

Experiences of PNH patients on eculizumab treatment with remote consultations

A generic explorative qualitative study

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ABSTRACT

Title: Experiences PNH patients on eculizumab treatment with remote consultations

Background: Due the COVID19-pandemic a shift took place from physical consultations to remote consultations in patients with Paroxysmal Nocturnal Hemoglobinuria (PNH). Currently, it is needful to gain knowledge about PNH patients' perspective of remote consultations, because patient involvement is crucial in remote consultations and an increase of the use of remote consultations is expected.

Research question: What are the experiences of PNH patients on eculizumab treatment with remote consultations?

Method: An explorative generic qualitative research was performed from January to June 2021. Data was collected through semi-structured interviews with purposefully sampled patients. The interview guide consisted of the following topics: experiences of physical and remote consultations, differences between physical and remote consultations, and the use of technology. Thematic analysis was used for data-analysis.

Results: Twelve patients with maximum variation applied to gender, age and travel distance were interviewed. Three main themes were derived from the data: *personal pros and cons*, *confidence with technology*, and *future perspective*. Patients described a remote consultation as a good addition to the physical consultations, but it is not seen as a definitive substitution. A preference has been expressed for an equal alternation between remote consultations and physical consultations.

Conclusion: Several advantages and disadvantages of remote consultations were identified within the main themes. A majority of the patients considered the remote consultations as a positive experience, even patients who did not believe the remote consultations provided the best care.

Recommendations: Remote consultations will be further implemented within PNH patients. Future longitudinal analysis regarding remote consultations should be performed in a larger group of patients. Also, research about the experiences of practitioners providing remote consultations and the economic factors is recommended.

Key words: Remote consultations, Paroxysmal Nocturnal Hemoglobinuria, Eculizumab, Qualitative research

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SUMMARY (DUTCH)

Titel: Ervaringen van PNH-patiënten onder behandeling met eculizumab van een consult op afstand.

Inleiding: De COVID19-pandemie heeft een verschuiving veroorzaakt van het gebruik van fysieke consulten naar het gebruik van consulten op afstand bij patiënten met Paroxysmale Nachtelijke Hemoglobinurie (PNH). Momenteel is er behoefte aan meer kennis over ervaringen van consulten op afstand van PNH-patiënten, omdat patiëntbetrokkenheid cruciaal is in het gebruik van consulten op afstand.

Onderzoeksvraag: Wat zijn de ervaringen van een consult op afstand bij PNH-patiënten met behandeling met eculizumab?

Methode: Een exploratieve generieke kwalitatieve studie werd uitgevoerd van januari tot juni 2021. Data werd verzameld met semigestructureerde interviews met doelbewust geselecteerde patiënten. De interview guide bevatte de volgende topics: ervaringen van fysieke consulten en consulten op afstand, verschillen tussen fysieke consulten en consulten op afstand en het gebruik van technologie. De analyse vond plaats met thematische analyse.

Resultaten: Twaalf patiënten, waarbinnen maximale variatie werd toegepast op geslacht, leeftijd en reisafstand, werden geïnterviewd. Drie hoofdthema's werden afgeleid uit de data: *persoonlijke voor- en nadelen*, *vertrouwen in technologie* en *toekomstperspectief*. Patiënten beschreven een consult op afstand als een goede aanvulling op een fysiek consult, maar het wordt niet gezien als een vervanger. Een voorkeur werd uitgesproken voor een gelijkmatige afwisseling tussen consulten op afstand en fysieke consulten.

Conclusie: Meerdere voor- en nadelen van consulten op afstand werden geïdentificeerd binnen de thema's. Een meerderheid van de patiënten ervaart consulten op afstand als positief, evenals patiënten die op voorhand negatief tegenover consulten op afstand stonden.

Aanbevelingen: Consulten op afstanden worden verder geïmplementeerd binnen PNH-patiënten. Verdere longitudinale analyse met betrekking tot consulten op afstand dienen bij een grotere groep patiënten te worden uitgevoerd. Ook onderzoek naar de ervaringen van behandelaren en de economische factoren is aanbevolen.

Kernwoorden: Consulten op afstand, Paroxysmale Nachtelijke Hemoglobinurie, Eculizumab, Kwalitatief onderzoek

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INTRODUCTION

Telemedicine has opened up a wide array of possibilities in communication provision in healthcare and is being driven by the need to improve patient empowerment¹⁻². Remote consultations, a form of telemedicine, brought new potential in healthcare, especially for outpatient resources³⁻⁴. A remote consultation is defined as: “*Consultation by remote telecommunications, generally for diagnosis or treatment of a patient at a site remote from the patient or physician*”⁵. The use of remote consultations for interactions allows patients to speak and/or see their physician whilst being in their comfort⁵.

The current literature states that remote consultations offer potential advantages to patients and healthcare system³. Reported benefits are improved access to healthcare while reducing traveling time preserving patient satisfaction⁶. In addition, at this time remote consultations are an important way for patients to access care in context of the COVID19-pandemic⁷. Consternation has been expressed that remote consultations may bring technical and logistical challenges⁶.

Currently, remote consultations are frequently used in hospitals for patients who have an outpatient appointment several times a year, a long travel distance to the hospital, and in patients with decreased immunity⁷⁻⁸. In the Netherlands, high-quality care for rare disorders is centralized in centers of excellence⁹. As a result, patients requiring specialized care may have a long travel distance to the hospital. This can be perceived as unpleasant, especially when several visits a year are necessary⁶. In the Netherlands, patients with Paroxysmal Nocturnal Hemoglobinuria (PNH) receive specialized care requiring several medical visits per year.

PNH is a rare acquired disorder of the hematopoietic stem cell that makes blood cells more sensitive to complement activation. The complement system is a part of the own innate immune system¹⁰. PNH is characterized by hemolysis causing hemoglobinuria and anemia, bone marrow failure, and an increased risk of thrombosis¹⁰⁻¹¹. The incidence of PNH is estimated at 0.1-0.2/100.000 persons a year worldwide and occurs in patients of all ages¹². The treatment of PNH consists of the intravenous administration of eculizumab every fourteen days. Eculizumab is a humanized antibody that prevents complement activation and thus reduces the degree of hemolysis and the risk of thrombosis^{10,12}. A disadvantage of eculizumab is a decreased immunity¹².

Eculizumab is administered at the patients' homes by home care nurses since 2015. Since then, patients no longer have to visit the hospital every fourteen days. Previous research has

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shown that this development resulted in an improvement of patients' psychological well-being¹¹. At this time, PNH patients only attend the hospital for an outpatient appointment every three months to ensure appropriate monitoring. The medical appointments are carried out by a hematologist or physician assistant (PA).

Since the COVID19-pandemic, in PNH patients, a shift took place from outpatient consultations to remote consultations. Current practice and previous research show indications that the experiences of remote consultations may depend on age and gender of the patient and the travel distance to the hospital¹³. These indications mainly seem to show that young patients with a long travel distance experience a video consultation more positively than older patients with a relatively short travel distance to the hospital¹³. But whether these indications are correct and what the further experiences of remote consultations are is unknown.

Given the lack of research concerning the experiences of remote consultations in PNH patients, the first step is to conduct a qualitative study to explore these experiences. Gaining insight into patients' experiences is important because patient involvement is crucial in remote consultations and the use of remote consultations is expected to increase.

Aim

The aim of this study is to explore the experiences of PNH patients on eculizumab treatment with remote consultations.

METHOD

Design

This study was performed using a generic explorative qualitative design. This design is the best-suited design for this study because this study focused on understanding the experiences of remote consultations of a small patient group with a lot of information about the topic¹⁴⁻¹⁶. An explorative approach was required because no previous qualitative research concerning remote consultations and PNH patients exists. To improve comprehensiveness, this study is reported using the consolidated criteria for reporting qualitative research (COREQ)¹⁷.

Population & Domain

This study was conducted on the hematology outpatient clinic of the Dutch PNH center of excellence. Patients who were included in this study: 1) were diagnosed with PNH, 2) were under treatment with eculizumab, 3) received a remote consultation since December 2020, and 4) were aged 18 years and older. Patients who were unable to read and speak the Dutch language, were excluded from this study. A heterogeneous purposive sampling method was selected to provide a diverse range of patients relevant to the objective^{15,18}. Since it was known that age, gender, and travel distance may influence patients' experiences maximum variation was applied on these characteristics^{13,19}.

Data collection

Data were collected through interviews using a peer-reviewed semi-structured interview guide (Appendix 1). The interview guide was developed using sensitizing concepts; previous studies aimed at remote consultations^{13,20-21} were studied to gain insight into relevant topics²². The interview guide consisted of the following topics: experiences of physical and remote consultations, differences between physical and remote consultations, and the use of technology. In response to emerging data, the interview guide was continuously adapted. Open-ended questions were used, so the patients were given freedom to answer, without limiting them with predefined answers¹⁵. Field notes were written to capture non-verbal communication and important perspectives directly after an interview to not disturb the interview¹⁵. The following demographic data were collected: 1) sex, 2) age, 3) job status, 4) treatment duration with eculizumab, 5) travel distance to the hospital, 6) means of transportation to the hospital, and 7) possibility to travel to the hospital independently. Interviews were conducted by one investigator and were audio-recorded. Interviews took place by video in context of the COVID19-pandemic and the interviewer was still able to pay attention to non-verbal communication.

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Data analysis

Thematic analysis was performed, because it is a flexible method and it offered a usable framework for doing the analysis²³⁻²⁴. Data was assigned to inductively formed themes, to ensure the data fully directed the analysis process²⁴. The analysis started with verbatim transcription of the audio-taped interviews and fieldnotes. During this process, memos were written containing thoughts about the data^{19,25}. After three interviews, transcripts were re-read to identify meaningful fragments. An in vivo coding process was performed twice by the investigator, once manually on printed transcripts and one week later on digitally transcripts to gain intra-rater reliability²⁶. In vivo coding prevented the investigator from imposing his own framework on the data¹⁵. The research question of this study was continuously kept in mind, to ensure the analysis finally led to answers to the research question. To gain inter-rater reliability three interviews were coded independently by two investigators²⁶. When this led to different codes discussion between the investigators took place until consensus was reached. Constant comparison of the codes was performed after a cluster of three interviews. After coding, codes were grouped into themes with similar meaning linked to the same phenomenon. Finally, two investigators identified which aspects of data were captured, what was interesting about the themes, and how the themes fit together. NVivo (version 12) was used during the iterative analysis process to manage the data accurately¹⁵.

Procedures

Eligible patients were asked by their practitioner whether they were interested in participating the study. Interested patients were contacted by the investigator by email for sending the patient information letter (PIF). The PIF described that the PA, known to the biggest part of the patients, was the investigator and interviewer of this study. All approached patients already filled in informed consent (IC) for participating in a non-Medical Research Involving Human Subjects Act (WMO) study in the center of excellence, so a signed IC form specifically for this study was not required. Two weeks after sending the PIF, the investigator contacted the patient by telephone whether the information was clear and whether the patient agreed to participate in the study. After agreement, an appointment for the interview was made. Interviews were planned during a home administration of eculizumab, so it leads to a minimum possible burden on the patients' time. All interviews were conducted in the home setting so it was not a factor that could lead to different interview data¹⁵.

Ethical issues

This study is conducted following the WMO and the principles of the Declaration of Helsinki. The Medical Research Ethics Committee (METC) of the Radboudumc declared this study not

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WMO mandatory because no invasion on the physical or psychological integrity of the patient was made. All data were handled anonymously and confidentially. Interviews were performed by the application Zaurus because it established a secure connection, so privacy was guaranteed²⁷.

RESULTS

Participants

Twelve interviews were taken from February to May 2021. The baseline characteristics are presented in Table 1. Six of the included patients were female, the age of the patients ranged from 26 to 80 years, and the travel distance to the hospital ranged from 53 to 211 km. Interview duration ranged from 29 to 45 minutes.

<Insert Table 1>

Themes

Based on the analysis of the interviews, three themes were identified: *personal pros and cons*, *confidence with technology*, and *future perspective*. See Figure 1 for the relation between the three themes and nine subthemes. New themes stopped emerging after twelve interviews, and thus meaning saturation was reached²⁸. Quotes were used to demonstrate the essence of the themes²⁹.

<Insert Figure 1>

Theme 1: Personal pros and cons

Subtheme 1: Impact on daily life

On the day of the remote consultation, less impact on daily life is experienced in comparison with a physical consultation. When a remote consultation takes place in the patients' environment no travel distance to the hospital has to be bridged. This leads to more time savings.

I don't spend a whole day when I have a remote consultation, it takes me only 30 minutes, so it does not have much impact on my work. When I have to go to the hospital for a physical consultation, it takes me a whole day. I always take a day off from work when I have a physical appointment. – Patient 08

However, several patients describe the travel distance to the hospital as relaxing, which makes it feel like escaping the daily routine. A remote consultation, compared to a physical consultation, has less impact on the physical condition for some patients. Because a remote
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consultation takes less time, the patients experience more energy and less fatigue on the day of and the day after the consultation. Additionally, less frequent hospital visits may cause PNH to be less prominent in daily life for some patients. This is perceived as positive and negative as illustrated in quotes underneath. The exact impact a patient experiences of a remote consultation on the day of the consultation depends on the patient work status and the travel distance to the hospital.

For me, the added benefit of remote consultations is that PNH is less present at the forefront of my life. However, every fourteen days I will still receive the eculizumab, so the reminder that I have PNH keeps coming back. – Patient 05

When I no longer have to come to the hospital, it may seem like I am no longer sick at all, but of course I am. PNH is just going on, and the day I have to come to the hospital will make me take PNH seriously and keep acknowledging that I have this disease. – Patient 10

Subtheme 2: Affection

During a remote consultation, patients perceived less bonding and contact with their practitioner. Because the practitioner was not directly physical opposite the patient during the consultation, patients missed some personal contact and the consultation, therefore, felt somewhat less official. During a physical consultation, the patient and the practitioner can pay more attention to non-verbal communication. Patients experienced that less attention was paid to informal chats during the remote consultation.

Umh... well, I think it is important to maintain physical consultations, because when there would be only remote consultations I think you will lose the affective bond with your practitioners. – Patient 02

Subtheme 3: Assessment of medical condition

When a remote consultation is performed, there is no possibility to perform a physical examination. A number of patients say this is a shame because they consider it as a required part of a consultation. Other patients argue that physical examination is mainly of added value if the patient has physical complaints.

When I feel good about myself, I have no reason to think that there is something physically wrong with my health and that physical examination should be performed. So in my opinion it depends on how I feel myself. – Patient 10

Patients expect that the content of a remote consultation should be comparable to the content of a physical consultation and thus physical complaints, the recent blood results, and new developments will be discussed. In a remote consultation, patients must have blood samples taken at the nearest blood collection point prior to the consultation. All patients think it is important that these blood results will be discussed during the consultation.

When the remote consultation is not combined with blood tests, there would be too little control over my illness. For me, the nearest blood collection point is only a few minutes from home or work, so it is only a short drive. – Patient 08

When having a physical consultation, many patients prepare the consultation during the travel time to the hospital. With a remote consultation, the patients do not have any travel time, which means that patients experienced to be less prepared for the consultation.

Subtheme 4: Contact with experience experts

More frequent use of remote consultations means that patients will be in the hospital less often. As a result, patients will have less contact with fellow PNH-patients and nurses from the hospital. Most patients experience this as a pity, but contact with other patients and nurses is not a reason to come to the hospital more often. Patients still maintain close contact with a healthcare provider by receiving eculizumab every fourteen days.

I have seen little of the fellow patients in recent years, in the early years I saw them more often. When I just got sick I was used to discuss questions about PNH and my treatment with the fellow PNH patients. At this point, where I am more experienced with PNH I don't mind that I see the fellow patients less now. – Patient 08

Theme 2: Confidence with technology

Subtheme 5: Handling technology

The Zaurus application, which was used during the remote consultations, was experienced as user-friendly. The installation of the application was simple and good quality of the video and sound were experienced. For some patients, it took some time to figure out how to establish a video connection. It is expected by the patients that this will improve when they use the application more often. Other experienced disadvantages were that the application does not work properly if there is a poor internet connection and when a user receives a phone call while using the application on the same phone.

My practitioner used the application on her own mobile phone. Because she received a phone call during the consultation, the conversation fell away for a moment. Those are the teething problems I think. The video and sound was of good quality. – Patient 01

Subtheme 6: Privacy

Because the secure application Zaurus is used, patients experience the privacy of their consultation was guaranteed. As a result, patients dare to discuss everything with their practitioner. It is found pleasant that a remote consultation can take place from the patients' own environment. A number of patients also see a challenge to ensure privacy during the consultation, the patient must ensure that the consultation can take place in a quiet environment.

Well I feel free enough to discuss private matters both during a physical consultation and during a remote consultation. To me it makes no difference at all. That is mainly because this application is well secured. The remote consultation felt like a confidential conversation. It actually just gave me the safe feeling of a consulting room, but in my own familiar environment. – Patient 04

Subtheme 7: Video versus phone

Patients expressed a clear preference for remote consultation by means of a video consultation over a telephone consultation. Because the patients can actually see their practitioner during a video consultation, more attention will be paid to the questions the practitioner asks. In addition, eye contact can be made so patients are less distracted and it ensures that a video consultation is considered more serious.

I will take the remote consultation more seriously when it is performed with a video connection. When there would not be a video connection, I would just take the phone call between two work activities and I think I will be quickly distracted. – Patient 07

Theme 3: Future perspective

Subtheme 8: Frequency remote consultation

Patients experienced that a remote consultation cannot completely replace a physical consultation. Patients expressed it is important to be involved in the planning in their appointments. Remote consultations are seen as a good addition to a physical consultation,

but not as a complete substitute. Ideally, patients would like to see that a physical consultation would be alternated with a remote consultation.

I am not in favor of that all my consultations with my practitioner will be a remote consultation. The continuous alternation between a physical consultation and a remote consultation seems an ideal balance to me, where I will have less travel distance, but still feel an affective bond with my practitioners. – Patient 01

The preferred frequency of a remote consultation depends on several factors, including the disease duration, duration of eculizumab treatment, content of the consultation, and the number of physical complaints. Patients with a long duration of illness and a long treatment duration prefer a remote consultation above a physical consultation. Patients prefer a physical consultation if new important developments will be discussed or if a patient has new physical complaints.

At the moment I am doing well. If my health was not going well, for example, if I had more physical complaints, it would become more important for me to come to the hospital to discuss my complaints with my practitioner. – Patient 06

Subtheme 9: New way of working

A number of patients are increasingly dealing with video calling in their daily lives. In an earlier phase, some patients have wondered whether a physical consultation could become digital, in order to keep up with the developments digitization offers. The call to digitize has become bigger among patients since the COVID19-pandemic.

I thought the idea of working with remote consultations was very smart and sensible. You know, with every travel movement you have a higher risk of contamination with the COVID19-virus. In my opinion it is a smart choice to control the patient flow in this way and to dose the patient flow a little. – Patient 04

DISCUSSION

To our knowledge, this is the first qualitative study identifying the experiences of PNH patients on eculizumab treatment with remote consultations. This study identified three different elements underlying the patients' experiences with remote consultations: 1) personal pros and cons, 2) confidence with technology, and 3) future perspective. Patients valued that they were given the option not to go the hospital for a consultation, so they could spend more time on leisure or work and they are therefore less reminded of their illness. Patients experienced less affection with their practitioner and less contact with fellow patients is experienced when using

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remote consultations. While using a new technology, it is perceived important that the privacy during the consultation is guaranteed. Patients clearly indicate that a remote consultation is a good addition to the physical consultation, but it is not a substitute.

A previous study focusing on the experiences of remote consultations in patients with other hematological diseases supports several aspects of this study¹³. The findings of that study are in line with the idea that patients experienced a valuable feeling of freedom when having a remote consultation because they did not have to spend time traveling to see their practitioner, and they could maintain their everyday lives. That study also indicates that patient involvement is important in the planning of their appointments. Additionally, in both studies handling a new technology is seen as a learning process. When the patients use the application more frequent, it is thought that the application will be easily integrated. Only the finding that intimacy is not about physical presence did not correspond to the findings identified in our study.

In the absence of other qualitative studies on the experiences of remote consultations in hematological patients, the findings were compared to a study in patients requiring secondary specialized care²¹. In that study patients expressed satisfaction with not having to take time off from work for a whole day. In line with this finding, our study concluded that patients experienced less impact on daily life when having a remote consultation. Our study confirmed that the preferred frequency of a remote consultation depends on the content of the consultations and the experienced physical complaints. This is also stated in the study in patients requiring specialized care.

Our study was performed in accordance with qualitative research methods^{14-15,17,19}. A generic explorative qualitative study was performed on the patients' perspectives. The richness of the data makes that the analysis has captured patients' most typical underlying aspects of their experiences with remote consultations. Results were exposed by quotes to provide thick description, to ensure transparency of the results and to enhance validity^{15,30}. Our study is strengthened by the fact that new insights that emerged during data collection were incorporated into the interview guide¹⁹. Although the results of this study were based on twelve patients, maximum variation sampling increased the diversity in the data. In addition, data were collected from a representative selection of patients which contributed to the transferability of the study¹⁸. Internal validity was increased by the use of a well-trained interviewer who participated in an interview training course. Moreover, recent literature³¹⁻³² about interviewing by video was studied by the interviewer prior interviewing. Rigour is ensured by discussion of analytical processes in depth throughout the study among two investigators^{14,19}.

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However, this study had a few limitations. The present findings are limited by the characteristics of selected patients and the treatment setting. Moreover, in this study, only Dutch adult PNH patients with eculizumab treatment who have stable disease were included, which limited the generalizability to other less stable PNH patients. To prevent overloading the patients, a member check was not performed. The lack of member check can lead to a reduced trustworthiness³³. Summaries were given by the interviewer after each interview, and the patients were asked whether the summary corresponded with the content of their interview to validate the patients' perspective.

One patient who was on the predetermined inclusion list was excluded from this study, because it was not accomplished to make a connection for a remote consultation. Therefore, it was not possible to collect the experiences of this patient. However, it is nevertheless a finding that no contact was made with this patient. Although, this finding was not included in the analysis of the results because the patient was excluded from this study. This may lead to a distorted view of the experiences of a remote consultation.

Maximum variation was not applied to the treatment duration. However, a reasonable variation within this characteristic has formed. No patients were included who had just started their treatment. This may bias the results, as some patients indicate that they currently have a different view on remote consultations than at the start of their treatment. If patients with a shorter treatment duration were included, this could possibly lead to less positive perspectives.

The investigator was already known to the patients prior to the study, due his role as PA in PNH care. According to current literature, it is not preferable for practitioners to also assume the role of investigator³⁴. This can potentially lead to a disturbed practitioner-patient relationship and multiple conflicts of a dual role³⁴. However, during this study, the dual role has had the advantage that patient recruitment has been straightforward and patients had clearly opened up during the interviews. The dual role was described in the PIF in order to minimize the negative consequences. The PIF also stated that participation or refusal of participation in the study did not affect the treatment relationship. Additionally, the investigator did not conduct the interviews in work uniform and at the start of an interview the investigator discussed that he assumed the role of investigator.

Efforts are being made to further implicate remote consultations within PNH patients. It is recommended to alternate remote consultations and physical consultations equally. Whether

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a consultation takes place physically or remotely should be discussed between practitioner and patient in order to provide patient-centered care. In order to continue using remote consultations in PNH outpatient practice, a number of conditions should be met: 1) improved technical infrastructure is required to allow remote consultations to become routine in PNH-care. 2) Purchasing a separate device for the use of remote consultations is recommended, so that an incoming call does not cause disturbance. 3) In order to maintain an adequate assessment of the medical condition, the remote consultations should stay combined with blood tests.

Future longitudinal research should be performed in a larger group of PNH patients. The included patient group should include multiple patients who had multiple video consultations as well as multiple telephone consultations, and have been treated with eculizumab for less than one year. Current study might indicate that the incidence of missed appointments increases as appointments changed from physical to remote consultations. So further research into patients who missed a remote consultation is needed. Also, research about the experiences of practitioners providing remote consultations and the economic and environmental factors is recommended.

CONCLUSION

This qualitative research explored the experiences of remote consultations of patients with PNH and eculizumab treatment. A majority of the patients considered the remote consultations as a positive experience, even patients who did not believe the remote consultations provided the best care. Several advantages and disadvantages of a remote consultation were identified on the impact of daily life, experienced affection, the assessment of medical condition and on the contact with experience experts. Additionally, the use of technology and the future perspective of a remote consultation were discussed. Patients describe a remote consultation is a good addition to the physical consultations, but it is not seen as a definitive replacement. A preference has been expressed for an equal alternation between a video consultation and a physical consultation. This study serves as a basis for a more patient-centered PNH outpatient care.

REFERENCE LIST

- 1 Lupton, D., & Maslen, S. (2017). Telemedicine and the senses: a review. *Sociology of Health & Illness*, 1-15.
- 2 Atzori, W. (2013, November 6). eHealth and patient empowerment: A patient perspective. NA. Brussel.
- 3 Totten, A., Hansen, R., & Wagner, J. (2019). *Telehealth for acute and chronic care consultations*. Rockville: Agency for Healthcare Research and Quality.
- 4 Thompson-Coon, J., Abdul-Rahman, A.-K., Whear, R., Bethel, A., Vaidya, B., Gericke, C., & Stein, K. (2013). Telephone consultations in place of face to face out-patient consultations for patients discharged from hospital following surgery: a systematic review. *BMC Health Services Research*, 1-9.
- 5 Deldar, K., Bahaadinbeigy, K., & Tara, S. (2016). Teleconsultation and Clinical Decision Making: a Systematic Review. *Acta Informatica Medica*, 286-292.
- 6 Barsom, E., Jansen, M., Tanis, P., van de Ven, A., Blussé van Oud-Alblas, M., Buskens, C., . . . Schijven, M. (2020). Video consultation during follow up care: effect on quality of care and patient- and provider attitude in patients with colorectal cancer. *Surgical Endoscopy*, 1-10.
- 7 Gilbert, A., Billany, J., Adam, R., Martin, L., Tobin, R., Bagdai, S., . . . Bateson, J. (2020). Rapid implementation of virtual clinics due to COVID-19: report and early evaluation of a quality improvement initiative. *British Medical Journal*, 1-8.
- 8 National Health Service. (2020). Clinical guide for the management of remote consultations and remote working in secondary care during the coronavirus pandemic. *National Health Service*, 1-12.
- 9 Vereniging Samenwerkende Ouder- en Patiëntenorganisaties. (2012). Expertisecentra voor zeldzame aandoeningen. Opgehaald van VSOP: <https://vsop.nl/wat-doen-wij/expertisecentra/>
- 10 Brodsky, R. (2014). Paroxysmal nocturnal hemoglobinuria. *Blood*, 2804-2811.
- 11 Arnold, L., Stephenson, J., Kelly, R., Buchanan, D., Jones, G., & Hillmen, P. (2008). Home Infusion of Eculizumab: A Unique and Innovative Model of Drug Delivery to Reduce Treatment-Associated Burden and Enhance Quality of Life for Patients with PNH. *Blood*, 1.
- 12 Devalet, B., Mullier, F., Chatelain, B., Dogné, J.-M., & Chatelain, C. (2015). Pathophysiology, diagnosis, and treatment of paroxysmal nocturnal hemoglobinuria: a review. *European Journal of Haematology*, 190-198.
- 13 Christensen, N., & Danbjørg, D. (2018). Use of video consultations for patients with hematological diseases from a patient perspective: Qualitative Study. *Journal of Participatory Medicine*, 1-10.
- 14 Creswell, J. (2013). *Qualitative inquiry & Research design*. London: SAGE Publications.
- 15 Holloway, I., & Wheeler, S. (2014). *Qualitative research in nursing and healthcare*. Oxford: Wiley-Blackwell.
- 16 Caelli, K., Ray, L., & Mill, J. (2003). 'Clear as Mud': Toward Greater Clarity in Generic Qualitative Research. *International Journal of Qualitative Methods*, 1-13.
- 17 Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): A 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*, 349-357.

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- 18 Palinkas, L., Horwitz, S., Green, C., Wisdom, J., Duan, N., & Hoagwood, K. (2016, September 1). Purposeful sampling for qualitative data collection and analysis in mixed method implementation research. *Administration and Policy in Mental Health*, pp. 533-544.
- 19 Boeije, H. (2010). *Analysis in Qualitative Research*. London: Sage.
- 20 Donaghy, E., Atherton, H., Hammersley, V., McNeilly, H., Bikker, A., Robbins, L., . . . McKinstry, B. (2019). Acceptability, benefits, and challenges of video consulting: a qualitative study in primary care. *British Journal of General Practice*, 586-594.
- 21 Johansson, A., Lindberg, I., & Söderberg, S. (2014). Patient's experiences with specialist care via video consultation in primary healthcare in rural areas. *International Journal of Telemedicine and Applications*, 1-7.
- 22 Wester, F. (1991). *Strategieën voor kwalitatief onderzoek*. Muiderberg: Coutinho.
- 23 Maguire, M., & Delahunt, B. (2017). Doing a thematic analysis: A practical, step-by-step guide for learning and teaching scholars. *All Ireland Journal of Teaching and Learning in Higher Education*, 3351-3364.
- 24 Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative research in psychology*, 77-101.
- 25 Briks, M., Chapman, Y., & Francis, K. (2009). Memoing in qualitative research. *Journal of Research in Nursing*, 68-75.
- 26 O'Connor, C., & Joffe, H. (2020). Intercoder Reliability in Qualitative Research: Debates and Practical Guidelines. *International Journal of Qualitative Methods*, 1-13.
- 27 Radboudumc. (2020, April 17). Videogesprek Zaurus met uw zorgverlener. Opgehaald van Radboudumc: <https://www.radboudumc.nl/patientenzorg/uw-afpraak/meer-informatie/videogesprek-zaurus/wat-is-een-videogesprek>
- 28 Saunders, B., Sim, J., Kingstone, T., Baker, S., Waterfield, J., Bartlam, B., . . . Jinks, C. (2018). Saturation in qualitative research: exploring its conceptualization and operationalization. *Quality & Quantity*, 1893 - 1907.
- 29 Eldh, A., Arestedt, L., & Bertero, C. (2020). Quotations in qualitative studies: Reflections on constituents, custom, and purpose. *International Journal of Qualitative Methods*, 1-6.
- 30 Moravcsik, A. (2019). Transparency in Qualitative Research. *Sage research methods*, 1-14.
- 31 Mirick, R., & Wladkowski, S. (2019). Skype in Qualitative Interviews: Participant and Researcher Perspectives. *The Qualitative Report*, 3061-3072.
- 32 Vadi, M., Malkin, M., Lenart, J., Stier, G., Gatling, J., & Applegate, R. (2016). Comparison of web-based and face-to-face interviews for application to an anesthesiology training program: a pilot study. *International Journal of Medical Education*, 102-108.
- 33 Birt, L., Scott, S., Cavers, D., Campbell, C., & Walter, F. (2016). Member Checking: A tool to enhance trustworthiness or merely a nod to validation. *Qualitative Health Research*, 1802-1811.
- 34 METc UMC Groningen. (2021, Mei 9). De rol van de behandelend arts en van de onderzoeker. Opgehaald van METc UMC Groningen: <https://metcgroningen.nl/themas/de-rol-van-de-behandelend-arts-en-van-de-onderzoeker/>

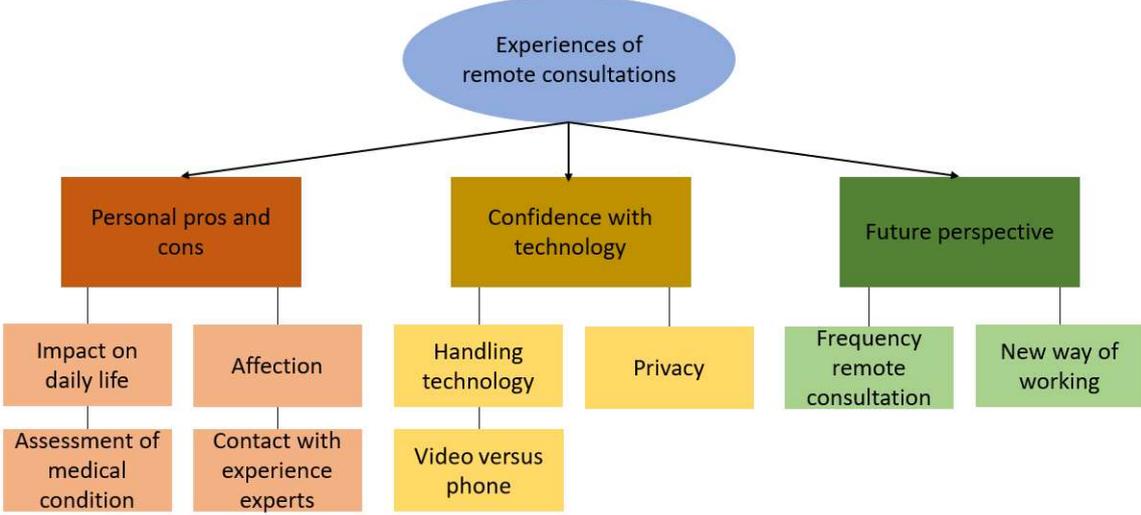
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Table 1 Baseline characteristics of the study population

ID	Sex	Age (years) [*]	Job status	Treatment duration [*]	Travel distance (km) ^{**}	Means of transport	Able to travel independently
P01	Female	62	Salaried employment	12 years, 2 months	98	Car	No
P02	Male	31	Salaried employment	5 years, 10 months	114	Car/Train	Yes
P03	Female	69	Retired	1 year, 7 months	211	Car	Yes
P04	Female	66	Unfit for work	1 year, 5 months	151	Car	No
P05	Male	43	Salaried employment	15 years, 4 months	64	Car	Yes
P06	Female	52	Salaried employment	15 years, 3 months	138	Car	Yes
P07	Male	43	Self-employed	3 years, 6 months	163	Car	Yes
P08	Male	61	Salaried employment	12 years, 7 months	60	Car	Yes
P09	Male	54	Unfit for work	6 years, 4 months	53	Car	Yes
P10	Female	41	Self-employed	4 years, 8 months	130	Train	Yes
P11	Male	26	Salaried employment	4 years, 10 months	107	Car	Yes
P12	Female	80	Retired	3 years, 11 months	75	Car	Yes

Notes: *: Data were collected at time of the interview; **: Distance of a one-way trip

Figure 1 Relation between themes and subthemes



APPENDIX 1: Interview guide

1. Ervaringen van fysiek consult

- Wat vindt u belangrijk in een fysiek poliklinisch consult?
- Wat is de meerwaarde van een fysiek poliklinisch consult?
- Wat is de invloed van een fysiek poliklinisch consult op uw dagelijks leven?
- Worden alle bloeduitslagen altijd al besproken tijdens het consult? *

2. Ervaringen van consult op afstand

- Wat is uw gedachte over een consult op afstand?
- Wat waren uw verwachtingen van een consult op afstand?
- Wat is de invloed van een consult op afstand op uw dagelijkse leven?
- Heeft u het gevoel dat u alles heeft kunnen bespreken met uw behandelaar?
- Heeft u het gevoel dat uw behandelaar uw toestand tijdens het consult heeft kunnen beoordelen?
- Wat vindt u ervan dat er bij een consult op afstand geen lichamelijk onderzoek plaats vindt? *
- Wat vindt u ervan dat u door een consult op afstand minder patiëntencontact heeft? *
- Wat zou u vinden van een consult op afstand wat telefonisch plaats vindt? *
- Hoe zag uw dag van het consult op afstand eruit? *
- Wat vond u ervan dat het consult wel/niet tegelijk met de eculizumab-toediening plaats vond? *
- Wat zou u vinden van een consult op afstand als u recentelijk begonnen zou zijn met de behandeling met eculizumab?*

3. Verschil tussen fysiek consult en consult op afstand

- Wat zijn volgens u de voordelen van een consult op afstand ten opzichte van een fysiek consult?
- Wat zijn volgens u de nadelen van een consult op afstand ten opzichte van een fysiek consult?
- Wat zijn de verschillen in de voorbereidingen die u treft voorafgaand aan een consult op afstand ten opzichte van een fysiek consult?
- Wat is de impact van de Corona-pandemie op uw mening van een fysiek consult dan wel een consult op afstand? *
- Wat is volgens u de impact van een fysiek consult danwel een consult op afstand op de omgang met PNH of uw behandeling?

4. Het gebruik van technologie

- Hoe heeft u het gebruik van de technologie ervaren?
 - o Hoe heeft u de kwaliteit van het geluid ervaren?
 - o Hoe heeft u de kwaliteit van het beeld ervaren?
- Wat vindt u ervan dat de Zaurus-applicatie alleen op de telefoon of tablet werkzaam is? *
- Hoe ging het installeren van de applicatie? *
- Wat was uw gevoel bij het gebruiken van een nieuwe applicatie? *

Note: *: *Formed questions in response to the emerging data*