

An Assessment of the Readability and Comprehensibility of Online Resources on Biotinidase Deficiency

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Abstract

Background: Biotinidase deficiency is a metabolic disorder that is included in newborn screening programmes. This condition prevents individuals from performing biotin metabolism properly and can lead to serious health problems when left untreated. It is important that health resources are understandable and accessible for families to be informed about the disease and to be directed to the right treatment.

Objective: This study evaluates the readability and understandability of online resources on biotinidase deficiency, a metabolic disorder included in newborn screening programs. The aim is to determine whether these materials meet health literacy standards.

Methods: Fifty online documents were initially identified via Google searches using “biotinidase deficiency.” After excluding academic articles, duplicates, and inaccessible resources, 21 documents were analyzed. They were categorized as nonprofit (13) or private (8) based on domain extensions. Readability was assessed using Readable.io, providing Flesch Reading Ease scores. The Patient Education Materials Assessment Tool (PEMAT) was used to evaluate understandability and actionability, with scores averaged by four reviewers. Statistical analyses compared group differences.

Results: Private articles had significantly higher Flesch Reading Ease scores, indicating more difficult readability (mean $\pm$ SD: 13.9 ± 2.2 vs. 10.7 ± 2.0 ; $p = 0.002$). PEMAT understanding (U) scores showed no significant difference between private and nonprofit articles (mean $\pm$ SD: 52.0 ± 10.5 vs. 42.3 ± 11.4 ; $p = 0.060$). Similarly, actionability (A) scores were not significantly different (mean $\pm$ SD: 29.1 ± 20.0 vs. 13.4 ± 18.0 ; $p = 0.063$). Articles with lower readability (levels D and E) had significantly lower actionability scores compared to higher readability articles (levels A to C).

Conclusions: Most online biotinidase deficiency resources fail to meet health literacy standards, particularly in readability and actionability. Improving clarity and usability is essential to better support families managing this condition. The study emphasizes patient-centered approaches for creating effective health education materials.

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Original Manuscript

Evaluation of Readability and Understandability of Biotinidase Deficiency Information Online

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ABSTRACT

Objective: This study evaluates the readability and understandability of online resources on biotinidase deficiency, a metabolic disorder included in newborn screening programs. The aim is to determine whether these materials meet health literacy standards.

Methods: Fifty online documents were initially identified via Google searches using “biotinidase deficiency.” After excluding academic articles, duplicates, and inaccessible resources, 21 documents were analyzed. They were categorized as nonprofit (13) or private (8) based on domain extensions. Readability was assessed using Readable.io, providing Flesch Reading Ease scores. The Patient Education Materials Assessment Tool (PEMAT) was used to evaluate understandability and actionability, with scores averaged by four reviewers. Statistical analyses compared group differences.

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Conclusion: Most online biotinidase deficiency resources fail to meet health literacy standards,

particularly in readability and actionability. Improving clarity and usability is essential to better support families managing this condition. The study emphasizes patient-centered approaches for creating effective health education materials.

INTRODUCTION

Biotinidase deficiency is an inherited metabolic disorder that prevents the body from effectively reusing the vitamin biotin. Biotin, also known as vitamin B7 or vitamin H, plays an essential role in carbohydrate, fat and protein metabolism. When there is a deficiency or dysfunction of the enzyme biotinidase in the body, free biotin cannot be broken down, causing disruptions in critical points of metabolism. If this deficiency is not treated early, it can lead to permanent damage such as neurological retardation, loss of muscle tone, skin lesions and hearing loss. (1)(2)

Turkey is one of the countries with the highest incidence of biotinidase deficiency. While the worldwide incidence of biotinidase deficiency is approximately 1:60,000, the rate in Turkey is much higher, ranging between 1:11,000 and 1:13,000 according to studies. The national newborn screening program in Turkey revealed high incidence. Based on published data from Ministry of Health, the incidence is approximately 1:7,116 (3) (4)

This high rate is a reflection of the higher prevalence of genetically inherited diseases in our society where consanguineous marriages are relatively common. Biotinidase deficiency is included in the newborn screening program in many countries other than Turkey. This program aims to detect various metabolic and genetic diseases with a few drops of blood taken in the first few days after birth. The inclusion of biotinidase deficiency in the screening program is an important step in preventing the serious consequences of this disease. Biotinidase deficiency diagnosed early can be completely controlled with daily biotin supplementation and the progression of the disease can be prevented. (5)

Although the inclusion of biotinidase deficiency in the newborn screening program is an important public health achievement, it can sometimes cause unnecessary concerns in families. Parents who encounter “positive” or “suspicious” statements in screening results often panic and search for information about the disease on the internet without understanding the subject in depth. At this point, it is vital that the articles encountered are readable, reliable and presented in a language that families can understand. Unfortunately, many resources are either written in complex language full of medical terms or contain incomplete and inaccurate information.

Parents' level of health literacy directly affects the comprehensibility of the information they encounter on the internet. Therefore, content that provides information about diseases screened in the newborn screening program, such as biotinidase deficiency, should be written in plain language, explain medical concepts and provide accurate information. As health professionals, our responsibility in this regard is not to create unnecessary fear while conveying the seriousness of the

disease and to inform families correctly.

The National Institution of Health (NIH) and the American Medical Association (AMA) advise that a medical information's reading level should be at least sixth grade level or lower. (6) (7) Many studies, meanwhile, have revealed that there is plenty of written material on the Internet with a language use above the advised levels. (8) These difficult-to-read materials cause misunderstandings and panic in patients (9)(10)

Readability tests use formulas to evaluate words without looking at the integrity of the text. So, readability tests alone can't be utilized to assess how easy it is to understand a text. Teaching the Patient the Agency for Healthcare Research and Quality made the Material Evaluation Tool (PEMAT) to test the extent to which audiovisual and written information is understood and used by patients as a whole. (11) It is very important that an article is not only easy to read but also easy to understand by those who read it

In this study, we evaluated the quality, readability, understandability, and actionability of articles on the subject biotinidase deficiency in web sites produced for patients. We hypothesize that most of the written materials have low quality, readability, and understandability.

METHOD

WEB SEARCH

In October 2024, the first 50 results of a combined Internet search for "biotinidase deficiency" were observed individually using the Google search engine. The investigation comprised solely websites that were prepared for users, including patients. The study did not encompass websites that were specifically designed for physicians, such as Medscape and Up to date, websites that requested subscriptions or fees, personal blogs, and Google Scholar and PubMed (Figure 1). Plain text from the 21 websites that survived the exclusion was replicated onto a basic Microsoft Office Word page. Websites of universities and those ending in ".edu" and Websites that ended in ".org" or ".gov" and were classified as nonprofit (np) and those of practice physicians and healthcare facilities were classified as "Private."

FIGURE:1 Flow diagram showing the elimination of the sources.

Total number of web
Pages analyzed
(n=50)

3 no information
3 duplication

2 can't be reached
2 test catalog

Numbers after removing unreachable
Or duplicated pages
(n=40)

13 journal
6 medical sources

↓
Numbers after extracting with
medical sources
(n=21)

EVALUATION OF READABILITY

The readability of each webpage was evaluated utilizing the online tool Readable.io. This tool offers metrics including the Flesch-Kincaid Grade Level, Gunning Fog Index, SMOG Index, and Automated Readability Index, which together assess the grade level necessary for text comprehension. A mean grade level was determined for each webpage. Furthermore, the Flesch Reading Ease score was documented, with elevated scores signifying enhanced readability. Websites were evaluated on a scale from A (superior readability) to D (inferior readability) according to these findings. As the grade levels increase, readability of the text decreases.

EVALUATION OF UNDERSTENDABILITY

The Patient Education Materials Assessment Tool for Printable Materials (PEMAT-P) was used to evaluate understanding and actionability. PEMAT assesses the understanding of the content and its presentation of obvious, actionable instructions. The instrument includes 17 questions related to understandability and 7 regarding actionability, evaluated by four assessors (two physicians, one dietitian, and one non-health professional). Each question was categorized as "agree," "disagree," or "not applicable," and the scores were converted into percentages. The Microsoft Office Excel program was utilized for data entry, while the PEMAT Auto Scoring Form was employed for score calculation. Elevated percentages indicated greater understanding and applicability. The total points have been added up and given as a percentage for both clarity and practicality. As the percentage

increases, both comprehensibility and applicability also rise.

STATISTICAL ANALYSIS

A statistical analysis was conducted to compare readability and comprehensibility among website categories. Mean, standard deviation, median, minimum, maximum value, frequency and percentage were used for descriptive statistics. The distribution of variables is measured by Kolmogorov Smirnov and Shapiro-Wilk tests. Independent samples t test was used to analyze independent quantitative data with normal distribution. Mann-Whitney u tests was used in the analysis of independent quantitative data with non-normal distribution. Friedman test was used for the repeated measurement analysis. The effect level was tested with logistic regression. Correlation between observers were tested with Intra Class Correlation. SPSS 28.0 was used for statistical analyses.

This methodology facilitated a thorough assessment of the quality and accessibility of online resources regarding biotinidase deficiency, aiming to identify deficiencies and opportunities for enhancing patient education.

RESULTS

A total of 21 articles related to biotinidase deficiency were analyzed after excluding academic articles and institutional websites (e.g., Medscape, NIH). These articles were categorized into two groups based on their domain extensions: 13 articles belonged to the **nonprofit group** (edu, org, gov domains), and 8 articles to the **private group** (commercial domains). The study aimed to evaluate the readability and understandability of these articles using the Flesch Reading Ease score and PEMAT (Patient Education Materials Assessment Tool), focusing on the U (understanding) and A (actionability) scores. Detailed readability analysis of the web pages is listed in Table 1.

TABLE:1 Average Readability Levels and Grades After Categorization

	All Web Pages	Non-Profit Web Pages	Private Web Pages
A	1	1	0
B	4	4	0
C	8	6	2
D	4	1	3
E	4	1	3

After readability calculations were done, 1 web pages graded as A level (NP: 1, P:0), 4 as B level (NP: 4,P:0), 8 as C level (NP:6 ,P:2), 4 as D level (NP: 1, P: 3) and 4 as E level (NP:1 P:3) for language use.

The **Flesch Reading Ease** score was significantly higher in the private group compared to the nonprofit group (mean $\pm$ SD: 13.9 ± 2.2 vs. 10.7 ± 2.0 , $p = 0.002$). This indicates that private group articles were more challenging to read. However, there was no significant difference between the nonprofit and private groups in terms of the **U Score** (mean $\pm$ SD: 52.0 ± 10.5 vs. 42.3 ± 11.4 , $p = 0.060$) and the **A Score** (mean $\pm$ SD: 29.1 ± 20.0 vs. 13.4 ± 18.0 , $P = 0.063$ (Figure:2)

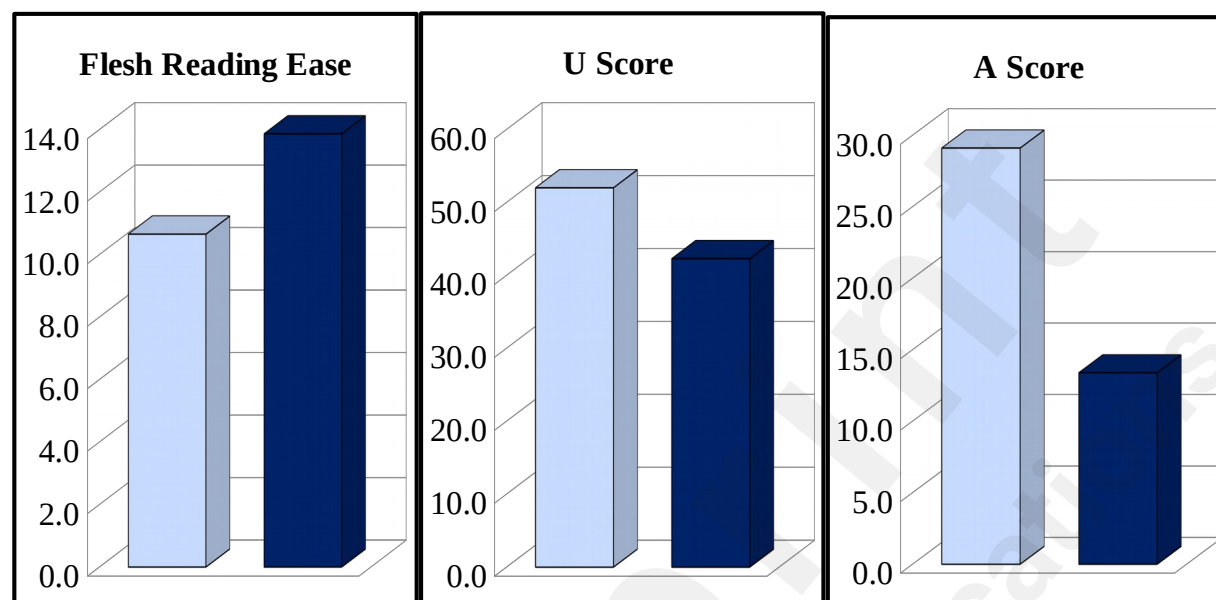


Figure 2: flesh reading ease, understandability and actionability score distribution of the groups.

Understanding (U Score) and Actionability (A Score) Analysis

There was no statistically significant difference in the **U Score** between the nonprofit and private groups (mean $\pm$ SD: 52.0 ± 10.5 vs. 42.3 ± 11.4 , $P = 0.060$). Similarly, the **A Score** did not show a significant difference between the two groups (mean $\pm$ SD: 29.1 ± 20.0 vs. 13.4 ± 18.0 , $p = 0.063$). These findings indicate that while private group articles were harder to read, their understandability and actionability were not significantly different from those of the nonprofit group.(table 2)

Table-2 : Readability and Understandability Scores, min-max and median values

		Min-Max	Median	Mean $\pm$ sd/n-%
Flesh Reading Ease		7.7 - 16.9	11.4	11.9 ± 2.6
U Score		23.7 - 73.7	50.0	48.3 ± 11.6
A Score		0.0 - 60.7	21.4	23.1 ± 20.4
Readability Score	A			1 4.8%
	B			4 19.0%
	C			8 38.1%
	D			4 19.0%
	E			4 19.0%

Categorization by Readability Levels

The articles were further classified into five readability levels (A to E), where A represents the easiest to read and E the most difficult. Articles categorized as D and E had significantly higher **Flesch Reading Ease** scores (mean $\pm$ SD: 14.5 ± 1.7) compared to those in categories A, B, and C (mean $\pm$ SD: 10.2 ± 1.3 , $p = 0.000$), confirming that readability decreases as the level moves from A to E.

While no significant difference in the **U Score** was observed between the A-B-C and D-E groups (mean $\pm$ SD: 51.8 ± 10.8 vs. 42.7 ± 11.1 , $p = 0.078$), the **A Score** was significantly lower in the D-E group compared to the A-B-C group (mean $\pm$ SD: 11.2 ± 16.3 vs. 30.5 ± 19.5 , $P = 0.018$). This indicates that while the overall readability might not correlate strongly with understandability, actionability tends to decline as readability worsens.

Observer Consistency in PEMAT Scoring

The PEMAT scoring for understanding (U) and actionability (A) was conducted by four independent observers. There was a high level of agreement among observers, with strong positive correlations observed for both the U Score (ICC = 0.832, $p = 0.000$) and A Score (ICC = 0.877, $P = 0.000$). This consistency highlights the reliability of the evaluation process.

DISCUSSION

The tendency of patients to use the Internet as a source of information has been increasing rapidly. Although this condition seems as an advantage for many, reliability and comprehensibility of the sources is the most important problem. Information presented to people on the Internet must be easy to read, understand, and apply. (12) In this study, we analyzed the readability and understandability of online documents on biotinidase deficiency, a disease screened in newborn screening.

The results of this study underline the significant need to enhance the readability and understandability of online health resources for biotinidase deficiency, as observed in similar studies conducted for other metabolic diseases such as phenylketonuria (PKU). (13) Accessible via internet searches, health information on biotinidase deficiency frequently reflects trends observed in other diseases like PKU, where readability grades exceeded the 6th-grade reading level advised by the NIH and AMA, so overcoming the recommended readability for the general population. (6)(7)

Research on Readability

Our data showed significant variations in readability scores between private and nonprofit group. Private source articles typically had higher Flesch Reading Ease scores, meaning more difficulty in reading than those from nonprofit sources. Such variations could result from the content's goal and target audience; private companies could be serving more medically educated customers. But since biotinidase deficiency mostly affects families with newly diagnosed children, too complicated

language can make comprehending and taking action difficult and possibly postpone required treatments.

Actionability and Understanding

Ideal averages for the PEMAT (Patient Education Materials Assessment Tool) index are generally considered to be 70% or higher. This indicates that the materials are sufficiently understandable and actionable for a wide audience. A comprehensibility score of around 50% for the articles screened in our study would be considered very low, especially for materials that provide health information. This may indicate that readers may have difficulty understanding the key messages in the material and that the content fails to provide actionable information.

Understandability (U) scores showed less variation while the actionability (A) scores for more difficult texts (D-E readability levels) were often lower. This means that although readers could understand the overall knowledge, they could find it difficult to come up with achievable strategies for addressing the disorder. These results coincide with observations in PKU studies, where low engagement and practical applicability of the material linked with high readability grades.

Challenges in Health Literacy

High readability levels have broader consequences for health literacy, especially for inherited metabolic diseases like biotinidase deficiency, where rapid understanding and action are absolutely vital. Families learning a new diagnosis often start their search for information online, thus it is crucial that online resources be easily available and practical. Inaccurate interpretation, increased anxiety, and delayed decision-making can all result from poorly designed resources.

Recommendations

The following approaches are suggested as solutions for these difficulties:

- 1. Simplify Language:** Reducing medical jargon and using simple sentences will help to increase readability and understandability of language. For instance, changing terms like "enzyme deficiency" for "lack of a protein that helps process vitamins" might help grasp.
- 2. Trying a more visual presentation:** Visual aids, headings, and bullet point summary of material help retention and understanding in structured layouts.
- 3. Prioritize actionable elements:** Clear directions, such "Consult your doctor about biotin supplements," says clear instructions that maximize the value of the material.

Research Limitations

This research has several limitations. With only 50 papers included and 21 articles analyzed, the sample size was rather small and might restrict the generalizability of the results. Second, the subjective nature of the understanding scores (U scores) underlined the reliance on human judgment since the viewpoint of the individual evaluator affected them. Third, the whole evaluation process was done by hand, which might bring prejudice and human mistake. Greater datasets and automated tools would help future research to reduce subjectivity and improve repeatability.

In general, this study has been done to evaluate the online written materials on the subject biotinidase deficiency presented to patients. To the best of our knowledge, no previous study on readability and understandability of biotinidase deficiency has been conducted.

According to the data collected in our study, the readability index of the texts written in this field is generally low and the comprehension index is fifty percent on average, which is below the recommended values. It has been shown that in different disciplines, same results were also found (14)(15) We recommend that when writing online written content, simpler words and sentences are utilized, medical terminology be used with caution (explanations must be provided), and objective details be comprised.

CONCLUSION

All things considered, empowering families and ensuring optimal management of the condition rely on providing online resources more easily available for biotinidase deficiency. Utilizing knowledge from related studies on PKU and other metabolic diseases, efforts should concentrate on producing patient-centered materials compliant with health literacy guidelines and so supportable actionable results.

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