

METHODS

This study was approved by the Ethics Committee of São João Local Health Unit as part of the protocol with the reference CES/CHUSJ: 334/2021, entitled "*ImpaHCta - Avaliação da qualidade de vida das portadoras de hemofilia*". All patients gave written informed consent.

A formal translation and cultural adaptation process was performed, as recommended in the International Society for Pharmacoeconomics and Outcomes Research Guidelines regarding patient-reported outcomes measures (PROMs).[15]

In the preparation phase, the researchers contacted the developer of the original version of the ISTH-BAT and obtained permission for the translation and cultural adaptation. A working group was assembled, including physicians with experience in the treatment of patients with coagulopathy, a coordinator and a team of Portuguese native speaker

translators.

The forward translation from English to Portuguese was carried out by three independent translators with professional fluency in English. They were instructed to produce colloquial translations that would be easily understood by the general population. Discrepancies were then discussed and reconciled by a specialist in the coagulopathy area, and a Portuguese native speaker who had not been involved in any of the forward translations.

Back translation of the reconciled version of the ISTH-BAT was then performed by an English translator. A literal translation was emphasized. This translator was neither involved in the previous steps, nor familiar with the original questionnaire. The working group then compared the back translation against the original ISTH-BAT in order to identify discrepancies in the concepts.

Since no conceptual discrepancies were found between the original English version and the European Portuguese translation, the newly translated ISTH-BAT was tested for cognitive equivalence in a group of 14 Portuguese-speaking respondents. We chose respondents drawn from the questionnaire target population: female hemophilia carriers and male hemophilia patients, with a mean age of 42 ± 10 years, all recruited in the coagulopathy reference center of São João Local Health Unit, in Porto, Portugal. They were asked to fill in the questionnaire, paying special attention to the construction and phrasing of the sentences, and ensuring that the concepts were interpreted appropriately. All patients' difficulties and opinions were analyzed and used as a basis for rephrasing. The final translated version developed during the previous steps was reviewed by the developers, who corrected any remaining spelling, grammatical, or other mistakes. The first author wrote the final report with the description of all translation and cultural adaptation decisions.

RESULTS

The authorization to translate and culturally adapt the original version of the ISTH-BAT [Appendix 1, Table 1 (Appendix 1: https://www.actamedicaportuguesa.com/revista/index.php/amp/article/view/22374/15587)] was obtained from the authors. In the translation process, several discrepancies occurred, namely, the expressions 'internal iliac ligation' and 'therapy'. All differences were discussed and resolved, and a final forward translation version was produced by consensus. The back translation was performed, and the resulting version showed no conceptual discrepancies with the original questionnaire.

Cognitive debriefing of the harmonized Portuguese version consisted of comprehensive reading and interpretation by seven hemophilia carriers and seven hemophilia patients from the coagulopathy reference center. We chose to implement a self-administered questionnaire to have a more realistic notion of the respondent's comprehension difficulties, thus avoiding procedural bias. The general opinion was that the questionnaire was easy to understand. Some medical terms, like 'epistaxis', 'antifibrinolytic', 'desmopressin', 'endometrial ablation', 'hysterectomy', 'menorrhagia', 'hemarthrosis', and 'hematuria' were pointed out as difficult to understand.

As the original ISTH-BAT is intended to be administered by a physician, we chose to culturally adapt the general terms used to define the site of bleeding, and we maintained drug classification terms because there was no way to simplify them. Another issue raised was the time frame of bleeding symptoms. The ISTH-BAT is intended to screen bleeding symptoms that may appear at any time, and we decided to include this information in the questionnaire header. The final European Portuguese version of the questionnaire [Appendix 1, Table 2 (Appendix 1: https://www.actamedicaportuguesa.com/revista/index.php/amp/article/view/22374/15587)] was reviewed and accepted by the working group.

DISCUSSION

To the best of our knowledge, no European Portuguese translation of the ISTH-BAT has been described in the literature.

We propose a European Portuguese version of the ISTH-BAT questionnaire, resulting from a formal translation and cultural adaptation process. Difficulties were identified after the first version was presented to patients. Some medical terms were difficult for patients to understand, which lead us to add colloquial terms next to medical terms in the European Portuguese version. Although this contributes to an easier comprehension of the questionnaire, it does not allow for this to be a self-ISTH-BAT version.

This study's main strength is the use of a formal methodology, which ensures that the obtained version maintains the same concepts as the original scale. The forward-backward translation process that we used is the most widely accepted method.[15] Similar methods have been used for the translation of this questionnaire into other languages.[11,16]

Study limitations include the absence of psychometric property analysis and its validation, including acceptability, test-retest reliability, internal consistency, and construct validity, which should be evaluated in the future. There is also a sampling bias, since all the individuals that tested the ISTH-BAT have the same bleeding disorder. In our opinion, this does not compromise questionnaire cognitive debriefing.

CONCLUSION

The present study reports the translation and cultural

adaptation of the ISTH-BAT to the European Portuguese language so it can be used in the diagnostic workup of suspected bleeding disorders in a primary care or hospital setting on the Portuguese population. Further studies are necessary for the validation of this new version.

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AUTHOR CONTRIBUTIONS

All authors contributed equally to this manuscript and approved the final version to be published.

PROTECTION OF HUMANS AND ANIMALS

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the Helsinki Declaration of the World Medical Association updated in October 2024.

DATA CONFIDENTIALITY

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

COMPETING INTERESTS

The authors have declared that no competing interests exist.

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