Qualitative final write up

Abstract

Introduction and Objective

. For patients with Haemophilia, in some patients activity has been shown to cause bleeding into joints leading to potential joint damage, while in others it protects the joint from bleeds and further destruction. The purpose of this study was to investigate the beliefs and experiences of exercise and activity in persons with Haemophilia (PWH).Method and materials We interviewed six men with severe haemophilia using a purposive sampling framework and the methods of Interpretative Phenomenological Analysis (IPA).semi structured interviews. .

Results

Five themes emerged from the analysis.

1) “Don’t think about Haemophilia, I’ve got to deal with it”
A level of acceptance that they are aware of their condition but they don’t want it to rule their lives.

2) ”I don’t let my limitations hold me back.”
Despite joint impairment striving to find activities they can participate in.

3) ”The worst thing anyone can do is stop being active”
Belief that activity helps to strengthen joints, gives confidence and improves both body and mind.

4) ”The best thing they did was educate me”
Knowledge of Haemophilia, how to treat and recognise bleeds and finding activities to suit their bodies.

5) “Time constraints at home”
Common barriers to exercise as the general population.

Conclusions

These findings provide the clinician with some insight into understanding the attitudes and experiences that patients with haemophiliahave towards activity

and exercise and how best to support and help individuals find the right activity/exercise for them.

Introduction

Hemophilia is an X-linked bleeding disorder characterized by deficiencies of Factor VIII or IX. With the advent of safe clotting factor preparations, most of those born with hemophilia survive into adulthood free of human immunodeficiency virus (HIV). As such, almost half of the approximately XXXX patients with hemophilia (PWH) living in the UK are now adults (ref).Encouraging activity and exercise is a major issue facing the world's population, with our ever increasing sedentary behaviour [ref]. There are many strategies to help encourage and promote the general population to take part in exercise and become more active. The World Health Organisation survey in 2002 stated that two thirds of the European Union were not achieving the recommended levels of activity. Look up UK levels

Lack of exercise and physical activity has been shown to increase the risk of many common conditions e.g. Diabetes Mellitus, cardiovascular disease, hypertension, osteoporosis, certain cancers and depression [ref].

Historically persons with Haemophilia (PWH) where unable to participate in many sports and physical activities due to their low factor levels, which in turn lead to bleeds into the joints and muscles. This bleeding caused joint damage and development of a haemophilia arthropathy.

With the recent advances in pharmacological treatment, PWH can now participate in regular exercise with protection against bleeds. Although this treatment has revolutionised their treatment and improved their quality of life, it doesn't offer full protection, as joint bleeds may still occur despite regular prophylaxis. There is still great debate as to what are the best forms of exercise for people with haem. The literature agrees that exercise will help with improving muscle strength, joint stability and joint proprioception. This in turn will help reduce the joint bleeds and subsequent haemophilic arthropathy.

There have been many papers discussing types of exercise and exercise prescriptions[refs] but limited studies looking into patients' experiences of being active with Haemophilia. Previous studies have also looked at many aspects of quality of life e.g. psychological wellbeing, coping strategies, family issues with haemophilia[refs]. These have used validated generic questionnaires e.g. SF 36, EQ-5D, together with disease specific questionnaires

e.g Haemo-QoL-A. Only a handful have interviewed patients directly and none to our knowledge have asked specifically about exercise and activity.

This study aimed to investigate the beliefs and experiences of PWH on being active and participating in exercise..

Methods

We used the methods of Interpretative Phenomenological Analysis (IPA) to explore the meaning of activity pacing for physiotherapists (Smith, Flowers and Larkin, 2009). Phenomenology is concerned with understanding the meaning attributed to a particular experience or phenomenon. It aims to explore the participants' view and to adopt as far as possible an insiders perspective (Smith and Osborn, 2003; Smith and Osborn, 2007).

Participants

We selected a purposive sample of patients with a diagnosis of severe haemophilia. We selected patients with severe….. All interviewees had English as their first language. In our sample the age range was … and the years of symptoms with haem was ….

Recruitment

Recruitment

We contacted all patients with Haemophilia registered at the Oxford Haemophilia and Thrombosis Centre between the ages of 18-69 yrs old. They were invited by post to take part the study to investigate their beliefs about in exercise and activity. We sent 256 invitations ; of these 43 participants with severe Haemophilia responded and ** gave consent to be contacted about being interviewed

As IPA aims to explore a detailed interpretative account of a small sample and thus gain 'insight into a particular experience', six subjects is considered a sufficient number to participate (Smith and Flowers, 2009; Shaw and Flowers, 2010).

Table of interviewees

	Age	Severity		

1 DM	46 yrs	Severe A	Male	Ian
2HM		Severe A	Male	Joe
3AS	18yrs	Severe A	Male	Andrew
4MP		Severe A	Male	John
5NW		Severe A	Male	Simon
6AC		Severe A	Male	Jim

SHOULD I GIVE EACH INTERVIEWEE A PSUEDONYM?

Interviews

The interviews were semi structured in nature and were conducted on a one-to-one basis. Participants were given to option of where to be interviewed, either at home, a place of their choice or the hospital setting. All participants chose the hospital setting and reasonable travel expenses were offered. Every attempt was made to put the person at ease and it was made clear that anything said in the interview would not affect their treatment. The interviewer also made it clear that the research was interested in the participant's experiences with exercise and activity and not to pass judgement.

Prior to interviewing participants the interview framework of questions was formulated and reviewed by other physiotherapists in the field. An example of the interview schedule is appended in App 1 [or add as table]Table ..

OR SHOULD I JUST SAY AN EXAMPLE OF QUESTIONS AND PROMPTS USED WERE….

Experiences

1) Can you tell me about your experiences of having haemophilia and trying to be active?
2) How do you feel about having haemophilia?
3) Would you describe yourself as active?
4) How would you describe someone who is active/not active?
5) Can you describe any benefits of being active for someone with haemophilia?

Barriers/ benefits to exercise

1) Can you describe anything that might hinder you being physically active? Can you describe any specific examples?
2) Can you talk to me about the last time you tried to exercise?

3) Can you tell me how you feel when you do exercise and are more physically active?

4) Can you tell me about your pain levels and exercise?
   Do you take any painkillers, other meds to help with pain e.g. anti-inflammatory from the centre?

History/timeline

1) What did you think when you were growing up about exercise and activity? Can you give me any examples of the types of activity you enjoyed? Health professionals' advice at the time? Parents' views about activity?

   Did you do anything different because of your Haemophilia?
   Did people treat you differently?

2) On your information it says you started prophylaxis at …yrs old. Did this have any impact on the way you active or exercised?

   Prophylaxis/on demand?

3) Can you describe what your parents thought about exercise and haemophilia?
4) Are your family active/sporty?

5) As you grew up, into your teens and twenties how did your exercising habits change?
6) As you have gone through life how has your activity changed?

7) Now as a …yr old, what activity would you like to participate in?

Health service provision.

1) We are trying to plan a service to encourage men with haemophilia to be active, what would you include in it to encourage increased activity?

If you were planning a service for PWH what would you include to help encourage activity?

How would you encourage someone with Haemophilia e.g. sore ankles to be active?

The interviews lasted approximately 45 minutes.

All interviews were recorded and transcribed verbatim and participants were given pseudonyms to allow anonymity. The researcher made notes after each interview recording initial impressions and other information gained once the recordings had stopped.

Data Analysis

IPA used! Interpretative Phenomenological Analysis

The lead researcher read and listened to each interview line by line. Each line was coded and the essence of the meaning was placed in the right hand column. A second stage was to make a list of emergent themes in the left hand column. The aim of this is to develop connections between the data.

I then did summary sheets of themes for each participant and I USED POST IT NOTES OF THE THEMES CONSENSUS!! How do I say that?

To promote rigour, ST discussed the themes with a second researcher FT an expert qualitative researcher to assist in thematic analysis.

(How to refer to ST / FT?)

Findings

This study explores the experiences and beliefs of being active with Haemophilia. We identified 5 themes

1) "Don't think about Haemophilia, I've got to deal with it"

2) "I don't let my limitations hold me back"

3) “The worst thing anyone can do is stop being active”

4) “The best thing they did was educate me”

5) “Time constraints at home” The same as everyone else.

Theme 1 “Don’t think about Haemophilia, I‘ve just got to deal with it”

ADD A DIAGRAM

Throughout the participants there was a consensus that they got on with their lives and didn’t give haemophilia much thought

“I don’t consider myself a Haemophiliac until I need to treat my joints for any reason, so I don’t go walking around with that consciously in my head, but subconsciously it’s always there” DM

“Most of the time I ignore it but not in a negligent way, but in a way that I won’t let it affect me” HM

The interviewees all expressed frustration with the bleeds into their joints but that it was just something that they had to deal with.

“Obviously there are times if I get a bleed it’s pretty demoralising and stuff like that but I try and do succeed I think in everyday life in not letting it be a major part of my life cos it’s not a conscious thing, it’s something I’ve done and if I did think about it the whole time and let it dictate things then it would just bring you down, which I strongly don’t think should happen.” HM p2

The frustration of having to be still and wait for bleeds to subside.

“I hate having a bleed, having to be still. I don’t like sitting at home as you have to rest it. It is frustrating.” AS p2

The subject of disclosing Haemophilia came up in all the interviews and there was generally the consensus that they didn’t want to tell everyone about their Haemophilia

I don’t tell people unless I consider them to be someone who needs to know, for example someone at work with lots of different people they don’t really know, my close friends know, they have been friends for years, there is no real reason to be telling anyone. You can manage being a haemophiliac without

having to have a warning sign on your forehead saying I'm a haemophiliac" DM p4

Others preferred to be like others and use others' behaviour e.g. being injured in football to cover a bleed.

"Because if I hurt myself playing football, everyone got hurt on a Sunday so it was normal. I don't tell loads of people." AS p 4

While others were open about their haemophilia

"Everyone pretty much knew, I think. There is a stage where children just are sort of curious and ask questions which I thought was fine." HM p 21

Theme 2 "I don't let my limitations hold me back"

All participants felt that it was important to be able to adapt as their bodies changed with age and ability.

"I am lucky I have got the aptitude to adapt" MP p 15

To be positive about what they can do and not to focus on what they can't and embrace new sports

"It sort of came to a point of trying to find things I could do, not so much trying to find things I couldn't do but things I could do." HM p 1

"I am a positive person, so I embraced cycling and jumped into that trying to allow that to be the focal point rather than the unhappiness of not being able to play golf." DM p 12

When certain activities they used to able to perform become difficult they are able to alter the movements to be able to continue.

"I played golf yesterday, 18 holes thoroughly enjoyed it, was quite sore at first and I had to change my swing to put less movement on the joints." DM p5

“I say kick a ball, I will throw it and my son kicks it back” AC

Theme 3 “The worst thing anyone can do is stop being active”

Activity was a good thing as it helped with joint strength and gave confidence in their body.

“Exercise gives you confidence, so sometimes what holds me back is just a lack of confidence, I think if I do this I feel my knee is going to go, whereas actually doing that and being out cycling not only gives you the confidence but afterwards your legs feel a bit stronger. It gives you the confidence to do something similar or more next time” HM p 5

Exercise helps with social inclusion and helped prevent the feelings of isolation and being different.

“I think everyone should be as active as possible, keeps you stronger. I think you have less bleeds and socially you can join in with everyone and don’t feel excluded.” AS p 19

Despite arthritic joints finding the right exercise for their bodies, they were able to participate with their families in activities and feel better.

“I went cycling earlier this year and I did feel better and my knees did get a bit better.” AC p 13

However not all activity suited everybody. Football was mentioned as a sport that caused problems with ankle joints.

“I don’t want to miss out on all the memories of playing football, but I would actually. I would swop that for healthy joints now.” DM p 14

“Probably why I have difficulties with my left ankle from kicking a football.” AC p5

“We tried to ignore the condition we had and it gave us lots of problems as a consequence. Half my problems with my ankles was because I played football and shouldn’t have done.” MP p16

Some were able to vary the amount and level of football to allow them to continue something they loved to participate in.

“I think that the ankle problems as such where when I was doing too much but since I have been sensible with what I do, it is all fine now.” AS p 14

Others expressed the opinion that if they had been warned of the potential damage that football could give that they would have not played as a child.

“If I was really given the choice now I wouldn’t have kicked a ball if I could have healthy ankles now and play sport that I can play into my old age!” DM p 15

Football was the only sport mention to potentially cause harm, most participants expressed the importance of finding an activity that will take you through life. One that you will always be able to participate in and enjoy.

“I would say is to find activities that will not give you long term effect on joints cos once you have got a damaged joint that is just painful, long term pain ,that is no fun for anyone.” DM p 17

Theme 4) “The best thing they did was educate me”

When asked about parents and the Multi-disciplinary team (MDT) and how they influenced their activity and beliefs, they responded that education was essential to be able to manage the condition correctly.

“I really can’t think of much my parents did stop me doing. I think the best thing they did was educate me in a way so I know what to look out for or what to do but when it came down to it, they were quite happy to let me get on and get on with it which I think is really important. For which I am incredibly grateful.” HM p 18

The element of trust from the parents and MDT was important and not to be wrapped up in cotton wool. The element of letting boys explore activity for themselves.

“I consider myself very lucky that my parents were very free with me and my brother and allowed us to be active as we would like and deal with the situations as they come rather than wrapping us up in cotton wool, as that would have changed our lives considerably for the worse I think.” DM p 1

Being given choice of activities and not focusing on what can’t be done but on what can be done.

“That was very important cos if my parents had been over bearing or say you can’t do this you can’t do that, then I don’t think I would have the same mentality about haemophilia now.” HM p8

“I think the advice has been fairly similar quite positive, there has never been that message of don’t do this don’t do that or this might happen.” DM p8

At school the importance of being included and the option to do alternative activities and not being singled out and excluded was raised.

“I think the teachers who made me feel different and I suppose it was them being nervous and stuff like that, but at the time I didn’t like that and I am sure now I would probably have sympathy with them, but when you are growing up you don’t want to be treated differently. So the best teachers were the ones who were aware and you could talk to but treated you like normal.” HM p20

“I was a referee for a while, I got involved in other ways”. MP p7

Theme 5 “Time constraints at home” The same as everyone else.

Activity and exercise were not always easy to fit in to life.

Time and opportunity were a problem

“Time constraints at home and finding a nice place to cycle. Like being near a nice path rather than on an A road. It would be nice to just cycle from the house. I can’t complain but the hassle of putting the bikes on the car …It is just another barrier” AC p13

“I haven’t been able to play football on a Sat because if the job, it is a bit annoying.” AS p5

Pain was also mentioned as a barrier to activity

“It is the strength and limited movement and pain that stops me doing exercise.” AC p8

Pain medication was mentioned to help but none of the interviewees took them regularly but on a need only basis. Preferring to live the pain

“The eterocoxib helped a lot but it seemed a habit to keep taking them. I still obviously got problems with the joints anyway. I just take them if my joints are really bad.” DM p7

“Arcoxia tablets seem to help.” NC

Discussion

There is little existing research which explores the experiences of exercise in PWH.

To our knowledge this is the first study to interview people with haemophilia about their experiences and attitudes towards exercise and activity using IPA.

There are many studies which investigate quality of life and the impact of Haemophilia but these articles tend to use validated questionnaires with fixed questions. Though their outcome is interesting it does not allow for more in-depth investigation into the real experiences.

SHOULD I LIST A FEW STUDIES HERE AND SUMMARISE THEIR FINDINGS?

EG SYSTEMATIC REVIEW IN 2012 24 STUDIES ON PSYCHOLGICAL ASPECTS OF HAEMOPHILIA 20 WERE QUESTIONNAIRE AND ONLY 3 WERE INTERVIEW.BUT NONE

The Haemophilia population are now living longer thanks to the development of recombinant replacement factors and are also more active than previous generations of PWH. Activity/exercise is now advocated for this population and with individualised prophylaxis many PWH can participate in most types of exercise. As previously discussed the general health benefits of being active to reduce the problems of obesity, reduction of developing type 2 diabetes to name only a few are now important to this group as they are now experiencing a similar life expectancy to the general population.

Understanding the experiences of being active with haemophilia is important for the clinician to help them advise and educate the most suitable form of activity. Baxbaum 2010 investigated the activity levels of adolescents with haemophilia and found they were equally inactive as the general population and suggested further research was needed into the psychosocial barriers of activity to help our understanding to encourage more activity.

Lots of studies advocate exercise but the type of exercise needs to be individualised as stated by Gomis 2009. Exercise needs to be assessed for risk, its biomechanical demands and also the individual's physical capacity. Gomis also discusses in the systematic review that activity not only has physical benefits but also psychological and social benefits too.

Theme 1

Beeton 2005 reports that persons with chronic health condition often report a positive quality of life. Following the interviews participants expressed a feeling of getting on with life and not focusing on the haemophilia. That they weren't to be defined by their condition. They tended to have strong internal locus of control and these favourable psychological characteristics appeared to reduce the perceived seriousness of haemophilia . Triemstra 1998.

Beeton talked about acceptance of condition (2005 Haemophilia An exploration of health related quality of life). The men interviewed all accepted their condition as they knew no different but they all expressed the need to get on with life, that they were treated similarly by friends and wanted to be treated like everyone else. This acceptance of their condition has also been described in other literature Reynolds F (The subjective experience of illness in physiotherapy A psychological Approach 3rd edition Oxford butterworth Heinemann 2004) From this study,their coping styles, acceptance and positive outlook may explain how they have a generally good experience with activity and exercise.

They expressed frustration when they had bleeds and being unable to go about normal life but acceptance that this was part of life and something that was not going to get them down

The issue of disclosure of their condition was discussed most considered to tell others on a need to know basis, while others were happy for everyone to

know. This tended to differ with age. The younger subjects were more relaxed about disclosure while the older participants were more wary often due to being co infected with HIV or Hepatitis C. However even the younger participants used sports injuries as a reason for not being able to play due the the joint bleeds sustained from football.

Theme 2

The ability to adapt through the natural life cycle of the condition was a common finding with all the participants. All the participants were very focused on what they could do SHOULD I LINK WITH QUOTES FROM THE RESULT SECTIONS?EG hm FOCUS ON WHAT I CAN DO .

They all expressed a positive outlook, being adaptable. It has been suggested that problem focused strategies can reduce stress and improve adaptive strategies to help improve quality of life.(Cassis 2012)

Our beliefs about what causes our actions then influence our behaviours and attitudes.

Theme 3

Activity was important to all, there is much in the Haemophilia literature about the importance of exercise in joint protection, general body health both physical and psychological. Subject expressed the confidence that exercise gave them and that it allowed them social inclusion. Despite some physical limitations the drive to find some activity to take part in. Football however was the only sport mention that had detrimental effects. While it gave them great pleasure, some blamed football for their joint damage. Others stated that if they had been warned about the potential effect of the bleeds on their joints that they would have chosen a different sport. In reality would this advice have been adhered to, perhaps this is something to investigate further.

Theme 4 Education

Choice, decision making working together was another theme that emerged Wittmeier 2006 Haemophilia also found this.

Having the knowledge about Haemophilia, the tools to make decisions about activity and exercise were expressed as very important. The MDT and parents were viewed as important to help inform, facilitate changes and educate . This was also found by Drake 2010 Am J Prev Med. The need for trust between

groups was noted and the choices of activity important. Not to focus on only the activities that are not advised but on those that are suitable.

Reasons not to exercise

The finding showed that PWH experienced many of the same barriers to exercise to those of the general public. One of lack of interest, ability to find time and opportunity to exercise. These finding were supported by Sherlock 2010 who reported pain, arthritis prevented participation in sport but also lack of interest, lack of time which are cited in for reasons in the general population too. Pain was mentioned and the use of pain medication was found to helpful to some but other actively chose to live with the pain. This belief would be interesting to explore further, as many PWH feel that just need to live with the pain from arthropathy and nothing can be done to help.

The research in this area in increasing but like Sherlock 2010 who evaluated the activity of the Irish Haemophilia population , we also conclude that more research into the beliefs around exercise are needed to gain actual information about how this cohorts experience of activity.

Clinical Message

Choice/options/flexibility to change sports/activity SHOULD WE GIVE MORE WARNING FOR FOOTBALL? Should we still take each individual and assess their abilities and tailor activity to suit?

Sherlock mentions need for choice of activity to suit the individual

Trust with MDT and parents

To get on with life and not focus on Haemophilia and that activity and exercise are helpful but find the right one that you can take through life.

Limitations

Due to the nature of qualitative research the finding are specific to persons with severe haemophilia. We did not explore the experiences of persons with moderate or mild severity types.

Due to the nature of the recruitment process for the study, only those persons happy to consent to talk about their experiences were included. This may affect the findings, as this group demonstrated characteristics of having a positive outlook, motivated to be active and altruistic. These results may not be able to be transferred across the whole severe severity group.

We plan further studies to explore the themes raised in more depth and to investigate attitudes and experiences across both moderate and mild severity types. Also what PWH experiences are towards football and the potential risks.

Conclusion

This study confirms the findings of many others that having an inherited condition doesn't always cause a poor quality of life. PWH accept their condition , they are positive towards exercise and find it of benefit both physically and psychologically. They are adaptable and can alter activities as required as their bodies change. They value education from both parents and the MDT to give the PWH the knowledge to make informed decisions regarding exercise and activity choice. And finally PWH are no different from the rest of the population regarding some of the barriers to activity. Pain from arthritis, together with lack of motivation and time.