

Assessment of thrombocytopenia symptoms using vignettes

DOI: 10.7365 / JHPOR.2015.2.9

Authors:

**Monika Szkulciecka-Dębek¹,
Mariola Drozd², Marta Bem³,
Nina Kiepuska¹, Joanna Mazur⁴**

*1- Department of Dermatology, Military
Institute of Medicine, Warsaw*

*2- Department of Applied Pharmacy, Medical
University of Lublin*

*3- Faculty of Social Science, Warsaw Uni-
versity of Life Sciences – SGGW*

*4- Department of Child and Adolescent
Health, Institute of Mother and Child
Warsaw*

Keywords:

quality of life, thrombocytopenia, vignettes

Abstract

Objective: Our aim was to create a vignette enabling the assessment of the impact of thrombocytopenia symptoms on patients' quality of life.

Methods: The vignettes which we created are focused on thrombocytopenia symptoms which may influence the quality of life of the patients. The vignettes designed in Polish language are to be used by adult patients and children caregivers and to be administered by an interviewer. There are separate vignettes for adult males and females as well as for children caregivers. Each vignette consists of two parts: the objective of the first one is to collect qualitative information about the major health problems related to the thrombocytopenia symptoms in each studied group, the second is related to specific symptoms and aims at measuring the intensity of impact on the quality of life in a quantitative way.

Results: We have created the vignettes with special focus on the most frequent thrombocytopenia symptoms and those which are most life threatening. The vignettes describe a hypothetical patient with thrombocytopenia and the symptoms which may impact the quality of life in their daily functioning.

In the first part the patients are asked to provide qualitative information in a descriptive way about symptoms influencing their daily functioning or the most threatening ones. The second part is using the Likert scale to assess quantitatively the impact of specific symptoms on patients' life.

Conclusions: The vignettes need to be tested in adult patients with thrombocytopenia and children caregivers in order to allow further comparisons.

Introduction

Quality of Life (QoL) questionnaires are used by researchers widely, however there are still areas for capturing additional information using other tools, such as vignettes. According to Collins, vignettes are short descriptions of hypothetical situations or scenarios, and are useful in understanding how respondents would answer questions about these situations and in showing whether the conceptual boundaries of the domain vary as between respondents^[1]. In our previous research we assessed the potential usage of vignettes in relation to quality of life^[2]. We found examples where a clinical vignette can be applied to a single patient, and the information giv-

en during an interview can provide additional explanation to patient's scores in the study of quality of life. The objective of our previous search was to define, in real life, the vignettes are used for quality of life or utilities assessment and if yes, how extensive their use is^[2]. We found that this is a promising additional technique to measure changes in QoL, however it is not very often used by the researchers. It can be considered as a supplementary method to standard QoL measurement methods. In view of the above findings we decided to work on creating a disease and symptoms specific vignette. We agreed to focus on thrombocytopenia, which as a congenital disease is relatively rare. However as a hematological problem it is quite common and there are many acquired diseases which may cause thrombocytopenia^[3].

The symptoms of thrombocytopenia influence patients' quality of life independently of the disease cause. Thrombocytopenia most characteristic symptoms are petechiae, purpura as well as skin and mucosa bleedings. Mostly the localization of the petechiae is on the extremities and the thorax, more rarely on the face^[4]. The most frequently bleedings are from the nose, gums, urinary tract and genital tract in women. The life threatening complication can be intracranial bleeding^[4]. Those symptoms can impact patients quality of life and with the vignettes we aimed to have better understanding of such influence.

Methods

By drawing our attention to a clinical condition we wanted to use the socio and psychological techniques to measure the impact of disease symptoms in qualitative way^[5,6,7]. To ensure proper understanding of the clinical situation we described a hypothetical situation in a clear manner, adapted to the patient level. That is mandatory in order to place the interviewed person in a clinical context allowing proper understanding of the situation by the patient. The first step was to define the hypothesis for the problem we plan to analyze with the vignettes. In this case we decided to assess the most frequent symptoms of the disease which may have impact on patients quality of life^[8,9]. We were trying to find out what the symptoms which most frequently produce fear and concern to patients with thrombocytopenia are. In order to keep the vignette understandable we decided to prepare separate vignettes for adult males, females and children caregivers and to concentrate on the more frequent symptoms: the appearance of small petechiae in the skin and mucous membranes of the mouth, easy bruising, bleeding from the gums, nose, genital tract in women and prolonged, heavy menstrual bleeding being the cause of the possibility of bleeding from the genital tract. Additionally, we added a description related to the life threatening complication of thrombocytopenia which is the risk of bleed-

ing to the central nervous system to the first part of the vignette. Patients are asked to provide descriptive information about those symptoms which cause limitations or influence their daily activities.

In the second part of the vignette the Likert scale is used in order to provide quantification to the impact of specific symptoms and clinical conditions related to thrombocytopenia on patients functioning^[7]. Patients are asked to rank on a 5 level scale (+2,+1,0, -1,-2) each of the symptoms mentioned in the first part, +2 meaning very significant impact, +1 mean impact, 0 - indifferent, -1 small impact, -2 no impact.

We have validated the vignettes within psychologists and clinicians in order to ensure proper context description. The vignettes are available only in Polish language. For other languages translation and validation process should be followed.

Results

We have created the vignettes with special focus on the most frequent thrombocytopenia symptoms and those most life threatening producing particular fear and concern to patients. The vignettes describe a hypothetical patient with thrombocytopenia and the symptoms which may impact the quality of life in daily functioning. We have not personalized the vignettes with details such as names in order to make the tool more flexible allowing the optimal identification and usage in a non-preselected group of patients.

In the first part of each vignette patients are asked to provide qualitative information in a descriptive way about symptoms influencing their daily functioning or threatening the most. The second part is using the Likert scale with 5 steps to assess quantitatively the impact of specific symptoms.

The vignette presented in **figure 1** is dedicated to adult male patients. The **figure 2** vignette presents the vignette dedicated to female patient. The **figure 3** vignette is for thrombocytopenia children caregivers.

Opis:
Mężczyzna w wieku średnim cierpi na małopłytkowość. Jest aktywny zawodowo i w wolnym czasie lubi uprawiać sport. Jednak występują objawy choroby, które utrudniają udział w codziennych aktywnościach. Od jakiegoś czasu zauważył pojawienie się drobnych wybroczyn na skórze i śluzówkach jamy ustnej. Dodatkowo stwierdza łatwe siniaczenie się i czasami nawet krwawienia z dziąseł lub nosa. W związku ze swoją chorobą odczuwał lek przed urazami, jako że wiąże się to z ryzykiem krwotoków wewnętrznych. Boi się urazów głowy, które mogą prowadzić do poważnych konsekwencji.

Część 1.
Proszę opisać, które z objawów związanych z chorobą utrudniają Panu w czynności dnia codziennego, ograniczając aktywność?
.....
Które powodują u Pana szczególne obawy?
.....

Część 2. Proszę wskazać stopień utrudnienia w wykonywaniu codziennych czynności w związku z następującymi objawami choroby (przy każdym objawie proszę wybrać jedną odpowiedź).
Wybroczyny na skórze i/lub śluzówkach
-2 duży wpływ
-1 średni wpływ
0 obojętne
+1 mały wpływ
+2 brak wpływu
Łatwe siniaczenie
-2 duży wpływ
-1 średni wpływ
0 obojętne
+1 mały wpływ
+2 brak wpływu
Krwawienia z dziąseł lub nosa
-2 duży wpływ
-1 średni wpływ
0 obojętne
+1 mały wpływ
+2 brak wpływu

Figure 1. Adult male's with thrombocytopenia vignette

Opis:
Kobieta w wieku średnim cierpi na małopłytkowość. Jest aktywna zawodowo i w wolnym czasie lubi uprawiać sport. Jednak występują objawy choroby, które utrudniają jej udział w codziennych aktywnościach. Od jakiegoś czasu zauważyła pojawienie się drobnych wybroczyn na skórze i śluzówkach jamy ustnej. Dodatkowo stwierdza łatwe siniaczenie się i czasami nawet krwawienia z dziąseł, nosa lub dróg rodnych, przedłużające się miesiączki. W związku ze swoją chorobą odczuwa lek przed urazami, jako że wiąże się to z ryzykiem krwotoków wewnętrznych. Boi się urazów głowy, które mogą prowadzić do poważnych konsekwencji. Również towarzyszy jej lek związany z przedłużającym się, obfitymi miesiączkami i istniejącą możliwością krwotoku z dróg rodnych.

Część 1.
Proszę opisać, które z objawów związanych z chorobą utrudniają Pani czynności dnia codziennego, ograniczając aktywność?
.....
Które powodują u Pani szczególne obawy?
.....

Część 2. Proszę wskazać stopień utrudnienia w wykonywaniu codziennych czynności w związku z następującymi objawami choroby (przy każdym objawie proszę wybrać jedną odpowiedź).
Wybroczyny na skórze i/lub śluzówkach
-2 duży wpływ
-1 średni wpływ
0 obojętne
+1 mały wpływ
+2 brak wpływu
Łatwe siniaczenie
-2 duży wpływ
-1 średni wpływ
0 obojętne
+1 mały wpływ
+2 brak wpływu
Krwawienia z dziąseł lub nosa
-2 duży wpływ
-1 średni wpływ
0 obojętne
+1 mały wpływ
+2 brak wpływu

Figure 2. Adult Female's with Thrombocytopenia vignette

Opis:

Dziecko chorujące na małopłytkowość jest bardzo aktywne, lubi biegać, grać w piłkę, jeździć na rowerze. Jednak występują objawy choroby, które utrudniają udział w codziennych aktywnościach. Od jakiegoś czasu rodzice zauważyli pojawienie się drobnych wybroczyn na skórze i śluzówkach jamy ustnej dziecka. Dodatkowo zaobserwowali łatwe siniaczenie się i czasami nawet krwawienia z dziąseł lub nosa. Rodzice zaobserwowali, że ich dziecko w związku z chorobą odczuwa lęk przed urazami, na przykład podczas zajęć z w-f. Rodzice dodatkowo boją się urazów głowy, które mogą prowadzić do poważnych konsekwencji w postaci krwotoków.

Część 1.

Proszę opisać, które z objawów związanych z chorobą utrudniają Pana/Pani dziecku czynności dnia codziennego, ograniczając jego aktywność?

.....

Które powodują u Pani/Pana lub u dziecka szczególne obawy?

.....

Część 2. Proszę wskazać stopień utrudnienia w wykonywaniu codziennych czynności przez dziecko w związku z następującymi objawami choroby (przy każdym objawie proszę wybrać jedną odpowiedź).

Wybroczyny na skórze i/lub śluzówkach

-2 duży wpływ
-1 średni wpływ
0 obojętne
+1 mały wpływ
+2 brak wpływu

Łatwe siniaczenie

-2 duży wpływ
-1 średni wpływ
0 obojętne
+1 mały wpływ
+2 brak wpływu

Krwawienia z dziąseł lub nosa

-2 duży wpływ
-1 średni wpływ
0 obojętne
+1 mały wpływ
+2 brak wpływu

Figure 3. Children with thrombocytopenia caregivers' vignette

Discussion

Patients with thrombocytopenia may suffer from the impact of the disease on their quality of life thus we looked for research already done to assess such impact. Despite the availability of numerous publications about diagnosis, treatment, and prognosis of the disease there are few publications related to quality of life, especially those related to children. QoL related to Idiopathic Thrombocytopenic Purpura (ITP) was mostly studied using Kids' ITP Tools (KIT) questionnaire, comparing the mean KIT score in the parents' group and in the children's group^[10]. Another example when KIT tool was used was to assess the quality of life in immune thrombocytopenia following the treatment. The authors concluded that the KIT parent-proxy scores were significantly worse for newly diagnosed children receiving treatment^[11]. In Egypt there was a QoL study performed with thrombocytopenia children participation and the results showed a positive correlation between scores of child/proxy and parent reports. The studied group consisted of children with immune thrombocytopenia and their parents. Authors assessed the QoL in relation to different disease parameters. The findings showed that newly diagnosed patients with immune thrombocytopenia had lower scores of both child/proxy reports and parent reports than chronic patients.

Longer duration of illness was correlated with higher scores of child/proxy reports^[12]. Another study in children population aimed to assess QoL using Pediatric Quality of Life Inventory™ and Kids' ITP Tools questionnaires at diagnosis and during standardized follow-up^[13]. The authors found that the quality of life of children with newly diagnosed immunothrombocytopenia was only influenced by clinical course of the disease and not by treatment modality or bleeding severity^[13].

While analyzing the available publications we focused not only on thrombocytopenia quality of life research but also we searched for the tools used by researchers. Taking into consideration that most symptoms are only experienced subjectively using self-report is crucial for early symptom detection and in their paper Kirsh et al analyzed the importance of patients reported outcomes and described what are the key aspects when developing new tools^[14]. Authors underlined that the treatment choice should rely more on symptoms underscoring the importance of incorporating the patient's perspective by using patient-reported outcomes (PROs) and when preparing a PRO tool the proper methodology should be followed as well as a collaboration between clinicians, scientists, statisticians, and patients is needed^[14].

Since communication with sick children may be particularly difficult, at the European Cancer Congress in 2005 Rodrigues presented a poem based communication project with regards to children having thrombocytopenia related to cancer. The one-page document containing a poem and cartoons was prepared in a discussion with target children and their parents. It explains to children what symptoms such as bruises to be alert to. It also urges children to notify parents of strong headaches. The intention was to reduce children's anxiety in relation to symptoms and not their evaluation. The poem was planned to be included in an information folder given to children with cancer and the folder would be used by the medical staff to explain to the children their symptoms and how to cope with them^[15]. However we have not identified any vignette studies used to provide information on quality of life in patients with thrombocytopenia. In view of the above our vignettes proposal can provide additional input into further research in that area.

Conclusions

The vignettes we have created are ready to use and validated in clinical and psychological context. The next step will be testing them in adult patients with thrombocytopenia and children caregivers in order to allow further comparisons and research. ■

References

1. Collins D., Pretesting survey instruments: An overview of cognitive methods. *Quality of Life Research* 12: 229–238, 2003
2. Szkultecka-Dębek M., Drozd M., Bem M., et al. Is the vignette method used to assess quality of life in practice? *Curr. Iss. Pharm. Med. Sci.* 2015 vol. 28 nr 1 s. 8-12
3. Burgdorf WHC., Plewig G., Wolff HH., Llandthler M. Braun Falco *Dermatologia*, red. Naukowa W.Gliński, tomII, 963-966
4. Kaszuba A., Adamski Z., *Leksykon Dermatologiczny*, tom I, 352-354
5. Babbie E. *Podstawy Badań Społecznych*, 2008 r. Wydawnictwo Naukowe PWN
6. *A Guide to Writing Multiple Choice Items*, American Academy of Neurology, 2013 American Academy of Neurology Institute
7. Université de Montreal, Etape3: rédiger le vignettes et items Available from: <https://www.cpass.umontreal.ca/recherche-et-developpement/test-de-concordance-de-script-/construire-un-tcs-/etape-3-rediger-le-vignettes-et-items.html>; [Accessed: 28.01.2016]
8. Zawilska K., *Płytkowe skazy krwi*, w *Podstawy hematologii*, praca zb. pod red. A. Dmoszyńskiej i T. Robaka, Czelej, Lublin 2008
9. Medeiros D., Buchanan GR. Major haemorrhage in children with idiopathic thrombocytopenic purpura: immediate response to therapy and long-term outcome. *J. Pediatr.* 1998, 133 (3), 334–339
10. Zilber R., Bortz AP., Yacobovich J et al. Analysis of health-related quality of life in children with immune thrombocytopenia and their parents using the kids' ITP tools.; *J Pediatr Hematol Oncol.* 2012 Jan; 34(1): 2-5
11. Grainger JD., Young NL., Blanchette VS., Klaassen RJ. Quality of life in immune thrombocytopenia following treatment. *Arch Dis Child.* 2013 Nov; 98(11): 895-7
12. Mokhtar GM., Farid SM., Shaker NM., Farrag KE. Health-related quality of life of Egyptian children with immune thrombocytopenia and their parents. *J Pediatr Hematol Oncol.* 2014 Apr; 36(3): 194-9
13. Heitink-Pollé KM., Haverman L., Annink KV., et al. Health-related quality of life in children with newly diagnosed immune thrombocytopenia. *Haematologica.* 2014 Sep; 99(9): 1525-31
14. Kirsch M., Klaassen RJ., De Geest S., et al. Understanding the importance of using patient-reported outcome measures in patients with immunothrombocytopenia. *Semin Hematol.* 2013 Jan; 50 Suppl 1: S39-42
15. S. Rodrigues. “Anaemia and me”, “Thrombocyte my friend”, “A little story about neutropenia” poems to explain thrombocytopenia, anaemia and neutropenia and their symptoms to children. Available from: https://www.europecancercongress.org/ecco_content/ECCO13_abstractbook/files/assets/seo/page506.html;