

BMJ Open Transitional care of adolescents with Multiple Osteochondromas: a convergent mixed-method study 'Patients', parents' and healthcare providers' perspectives on the transfer process'

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ABSTRACT

Objectives Multiple osteochondromas (MO) is a rare hereditary disease characterised by numerous benign bone tumours. Its chronic aspect requires a well-organised transition from paediatric care to adult care; however, little is known on organising this care specific for patients with MO. This study aims to gain insight on this topic.

Design Convergent mixed-method study.

Setting This study was conducted at the orthopaedic and paediatrics department of an MO-expertise centre in the Netherlands.

Participants 12 patients, 10 parents and 10 healthcare professionals were interviewed. An additional survey was completed by 2 young adults.

Primary and secondary outcomes The (1) themes on transition, identified through template analysis and (2) transfer experience and satisfaction assessed by an 18-item On Your Own Feet-Transfer Experience Scale (OYOF-TES, range: 18–90) and by Numeric Rating Scale (NRS, range: 1–10).

Results The following three key themes were identified in the qualitative analysis: (1) patient and parent in the lead can be encouraged by self-management tools, (2) successful transfers need interprofessional collaboration and communication and (3) how can we prepare patients for the transitional process? Stakeholders' insights to improve transition were listed and divided into these three themes. Several important aspects were underlined, particularly within the first theme; speaking-up was difficult for patients especially when parents were not directly involved. Moreover, the high psychological impact of the disease requires coaching of self-management and psychological counselling to facilitate stakeholders in their changing roles.

Twenty patients completed the quantitative survey. Mean satisfaction score with the transfer process was poor, which was assessed with the NRS (mean=5.7±2.1; range: 1–9) and the OYOF-TES (mean=56.3±14.2; range: 32–85). The OYOF-TES only showed a negative correlation ($R^2=0.25$; $p=0.026$) with the number of surgical interventions in the past.

Strengths and limitations of this study

- To our knowledge, this is the first study to evaluate transitional care of patients with multiple osteochondromas.
- This study provides a broad range of perspectives from different stakeholders in different settings.
- The convergent parallel mixed-method design provided us with different but complementary data on the same topic, and allowed us to bypass any social desirability bias from semi-structured interviews.
- This study is first limited by a small number of survey respondents due to the rare aspect of the disease.
- Second, not all patients could be reached, therefore, it is not known whether they have dropped out of care or whether these patients experienced no problems during transition. Therefore, selection bias cannot be excluded.

Conclusion Overall, the transfer process was found unsatisfactory. Improvement can be achieved by supporting and guiding the patients to be in the lead of their care. Moreover, preparation for transfer and a multidisciplinary approach may enhance successful transition.

INTRODUCTION

Transitional care is 'the purposeful, planned movement of adolescents and young adults with chronic physical and medical conditions from child-centred to adult-oriented healthcare systems'.¹ Several studies have shown that adolescents (ADs) with chronic disease are potentially vulnerable when transitioning to adult care, and that inadequate guidance of the transition process is associated with deterioration in health of ADs with chronic diseases.^{2–9} Research to date has not

yet explored effective transitional care services for ADs and young adults (YAs) with multiple osteochondromas (MO).

MO is a rare autosomal dominant inherited skeletal disorder with an incidence of approximately 1:50 000 in the general population.¹⁰ MO is characterised by the presence of numerous benign bone tumours, osteochondromas, that develop at the metaphysis of long bones. Surgical removal of symptomatic osteochondromas and limb reconstruction procedures in children and adults with MO is necessary in 66%–74% of the entire population.^{10–12} Goud *et al*¹¹ reported that 48% of children with MO already underwent one or more surgical procedures.

ADs with chronic diseases such as MO face a lot of challenges; they do not only have to deal with becoming an adult and with their disease, they also have to cope with transferring from paediatric care to adult care. If the transitional care is well managed, this could be an opportunity to influence and promote healthy behaviour during this specific period as well as later in life.^{13 14} If not, patients could withdraw from healthcare, which could have major consequences. More specific, ADs and YAs with MO are at risk of developing severe growth disorders (80%),¹¹ nerve compressions, impairment of joint function and development of chondrosarcomas (3%–6%).^{15–18} Malignant degeneration of osteochondroma to chondrosarcoma in MO usually occurs at a relatively early age, with a peak incidence between 20 and 40 years. If these patients consult their physician regularly, the risk of poor outcome can be reduced.

In the transitional literature, an extensive amount of studies reported on specific patient groups such as ADs with rheumatic musculoskeletal disease (RMD) or childhood cancer survivors.^{19–21} Although patients with MO might have certain disease-related problems in common with these group of patients, there are also specific MO-related problems and complications that could affect the study outcome. Important factors in this are the lifelong risk of malignant degeneration in MO, the multiple surgical procedures that are often necessary throughout life and the fact that in most cases, one parent has the same disease. Therefore, the current literature on other chronic disease should not be extrapolated to this patient group without any further research.

This study aims to elicit patients', parents' and healthcare professionals' perspectives on the transfer to adult care to understand perceived barriers and facilitators, and to make recommendations for future transition pathways.

METHODS

Setting

This study was conducted at the orthopaedic and paediatrics department of an expertise centre for MO in the Netherlands. In this centre, consultations at the orthopaedic outpatient clinic, for both paediatric and adult patients take place at one location of the hospital. Clinical admissions and surgical procedures for children are carried out

Table 1 Characteristics of patients, parents and health professionals taking part in the semi-structured interviews

Semi-structured interviews	N=32
Adolescents (16–18 years)	n=6
Sex, female, n (%)	4 (67%)
Age, mean (SD)	17.5 (±0.84)
Parents of adolescents	n=5
Sex, female, n (%)	4 (80%)
Young adults (18–25 years)	n=6
Sex, female, n (%)	4 (67%)
Age, mean (SD)	21.3 (±2.07)
Parents of young adults	n=5
Sex, female, n (%)	5 (100%)
Healthcare professionals	n=10
Child care services	6 (60%)
Adult care services	3 (30%)
Both services*	1 (10%)

*The physician is involved in both paediatric care and adult care services.

at the same location, adult patients, however, are referred for this to the other location. Patients transfer from the paediatric to the adult care services at the age of 18 years. The care of adult and paediatric patients is provided by the same orthopaedic surgeons, who are specialised in paediatric and adult orthopaedic surgery with a focus on MO. They are the only stable factor during and after the transfer to the adult orthopaedic ward. The outpatient care location does not change after transfer.

Study design

A parallel convergent mixed-method study was used combining a qualitative design with a cross-sectional quantitative design.^{22–24} The main focus relied on the qualitative data, which was designed to capture in-depth perspectives of stakeholders. The quantitative part was conducted to verify the qualitative findings without interpretive bias. Key findings from both methods were integrated in the discussion section. Semi-structured interviews were conducted with the following stakeholders: ADs with MO (16–18 years), YAs with MO (18–25 years), parents and healthcare professionals (table 1). The ADs were yet to transfer to the adult care services. The YAs already transferred to the adult care services and were asked to fill out the validated On Your Own Feet-Transfer Experiences Scale (OYOF-TES) questionnaire.²⁵ This questionnaire assesses transfer experience from paediatric care to adult care and consists of 18 items rated on a 5-point Likert scale. The computed score ranges from 18 to 90, with 90 being the highest possible satisfaction score. A specific cut-off point is not defined. In addition, a Numeric Rating Scale (NRS) ranging from 1 to 10 (1=completely unsatisfied and 10=completely satisfied) was used to find out how satisfied patients were with the

Table 2 Characteristics of survey respondents

Survey participants	n=20
Sex, female, n (%)	10 (50%)
Age, mean (SD)	21.6 (\pm 1.88)
Age diagnosis, mean (SD)	5.0 (\pm 4.5)
Age first surgery, mean (SD)	10.2 (\pm 3.9)
Total number of surgeries, mean (SD)	11.6 (\pm 7.4)
Hospital admissions in past 2 years, mean (SD)	1.4 (\pm 0.8)
Outpatient clinic visits in past 2 years, mean (SD)	5.3 (\pm 2.9)

overall process of transfer.²⁵ Furthermore, demographics and clinical data were obtained, such as the number of hospital visits, surgeries and age at diagnosis (table 2).

Sampling and recruitment

Patients aged 16–25 years who had been admitted to the paediatric orthopaedic ward for MO-related surgical procedures were recruited from the hospital database and the national patient association. A total of 75 patients (39 ADs and 36 YAs) met the inclusion criteria, of which 6 ADs and 6 YAs were randomly selected for the interviews. Ten of the parents of these patients agreed to participate in the interview, two declined due to lack of time. Seven of the participating parents were diagnosed with MO themselves. The following 10 healthcare professionals from both departments were selected using purposeful sampling and all participated: paediatric nurses (n=3), pedagogical workers (n=2), psychologist (n=1), orthopaedic nurses (n=3) and physician (n=1) (table 1). For the survey, 20 of the 36 eligible YAs participated.

Data collection

All semi-structured interviews were audiotaped and carried out by the first author. Patients and their parents could choose to be interviewed at the outpatient clinic or at home, and were interviewed separately with the exception of one YA who preferred to have their parent present. Healthcare professionals were interviewed at their working place. Mean duration of the interviews was 30 min (range, 20–64 min). The interview guide was based on the available literature and clinical experience of the research group (online supplemental appendix 1). Questions were mainly open-ended because of the exploratory goal of the interview. The questions addressed the expectations and experiences of transfer and the overall satisfaction of care.

The quantitative data were collected through a web-based survey.

Analysis

The interviews were transcribed verbatim and analysed using a template analysis technique.²⁶ The available literature on transitional processes in other chronic diseases was used to form a priori themes (online supplemental

appendix 2).^{27–29} The first author coded the first three interviews with the a priori themes using a qualitative data analysis software programme (MAX.QDA 2007). The second author coded these three interviews separately and subsequently the two authors compared these coded fragments to verify the themes. Potential newly emerged themes and discrepancies between the two authors were discussed and this resulted in the final draft of the template. All interviews were analysed by the first author with the final template.

The quantitative data were analysed using IBM SPSS Statistics for Windows V.22.0 (IBM Corp., Armonk, NY, USA). Descriptive statistics were performed to describe the study sample and to present the results of OYOF-TES. The 5-point Likert scale of the OYOF-TES was recoded to reflect agreement more easily (5=strongly agree and 4=agree). Bivariate correlations (Pearson's r or Spearman's r) between the OYOF-TES and other variables were calculated. Overall significance was set at a p value of <0.05.

Consent

Written and verbally recorded informed consent was obtained for all participants.

Patient and public involvement

The study was supported by the national patient association 'HME-MO vereniging Nederland'. The association recruited patients through placing calls on their website and newsletters. The association also cooperated in the assessment of the burden of the study. Once the study has been published, we intend to inform participants of the results through the website of the national patient association and will send details of the results in a study newsletter suitable for a non-specialist audience.

RESULTS

Qualitative data

The following three key themes were derived from the analysis of the stakeholders' perspectives: (1) patient and parent in the lead can be encouraged by self-management tools, (2) successful transfers need interprofessional collaboration and communication and (3) how can we prepare patients for the transitional process? Several recommendations for transition were given by the stakeholders which could be divided into the above-mentioned three key themes. The recommendations are outlined in table 3.

Patient and parent in the lead can be encouraged by self-management tools

Speaking-up

Patients and parents mentioned that over the years they became increasingly outspoken towards their healthcare professionals. They gained knowledge through experience and developed 'rituals' on how everything should go before and after surgery, for example, what kind of

Table 3 Stakeholders' recommendations**Stakeholders' recommendations**

Good preparation for transition*	<ol style="list-style-type: none"> 1. Start talking about transition around the age of 15 or 16 years and provide detailed information in writing on the differences between the paediatric care services and adult care services. 2. Make a transition plan and discuss with patient and parents which could be the pitfalls of transition and how to resolve them. 3. The patient and parents should get familiar with the adult care services prior to transfer and meet the nurses and physicians working there. 4. Physicians and parents need to coach the self-management of the patient (encourage the patient to visit the outpatient clinic on its own or sleep one night without their parent on the paediatric ward).
Interprofessional collaboration and communication	<ol style="list-style-type: none"> 1. Handover between paediatric and adult care services to exchange valuable information about patients' care and best practices that were gained during the paediatric care services. 2. Monitor the transitional process of patients in their electronic file to be able to provide customised support. This includes recording information about how to deal with certain anxieties or behaviour. 3. Designated transition teams; nurses and physiotherapists who have a profound knowledge about MO, a pedagogical worker or a psychologist who can support the patient in their anxieties and empower them in self-management. 4. A case manager who can follow the patients' progress and make adjustments if necessary.
Patient and parent in the lead	<ol style="list-style-type: none"> 1. Healthcare professionals should encourage and stimulate speaking up of parents and patients regarding their care. 2. Psychosocial guidance for parents and patients should be offered in and outside the hospital setting to address fears and encourage patients to detach from their parents. 3. Let patients decide on the presence of parents prior to and after surgery; this may stimulate a quicker recovery. 4. Extend visiting hours for parents if asked for by patients.
Artefacts	<ol style="list-style-type: none"> 1. Redesign the interior of the orthopaedic ward; rooms with attributes appropriate to their age such as state-of-the-art televisions, a gaming corner for distraction and clustered rooms with YAs so they can exchange stories and support each other.

*A uniform description of 'good preparation' was given by all stakeholders resulting in four recommendations. In addition to these recommendations, improvement of self-management skills of patients was pointed out, exchange of knowledge between paediatric and adult care services was encouraged and some house rules of the orthopaedic ward were addressed. MO, multiple osteochondromas; YA, young adults.

medication should be administered and what kind of sutures should be used. Some of these routines did not comply with the protocols of the orthopaedic adult ward, which sometimes caused conflicts between nurses and patients or parents.

The nurse said it was better to leave the intravenous catheter in because I might get more pain later. And I thought well that's my own responsibility and if that happens, you can give me another intravenous catheter. (...) I know exactly what's necessary and that's removing the intravenous catheter. (YA)

Most of the time the healthcare professionals supported this outspokenness and confirmed that these patients should manage their care themselves. Most patients seemed to know what is best for them; yet, not everybody spoke up as they were not always comfortable to discuss problems. Some patients still needed a little encouragement to speak-up their mind, in particular on the adult orthopaedic ward, since parents and pedagogical workers who usually recognise the smallest discomfort, are increasingly absent on the adult ward. ADs

on the paediatric ward were coached by pedagogical workers to ask questions and discuss matters they do not agree on.

You miss a counsellor or someone you can talk to so you can say you do not like something. (...) I get the feeling it is better to say nothing so you are not a burden. When I enter the hospital, (...) I become a small child who wants to hold his parent's hands. (YA)

Psychological impact

The interviewed ADs and YAs reported many anxieties which had developed during their childhood. The most common fears were fear of needles, swallowing and hospitals. Extensive psychological and sometimes even psychiatric counselling were necessary for these patients. Two YAs were traumatised during transition and underwent eye movement desensitisation and reprocessing. These two YAs felt uncomfortable and unsafe by other patients having an early onset of delirium on the orthopaedic ward.

Patients expressed different feelings about the transfer to adult care services. Some YAs perceived it as normal, while the majority felt lonely, anxious, afraid or nervous.

It was nerve wrecking, (...) It is a different ambience but cosy, well, very different. (...) I had to get used to it. (YA)

Pedagogical workers and paediatric nurses mentioned that they are more experienced with these types of fears and might use a different approach than the orthopaedic nursing staff. This was recognised by patients, parents and orthopaedic nurses.

Some parents and patients reported about the lack of psychological support and would have appreciated more attention towards themselves. Parents felt overlooked and guilty for passing on this disease to their children and felt that healthcare professionals focused more on the medical part and less on the psychosocial impact of MO.

A psychologist or something, to learn how to deal with it, (...) all those bells and whistles attached to my child. I found that very difficult in the beginning. And still, I do struggle with it, how much I have gone through. (Parent)

Changing roles between parents, patients and healthcare professionals

Patients had different views on what becoming an adult means and how the responsibility should gradually shift from the parents to the YAs. This shift did not always take place as expected.

I think parents should be supportive, not leading. (...) My mother made a lot of the decisions on her own, not much was discussed. Of course, it is also my fault I just let it happen. (YA)

The majority of the ADs and YAs did not mind their parents remaining responsible for their healthcare. They preferred their parents to be continuously present on the orthopaedic ward because they did not feel comfortable with the orthopaedic nurses. They were reluctant to ask nurses small favours, for example, asking for a glass of water or adjusting their legs in bed. Two YAs still let their parents arrange their doctor's appointments and accompany them to the visits; it had always been that way and they did not feel any need for change.

Healthcare professionals emphasised that many parents tried to mediate and assisted in explaining things to their child. The experience that parents had gained from having MO themselves is something healthcare professionals viewed as very useful when addressing MO-related problems with patients. However, healthcare professionals often perceived the interaction between child and parent as being overprotective, too much involved and very controlling.

I usually do not discuss this, the family dynamics, I know it is a bit cowardly but I do not have enough

time to address it. However, I do think if there was a transition protocol it would be a good topic and a great opportunity to discuss. (Physician)

Physicians would discuss the young patient's health with parents first, nevertheless the roles reversed at a certain age at which patients would be addressed first. Some parents encouraged this, while other parents had difficulties trusting their child to self-manage care. One parent mentioned that she has never been able to give full responsibility to her child.

Successful transfers need interprofessional collaboration and communication

Collaboration

Patients and parents observed a different approach between paediatric and orthopaedic nurses. Some of the patients underwent as much as 30 surgical procedures before transfer to the adult ward took place. During this period, the paediatric nurses got to know these patients and their parents well and developed a good understanding on how to support them. However, this information was not transferred to the adult care, which was considered a missed opportunity. Healthcare professionals of the orthopaedic ward frequently reported their desire for better training of staff on the disease MO as well as how to care for this group of patients. All involved medical professionals encouraged more collaboration between the paediatric and orthopaedic departments; they encouraged exchange of knowledge and experience, and suggested to continue the accessibility of pedagogical workers and psychologist on the orthopaedic ward after transfer.

Communication

Communication and its challenges were often discussed while addressing transfer. Patients and their parents implied that adult and paediatric care providers communicate differently.

According to some patients, paediatric nurses would elaborate on what is scheduled, while the orthopaedic nurses would not inform the patient and families well enough, nor give them time to acclimatise to the new environment.

Orthopaedic nurses are so attached to their protocols that they do not look at you as a human, but as a protocol. (Parent)

The style of communication of paediatric professionals was described as kind, caring, comforting, yet sometimes as too childish. The orthopaedic professionals were perceived as straightforward and clear, and sometimes described as curtly, but also as more mature which was considered a welcome change. The orthopaedic healthcare providers agreed on the sometimes childish way of communicating on the paediatric services. However, they also reported that this patient population generally acts more youthful compared with their peers which was maintained by the parents, consequently leading to a childish

way of communicating. A gradual transition process based on the development of the child was suggested.

How can we prepare patients for the transitional process?

Expectations of transferring to adult care

A frequently mentioned concern was that patients expected to receive less attention from nurses on the orthopaedic adult ward compared with the nurses on the paediatric ward. Another concern was the absence of parents by their bedside. Both parents and patients feared that the nurses of the orthopaedic ward would not arrive in time when they needed help, or that they would be forgotten.

Once, I had to wait for one hour at the paediatric ward after I called for assistance. Yes, and that frightens me of the orthopaedic adult ward. Nor my mother nor my father will be there to see whether the nurses are coming, then you are lying in a bed and can't do anything about it. (AD)

Children of parents who expected the transfer to be a challenging and potentially traumatising period, perceived it in the same way as their parents. Conversely, two ADs who had not transferred yet perceived the transfer as being something normal, as did their parents.

Preparation for transition

All stakeholders stated a lack of guidance and preparation for transfer. All nurses indicated the need for a protocol to facilitate transition. Some paediatric nurses tried to inform patients and parents about the scheduled transfer; some nurses tried to coach these ADs to be more independent and addressed the patient more than their parents during this transition period. However, these paediatric nurses also knew that this was not common practice among their colleagues.

Terms like 'abrupt', 'sudden', 'lack of guidance/information' were frequently used by parents and patients during the interviews to describe the transfer from the paediatric care to adult care.

Personally, I think it is very sudden. Like yes, you are eighteen, you are officially an adult, but it is really too fast, like from suddenly seventeen and click you're eighteen, adult and now you should be able to do everything. (YA)

Good preparation is of key importance according to the stakeholders. To the YAs, this meant the possibility to visit the adult ward once or twice before transfer took place as well as receiving information about the adult ward, such as the 'house rules'. YAs who visited the orthopaedic ward prior to transfer expressed that they were less anxious afterwards. The main difference between adult care and paediatric care services and also the most important bottleneck, is that parents will not be around as much as they used to. After being guided by their parents for years, the patients are supposed to have become independent at once, which was experienced as challenging.

It was also challenging for parents to let their children go. Some ADs were thinking about gradually making steps towards independence by sleeping without their parents while being admitted on the paediatric ward. In addition, they expected to benefit from an interprofessional team that is able to provide guidance regarding the transition.

A counsellor or special team who prepares you for the transfer, a conversation for both, parents and children. And maybe, it could be useful to have separate conversations for the patients, without their parents, because that might also help, so you won't be suppressed by your parents. (YA)

Timing of transfer

The age limit for the paediatric care had been discussed with all stakeholders. The ADs had different opinions on what the optimal age for transferring would be. Half of the group would not change the age limit of 18 years old: 'once you have reached the age of 18 years you should get the same service as any other adult'. The other half of the group would prefer to extend the age limit to 20–23 years. Two of these patients would rather withdraw from healthcare services than transferring when becoming 18 years of age.

This would be a reason why I would rather delay my surgery. Even if the surgery would be necessary. (AD)

The parents of the ADs, who had not transferred yet, seemed to have similar opinions as their children. The group of YAs was also divided on what would be the best age to make the transfer. Half of the YAs was not ready for transfer and therefore recommended to extend the age limit to 21–23 years of age, which was also the opinion of their parents. Furthermore, the timing of transfer should not be defined by age solely but by developed skills of the patient. Most of the healthcare professionals agreed on this matter; however, they also expressed that there should still be an age limit. If patients were not capable of taking care of themselves or if they were not ready to transfer, additional support should be provided. The majority of healthcare professionals would stick to the age limit of 18 years except for one paediatric psychologist who would raise the age limit to 23 years, since the mental healthcare system generally uses this age to distinguish between ADs and adults.

Familiar to unknown ward culture

Parents and their children were very concerned about the prospect of turning 18 years old and therefore being admitted to the other location of the hospital. They had familiarised themselves with the paediatric ward and sometimes even defined it as their second home, as the services were adjusted to all their needs and they doubted that it would be the same for the adult care services. This was particularly mentioned for the fact that parents' visits were restricted to official visiting hours and the lack of facilities to stay and relax as well.

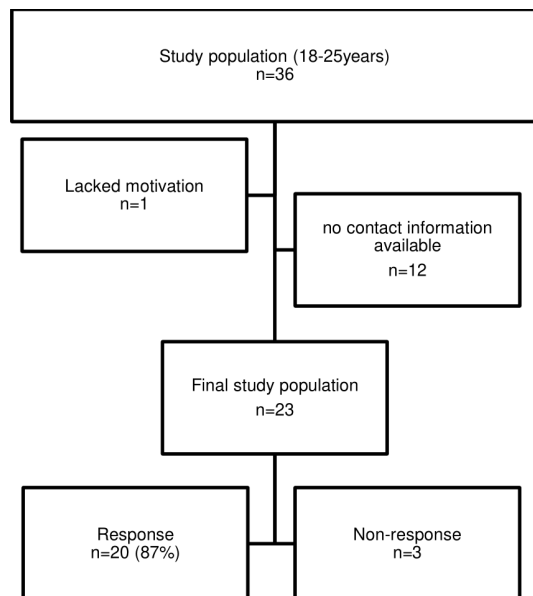


Figure 1 Study sample and response.

It is a totally different building, so totally unknown, so that is a big shock. You do not know what to expect, what kind of ambiance, because here you know everything, you know the nurses and doctors and that is different for the other building where other people work with a different mindset perhaps. (YA)

The views of the patients on the two different locations of the hospital mainly focused on material surroundings. The paediatric ward was described as colourful, with beautiful rooms which you did not have to share and filled with luxurious amenities like games, well-functioning internet, the newest television sets and modern hospital beds. These amenities were lacking at the adult ward. The patients would often be admitted in quadruple rooms and therefore felt they had less privacy. They felt less comfortable and even awkward to be admitted with elderly patients in one room. Although not having met the staff members of the adult care services before transfer, the contact afterwards was described as understanding and caring. With regard to the quality of care provided by the adult and paediatric healthcare professionals, not much difference was experienced.

Quantitative data

Twenty patients completed the survey. Three patients declined to fill out the survey due to lack of time. [Figure 1](#) illustrates the participant flow and [table 2](#) outlines the characteristics of the respondents.

The mean score of the OYOF-TES was 56.3 (SD 14.2; range: 32–85). The most appreciated items were: ‘I can manage well on my own during hospital consultations, also without my parents’ (85% agreed), ‘I am happy with the care I receive in the adult care setting’ (85% agreed). The least appreciated items were: ‘I had a say in the timing of the transfer’ (5% agreed), ‘Before the transfer, I had already met my new healthcare providers’ (15% agreed),

‘There was good collaboration between paediatric and adult care’ (25% agreed). The transfer to adult care was prepared well according to only 30% of the patients. Online supplemental appendix 3 lists all OYOF-TES items, mean values and percentage of agreement.

The mean satisfaction score for the transfer process assessed by the NRS was 5.7 (SD 2.1; range: 1–9) and 65% of the patients scored ≥ 6 . The association between OYOF-TES score and patients’ characteristics such as admissions rate, total amount of surgery and clinic visits, only showed a negative correlation ($R^2=0.25$; $p=0.026$) between the OYOF-TES and the number of surgical interventions in the past.

DISCUSSION

This mixed-methods study explored stakeholders’ perspectives on the transfer from adolescent to adult care for patients with MO. Several bottlenecks and recommendations have been identified for this transitional process. The majority of the quantitative data confirmed the qualitative data and revealed that the overall transfer process was unsatisfactory. Three key themes emerged from the qualitative data collected. Although all themes were consistent with the literature on organising transitional care for other chronic diseases, the theme ‘patient and parent in the lead’ is slightly different as in MO there often seems to be a more complex relationship between parent and child due to the autosomal dominant inheritance of the disorder, potentially resulting in a parent feeling guilty about transferring MO to his/her child.

Unlike in other chronic diseases (eg, cystic fibrosis, juvenile idiopathic arthritis, sickle cell disease and type 1 diabetes), almost 90% of the patients has a parent that suffers from MO themselves.¹⁰ Consequently, these parents already have a certain expertise, experience and leadership of their child’s care process. This can have both advantages and disadvantages. The majority of parents and patients with MO had little difficulty speaking-up regarding their care. However, some patients felt burdened to ask for help and therefore were too afraid to speak-up without the help of their parents or a pedagogical worker. In the transition literature, the concerns of ADs about becoming independent are often addressed but the fear of speaking-up has not been mentioned. Although only a few patients had difficulty speaking up, attention is warranted as this is an important aspect of self-management.

The development of self-management skills necessary for transition is not facilitated in daily practice,³⁰ which was confirmed by the healthcare professionals in our study. The group comprising of patients with MO needs more support and guidance in developing self-management skills. About 85% of the survey respondents reported they could manage well on their own during hospital consultations without parents, however, in the interviews one-third of YAs mentioned difficulties with self-management once transferred; they were used to

their parents managing their care and did not feel the need to be in the lead themselves. At the same time, this behaviour seemed to be stimulated by the parents as they did not expect their child to be capable of managing his own care. This finding is consistent with the literature on transitional care of patients with other chronic diseases.^{18 29 31 32} The lack of responsibility and the difficulty to detach from parents has been reported in several studies.^{27 29 33} Nevertheless, in MO, the experience of the parent with the disease himself/herself, might have a high impact on the self-management skills of the patient. More research is necessary to investigate this potentially complicating factor.

In contrast with previous findings in the literature, the majority of our patients did not perceive transferring as something normal.^{27 29 34} The transition of adolescence to adulthood is already a challenging period.³⁵ Little to no attention has been paid to the psychological impact of transition in healthcare of these vulnerable youngsters with MO. One-third of YAs had suffered from a traumatising experience during transfer and needed psychiatric or psychological counselling, which was striking since such extreme experiences have not yet been reported in the transition literature. It would be of further interest to investigate which patient and disease characteristics in this specific patient group makes them more susceptible for such psychological distress. It seems that transfer might be a tipping point for these vulnerable patients who are already dealing with many changes. In our study population, several fears already existed at childhood, but the most traumatic experiences occurred during admissions on the adult orthopaedic ward. Within our study design, we have not investigated this phenomenon further and merely identified it. Future research with a larger population should focus on this psychological impact of transfer to confirm that this is an issue and to create awareness to provide proper counselling.

In this study, patients and parents had similar views on transfer, whereas in several other studies, ADs with other chronic diseases were less satisfied and more critical about the adult care setting than their parents.^{33 36–38} A possible explanation for this could be that the majority of our patients had a parent suffering from MO as well, and hence might have similar perceptions.

Patient satisfaction is associated with quality of delivered healthcare and can be of added value in evaluating and improving healthcare.³⁹ Van Staa *et al* explored the satisfaction of transfer for a broad range of chronic diseases using the OYOF-TES and NRS (mean 61.8, SD 13.21 and 6.63, SD 1.79, respectively).²⁵ They reported that almost 20% of the patients were unsatisfied with their transfer (NRS <6). Our study showed that 35% of the patients rated the transfer process as unsatisfactory. About 45% of respondents felt that they were ready for transfer which is less than reported by Staa *et al*²⁵ (64%) and in other literature.^{40 41} Of the respondents in our series, 30% reported that they were well-prepared, which was consistent with the qualitative data but again less

when compared with Staa *et al* (49%). Furthermore, Staa *et al* reported that the male gender perceived transfer more positively than their female counterparts, while we found no gender differences. The only association found was a negative correlation ($R^2=0.25$; $p=0.026$) between the OYOF-TES and the number of surgical interventions in the past. We hypothesise that the more surgical procedures patients have had in the past, the more hospitalised they become and the more difficult it could be for them to leave the familiar surroundings of the paediatric ward. Patients reported that the paediatric ward became more of a second home the more often they were admitted there.

The timing and preparation for transfer is an issue in most studies.^{27 42 43} The healthcare providers would not extend the age limit but prefer to assist patients in developing skills to prepare them for transfer. They also emphasised the need for improvement of interprofessional collaboration to make transition successful. Only 25% of the survey respondents perceived the collaboration between paediatric and adult healthcare services as good, which supports the findings from the interviews. Early and sufficient preparation, getting familiar with the adult care services prior to transfer, discussing the possible pitfalls and improving self-management of the patient could all be helpful in closing the gap of transfer.⁴⁴

The transitional care has a tri-phasic nature with the first phase being preparation, the second phase being the event of transfer and the third phase following after transfer. Previous studies on transitional care have tended to focus on the event of transfer rather than this third phase.²⁰ In MO, this last phase might be even of more importance because of the chronic aspect of the disease, the many surgical procedures generally performed in adulthood and the life-long burden of possible malignant degeneration. Considering these aspects, further research is needed on how these youngsters are affected in the third phase and whether providing developmentally appropriate healthcare and tools to obtain transitional skills can affect this phase.

To our knowledge, no studies have been performed on transitional care of patients with MO.

However, several other studies with a variety of chronic diseases, such as RMD and childhood cancer, have been performed and have actually promoted similar approaches for transitional care across these different diseases.^{20 45} Patients with MO may experience a lot of problems that are also encountered by patients with RMD and childhood cancer, such as chronic pain, functional impairment, chronic use of medication and the fear of relapse. On the contrary, patients with MO differ on several aspects, with MO being a chronic disease with a life-long risk of malignant degeneration, the necessity of multiple surgical procedures throughout life, but also on having a parent with the disease. Therefore, a different approach might be warranted and the disease-specific conditions should be an important part of the transition protocol.

Even though the majority of studies in literature addressed a variety of chronic diseases, different chronic illnesses seem to require specific approaches; a ‘one size fits all’ transition programme’ will be insufficient.^{33 46 47}

Moreover, the majority of studies have focused on the paediatric setting and single perspectives of patients.^{28 48–53} Only a few studies investigated the views of healthcare providers in the adult care setting.^{29 54 55} This study provides a broad range of perspectives from different stakeholders in different settings. With the convergent parallel mixed-method design, we were able to provide different but complementary data on the same topic. Moreover, this design allowed us to bypass any social desirability bias from semi-structured interviews.

This study is limited first by the small number of survey respondents due to the rare aspect of the disease. Second, not all patients could be reached, therefore it is not known whether they have dropped out of care or whether these patients experienced no problems during transition. Therefore, selection bias cannot be excluded. Finally, the hospital setting in this study is not generalisable. Studies showed that patients with other chronic diseases who had been transferred to the same hospital and same building were more satisfied with their transitional care.³ Although our patients were transferred within the same hospital but to a different building for their clinical admissions and surgical procedures, they still viewed it as a different hospital.

CONCLUSIONS

Overall, the transfer process for patients with MO was unsatisfactory. Patients should be more in the lead of their care and need to become independent from their parents. Self-management skills are underdeveloped in this group of patients and warrant more attention and support from healthcare providers. If not guided properly, transition can be a traumatising period especially for these patients.

Therefore, an early and good preparation using an individualised transition plan, improving self-management skills and interprofessional collaboration are key elements for a successful transition process. Future research should point out whether translation of these recommendations to a transition protocol are effective in improving the quality of the transfer process in transitional care.

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