Patient experiences in care of Chronic Recurrent Multifocal Osteomyelitis

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**Patient experiences in care of Chronic Recurrent Multifocal Osteomyelitis**

The aim of the qualitative research was to find out the experiences of patients in the care of CRMO. The purpose of the research was to increase understanding of the disease among nurses, and thus, develop the care given to patients with rare diseases.

The research was implemented using a qualitative approach. A narrative approach was applied to collect the data. The data was collected using a virtual interview method. The sample consisted of one American woman and two Finnish women suffering from CRMO. Inductive content analysis method was used to analyze the data.

The study results showed that the patients were mainly dissatisfied with the care they had been given since the care did not meet the patients’ needs. However, the care experiences had often been positive when care was provided by healthcare professionals familiar with the condition.

As a conclusion, patients suffering from CRMO had experienced that the care they had received had been insufficient. However, the quality of care had been better when healthcare providers were familiar with the disease and its treatment. Thus, increasing understanding might increase the quality of care.
1 Introduction

This bachelor’s thesis investigates patients’ experiences in care of chronic recurrent multifocal osteomyelitis (CRMO). CRMO is a rare autoinflammatory bone disease that is considered a pediatric disease since it mainly occurs in children and adolescents. However, the condition has also been found in adult patients. (Finn, Ramanan, Roderick, Rogers & Shah, 2016; Keskitalo, Kröger, Niinimäki, Remes-Pakarinen & Vähäsalo, 2016; Hasegawa, Ito, Kuroda, Nakano, Nakatsue, Narita, Nozawa, Saeki, Sato, Suzuki, Umez u & Wada, 2016.) Severe pain caused by recurrent and chronic inflammation in bones is a common symptom. Chronic pain and changes in the bones caused by the disease often have a negative effect on the quality of life of patients. In addition to physical complications, the disease affects patients’ mental health. Rare diseases are often life-threatening, meaning that the right and suitable care is essential. (Rare Disease Day, 2016.)

The needs of care within patients suffering from rare diseases are not well fulfilled since the needs are not the same as for the general population (Dawkins, et al., 2016). The symptoms of rare diseases often vary significantly from person to person, even when the disease is the same. Rarity and lack of knowledge may lead to a delay of diagnosis, and misdiagnosis, which again may lead to worsening of the disease and increased need for care. (Berven, Burr, Caskey, Elderkin, Esposito, Gall, Goldring, Jacobs, King, Klippel, Pollak, Sandborg & Templeton, 2013; Rare Diseases Europe, 2014.) Misdiagnosis often leads to wrong kind of treatment and prescriptions of wrong medicines, possibly causing further problems (Dawkins, Di Pietro, Fookes, Molster, Petrie, Urwin & van der Laan, 2016).

Patients with rare diseases often lack quality care, and it is difficult for them to access treatment and care. Their situation can be improved for instance by sharing scientific knowledge about rare diseases and raising awareness. (Rare Diseases Europe, 2014.) Chronic recurrent multifocal osteomyelitis is a disease unknown to many. Some studies have been done about rare diseases in general, but only a little information is avail-
able about the care of CRMO and patients experiences in care of the disease particularly. Understanding patients’ experiences in care of chronic recurrent multifocal osteomyelitis can help to improve the care and patients’ quality of life in the future. The aim of this study is to find out what kind of experiences patients suffering from chronic recurrent multifocal osteomyelitis have of the care they have received. The purpose of this Bachelor’s thesis is to increase understanding of CRMO, and thus, develop the care given to patients with CRMO.

2 The care of patients with rare diseases

2.1 Rare diseases

Rare diseases are diseases affecting a small number of people. In Europe, a rare disease is defined as a disorder that affects less than 1 of 2,000 in a population, while in the United States, it is defined as a disorder affecting fewer than 200,000 inhabitants. (Dawkins, et al., 2016.) The prevalence of some rare diseases can be higher or lower in some areas, due to different sizes of populations. Sometimes a rare disease may affect only a few people of a population. The causes of most of the rare diseases are unknown. Though, the majority of 80% of rare diseases are genetic. Genetic rare diseases are often caused by a mutation of one single gene. Thus, genetic rare diseases can run in families. (National Human Genome Research Institute, 2012; Rare Disease Day, 2016.) A mutated gene can be transmitted dominantly, recessively, or it can be X-linked. A mutation can happen also on a chromosomal level when there are structural changes in the chromosomes, or the amount of chromosomes is abnormal (more or less than 46 chromosomes). (Harvinaiset-verkosto.) Other causes for rare diseases include bacterial and viral infections, allergies, and environmental factors. The lack of knowledge in rare diseases is often seen as a delay of diagnosis or a misdiagnosis. Patients suffering from rare diseases can receive unequal care since the needs for care are different. Treatment and care can be difficult to access. (Dawkins et al., 2016; Rare Disease Day, 2016.) According to Anderson, Elliot, & Zurynski (2013), studies have shown that patients suffering from rare diseases sometimes face rejection by
healthcare professionals. Poorly provided care of rare diseases is common due to mis-diagnosis and delays of diagnosis. The health outcomes of patients suffering from rare diseases are poor, as well as the quality of life. (Dawkins, et al., 2016; rare Diseases Europe, 2014.)

2.2 Chronic Recurrent Multifocal Osteomyelitis

Chronic recurrent multifocal osteomyelitis, or CRMO, is a rare autoinflammatory disease affecting the bones. The disease is considered a pediatric disease since it primarily occurs in children and adolescents. (Finn, Ramanan, Roderick, Rogers & Shah, 2016; Keskitalo, Kröger, Niinimäki, Remes-Pakarinen & Vähäsalo, 2016.) The prevalence of the disease is unclear due to the disease’s unfamiliarity, but a study made in Germany claims that 4,5 out of a million children suffer from CRMO. (Finn, et al., 2016; Grote, Jansson, Silier, & Voit, 2017). Also, the disease is more common in females than males (Amini, Aslam, Elshikh, Haygood, Madewell, Mujtaba, Synghal, 2017). The symptoms of the disease include pain in at least one bone and swelling and tenderness in the soft tissue around the affected bone. Lesions are commonly found in long bones, but it can affect any bone. (Finn, et al., 2016; Keskitalo, et al., 2016.) Additionally, arthritis and skin, bowel, and eye symptoms can occur (Keskitalo, et al., 2016). Due to the rarity of the condition, there are no specific recommendations for treatment. The aim of care is to alleviate symptoms, especially to ease the pain, to speed up the recovery of the bones, and to prevent bone changes from increasing and the disease from reactivating. (Keskitalo, et al., 2016.) It is important to recognize the disease in an early stage to avoid the use of antimicrobial drugs, unnecessary examinations such as imaging which predisposes patients to excessive radiation exposure, and unnecessary bone biopsies (Finn, et al., 2016; Keskitalo, et al., 2016; Amini, et al., 2017). Pain and changes in the bones can affect the quality of life. Chronic pain can have a negative effect on one’s mental health by causing depression, for instance. Even half of the patients suffering from chronic recurrent multifocal osteomyelitis experience that their life quality has gotten worse. Thus, early diagnosis may also enhance the quality of patients’ life. (Keskitalo, et al., 2016.)
2.3 Caring for patients with rare diseases

Patients who suffer from rare diseases often perceive a lack of medical solutions for their disease, which may lead to patients feeling abandoned. The feeling of abandonment increases their need for emotional support and thus they tend to need holistic care (Caputo, 2013). In holistic care, a patient is seen as a whole, and one's physical, social, psychological and spiritual aspects are considered when providing care. Holistic care enables care givers to understand patients and their needs. (Jasemi, Keogh, Taleghani, Valizadeh & Zamanzadeh, 2015.) Patients may be skeptic and lack trust in healthcare making them feel helpless and vulnerable. Consequently, health providers should guide patients and encourage them to join support groups which may enhance their psychological wellbeing, and ensure that the patients feel like they can trust in the healthcare system. (Caputo, 2013.) Since rare physical diseases strongly affect patients' emotional and mental health, care givers should give patients the opportunity to talk and be listened to (Frigerio, Invernizzi, Montali & Riva, 2011). Although finding medical solutions is a major part in improving the quality of life of patients with rare diseases, patients find their moral needs being considered more important. They want to be treated well morally, including being taken seriously, being listened to, and being supported and guided in terms of their needs. (Huyard, 2009.)

2.4 Nursing care of Chronic Recurrent Multifocal Osteomyelitis

There are no specific care recommendations for CRMO and thus the treatment is symptomatic. The care of CRMO patients is teamwork of several professions which include nurses, doctors, physiotherapists, occupational therapists, and social workers. (Keskitalo, et al., 2016; About Kids Health, 2010.) Pain in bones being a typical symptom, pain management plays a big role. (Keskitalo, et al., 2016.) The nurses’ role in care of CRMO is providing information about the treatment to patients, their families, and care givers, supporting patients and their families, and helping in treatment. The aims of nursing care include alleviating pain, preventing possible complications and providing information about the disease and its treatment to the patients and their families. If a patient needs assistive devices, such as crutches, nurses teach the patient
how to safely use them. (Çetinkaya & Kusdemir, 2015.) Medicines used for CRMO include drugs that have to be administered subcutaneously or intravenously. Nurses guide patients and their family members to administer subcutaneous injections safely at home, and they take care of patients when they come to health centers and hospitals for intravenous drug administration. The disease is a physical burden, but it also has a negative effect on the patients’ health from psychological and social aspects due to the disease restricting patients’ life. Thus, nurses have to offer patients psychosocial support as well. In psychosocial care nurses motivate and support patients in coping and living with the disease, and guide them with their challenges. (Kinder-Rheumahilfe München, 2015.)

2.5 Patient experiences

Patients’ experiences have become an important measurement tool in quality of care (Bell, Doyle & Lennox, 2013; LaVela, Niederhauser, Marshburn & Wolf, 2014). Other indicators of quality of care are patient safety and clinical effectiveness. Patient experiences may bring up positives and negatives in patient safety and clinical effectiveness, thus, the two areas can be improved by improving patients’ experiences in care. (Bell, Doyle & Lennox, 2013.) Patient experience is a board concept. It is about how a patient feels when undergoing care from the first encounter in a healthcare organization throughout the whole continuum of care. The experiences are formed through patients’ perceptions of received care. Some important aspects are interactions, patient-centeredness, individualized care, and responsiveness. All interactions, direct and indirect, clinical and non-clinical, and their quality and value, inside an organization during a course of treatment influence patients’ perceptions of care. A patient’s experience is ideal when the care is patient-centered. Patient-centeredness consists of personalized, competent and high-quality care, timely responses and care coordination, and the care is responsive and reliable. Individualized care is an aspect similar to patient-centeredness. It consists of compassion, empathy, responsiveness, patient education, physical comfort, and emotional and mental support. Care is personalized for individual patients, and patients have a chance to participate in their own care actively. Responsiveness is referred as the health system’s manners and environment in
which patients are treated when in need of care. Patients’ experiences from the perspective of responsiveness are affected by its domains which are autonomy, confidentiality, dignity, choice, speedy attention, and the quality of basic services. Patients’ experiences are also affected by expectations. Patients have expectations of the care they are going to receive, and their experiences are affected by how their expectations are met. Patients’ expectations of care include convenient and punctual appointments, informative counseling on one’s disease and treatment, alleviating suffering, professionally acting and respectful healthcare staff who treat their patients with dignity, and a chance to talk about one’s problems. Additionally, patients expect their emotional needs to be met, which are confidence, integrity, pride, and passion. (LaVela, et al., 2014.)

3 Aim, Purpose and Research Questions

The aim of this research is to find out the experiences of patients in care of chronic recurrent multifocal osteomyelitis. The purpose of this Bachelor’s thesis is to increase understanding of the disease among nurses and to develop the care given to patients with chronic recurrent multifocal osteomyelitis.

Research questions:

1. How do patients with chronic recurrent multifocal osteomyelitis experience the care they are given?

4 Methodology

4.1 Research Method

A qualitative interview method was used in this Bachelor's thesis. Qualitative approach is often used among nursing researchers, and the utilization of qualitative research methods has started to increase in the field of health and healthcare (Pope & May,
Qualitative research method allows the researcher to study the participants' experiences fully by using a number of qualitative research methods (Bailey, Hennink, & Hutter, 2011). It is aiming to elaborate ideas that can be helpful when it comes to understanding, for instance, the experiences and views of patients (Al-Busaidi, 2008). Qualitative research method enables the participants to tell their stories and experiences to the researchers. The stories told by individuals again give the researcher a wider perspective of the issue that is being studied (Creswell & Poth, 2017). In nursing research, a qualitative method allows the researcher to get a more holistic picture of the experiences of a patient (Aveyard, et al., 2017). A narrative approach was applied in the qualitative interviews. In a narrative research, the participants write their own individual story about their experiences. The narrative approach is an excellent way for an individual to describe one's experiences in detail, in this case, the story of their illness and the experience in care of one's disorder specifically. (Creswell & Poth, 2017.) A qualitative interview applying a narrative approach was the most effective way to collect detailed information about the patients' personal experiences in the care of their disease for the research.

4.2 Participants

To find interviewees, the Finnish Network for Rare Diseases was contacted and they agreed to help to find participants, but none were found. Additionally, a message introducing this Bachelor’s thesis was posted in a Facebook support group created for people suffering from chronic recurrent multifocal osteomyelitis. The interviewer did not contact anyone, but the members of the group who were willing to volunteer contacted the interviewer. Purposive sampling was used to select participants. The goal of purposive sampling technique is to find people who have some particular characteristics or experiences and it a common technique used in qualitative research (Aveyard, et al., 2017). In this research, the target group was people with experiences of a specific disease. The inclusion criteria were that the participants are 18 years old or older, they have a chronic recurrent multifocal osteomyelitis diagnosis, and they are willing to take part in the research. Chronic recurrent multifocal osteomyelitis is considered a pediatric disease, but adult patients were chosen since they have had the disease for a longer period of time, and they more likely have more experience in care...
of the disease and can describe their experiences in more detail than young patients. Eight people were willing to volunteer. Though, by the time of the interviews, only four of the eight volunteers filled in the questionnaire form. When going through the collected data, it was noticed that one of the interviewees did not meet the inclusion criteria because the person had not yet been diagnosed with CRMO. Thus, this Bachelor’s thesis was written based on three participants’ experiences. One participant was from the United States of America, and the rest were from Finland. Their average age was 29.3, and they had suffered from CRMO for an average of seventeen years. Participants from both genders were wished to participate in the study, but all participants were females.

4.3 Data Collection

The data collection was conducted using a virtual interview method. The internet is a fast-growing phenomenon that was originally introduced as a research tool. Today, the use of internet and web-based techniques in research is increasing (Aveyard, et al., 2017). The data for this Bachelor’s thesis was collected virtually because some of the participants were from another country, thus face to face and phone interviews would have been challenging to arrange. A virtual interview differs a lot from a face to face interview and it has its advantages and disadvantages. In a virtual interview, the participants have more time to answer the questions and they are able to make changes to their answers. Though, the participants’ ability to edit one’s responses makes the data less spontaneous. People may feel more convenient answering in their own homes in an environment they know, and it may give a feeling of anonymity. Virtual interviews reduce the feeling of social pressure, and people with less confidence may feel more comfortable writing their answers rather than talking to an interviewer face to face. In virtual interviews, there is always a higher risk of the data being accessed by outside sources, which again would jeopardize the anonymity of the participants. (Braun & Clarke, 2013.) To protect the participants’ anonymity, the collected data was saved behind passwords and only the researcher had access to the data. The data collection applied a narrative approach. The questionnaire form used to collect the data included one narrative question that asked the participants to describe their
experiences in care of their disease. It allowed them to write their own narrative describing the care they had received over the years and how they had experienced the care. The participants were sent a questionnaire form as a Word file privately via Facebook. (Appendix 1. and Appendix 2.) The participants were then given about one month to write their answers. The answers were then sent to the researcher as Word files privately on Facebook or via email. The files were saved and the messages were deleted to ensure the participants’ privacy. In this Bachelor’s thesis, applying the narrative method let the participants freely and in more detail write about their experiences in care of their rare disease. Online interviews make it possible for the interviewee to interview participants from different locations around the world. (Braun, et al. 2013; Aveyard, et al. 2017.) The data were collected in summer in 2017. When the data was no longer needed, the collected data was destroyed to ensure the privacy of the participants.

4.4 Data Analysis

The data analysis process started in summer in 2018. Inductive content analysis method was used to analyze the collected data. It is commonly used in qualitative and quantitative nursing studies. It can be used to analyze written, verbal or visual communication messages. (Elo & Kyngäs, 2007.) Previous research, concerning patient experiences in care of chronic recurrent multifocal osteomyelitis particularly, have not been done, thus, inductive content analysis was a suitable method for this research. Elo & Kyngäs (2007) state that an inductive approach is useful when there is no previous research concerning the phenomenon or if the previous studies are fragmented. Inductive content analyzing process starts with interpreting the collected data and developing concepts and themes (Aveyard, et al., 2017). The data is gone through and similar pieces are collected together. The next step is to combine the material together into a whole that answers the questions of the research. (Juvakka, et al., 2007.)

The participants included Finns and Americans, thus the interviews were done in Finnish or in English depending on the first language of the participant. Since a narrative approach was applied in this research, the material was already in a written form. The data was gone through several times to make sure that the data was properly understood. The data collected from Finnish speaking participants was then translated to
English. After translating, the data was carefully read through again, and main points were collected from the data and was then combined together into a few categories. The combined information was then analyzed, and the answers to the research question were found. Additionally, tables of the main points were created. (Table 1. and Table 2.)

5 Study results

Each participant described their experiences in care of chronic recurrent multifocal osteomyelitis from when their symptoms first started and when they first sought care until present. The experiences of all the three participants were very similar. The findings about the patients’ experiences were divided into two simple categories: negative experiences and positive experiences.

5.1 Negative experiences

The findings of the research indicate that the patients had mainly had negative experiences in care of their disease. As a result, patients were highly dissatisfied with the care they had received.

Table 1. Negative experiences

<table>
<thead>
<tr>
<th>Codes</th>
<th>Subcategory</th>
<th>Generic category</th>
<th>Main category</th>
</tr>
</thead>
<tbody>
<tr>
<td>Patients were told they were seeking attention if nothing was found</td>
<td>Care did not meet the patients’ needs</td>
<td>Negative experiences</td>
<td>Patient experiences in care of chronic recurrent multifocal osteomyelitis</td>
</tr>
<tr>
<td>Doctor laughed at a crying patient</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Patients felt like they were not treated as human beings</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Patients said directly that the care was not enough

Patient felt like she was not a patient but a case

Patient was referred to see a wrong specialist

Patient questioned doctor’s knowledge

Wrong medicines prescribed

Incorrect diagnoses

Wrong information given to patients

Lack of knowledge

5.1.1 Care did not meet the patients’ needs

All participants stated how neither their medical or non-medical needs had been considered in the past. Healthcare professionals’ behavior towards the patients had been cold. Healthcare professionals did not take the patients and their worries seriously. The patients’ symptoms had often been doubted and it was common that the patients’ were blamed for pretending to be ill.

“Nothing was found so I was told I’m just seeking attention.”

“I was totally broken, tearful and scared, and he [the doctor] just laughed dismissively and said that I look completely healthy. I was shocked.”
Patients expressed how they felt like they were not treated as patients or even human beings, and that the care they received did not meet their needs.

“I feel like they didn’t see me as a patient, but as just some “case number” who suffers from something rare.”

“At first, I was treated very badly. I wasn’t treated like a human being. The care I received was not enough or what I would have needed.”

“The care I have received has not been enough and I have been treated as a case, not as a human being that is able to think and feel, and is worried about her own health.”

5.1.2 Lack of knowledge

Every participant noted that the healthcare staff often did not know what to do, how to treat the patients or what kind of care should be offered. This raised frustration in participants.

“He suggested I see an infectious disease doctor. The fact that he made that recommendation made me realize that he really had no idea about my disease and didn’t bother doing any research before I came in every year, because the first thing to know about CRMO is that it is non-infectious.”

“...you’d think doctors would at least know what type of specialist I should be seeing.”

All patients had gone through a lot of examinations. The lack of knowledge had led to incorrect and delayed diagnoses. Misdiagnoses and delay of diagnosis were reported by the majority of the patients. The patients who mentioned being misdiagnosed had received a wrong diagnosis at least once. The wrong diagnoses included both physical and mental illnesses. Misdiagnosis had often lead to doctors prescribing wrong kind of medicine and giving incorrect guidance.

“After the examinations I saw my doctor and he had come to the conclusion that I’m imagining everything. He offered me diazepam...”
“…the doctor thought that I’m a drug abuser, and he threatened to call the security guard because I cried and was angry.”

“I got an osteopenia diagnosis. I wasn’t given any painkillers even though I told I’m unable to sleep because of the pain. The doctor said that I’m stressed for nothing and that doing sports and taking vitamin D will help, and he told me to come back in 2 years.”

“Back then it was believed that CRMO was caused by Staphylococcus aureus … all bacterial cultures of my [bone] biopsy were negative, nevertheless I was prescribed antibiotics for the next ten months … no antibiotics had notable effect.”

5.2 Positive experiences

Table 2. Positive experiences

<table>
<thead>
<tr>
<th>Codes</th>
<th>Subcategory</th>
<th>Generic category</th>
<th>Main category</th>
</tr>
</thead>
<tbody>
<tr>
<td>Patient was taken seriously even though nothing was found</td>
<td>Patients needs were taken into account</td>
<td>Positive experiences</td>
<td>Patient experiences in care of Chronic recurrent multifocal osteomyelitis</td>
</tr>
<tr>
<td>Patient found a doctor who took the patient’s symptoms seriously</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Patient’s treatment was transferred to a university hospital and she found a good doctor</td>
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</tr>
</tbody>
</table>
Patient found a doctor who was familiar with CRMO and an effective medication was prescribed to the patient

Patient’s medical needs fulfilled

Although the patients’ experiences were mostly negative, all patients expressed that there have been times when they have been happy and satisfied with their care. The patients were more satisfied with their care when they were treated in healthcare organizations where their needs and expectations were met and when their care givers had experience of their condition. They were also satisfied when care givers without knowledge of the disease were concerned and took their patients seriously. Even if nothing was found, they were satisfied in cases when they were referred to see a different doctor, or if they were transferred to another healthcare organization.

“We went to a different health center and there I was taken seriously and I was prescribed pain killers. Again, nothing was found but I was sent to see a more experienced doctor.”

“I was transferred to X, I met a good doctor who believed me and my pain.”

Finding good doctors led to patients’ receiving the correct diagnosis and some had also found an effective medication. By the time of the interview, an effective treatment had not been found for one of the participants, but she was grateful for the doctor for trying to make her life easier.

“I found another one [a doctor] who treats both adults and children so he was very familiar with CRMO. He is fantastic. He was much more aggressive in his treatment plan and immediately put me on X (TNF-α antagonist) and X (DMARDs). I haven’t had any pain since.”
“... I started seeing a doctor who believed me and that I was in pain.”

6 Discussion

6.1 Ethical considerations

A research has to be conducted ethically. Ethics is discussion of morality; what is right and what is wrong. Nurse researchers are essentially guided by the same ethical principles as nursing practice. (Galvin, & Holloway, 2017; Aveyard, et al. 2017.) Informed consent, justice, non-maleficence, beneficence, and confidentiality are ethical principles often associated with nursing research (Aveyard, et al., 2017). Human rights have to be protected and respected when conducting a research. The human rights of the participants can be protected by obeying the ethical principles. The researcher must make sure that participants are informed when it comes to the research they take part in, including any possible risks or benefits, and about their right to decide about participating or withdrawing from the research. Enough time has to be given to the possible participants to make the decision of participating or not participating. (Braun, et al., 2013; Aveyard, 2017.) The first step of finding volunteers to participate in this research was writing a message public to all the members of the Facebook support group for people suffering from chronic recurrent multifocal osteomyelitis, and their families. The topic of this Bachelor’s thesis and the research method were introduced in detail. The people of the group who were willing to participate contacted the researcher. The researcher did not personally ask anyone to participate. Thus, the participants had the right to choose whether or not to participate. When the research questionnaire was sent to the participants, they were again reminded and informed that they have the right to withdraw from the research at any time. Justice in research means that the participant has the right to be treated fairly. The researcher ought not have preferences, or be discriminative when it comes to participants. (Braun, et al., 2013; Aveyard, 2017.) In this study, all participants were treated equally, and their patient stories were taken into account equally. Non-maleficence and beneficence in research mean that the participants have to be protected from any harm and discomfort.
Causing physical, psychological, emotional, social and economic harm must be avoided. The research should be beneficial for the participants as well as for the society. (Aveyard, et al., 2017; Galvin, et al. 2017.) No harm was caused to the participants of this study. The research data, as well as the identities of participants, are confidential information. A researcher must always respect the participants' confidentiality. Data can be given to outside sources only if the participant has given permission. (Aveyard, et al., 2017.) Only the implementer of this Bachelor's thesis had access to the collected data. The collected data was destroyed when it was no longer needed. The identities of the participants cannot be recognized by readers.

6.2 Validity and reliability

In qualitative research, validity means that the tools, processes, and data of the research are appropriate. Validity measures whether the research shows what it claims to prove. (Braun, et al., 2013; Aveyard, et al., 2017.) To confirm the validity of this research, the whole research process was strived to be described clearly. In qualitative research, a form of validity called ecological validity can be considered when assessing the validity of a research. Ecological validity expresses if the data collection context resembles the real world context and if the findings of the study can be applied to real-world settings. (Braun, et al., 2013.) The study results were combined together from the participants’ narratives about their care experiences that had happened in real world settings, which gives the results ecological validity. A scientific research strives to conduct as reliable information as possible of the phenomenon that is being studied (Juvakka & Kylmälä, 2007). It measures whether similar results would be found if a different researcher did the same research with a different group of participants (Braun, et al. 2013). The reliability of the results could be measured by providing a similar research with a different group of participants. The results of another similar study might differ in the future if development will happen in the care, and also if the sample was bigger and included both female and male patients. Assessing the reliability of a research is essential for scientific knowledge and utilizing the results. The reliability of a qualitative research can be measured by using criteria, such as credibility, dependability, and transferability. Credibility means that the research and the results are reliable and that it can be verified in the research. (Juvakka, et al., 2007; Aveyard,
Comparing the results to previous studies about the phenomenon enhance the credibility of the results. The results could not be compared to another study considering patients’ experiences in care of CRMO since previous studies of patients’ experiences in care of the disease particularly have not been done, but they were compared to similar studies discussing patients’ experiences in care of other rare diseases and living with rare diseases. Factors such as the amount of time invested in the phenomenon being studied, triangulation, and discussing with other researchers may enhance credibility. (Juvakka, et al., 2007; Aveyard, et al. 2017.) This research was conducted by only one person. The credibility of the research could have been enhanced if another researcher had additionally been conducting the study and analyzing the results. However, a lot of time was spent on going through the data and analyzing the results. Dependability means that the research process has been reported precisely enough for another researcher to be able to follow the research process (Juvakka, et al., 2007; Aveyard, et al. 2017). All phases of the research process have been reported to make it possible for others to follow the process and to enhance dependability. Transferability measures whether the findings of the study can be transferred to a similar situation. The researcher has to provide descriptive information of the participants, sampling, and study design so that others can assess the transferability of the results. (Juvakka, et al., 2007; Braun, 2013; Aveyard, et al. 2017.) The participants, sampling and study design have been described in the research. Only three people were interviewed, thus the results of this study are based on a narrow sample. There could have been more differences in the answers if the sample had been bigger. However, the answers of the three participants were very similar, the participants came from two different countries and had received care in three different countries and several different cities which give the results reliability. Also, the disease is rare which means that a small amount of people suffers from the disease and thus a smaller sample might be enough to get reliable results. Additionally, there could have been differences in the results if there had been male participants as well.
6.3 Discussion of the results

The patients were inhabitants of Finland and the United States of America. Additionally, one participant had received care in the United Kingdom. The stories and experiences of the patients were very similar compared to each other. The participants had received care in different cities and a few different countries, thus larger differences between their experiences could have been possible. The study shows that there was mainly dissatisfaction among the participants when it comes to the care they have received. However, patients also had positive experiences.

The main reason for dissatisfaction in care was that the patients’ needs and expectations were not met. The care the patients were given was not patient-centered or individualized. Findings of previous studies confirm that people with rare diseases suffer from their healthcare needs not being met since their healthcare needs and expectations differ from the needs and expectations of the general population. (Dawkins, et al. 2016). Patients with rare diseases expect their moral and emotional needs to be met when they seek care. They want to be recognized as human beings instead of patients and they want to be treated with dignity. (Huyard, 2009; Caputo, 2013.) The participants had experienced rude and inappropriate behavior from care providers. Their complaints of pain were often belittled and not taken seriously, and they were told that they were seeking attention. The negative behavior left patients feeling like they were not treated as human beings. Patients consider negative behavior, such as rudeness of healthcare professionals towards patients as neglect (Gillespie & Reader, 2013). The quality of communication with healthcare professionals, especially with doctors, has a lot of effect on how satisfied patients are with the care they are given (Abel, Campbell, Elliot, Lyrazopoulos, Paddison & Roland, 2013).

Lack of knowledge was reported as a factor that affected negatively in the care experiences. It was seen as a major reason for misdiagnoses and delayed diagnoses. Majority of the participants had been misdiagnosed at least once. Delayed and incorrect diagnosis is a common feature of rare diseases (Rare Diseases Europe, 2014; Dawkins, et al., 2016). The delay of the diagnosis is not always a problem to patients, but a delay of diagnosis is often associated with the health professionals' lack of knowledge. In case of lack of knowledge, rare disease patients expect to be referred to see another
more an experienced professional, but care providers often fail to do so. Patients may consider such behavior as ignorance. (Huyard, 2009.) In Australia, nearly half of the patients with rare diseases have been misdiagnosed at least once before receiving the correct diagnosis. Being diagnosed had taken from one year to over 5 years. (Dawkins, et al., 2016.) The participants of this study reported that it had taken even up to 10 years to get the correct diagnosis. The misdiagnoses included both physical and mental illnesses. Those who had been misdiagnosed were told by several healthcare professionals that they are imagining their symptoms. Mental misdiagnoses included, for instance, drug addiction. Physical misdiagnoses included bacterial osteomyelitis, thoracic outlet syndrome, and osteopenia. Misdiagnoses can cause harm to patients since it may lead to delay of appropriate treatment and thus lead to reduced quality of life (Dawkins, et al., 2016).

Bacterial osteomyelitis is a common misdiagnosis for chronic recurrent multifocal osteomyelitis. Thus, patients suffering from CRMO are often prescribed antibiotics, causing an unnecessary medical burden and unnecessary hospitalization in case intravenous antibiotics are prescribed. (Amini, et al., 2017.) One patient noted that she had been prescribed antibiotic drugs even after being diagnosed with CRMO. Instead of antibiotics, non-steroid anti-inflammatory drugs (NSAIDs), TNF-α antagonists, disease-modifying antirheumatic drugs (DMARDs), bisphosphonates and corticosteroids have been found to be effective drugs in treatment of CRMO. (Amini, et al., 2017.) Majority of the patients had experienced a delayed diagnosis. A delayed diagnosis may lead to further complications in the bones. CRMO can cause discrepancy of limb-length, which is a result of premature closure of growth plates of the affected bones. Other complications related to a delayed diagnosis and inappropriate treatment include pathological fractures, and complications of the spine such as scoliosis. A delayed diagnosis also leads to patients’ having to go through unnecessary examinations. (Keskitalo, et al., 2016; Amini, et al., 2017.)

Positive experiences were also reported by the participants. The positive experiences were connected to the quality of communication with health providers, and the way the participants had been treated by them. The positive experiences took place mainly after being diagnosed, or in situations where their worries were taken seriously, and when efforts were made to alleviate symptoms or when a referral to another doctor
was made. Previous studies about patient experiences in care of other rare diseases state that patients suffering from rare diseases expect their moral and emotional needs to be considered, and find those needs more important being met rather than their medical needs being met (Huyard, 2009; Caputo, 2013). All patients had seen doctors who did not take them seriously and belittled their symptoms, but also doctors who took them seriously and were concerned and willing to find what is causing their symptoms, and how to treat them. Rare disease patients are often happy with honesty and efforts to improve their health gradually, and they appreciate guidance and non-medical advice that helps in their daily lives. (Huyard, 2009.) The care experiences of the participants clearly depended on who had taken care of them. Lack of knowledge itself caused frustration in patients but it alone was not a reason for dissatisfaction. The way healthcare professionals acted towards patients and responded to their needs mattered more.

6.4 Conclusion and recommendations

As a conclusion, patients suffering from chronic recurrent multifocal osteomyelitis experience that the care they receive is mainly insufficient in consideration of their medical and non-medical needs. The insufficiency results from healthcare professionals' lack of knowledge of the condition, and lack of patient-centeredness and individualized care. Patients experience their care more sufficient when they receive care in healthcare organizations where their needs are being met and where their caregivers are more experienced with the disease and its treatment. Thus, increasing understanding of chronic recurrent multifocal osteomyelitis, the quality of care given to patients suffering from the disease might improve, and their care needs would more likely be fulfilled.

Raising awareness and educating nursing students and care providers already in profession could improve the care given to CRMO patients in the future and thus improve their health and quality of life. The rare disease in question is considered a pediatric diseases and thus, the care experiences of pediatric patients suffering from CRMO could be studied since the needs of pediatric patients suffering from rare diseases are again different from adult patients or pediatric patients in general. A similar research
with a bigger sample including both female and male patients could bring up possible differences in the care experiences between the genders.
References


Anderson, M., Elliot, E. & Zurynski, Y. 2013. Australian families living with rare disease: experiences of diagnosis, health services use and needs for psychosocial support. Orphanet Journal of Rare Diseases, 8, 22


Caputo, A. 2013. Exploring quality of life in Italian patients with rare disease: a computer-aided content analysis of illness stories. Psychology, Health & Medicine, 19, 2, 211-221


Finn, A., Ramanan, A., Roderick, M., Rogers, V. & Shah, R. 2016. Chronic recurrent multifocal osteomyelitis (CRMO) – advancing the diagnosis. Pediatric Rheumatology Online Journal, 14, 1


Frigerio, A., Invernizzi, P., Montali, L. & Riva, P. 2011. ‘It’s as if PBC didn’t exist’: The illness experience of women affected by primary biliary cirrhosis, Psychology and Health, 26, 11, 1429-1445


Gillespie, A. & Reader, T. 2013. Patient neglect in healthcare institutions: a systematic review and conceptual model, BMC Health Services Research, 13, 156


Huyard, C. 2009. What, if anything, is specific about having a rare disorder? Patients’ judgments on being ill and being rare. Health Expectations, 12, 361-370


Kyselylomake CRMO potilaille

1. Perus tietoja

Ikä: Sukupuoli:

Kansalaisuus: Kuinka kauan olet sairastanut CRMO:ta?

2. Kokemuksia CRMO:n hoidosta

Kuvailkaa mahdollisimman tarkasti ja laajasti kokemuksianne saamastanne hoidosta sairauden alkuajoista tähän päivään asti.
Questionnaire for Patients with Chronic Recurrent Multifocal Osteomyelitis

1. Basic information

Age: Sex:

Nationality: How long have you had CRMO?

2. Experiences of the care of CRMO

Please describe in detail your experiences of the care you have received since your symptoms started until this day.