Witness Name: Katherine Victoria Burt
Statement No: WITN6392001
Exhibits: WITN6392002 - WITN6392267

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WITN6392253

The social and economic impact of Hepatitis C in people with Haemophilia

Report of a pilot study

Prepared for the Haemophilia Society

Sarah Bond and Jennifer A Roberts

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March 2000

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Executive Summary

1. **The Aims of the study were:**

to test the feasibility of exploring the impact of Hepatitis C on those with haemophilia using a semi-structured interview and postal questionnaires to measure health status.

- to report on the findings of the pilot study, and
- to provide a revised research instrument that could be used later in more comprehensive studies.

2. **Study population and methodology**

Twenty five names and telephone numbers of members who had agreed to be interviewed were provided by the Haemophilia Society. They included men and mothers of children with haemophilia and women with von Willebrand's disease. Twenty two persons were interviewed, two could not be contacted and one could find no suitable time for a confidential interview. Complete information was available on 20 people. Nineteen people who agreed to provided an address were subsequently sent a SF36 health status questionnaire to enable their health to be compared with people with other diseases and those of the general population. Thirteen of the 19 (68%) returned fully completed questionnaires.

3. **Testing the Feasibility of exploring the impact of Hepatitis C**

- The major difficulty in assessing the impact of hepatitis C in persons with haemophilia is the lack of a control group of people with haemophilia who do not suffer from hepatitis C. This problem is confounded by the co-infection of some cases with other blood borne diseases including HIV and hepatitis B.
- Standard questions about the impact on activities of daily living did not seem to be sufficiently sensitive to attribute the impact of the disease to the likely cause, i.e. haemophilia or hepatitis C. Full discussion with cases, however, did enable the more extreme symptoms of haemophilia and hepatitis C to be distinguished. The range of symptoms attributed to hepatitis C was compatible with those found in the literature.

- The summary scores on the health status measures were low compared with the general population and others with hepatitis C but the sample was small.
- The problem of attribution could be addressed by comparing cases with different severity of haemophilia and at different stages of hepatitis C. Health status for each combination of severity can be compared to others at different stages of hepatitis C who have acquired hepatitis C from blood products and who do not have haemophilia.

Usefulness of telephone interview to assess the impact of hepatitis C

- The telephone interview was very effective in obtaining information about the factors that led to the diagnosis or the disclosure of diagnosis, information about whether or not they had obtained treatment for hepatitis C and whether it had worked or not. Details of the impact of hepatitis C on their lives and the lives of their families was discussed fully.
- It was difficult to obtain precise clinical details of hepatitis, comorbidity and investigations and treatments from all respondents systematically using a telephone interview. Future research should include a survey of case notes to obtain clinical details of haemophilia and hepatitis C.
- It was possible to obtain rich descriptive information relating to education and employment history but precise financial details were not easily obtained by telephone. A structured postal questionnaire would probably be a better way to obtain these data.

Information on health status measure (SF36) was successfully obtained using a postal questionnaire.

4. Report on the findings of the pilot study

The numbers in this study were small and care should be taken in drawing conclusions from them. From this small sample, however, some worrying aspects emerged which merit further inquiries.

Clinical Care and Management of Hepatitis C

- Many cases were not told that tests had been taken. The way cases were told that they were infected with hepatitis C was not satisfactory.
- General practitioners and some centre staff (including consultants) either were not fully aware of the illness and its likely progression or were unable to convey this information.
- Lack of knowledge of the likely progression of the illness was causing extreme anxiety amongst many patients. and caused some to adopt extreme precautions, whilst others seemed unaware of any dangers.

- There was a general lack of counselling for those affected. Parents of teenage children could not obtain counselling help either for themselves or their children.
- Access to interferon α was varied across the country and information about its likely impact and side effects was rarely given.
- The Haemophilia Society and the British Liver Trust rather than the NHS had been the source of information.
- Some self-help groups had been set up to deal with the lack of information and support.

Impact on life

- Interruption of work and school was reported. This was sometimes directly attributed to hepatitis C, but especially amongst the older group, was caused by the underlying haemophilia.
- Family life had been affected in most cases. Inability to plan for the future, worries and uncertainty about life expectancy and morbidity were blighting the lives of many respondents. Leisure activities were curtailed or limited and some times the illness was implicated in marriage failures.
- Many people felt stigmatised by the illness. A number did not want their immediate family to know they had the disease; others did not want work colleagues to know.
- Most had experienced problems with mortgages and life insurance.

5. Conclusion and Recommendations

- A study of the impact of hepatitis C should be conducted from a sound epidemiologically based sample of cases at each stage of the disease. Clinical records should be used to obtain details of the severity of the disease, dates of investigation and treatments. The telephone interview should be used to assess the experience and impact of hepatitis C on the lives of cases supplemented by a postal questionnaire to measure health status and details of income and expenditure.
- The NHS should make provision for high risk groups exposed to repeated doses of blood based products. Informed consent to tests should be obtained and sensitive reporting of test results with advice and information should be made available. Access to counselling and therapy where appropriate should be made available.
- Clinical governance and control assurance should ensure adherence to guidelines.

The social and economic impact
Hepatitis C on people with Haemophilia
A Pilot Study

Introduction

Hepatitis C (HCV) has become a major issue for people with haemophilia and Willebrand's disease. Little work has been undertaken to assess the impact of the infection on people with haemophilia and those who care for them. This work, undertaken for the Haemophilia Society, has piloted a research instrument to assess the feasibility of exploring these issues.

This report begins with background information on hepatitis C and its consequences and the way in which therapies may have an impact upon the course of the disease. The aims and methods used are described and the findings are discussed. Recommendations for methods that might be used in further research are outlined and suggestions for research instruments that may be used are provided.

Background

Although some information is emerging about the natural history of Hepatitis C much remains to be discovered. Evidence is accumulating but is complex. Hoofnagle, (1997) recently described the natural history of hepatitis as having 'no single typical course...but rather a broad clinical spectrum of the disease presentations and outcomes.' Presentation with acute hepatitis is not typical. The disease usually has few symptoms and may be sub-clinical. The disease progression is also complex. It appears to resolve in about 15% of cases whilst the remainder have chronic viral hepatitis infection. A proportion of these cases proceed to cirrhosis of which a further proportion (20% -30%) will have liver cancer (HCC) or end stage liver failure over a period of ten to thirty years, (Seeff et al 1992, Tong et al 1995, Poynard et al 1997, Darby et al 1997). In his review Hoofnagle (1997) predicts that there are 'multiple typical courses, from rapidly progressive to slowly progressive to non-progressive.' The progression is thought to be affected by genotype, co-morbidity and immune status - particularly HIV status - age at infection, viral load and alcohol use. Hepatitis C is one of the emerging issues for public health world wide. In the USA 20% of acute hepatitis is attributable to hepatitis C (Alter and Mast, 1994, quoted by Hoofnagle, 1997). 'Deaths from Hepatitis C are expected to triple over the next two decades becoming responsible for greater mortality than AIDS,' (Di Bisceglie 1998.) Infections are decreasing as a result of successful screening of donated blood but deaths of those already infected with hepatitis in the UK will rise from 8,000 to 24,000 in the next 20 years (Tanne 1997).

Hepatitis C is of particular significance for those with haemophilia. Lee (1995) summarises the history of transmission of hepatitis through blood products. Since Beeson (1943) first postulated a link between blood transfusions and jaundice in seven cases, the practice of retaining a portion of blood or serum used in transfusions was recommended in order to assess the transmission of hepatitis. Because of this practice Kasper and Kipins (1972) were able to undertake an investigation that noted a high incidence of large-pool clotting-factor concentrates amongst haemophilia cases treated with factor VIII and it was established that prior to the introduction of heat treated clotting factor almost all patients developed hepatitis C. The high transmission rates were attributed to the pooling of 1,500 donor plasmas during a period in the USA when the incidence of HCV in volunteers was 7% and in commercial blood donors was 28%. A cohort of haemophilic patients in the USA reported a 98% prevalence rate of hepatitis C in those who had received untreated clotting-factor concentrates, (Dhillon, 1995).

The period from exposure to the development of liver disease has been estimated and it appears that co-infection with HIV accelerates the development of liver failure and that the viral load increases as the immune deficiency advances, (Telfer et al 1995). '15% of patients with co-infection have liver cirrhosis at 10 years after infection compared with about 2.5% with HCV alone,' Dhillon (1995) quoting Soto et al (1994). This possibly explains the poor response rate to interferon that was observed in this group although serotypes would also modify the effectiveness of treatment. The treatment of hepatitis C involves dealing with symptoms as they occur. The interruption of the disease has been made possible for a proportion of cases by treatment with interferon. The availability of this treatment has been uneven across the UK. The current issues relate to the usefulness of combined therapy - interferon α and ribavirin - and about the appropriate stage of the illness at which to treat cases and the possible selection of genotypes that are susceptible to treatment, (Poynard 1998, Dusheiko, Khakoo, Soni et al. 1996). The evidence from randomised multi-centred trials reported by Poynard indicate that for all genotypes combined therapies - interferon α 2b and ribravirin - had higher response rates than interferon α 2b alone. The differential response varied significantly by genotype. The response increased from 33% to 64% for genotype 2 and 3 and from 11% to 18% for genotypes 1, 4, 5, 6 - this response rate was increased further to 31% when the treatment period was increased to 48 weeks. The most favourable response occurs in women, under 40 with genotype 2 and 3. In this group the response rate increased from 33% for interferon α 2b to 80% for combined therapy of 48 weeks or more.

There are side-effects to treatment. Interferon needs to be injected - not a very pleasant process. Commonly reported side effects of interferon include malaise, muscle aches, headaches, poor appetite, weight loss, sleepiness and depression. These adverse effects of interferon α noted in a clinical trial were mostly mild and common to all groups, but those on combined therapy (interferon α and ribavirin) also mentioned dyspnoea, pharyngitis, puritius, rash, nausea, insomnia and anorexia, (Poynard 1998).

Resources are scarce so questions arise about whether interventions are worthwhile and which are the most cost-effective treatment regimes. To answer these questions we need to assess the impact of therapy on disease progression and to compare the costs of managing the illness using alternative strategies. Some exploratory models have been undertaken to assess the likely cost effectiveness of therapies, (de Ancos et al. 1990; Dusheiko and Roberts 1995, Reinne 1997, Wold 1998). As knowledge of the disease is still accruing, and given the long course of the illness, the most useful approach is to model the likely scenarios, and to project the outcome over time. Previous studies have taken this approach (Garcia, Roberts and Dusheiko, 1990; Dusheiko and Roberts, 1995). The Dusheiko and Roberts, (1995) model, based on a hypothetical cohort of 1000 cases, predicted that intervention with a thirteen week course of interferon α would result in fewer deaths (13 and 43 fewer depending on the transitional probabilities used), fewer cases would have cirrhosis, fewer cases would decompensate and there would be need for fewer transplantations. There are no specific studies that have systematically assessed the effectiveness of interferon therapy amongst haemophilia cases, Roberts, (1999).

Whilst much is still to be discovered about the disease, the economic implications for cases and their families and the health sector has been grossly neglected. Few studies have explored the impact of the infection on the health status of cases, Davis et al. (1994) and Foster et al. (1997). Little is known of the impact of hepatitis C on the daily life of those affected. The health status of those with mild chronic viral hepatitis C has been measured using SF 36. Foster et al. (1997), studied seventy two patients (39 women and 33 men) with a mean age of 39 years who suffered from chronic viral hepatitis C, the patients in this study had been given the same information and had obtained counselling from experienced nurses. The health status scores for these people were compared with those of the general population of the UK and with a sample of cases with hepatitis B. It was established that those with chronic viral hepatitis C had lower scores on all dimensions of illness. Those who had been diagnosed after presenting with symptoms had similar scores to those who were identified by routine tests. Those with more 'severe' disease, identified by tests to indicate the degree of hepatic inflammation, did not score higher than those with less. Those who had ever been intravenous drug users had lower scores on all variables except pain and general health perception than others with hepatitis C. Those with chronic hepatitis C scored much lower than the general population on all dimensions. The authors (Foster et al 1997) considered the possibility that the measurement was picking up a general response to living with a serious illness. To test this hypothesis the hepatitis C cases were compared to a similar group of cases who had hepatitis B. Whilst this group of cases had reduced scores for mental health and general health perceptions, they did not show such reduced scores on physical function as groups with hepatitis C. The authors concluded that this indicated that hepatitis C caused substantial and significant reductions in physical function.

Few studies have explored the implications of hepatitis C on those with haemophilia. Miners (1999) measured the health status of haemophilia cases. The findings of this study indicate that those with haemophilia, of all levels of severity, had lower health status scores than the general population. These scores were lower than those obtained for cases with mild hepatitis C (Foster et al. 1997) who did not have haemophilia.

Fewer studies have assessed the more general socio-economic impact of hepatitis C on those with haemophilia. A notable exception is the comprehensive descriptive study by Cheetham (1996) funded by the Haemophilia Society. This work used a number of qualitative research methods including focus groups, semi-structured questionnaires and interviews. In addition the study included a component of participant observation as the researcher took calls from people seeking information on hepatitis from the Haemophilia Society and visited Haemophilia Centres and attended conferences on hepatitis C. Extensive policy recommendations were made. The present study aims to extend this work by developing a research instrument to assess the social and economic impact of the infection systematically.

The Study

The Purpose of the study

The present study was set up to explore the feasibility of assessing the socio-economic impact of hepatitis C upon people with haemophilia; to develop a population based instrument to quantify and describe the impact of hepatitis C on cases and their families, and to report the findings of the pilot study. A revised research instrument was to be produced.

Method

The method chosen to assess the socio-economic impact involved using a telephone interview that was based upon a semi-structured questionnaire design. The questionnaire (Appendix A), enquired about the characteristics of the respondent's age, sex, marital status, household composition and employment status. The survey also collected information on the type of haemophilia, the effect of hepatitis C on the health status of cases, the management of the illness including components of the way cases were told about their hepatitis C status, and the medical management and support given, both at the time of diagnosis and later, whether any treatment was given and if so with what effect. Side effects of interferon were documented and the reasons for the interruption or cancellation of the therapy noted. It was intended to discover how far hepatitis C had affected the life-style of the case. The effect of hepatitis C on the family and friends of the respondent was also explored. Finally, the respondents were asked what would be most helpful to them in coping with the infection. A health status questionnaire was sent to those who agreed to be contacted and provided their address.

Sample

Twenty five persons were selected by the Haemophilia Society from people known to have hepatitis C who had agreed to participate. The purpose of the study was described and each case contacted was asked to consent to the interview. The interview lasted on average for one hour. The interview was conducted by telephone during the summer and early autumn of 1998. Repeat interviews took place with a sub-sample to test the repeatability of the interview technique. Twenty two out of the 25 persons who had agreed to be interviewed were interviewed: a response rate of 88%. The remainder either could not be contacted at all or could not find an appropriate time to be interviewed privately either at work or at home. Complete data were available for twenty cases. These cases were analysed. Two interviews with mothers collected information about mothers and their sons. All other interviews were with cases. Information about activities of daily living was collected and the health status measure SF36 that had been used in other studies of hepatitis C was administered as a postal survey. At the end of each interview, each respondent was asked if a questionnaire to measure their health status could be sent to them. If they agreed they were asked for their address. Nineteen persons interviewed provided their address and were sent an SF36 form and a stamped addressed envelope for its return. Thirteen responses (68%) were received.

Results

The results will be discussed for each objective in turn.

Feasibility of exploring the socio-economic impact of hepatitis C

- **General Problems.**

The major difficulty in assessing the impact of hepatitis C in persons with haemophilia is the lack of a control group of people with haemophilia who do not suffer from hepatitis C. This problem is confounded by the co-infection of many cases with other blood borne diseases including HIV and hepatitis B. It is difficult to attribute illness directly to hepatitis C. The respondents were aware of this problem and referred to it directly in the course of the interview.

The use of a semi-structured telephone interview to assess the impact

- **Achieving a response.**

The people telephoned had all agreed to be contacted. Most people could be reached after one or two telephone calls and were either interviewed then or at a later agreed time. An appropriate time was sometimes difficult to negotiate and two could not be contacted. Some could only be contacted at work, some wanted never to be contacted at work, some could not be contacted at home at certain times or if certain family members were present. Not all agreed to receive a postal questionnaire. This was largely because they did not want others in the household to know that they had hepatitis C. The questionnaires that were sent did not mention the hepatitis C. The response rate to the postal questionnaire was 68% and a discrete reminder may have increased this response.

- **Quality of the response.**

Information that was successfully obtained from the telephone interview. All those who agreed to be interviewed gave up a considerable amount of time, and were prepared to discuss the impact of the illness upon them in great detail. Information about the diagnosis of hepatitis C, the factors that led to the diagnosis or the disclosure of diagnosis was clearly conveyed. The respondents were also able to give information about whether or not they had been offered or had obtained any therapy for hepatitis C and explain whether it had worked or not. The impact of hepatitis C on their lives and the lives of their families was discussed fully.

Information that was less easy to assess by telephone interview. Most respondents could give details of the severity of haemophilia, some were able to describe the stage of hepatitis C but the clinical information about the illness, investigations made and treatments given may have been more accurately and systematically collected from clinical records.

Precise information about the financial implications of the illness may have been better addressed using very focused questions, probably in a self-administered postal questionnaire that would have allowed time for respondents to consider their response. The items that would have benefited from such an approach related to source of income, loss of earnings and out of pocket expenses both for day to day expenses and capital expenses. Similarly, questions about insurance would have been more accurately ascertained from a self-administered structured questionnaire. The pilot study provided some insights into how such questions should be framed. Thus the data have to be regarded as illustrative of expenses rather than a systematic estimation of the financial burden. There is always a problem in assessing the costs of living with a chronic illness because the possibility of spending sums that might be required to compensate for the disabilities encountered because of the illness are reduced as incomes are usually low.

It was sometimes difficult to distinguish the impact of hepatitis C and the impact of haemophilia on education, employment and activities of daily living.

The pilot study provided a case study of the perceptions of hepatitis C on people with haemophilia and their families. A study that allows comparison by severity of haemophilia and stages of hepatitis C with cases who have hepatitis C who do not have haemophilia is required. Details of the health status for each stage of hepatitis C will be available from the Mild hepatitis C trial, (Thomas 1998).

Description of the findings of the pilot study

The characteristics of the sample: the age, type of haemophilia, stage of hepatitis C, co-morbidity, are described first, followed by the diagnosis, management and treatment of hepatitis C and a discussion of the perceived impact of the infection on the health and lives of cases. The interview provides insights into treatment and management of hepatitis C and the impact of the infection on the cases and their families. The particular themes that have emerged strongly from the interviews are those surrounding diagnosis, availability of information; attribution of symptoms to haemophilia or hepatitis C, and the anxieties and worries that hepatitis brings to cases and their families. The sample is small and findings need to be interpreted with care.

Characteristics of the Sample

Age and sex distribution

Twenty cases provide full information on age. 65% of the sample were adults between 25 and 60, see Table One. Four respondents were women.

Table One Age distribution of the sample

Age Band	Frequency	Percentage
under 25	3	15
2 6 - 45	6	30
46 - 60	7	35
60+	4	20
Total	20	100

Marital Status

The majority of the sample, 60%, were married at the time of interview. Two persons were either separated or divorced. Eleven respondents lived with partners, two with their children and four lived alone. Five lived with just one other person and the remainder lived in family groups of up to five members. During the interviews several respondents mentioned that they had been in previous marriages that had been dissolved. Separation and failed relationships were attributed to the hepatitis by two respondents, others reported difficulty in establishing or maintaining relationships.

Health Status

Type of Haemophilia

In the sample 70% of those interviewed had Haemphelia A 10% Haemophilia B and 20% von Willebrand's Disease. Eighteen (90%) described their haemophilia as stable or controlled and 10% described it as unstable. Many considered haemophilia was the 'least of *their* problems'..'controlled as it has been all my life'. One person with unstable haemophilia reported more bleeding since acquiring hepatitis C.

Co-morbidity

Eight people reported other illnesses. These were AIDS related illness (2 cases), hepatitis B (2 cases), fibromyalgia, gall bladder, growth on liver, kidney stones.

Knowledge of Hepatitis C

Information was sought about the date of diagnosis and the date when the respondents learnt they were infected, the way in which the news was conveyed to them, and the subsequent management of the illness. All the people interviewed were aware that they were infected with hepatitis C. Hepatitis was described as mild to moderate by most cases, one case had cirrhosis and one described the hepatitis as severe, this person already had liver cancer.

Diagnosis

The respondents were asked how long it was since they learnt that they had hepatitis C. Four of the sample could not remember or had assumed they were positive for many years. One had learnt quite recently, six between two and four years ago and a further six had learnt of their condition more than six years ago, see Table Two.

Table Two Years since Diagnosis of Hepatitis C

Years since diagnosis known	Frequency	Percentage
less than 2	1	5
2 - 4	6	30
5 - 6	3	15
7 - 10	6	30
not known	4	20
Total	20	100

Pathways to diagnosis

There appeared to be different pathways to diagnosis - many of which were far from satisfactory. Some people had experienced problems in getting a test, some had not been informed that they had been tested and had not been informed of the result for some time afterwards. The manner in which results were provided and the information supplied was a concern for many people.

Only two respondents reported that hepatitis had been recognised because they had symptoms. One of these cases had reported feeling ill between 1984 and 1989 and was eventually tested and found to have, what was then termed, non-a non-b hepatitis. Two reported that the Haemophilia Centre had suggested the test and one person had been tested following a car accident. Eight people reported that they had discovered they had hepatitis C following what they understood to have been routine tests. One case tested in 1991 already assumed he had hepatitis but most were neither aware of the disease or that they had been tested when the test had been taken.

Six people reported that they had requested a test. One person initiated the test after his brother died of hepatitis C. A mother initiated the tests for herself and son after reading an article about hepatitis. She became concerned because of the amount of factor VIII her son had had after having his tonsils out. She sought a test through the family GP, 'who didn't seem to know what to do', a local hospital refused to do a test but a test was taken at a tertiary centre. One person initiated the test herself after getting information from the Liver Trust and after reading information from the Haemophilia Society.

Breaking the news

Respondents were asked who had told them of their hepatitis C status. Five had been told by the heamophilia specialist, six by the clinic nurse or other person at the Haemphilia Centre and one by the general practitioner. Eight where not clear who had told them.

The manner of the disclosure was often far from satisfactory. One mother recounted how she and her young son had learnt of the illness. The child asked what all the tests were for during a routine clinic appointment and a doctor (not usually working in the Centre) told him about each test. She mentioned hepatitis C. They asked what hepatitis C was. 'Don't you know about hepatitis C? .. She realised she had dropped a bombshell and sent for a consultant.' The consultant came across - but could not give much information. It was very technical, but she felt 'he had told her all he knew.'

One person reported a telephone call from a clinic nurse, 'not to worry but you've got hepatitis. If you want to know any more make an appointment'. When he arrived for the appointment he was put in a room without much light and told 'it leads to cirrhosis and liver cancer'. Another case was told in the last moments of a routine hospital appointment on a friday evening. On asking what it was he was told 'it could be life threatening but it would probably be all right'. He described his distress on leaving the clinic and traveling home, nearly thirty miles away, in a shocked state. He was not able to learn any more about the condition until he contacted the Haemophilia Society the following monday.

Two cases had learnt by chance when a new doctor had been involved in their case. One person complaining of indigestion learnt about hepatitis, almost by accident, four and a half years after the tests had first been taken. He was told that his symptoms were to be expected 'with your condition - you know you've got hepatitis? Don't you know you've got hep C? You'd better talk with the consultant'. Another learnt from a doctor who he saw following an accident, 'How are you getting on with non A non B?' 'He then realised I had not been told.' One man was informed, 'By the way do you know you have hep C? Can be life threatening but nothing to worry about.' One was told he had 'a bit of an infection. Don't worry.'

Anger was expressed by some respondents about the failure of professionals to inform them properly both that the test had been taken and of the result. One man felt he had been cheated of an opportunity to make changes in his drinking habits that might have delayed the progression of the disease.

Information about the disease.

Respondents were asked what information about hepatitis C and its management had been given following the diagnosis. Fifty percent reported receiving information and 40 % said they had had none. 10 % could not remember. Of those who received information seven mentioned the Haemophilia Society, one the Haemophilia Centre and one had been given information from a hospital doctor. Another case had received no information and felt it had been taken for granted that he needed none.

Three people reported that the information had been *very helpful*. One person mentioned the Haemophilia Society fact sheets. Another mentioned a consultant by name, 'He was a good doctor, you could tell by the way he spoke - an NHS doctor who knows'. One reported, 'All my questions were answered but they didn't seem to know much'. One person, who had not received much information from the consultant initially, reported similar deficiencies at subsequent appointments. He found the information too technical A mother of a young child did not find the information from the Liver Trust and the Haemophilia Centre helpful but had found a TV programme about the Scottish Blood transfusion service most helpful. The information received was classified as being *quite helpful* by six people and *not helpful* by one person.

Attempts by cases to circulate information was rebuffed. The Haemophilia Centre Director objected to the leaflet brought in by a person on hepatitis C, she had folded it away saying, ' you don't want any one to know you have this do you?' One person had been more or less ushered out by her GP when she had offered him a pamphlet about the disease, 'I've no time to read this'. This was a comment from a doctor who had previously admitted not knowing about the infection.

Information for patients within the NHS seemed either sparse or non-existent and it was feared it might 'worry the patients'. One person objected to the British Liver Trust's leaflet that he felt was targeted to drug addicts.This vacuum left the task of information provision largely to the Haemphilia Society, British Liver Trust or the media.

The Manor House group was founded by persons dissatisfied with the information available to them. One person in the study who had been involved with this group since its inception and two other members of that group were interviewed. The Manor House group appeared to be functioning as both a supportive net-work and as a pressure group on issues related to hepatitis C.

Information about the disease is still being amassed and it is perhaps not surprising that ignorance was widespread, but the failure of professionals responsible for cases to engage with the issue and become informed is less understandable.

Counselling

Two cases (10%) reported having counselling whilst 17 (85%) had had none. Those who were counselled were counselled by a nurse at the Haemophilia Centre. Eleven cases (55%) said they would have liked counselling. The aspects of counselling that would have been most useful included: information about hepatitis C, information about how to cope with the illness, information about symptoms, 'what symptoms to worry about' and information about interferon, how to cope with depression, information about prognosis. The mothers of teenage boys were particularly concerned with lack of information and counselling for young adolescents. One felt that her son had been badly affected by the 'uncertainties and stigma of the illness'. He was rebellious and saw no future for himself, 'he had nothing to plan for'. She had sought help for months but had received no support. Another woman had sought counselling for her daughter to help with the anxiety she was suffering because of her mother's condition, at the time of the interview she had been unsuccessful at obtaining any help.

Knowledge of others with the disease and isolation

There was a dichotomy between those who had close family experience of the illness and those who were isolated. One person had lost two brothers with the disease and had other family members infected with hepatitis C and HIV. One person's mother who had hepatitis C and AIDS died, 'mother had been through hell - I don't wish to go through the same'. Two mentioned either a penpal or a friend, three mentioned contacts from the Liver Centre or the Haemophilia Centre and two mentioned contacts with the Manor House Group. Ten belonged to a hepatitis C support group of some kind. Conversely, some felt very alone. One said she felt as if she was 'the only one in the world' suffering from hepatitis C. Mothers of teenage boys felt isolated from others facing similar problems.

Treatment

Respondents were asked what treatment had been made available to cases. Seven (35%) had received interferon and/or ribavirin. Six (30%) cases reported declining treatment offered. One had declined ribavirin and interferon, and three interferon alone. Three had discontinued treatment, one because of the side effects and one because he did not like injections. One had had treatment for one month, one for three months, and one for 12 months

Side Effects of the Treatment

One person reported four symptoms, two cases reported three or more symptoms from treatment. Depression was the most common side effect (5), followed by hair loss (4) other symptoms each mentioned by one person included irritability, headaches, muscle aches and anaemia. One person considered that there was no quality of life whilst on interferon - and that he couldn't persist for twelve months for 'family reasons'. Side effects were not always sympathetically discussed by professionals. When one person complained about side effects he had been told 'not to be stupid'; it was his 'diet not the treatment'. In response he photocopied literature for the clinic on the side effects of interferon. He was resentful because he had no insights into the violent mood swings he experienced. He attributed loss of hearing to the treatment and considered that it had taken nine months to get his taste back.

A 64 year old man who had been on interferon therapy for three months and a 26 year old who had been on therapy for six months discontinued therapy because of side effects - 'hair falling out', 'flu like symptoms' and ' bad depression'. One person who had been treated for six months with interferon had stopped because he had suffered depression, nausea, itching, lack of sex drive. He had not respond to treatment. Another person had discontinued because he had learnt that a friend had suffered from bleeding whilst on interferon and he believed that in any case he might have only a 5% chance of clearing the virus and that it might recur. A young man of sixteen had given up interferon therapy after 4 weeks because of side effects - 'felt rotten', 'irritable', 'flu like symptoms' and headaches. This therapy had been administered whilst he was studying for his GCSE's.

Some had been denied treatment or had not been offered it. One person had been told that he was 'not ill enough' although he would probably have had the treatment had it been offered. One woman was told it was 'at the moment unnecessary'. Another was told that interferon would not help her though she could have had it if she had wanted it. One was told that interferon would not be given unless she had a liver biopsy. One woman was offered it but declined because 'it was unknown, untested and might make things worse'. None of the four women in the sample had received treatment.

Impact on Health

'They just say hepatitis C affects the liver - I think it affects the whole person.'

Three measures of health impact were taken: a subjective categorical ranking, one based on activities of daily living and another on SF36. The subjective categorical description of their health by individuals showed that 10 (50%) described their health as quite good, six (30%) described their health as poor and one as good, three did not answer. The stage of hepatitis C was described by 60% mild to moderate, 15% had cirrhosis, 5% had severe disease. The clinical details on the stage, genotype and duration in the stage were not obtained.

The SF36 was administered by a postal survey to nineteen who agreed to give their address. Thirteen (68%) returned this form and were found to have significantly lower scores than those of the general population. Compared with the control population used by Foster et. al (1997) the physical functioning of the cases in the sample was very much lower, 33% compared with 92%. It was also much lower than the score for cases with hepatitis C, 79% and much lower than the hepatitis B score, 88%. Social functioning had a score of 31% compared with a score of 65% amongst chronic hepatitis C cases. Miners et al (1999), calculated scores for those suffering from haemophilia (most of whom would have hepatitis C) and estimated a score of 54% for physical functioning and 70% for social functioning. These SF36 measurements that can, when administered in a controlled way to well defined populations, provide robust estimates of group means, indicate that the sample was considerably less well than other groups. The instrument seemed to be sensitive to the health state of the group. The sample was tiny, the group was heterogenous in terms of disease severity, age and co-morbidity so no judgment can be made on the basis of this estimate. It does, however, indicate the need for further controlled studies on well defined populations.

The results were consistent with health as measured by symptoms, ability to conduct normal activities and the use of health services, all of which indicated considerable morbidity in the sample population.

Symptoms were described by all cases. 85% reported having had symptoms in the past two weeks. The predominant symptoms were tiredness, 11 (55%) persons mentioned this and general aches and pains were described by five (20%) persons. Three mentioned nightsweats. Depression, 'flu, indigestion, nausea, headaches and diarrhoea were each mentioned by two people. Feeling cold, loss of hair, itching, bladder infection and palpitations and stomach swelling were each mentioned by one person.

Eight persons (40%) reported having spent some time ill confined to bed during the past three months. Three had spent seven days or fewer in bed, one had spent 14 days in bed and one 90 days in bed. Eleven people were confined to their home but able to do ordinary household duties. Twelve people had spent time at home unable to do normal activities. Three had been unable to carry on normal day to day activities inside the home - one for the whole period, one for ten days and one for three days.

Eleven people (55%) reported that their illness had affected their leisure activities. Four reported not being able to go for walks, others reported that it had made participating in holidays, sports, shopping and social occasions problematic. Holidays abroad had been abandoned by two people, family holidays were becoming difficult for others.

Attribution of symptoms is difficult to establish. Some of those interviewed seemed clear about how to attribute their symptoms; others were less certain. The symptoms described are attributed to hepatitis C by 12 (60%) people. One person complaining of 'fullness and pains in the stomach' was less sure, ' it is difficult to tell - the symptoms of HIV and hepatitis are so similar'. Another person who had described his health state at present as poor had cirrhosis. In the past two weeks he had experienced 'tiredness and flu like symptoms. These he blamed on having done 'too much' at the weekend explaining that 'it is easy to blame everything on hep C.' Others still felt unsure what the symptoms of hepatitis C were. One reported that the symptoms were due to haemophilia, and a further two people attributed symptoms to other events or illnesses.

This methodological issue of attribution could not be resolved in this study. Indeed the heterogeneity of the group made it almost impossible to determine likely causes of symptoms. Yet most symptoms reported were mentioned in the list of symptoms described by Hoofnagle (1995) as likely to occur in those with hepatitis C and the low scores on the SF 36 were also reflected in other studies of hepatitis C. Treatment of these symptoms and the continuing monitoring of diseases had led to use of hospital services and community health services.

Use of hospital and community services in last six months

Two persons had been admitted to hospital, one twice, in the past six months. These admissions were concerned with treating bleeds and replacing joints. 40% (8) had visited the hospital outpatients once in the past six months, three had visited more than 10 times. Nine cases (45%) had visited or been visited by a doctor in the past six months. Four cases had had visited or been visited by other health sector professionals up to eight times. The use of these services could be costed in a more extensive study. Had this information been obtained from case notes a more complete data set could have been derived.

Employment Status

Eight people (40%) described themselves as retired - including those who had retired early (only four of the sample were over 60 years of age). Five (25%) were at work, of these two were working full time. Three (15%) were sick, two (10%) were at school or college.

Those working described their jobs as: a warden at a historical site, a salesman, a research assistant, a computer programmer and a telephone technician. Ten out of the twenty cases had been able to work at some point in their lives. One, who had driven buses for 30 years, had not worked since 1986/7; one had been a salesman in a shop and had had a kitchen job, he had retired sick in 1991; one had been a taxi driver for a health authority until 18 months ago, one a clerical assistant until an incident 7 years ago. One had been a runner for an art company, another a fashion buyer. One retired person had owned a painting and decorating firm.

Five (25%) felt that their career choice had been affected by the illness; one did not. Six (30%) commented that their illness had affected their education. Education of one respondent had been affected by having attended a school for the handicapped taught by teachers that he considered were poor. One was disappointed in not being able to join the police. Few reported formal qualifications - one reported having O levels, one had a degree and one had a public service vehicle heavy goods driving license.

In general, career choices were affected differently for the older respondents than for younger ones. The older cases were more greatly affected than the younger ones. This was partly because of improved educational provision and partly because of the improved control of the disease. It was noted, however, that a number of younger people with haemophilia had suffered major bleeds either because of accidents, failure to obtain correct treatment in Accident and Emergency Departments or as side effects of other medical interventions. And although educational provision had improved the stigma and uncertainties relating to hepatitis C was said to be affecting the achievements of the teenagers in the study.

Carers' work had been affected - mainly the employment of mothers who had either never taken on full-time employment or who had given up work. One mother reported that her partner had stopped working for a time to look after her and their child, this had cost the family £5000 in lost wages the time over which such losses had occurred were not clear. One mother who had returned to work reported that she regarded work as a sanctuary from the stresses of caring for others at home.

The great variety of responses to eduction and employment questions made quantitative analysis difficult. More systematic material would have been required and would need to be collected by instruments that allowed respondents time to recall the relevant dates and amounts.

In response to a question about whether the infection had affected their regular income and expenditure Income was thought to have been reduced on a regular basis by £6,000 by one family and by £80,000 in total by another. The reduction by £6,000 was caused by failure to work regularly. He was self-employed. The source of income from employment, state or private pension schemes, social security was difficult to collect on a systematic basis in the telephone interview.

The effects of the illness on regular expenditure was mentioned by 15 (75%) people. Ten people mentioned costs of heating, seven reported travel costs, three reported expenditure on special food and alternative medicines. Extra heating was needed because some people could not move around because of arthritis and so felt the cold. Some younger cases felt constantly chilled, they attributed this to hepatitis C. Five people said that they regularly spent £15 per week on expenses relating to hepatitis C. Three reported spending up to £4 a week, one person £30 and one person quoted £50 per week. (This was associated with a case who had to take a taxi - even to buy a loaf of bread, and who has to pay for a cleaner because she is too fatigued to do her own housework). Travel to appointments was frequently mentioned as an expense, one person had reduced visits to hospital because of this cost. Some extra costs were attributed by some people to their attempts to buy a healthy diet. 'Thistle milk' taken as a food supplement was mentioned by two people and others were on low fat diets. Two people reported large exceptional expenses changes to premises. One had spent £5,000 and one £20,000. In a larger study such costs could be explored in detail distinguising between those expenses incurred regularly and those that occurred irregularly or were incurred to invest in long term adjustments to living standards. There are problems of collecting this information systematically in a telephone interview as respondents often need time to recollect dates and items. The data collected was not sufficiently systematic for quantative analysis. Reported expenditure was correlated with severity of illness, employment status or age: none were significantly related. Chronic illness poses problems for those attempting to assess the additional costs of the illness as expenditure on items to compensate for disabilities are limited by available income. More detailed discussion would be required in order to assess what expenditure would have been required to compensate for some health deficits.

Insurance

Insurance questions differentiated between life insurance and insurance based pensions. Eight people reported having trouble obtaining insurance, seven had not. One reported that a pension was too expensive because of the infection and another expressed worries about lack of a pension. One woman had cashed in insurance, now all insurance was taken out in her partner's name. Another woman was insured through her husband. One reported that his wife was 'insured up to her eye balls' whilst he could not obtain insurance. One person had solved his problems by changing to Saga, 'they know about hepatitis C but had been very helpful.' One had a funeral policy for £2000. Two reported that they were not able to obtain insurance, one person couldn't obtain insurance because of infection with HIV and Hepatitis C.

Housing

Eight (40%) persons owned their own home, five (25%) had a mortgage and five rented their home. Six (30%) reported that their illness had affected their housing. Nine (45%) said it had not. Two were trying to sell, one person was unable to get a mortgage, one was moving to a smaller house. Housing was often held in the wife's name. One family had considered raising money by selling or getting a second mortgage in order to buy their son a flat. One family had moved house twice to be located nearer to a 'good centre' (Haemophilia Centre).

Leisure

Many reported that the infection had affected their leisure activities. It was difficult to attribute problems to hepatitis C. The case who was infected with both HIV and hepatitis C had given up holidays, and went out less, another reported that holidays had been given up on the advice of a doctor, this person also mentioned having to give up family holidays. Others reported that they were able to enjoy little social life. One person badly crippled from bleeding into joints could no longer walk, climb stairs or stand. He could no longer go to art classes because they had been moved from the ground floor of the college. Holidays abroad had been a disaster for two people and one reported family holidays had become impossible.

Expressed concerns and worries

The final part of the interview related to the worries and concerns that the respondents felt in regard to hepatitis C. The concerns of the families with dependents obviously differed from the concerns of younger respondents. But certain dominant themes emerged: Information, worry and concern about the family, stigma, dealing with the uncertainty the infection brought and anger, both about becoming infected and the subsequent management of the disease. There was also concern about reduction of services in the home.

What would have helped you most?

'If I could ask a hepatologist one question, it would be 'Will this get worse?' '.

Information about the illness in all its dimensions was of crucial importance to most people. What were the consequences of the disease, what symptoms could be attributed to it, how could it be transmitted. Sexual transmission was a particular concern for mothers of teenage boys and young men but all age groups were concerned to know more. Respondents stated that it was the 'uncertain nature of hep C' particularly in terms of likely morbidity and mortality, that was one of the most difficult aspects of living with the illness. 'What will happen to me?' 'Will it shorten my life, by how much?' 'How long have I got? I've had it for 18 years and they say most people don't live past 20.'

The uncertainty about the progression of the illness made planning for the future difficult and for many, anything other than living 'week by week' was problematic. This could lead to feelings of depression even for those respondents who were physically feeling very well as they waited for, 'hepatitis C to explode into life'. For those with young children this obviously generated much anxiety. 'The point is I struggle each day to provide an income for my family and I don't know how long I've got.'

Many tried to deal with the uncertain nature of the illness by obtaining as much information as they could about hepatitis C and possible therapies, orthodox and alternative. This gave some a positive feeling of being in control. But also could leave respondents feeling that they knew more about the condition than many of the health professionals they came in contact with. This could be frustrating or, more disturbingly, leave them feeling vulnerable and insecure.

A person who had been very active in attempting to obtain and distribute information was very depressed about progress. 'Sorry I started the research - disgusted that medical profession will not come out to support you. He no longer trusts the medical profession who he feels are engaged in 'a cover up.' Although recognising the unknown nature of hepatitis C many felt the way the medical profession conveyed this uncertainty was not always well judged. When information was given it was too technical, 'if you ask for results they give figures but don't explain what they mean'. Or too little information was provided and patients seeking more were made to feel they shouldn't be there. This led some to feel that knowledge was being withheld and, given that some respondents had not been informed promptly of their hepatitis C status, they became ' suspicious about what else is out there.' Often the information was either too complex or given in a perfunctory fashion. One respondent offered advice about information needs, 'Speak of hepatitis and speak frankly. Speak to haemophiliacs about all the information they have or would like to have'.

Impingement on family life

'Mummy wants to kiss you but doesn't want to give you hep C'

Concerns for the welfare of the family, whether for their own children or for their parents or others who looked after them were paramount for many people. A woman whose children were 'very worried about her' was trying to get counselling for her ten year old daughter but had been unsuccessful. This woman had become anxious about transmitting the disease to her children and worried about kissing them, 'Mummy wants to kiss you but doesn't want to give you hep C.' One respondent worried about his parents who had found his diagnosis very difficult to cope with as they felt responsible as they had taken the decision for him to have blood products as a child. A mother and son who both had hepatitis C faced particular anxieties as they worried about each other and the progression of the illness.

The interviews provided many illustrations of how the condition disrupted family life. A father stated that he had felt that he had 'missed out on his daughters growing up 'because he had been 'ill a lot'. Most respondents regretted not being able to participate fully in family occasions and holidays. A grandmother regretted not being able to look after her grand children as much as she would like. Many expressed concern for family members who had taken on extra duties looking after them or their children. A mother expressed concern that because of her condition her daughters were 'taking on responsibilities they were too young for.'

The stigma surrounding a diagnosis of hepatitis C was an issue for many of the respondents in the study.

Stigma

'You don't want any one to know you have this do you?'

This was the remark of Haemophilia Centre Director as she crumpled up a leaflet giving information about the disease that the person had brought into the clinic. One person seeking more information had sought a contact with some one else with the disease via the Liver Trust but although a person in the north of Scotland was found, that person didn't wish to be identified because of the 'stigma' of the disease. This left the person feeling even more isolated and she now fears talking about it to any one. She felt the stigma had been transferred to her. A woman and her son were not allowed to use either cups or towels when they visited her mother. 'You find out who your friends are', said one person whose sister could not come to terms with his infection. 'How's the thing?' was the nearest she got to discussing hepatitis C. 'You see' he said, 'people shy away'. When this man was drinking with friends in his pub he recounted that one person in the group would hold on to his glass and have it refilled all night in case he had the glass that had been used by the respondent. Another reported a neighbour growing a hedge to stop the virus contaminating their property; others reported graffiti. One person reported that though he had not lost a job because of illness he hasn't felt able to talk openly about it because of the stigma, 'They'll think I've got AIDS'.

Other items that were commonly mentioned were service provision and compensation.

Provision of Services and Treatments

Service provision was a dominant theme. This related to the provision of recombinant interventions - fears that it would not to be distributed as it was too expensive. Another wanted reassurance that his supplies would not be curtailed because he was in a 'Fund holding practice'. The continuance of this therapy was of particular concern to one whose religion forbade the use of blood products. One respondent commented that cuts in budgets had caused the withdrawal of support, 'nurse visiting stopped a few years ago,' and wanted them restored. A mother urgently sought support for her children and help with the housework.

Many believed that some professional support that would offer counselling for themselves and their families would be helpful. They felt, that in comparison to the response to HIV infection, there had been little support offered to families with Hepatitis C, 'family weekends for HIV not for hepatitis C'. Many felt that a more generous benefit system would have helped them to cover the day to day expenses they incurred because of the condition. The present social security support was considered to be unfair.

'Compensation, compensation, compensation'... Incorporated in the desire for compensation was a desire for justice: the desire that 'Injustice to be brought to the front' of the policy agenda. 'Haemophilia would have allowed a normal life. Hepatitis C - shortens life and quality of life'. Financial awards were desired both as compensation for the transmission of the disease and to cover the extra costs of coping with the disease and providing security for dependents.

Discussion and Recommendations

Attributing outcomes to hepatitis C is the most difficult research problem as there are no control groups of people with haemophilia who do not have hepatitis C. A research design that allows cases at similar disease stages, for both haemophilia and hepatitis to be compared and these groups to be compared to those without haemophilia with hepatitis C acquired from blood products.

Whilst the pilot study demonstrated the feasibility of obtaining information about the impact of hepatitis C on those with haemophilia the telephone interview was not the most effective method of obtaining all the components accurately. The telephone interview was very effective in obtaining information about the factors that led to the diagnosis or the disclosure of diagnosis. It was also effective in obtaining Information about whether or not treatment for hepatitis C had been given and whether it had worked or not. Details of the impact of hepatitis C on their lives and the lives of their families was discussed fully. It was difficult to obtain precise clinical details of hepatitis, comorbidity and investigations and treatments from all respondents systematically using a telephone interview. Precise financial details of the economic impact of hepatitis C was not easily obtained by telephone but it was possible to obtain rich descriptive information relating to education and employment history. It suggested that the issues can be explored best using a combination of methods that include the details from patients' casenotes, telephone interviews and postal or administered questionnaires structured according to the severity of illness. An epidemiologically robust study should be undertaken that explores the impact of hepatitis C on the lives of those with haemophilia and von Willebrand's disease.

In considering the findings of this pilot study it is important to remember that it is a small non-random sample. This small study did however reveal some important issues that even if not confirmed generally are a matter of concern. Hepatitis C has not been handled well in the care of haemophilia in many parts of the country. There appeared to have been no national strategy to inform staff about the appropriate way of dealing with the issue with respect to testing, informing people about the illness or conveying information about treatment options. Health care professionals in many cases have not appeared to have been fully aware of the disease or its implications. Access to treatment was patchy and no women, the group most likely to respond to therapy, had received treatment. Provision of information was largely left to voluntary agencies. Risks should be explained, informed consent for tests should be obtained and sensitive reporting of test results should be

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Appendix A

Pilot questionnaire

First confirm that the person answering the phone is the person listed.

Consent statement

We are undertaking as study on behalf of the Haemophilia Society to explore the impact of hepatitis C upon those with haemophilia.
We understand that you have agreed to participate?
Is that so?
Thank you?

The information that you give us will be treated in strictest confidence. The questionnaires will be coded and we can assure you that your name will not be recorded on the record of the interview or on any written or computerised material reporting this study.

The purpose is to develop a questionnaire that can be used to assess the impact of hepatitis C and compare it with other cases with the illness and with the general population.

First of all we would like to ask you some questions about the illness. Please feel able to decline to answer any question

This study is a preliminary survey we would like you to ask for your advice about the pilot questionnaire.

Haemophilia Society Survey

1. **Identification Number** -- -- --

2. **Is the respondent the case or carer?** **case/carer** --

To begin with I would like to ask you some questions about yourself (or your child)

3. **Date of Birth** --/--/-- -- --

4. **Marital Status**
single/married/with partner/separated or divorced/widowed --

If the respondent is not the case go to question 6

5. **Occupational Status**
at work/not at work/ sick/unemployed/retired/school or college --
Please tick

If at work, what is your job... -- --
please get enough information to estimate social class
Are you employed full-time or part time? Pt/ft --

If you are retired or have had to leave work for health grounds
what was your previous job or jobs. --

If your are unemployed, for how long have you been unemployed?
.... weeks -- --

If you are unable to work because of sickness , have you ever
been able to work?..yes/no --

If yes, for how long have you been unable to find work
because of illness?... -- --

If the respondent is not the case

6. **What is your employment status?**
At work/not at work/sick/unemployed/retired/school or college --
other
Please tick

If at work, what is your job... -- --
please get enough information to estimate social class

Are you employed full-time or part time? Pt/ft --

If your are unemployed, for how long have you been unemployed?
.... weeks -- --

If you are unable to work because of sickness , have you ever been able to work?..yes/no --

If yes, for how long have you been unable to find work because of illness?.. --

7. **Household structure**

Do you live alone?...Yes/no --

If no, how many are in the household .... -- --

How are they related to you? .... -- --

.... -- --

.... -- --

Please specify family relationship or friends

Now I would like to ask you some questions about your illness

8. **What type of Haemophilia do you have?** --

Haemophilia A (factor IIX) ....

Haemophilia B (factor IX) ....

Willebrands disease ....

How would you describe the management of the haemophilia at present e.g. controlled/ unstable -- --

....

9. **When did you first learn that you had Hepatitis C?** .... -- --

Who told you? .... --

....

Was it recognised because you had symptoms of the illness? Yes/no --

Routine checks? Yes/no........

Other? explain...

....

....

....

10. **Did you receive any information about the illness?** Yes/no --

If yes, from whom?.................................... --

How helpful was this information?

not helpful/quite helpful/not helpful --

Do you know any one else with Hep C? ....*Yes/no*................... -- --

If yes who is this?...

Do you belong to any Hepatitis C support groups? ..Yes/no.......... -- --
If yes, what? .... -- --
....
....

11. **Did you have any counselling about the hepatitis c?** Yes/no --

If yes, who provided it? ....
.... --
e.g. hepatologist, centre staff, GP, other

If not, would you have liked to have had the opportunity to have
discussed the illness with a counsellor or other professional? Yes/no --
If yes what information would have been most useful?...................
....
....
Do you have any other illness? Yes/no --
Allow them not to answer this question

12. **Symptoms**
How would you describe your health at present? ....
good/quite good/poor? --

Is the Hepatitis C at this stage
mild/moderate - with cirrhosis - or severe?... --

Have you experienced any symptoms in the past two weeks?
Yes/no --
If yes, what are these? Please specify.. -- --
.... -- --
.... -- --
.... -- --
.... -- --
.Do you attribute these symptoms to hepatitis c, haemophilia or other
illnesses..
-- --

Don't prompt for symptoms but after each one ask if it was mild moderate or severe. Record as e.g. tired, mild.

13. **What impact have these had on your use of health care and on your day to day activities?**

In the past three months
Have you been at home confined to bed? Yes/no --
If yes, for how many days? .... -- --

Have you been at home unable to undertake normal activities? yes/no --
If yes, for how many days? .... --

Have you been confined to your home but able to do normal activities?

Yes/no --
If yes, for how many days? .... --

Have you been unable to go to work or school? Yes/no........ --
If yes, for how many days? .... --

Have you been unable to take part in normal leisure activities? ..... --
If yes, what were you not able to do?... -- --

14. **Treatment**
Have you had any interferon therapy and/or ribovirin therapy?
Yes/no --
If no, did you decline treatment that was offered? yes/no --
....
If yes, what treatment did you have?........................ --
For how long?...........In months (or weeks).............. -- --
Did you complete the course of treatment? .....Yes/no --

How did you feel whilst you were having treatment ? -- --
List symptoms .... -- --
.... -- --
.... -- --
.... -- --
Do not prompt but if they mention a symptom ask whether it was mild moderate or severe and how long it lasted.

Did you have to discontinue treatment? .... -- --
If yes, why? .... -- --
.... -- --
.... -- --
What was the result of the treatment? .... --
Expect cleared virus, remission, no response?.. —

15. **Use of Services**
In the past six months?
Have you been admitted to hospital.........Yes/no .... --
If yes, how many times? .... --
For how long on each occasion? days .... -- --
.... -- --
Have you visited a hospital out-patients' department?...Yes/no --
If yes, how many times? .... --

Have you visited or been visited by your GP?...........*(Please specify)* --
....
If yes, how many times?.. --

Have you visited or been visited by any other person from health

or social services? Yes/no......... --
If yes, how many times?.................................... --

Now I would like to ask you about the wider impact of the illness on yourself and family

16. Education

Has the illness affected you education? Yes/no --
If yes, how? .... -- --
....
....
....
Don't probe but expect time off school, tired, unable to cope
Have any achievements/career choices affected by our illness?
Distinguish aemophilia from hepatitis or others
Yes/no .... --
If yes what were they?.....................................
....
.... --
What qualifications do you have?..
....
....
.... --

17. Work

If adult responding for child ask about the impact on their work
Have you had to give up a job because of illness? --
If yes, please explain?.. -- --
....
....
....
....
What is your present job?... -- --
....
How long have you been in your present job?...........................*months*...... -- --
Has the illness affected the job you do?...........Yes/no.................... -- --
If yes, how?....................................... -- --

Have you ever feared losing a job because of your illness? Yes/no --
If yes, explain.. -- --
.... -- --
....
Has any one else had to give up or change a job because of your illness? Yes/no................................ --
If yes, expand... -- --
....
....

18. **Housing**

Do you own, rent or mortgage your house? ....Please tick..... --

Did your illness affect your choice of housing?.....Yes/no...... --

If yes, how.. -- --

....

....

19. **Financial Circumstances**

Has your illness had any impact on your regular expenses?...*Yes/no* --

If yes, how?.. -- --

....

....

....

Can you estimate how much these expenses cost each week?.......... --

Has your illness had any impact on your regular income?...*Yes/no* -- --

If yes, can you estimate the impact? .... -- --

....

Have you had any exceptional (large) expenses or losses of income associated with the illness? .... -- --

....

If yes, what was this for ?.. -- --

How much did it cost?... -- --

Have you ever had problems obtaining insurance or entering a pension scheme because of your illness? .... -- --

....

....

....

If yes, what affect has this had upon your plans for the future?.......... -- --

....

....

20. **Leisure activities**

Has the illness affected your leisure activities? Yes/no --

If yes, how? .... -- --

....

....

Don't prompt but expect holidays sports, family life etc.

If carer is being interviewed ask about impact on leisure time of carer or family

Has the illness affected your leisure activities? Yes/no --

If yes, how? ... *Don't prompt but expect holidays sports, family life etc.*

.... -- --

....

....

....

21. Impact of the illness

Is there anything you would like to add about the impact of the illness upon your life or that of your family? ....

....

....

....

....

....

....

....

What are your most pressing anxieties and worries about the condition?..

....

....

....

....

....

....

What would help you most? ....

....

....

Thank you very much for your help.

Would you be prepared to fill in a questionnaire that would compare your health with that of other cases with Hepatitis C and with the general population? ....

If so could we send them to you or would you prefer to answer such questions by telephone?

If you would prefer a telephone call would be a good time to phone again?

....

If we are to send them to you could we have an address to which the forms could be sent. ....

....

....

Appendix B

SF36

Survey on Health Status undertaken for The Haemophilia Society

by Dr J.A.ROBERTS

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Thank you for agreeing to fill in this questionnaire.

Please return this to the above address in the envelope provided. Please answer each question as accurately as you can. You should place a cross inside the relevant box as indicated below.

☒

There are specific instructions for each question such as:

Please put a cross in one box or *Please put a cross in one box on each line*

Q1 In general would you say your health is:

Please put a cross in one box

☐ Excellent
☐ Very good
☐ Good
☐ Fair
☐ Poor

Q2 **Compared to one year ago,** how would you rate your health in general now?

Please put a cross in one box

☐ Much better than one year ago
☐ Somewhat better now than one year ago
☐ About the same
☐ Somewhat worse now than one year ago
☐ Much worse now than one year ago

Q3 The following questions are about activities you might do during a typical day. Does your health limit you in these activities? If so, how much?

Please put a cross in one box on each line

	Yes limited a lot	**Yes limited a little**	**No not limited at all**
a) **Vigorous activities** such as running, lifting heavy objects, participating in strenuous sports	☐	☐	☐
b) **Moderate activities** such as moving a table, pushing a vacuum cleaner, bowling or playing golf	☐	☐	☐
c) Lifting or carrying shopping	☐	☐	☐
d) Climbing **several** flights of stairs	☐	☐	☐
e) Climbing **one** flight of stairs	☐	☐	☐
f) Bending, kneeling or stooping	☐	☐	☐
g) Walking **more than a mile**	☐	☐	☐
h) Walking **half a mile**	☐	☐	☐
i) Walking **100 yards**	☐	☐	☐
j) Bathing and dressing yourself	☐	☐	☐

Q4 During the **past 4 weeks,** have you had any of the following problems with your work or other regular daily activities **as a result of your physical health?**

Answer Yes or No to each question

	Yes	**No**
a) Cut down on the **amount of time** you spent on work or other activities	☐	☐
b) Accomplished less than you would like	☐	☐
c) Were limited in the **kind** of work or other activities	☐	☐
d) Had **difficulty** performing the work or other activities (e.g. it took extra effort)	☐	☐

Q5 During the **past 4 weeks,** have you had any of the following problems with your work or other regular daily activities **as a result of any emotional problems** (such as feeling depressed or anxious)?

Please answer Yes or No to each question

	Yes	**No**
a). Cut down on the **amount of time** you spent on work or other activities	☐	☐
b). **Accomplished less** than you would like	☐	☐
c). Didn't do work or other activities as **carefully** as usual	☐	☐

Q6 During the past 4 weeks, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbours or groups?

Please put a cross in one box

☐ Not at all
☐ Slightly
☐ Moderately
☐ Quite a bit
☐ Extremely

Q7 How much bodily pain have you had during the **past four weeks?**

Please put a cross in one box

☐ None
☐ Very mild
☐ Mild
☐ Moderate
☐ Severe
☐ Very severe

Q8 During the past 4 weeks, how much did pain interfere with your normal work (including work both outside the home and housework)?

Please put a cross in one box

☐ Not at all
☐ A little bit
☐ Moderately
☐ Quite a bit
☐ Extremely

YOUR FEELINGS

Q9 **These questions are about how you feel and how things have been with you during the past month.** (for each question, please indicate the one answer that comes closest to the way you have been feeling).

Please put a cross in one box on each line

During the past month:	**All of the time**	**Most of the time**	**A good bit of time**	**Some of the time**	**A little of the time**	**None of the time**
a) Did you feel full of life?	☐	☐	☐	☐	☐	☐
b) Have you been a very nervous person?	☐	☐	☐	☐	☐	☐
c) Have you felt so down in the dumps that nothing would cheer you up?	☐	☐	☐	☐	☐	☐
d) Have you felt calm and peaceful?	☐	☐	☐	☐	☐	☐
e) Did you have a lot of energy?	☐	☐	☐	☐	☐	☐
f) Have you felt downhearted and low?	☐	☐	☐	☐	☐	☐
g) Did you feel worn out?	☐	☐	☐	☐	☐	☐
h) Have you felt a happy person?	☐	☐	☐	☐	☐	☐
i) Did you feel tired?	☐	☐	☐	☐	☐	☐
j) Has your **health limited your social activities?** (like visiting friends or close relatives)	☐	☐	☐	☐	☐	☐

Q10 Please choose the answer that best describes how **true** or **false** each of the following statements is for you.

Please cross one box on each line

	Definitely true	**Mostly true**	**Not sure**	**Mostly false**	**Definitely false**
a) I seem to get ill more easily than other people	☐	☐	☐	☐	☐
b) I am as healthy as anybody I know	☐	☐	☐	☐	☐
c) I expect my health to get worse	☐	☐	☐	☐	☐
d) My health is excellent	☐	☐	☐	☐	☐

Appendix C

Haemophilia Society Survey

(Patient questionnaire - Postal questionnaire should be supplemented by face to face or telephone interviews on a sub-sample to explore attitudes and experiences.)

Part One - Personal Characteristics

To begin with we would like to ask some questions about yourself (or your child)

1. **Identification Number** ....

2. **Case or carer..................If carer, relationship to case...............................**

3. **Date of Birth** --/--/----

4. **Marital status** .... single /married (include living with partner)/ /separated or divorced / widowed (Please tick one)

5. Household structure
Do you live alone or with others? yes /no
If with others, how many are there in the household?.............................
How are they related to you, e.g. family, friends

Part Two About you illness

6 Type of Haemophilia
a. Type of Haemophilia A; Haemophilia B; and von Willebrand
b. Level of a.n activity ....
c. How would you describe the management of haemophilia at present?
e.g. well controlled, some problems, major problems

7. When did you first learn that you had hepatitis C? date ....
a. Who told you?........................
b. Were tests taken because of symptoms?.....
c. Were tests part of routine checks?...............
d. Other..................................

8. Did you receive any information about hepatitis C? Yes/ No
a. If yes, how helpful was this information? not helpful /quite helpful/ very helpful
b. Who provided this information?
c. Did you have any counselling about the illness? yes / no
d. If yes, who provided it?

e. If no, would you have liked to have had the opportunity to discuss the illness with a counsellor or other professional?...

9. Other illnesses?

a. Caused by blood products? Yes /No
Specify if you wish to......................
b. Do you have any other illnesses? Yes /No
Specify if you wish to......................

10. Were you offered any treatment for hepatitis C? Yes/No

a. If yes, did you accept treatment
b. If so, what treatment did you have
Interferon α
Combined interferon α and ribavirin therapy?
c. For how long?..............................
d. Did you complete the course? Yes/ No ....
e. If no why not?..
f. What was the result of the treatment?
g. Did you decline treatment offered?
h. If you declined treatment what were your reasons for this?...........

11. How did you feel whilst you were having the treatment?

List symptoms, duration and severity

....

....

12. Is hepatitis C followed up?

a. If so where?
b. How frequently?

13. How does the illness affect you? Please tick

No noticeable effects

fatigue
tiredness
malaise
appetite loss
nausea
vomiting
abdominal pain
dizziness
joint pains
muscle pains
muscle cramps

dyspnoea
tingling sensations in hands
headaches
rashes
itching
depression
dry eyes
tongue changes
weight loss
weight gain
sexual dysfunction

very tired
jaundice
dark urine
swellings of legs and/or abdomen
bruising
bleeding from stomach
bleeding from bowel
changes in mental function
sleeping during the day
kidney disease
intellectual functions

Other please add any other symptoms..

Add SF 36 to Measure Health Status

Part Three Use of services (*This section could be used to validate data from clinical records and to trace hospitals that would need to be contacted for information)*

14. Have you used hospital services in past six months?

a. Have you had any hospital admissions? ...
b. If yes, for what for (please specify)Haemophilia /hepatitis C / other..
c. Which hospitals were they?...
d. How long were you in hospital.? ....
e. How many hospital visits have you made to out-patients' departments?
f. If yes, what for (please specify)Haemophilia /hepatitis C/ other
g. Have you visited the Accident and Emergency clinics?
h. If yes, how many visits have you made to Accident and Emergency clinics?
i. If yes, for what for (please specify)Haemophilia /hepatitis C/ other
j. How many visits have you made to a Haemophilia Centre?
k.How far was it from your home to the hospital? miles
l. How long does it take to reach the hospital and return home by
 public transport?
 private car?
 health authority transport?
m. How much did it cost you £ and your companion if you were accompanied £...............
n. How far is it to the Haemophilia Centre? miles
o. How long does it take to reach the centre and return home by
 public transport
 private car?
 health authority transport
p.. How much does it cost you?......... and your companion if you were accompanied £...........
q. Have you visited or been visited by your GP in past six months?.................................
r. How many times?
s. Was the visit related to haemophilia, hepatitis C, another disease?.
t. Have you been visited by or visited any other professional from health care or social services?..
u. Was the visit associated with, haemophilia, hepatitis C, another matter?.

15. How has your illness affected you in the past six months

a. Have you been at home confined to bed? On how many occasions?.........
b. How long were you ill ?..
c. If yes, what was the reason for this (please specify) Haemophilia /hepatitis C/ other.
d Have you been ill at home unable to undertake normal activities?........If yes, for how long?.....
e. Have you been ill at home able to do most things but unable to go out to shops etc.? If yes, for how long.
f. If yes, for what (please specify)Haemophilia /hepatitis C/ other
g. Have you had any time off work ? ....
h. How many times.......and for how long?..

i. If yes, for what for (please specify) Haemophilia /hepatitis C/ other
j. Have you been unable to undertake normal leisure activities? ....
k. If yes, what were you not able to do?...
l. If yes, for what reason (please specify)Haemophilia /hepatitis C/ other

Part Four Employment and Income

17. Occupational status

a. Are you at present at work/ not in work / sick/ unemployed/ retired/ school or college (tick one)
b. If at work, what is your job?...
c. Are you employed full time or part time? Pt / Ft
d. If unemployed, for how long have you been unemployed?................Months
e. If you retired early what was you last job?
f. When did you last work?
g. Has haemophilia/hepatitis C/other affect the jobs you were able to do? ....
h. Has haemophilia/hepatitis C/other affected how you did your jobs?.........
i. Have you had to change jobs because of illness?...
j. Have you ever lost a job or feared losing a job because of Hepatitis C?......................
k. Has any one else had to give up or change jobs to take care of you?.....................
l. Has any one else had to take time off work to care for you or those who you are responsible for?
m. If yes, who? How much time did they take off?
n. If yes, was this because of haemophilia/hepatitis C/other.

18. Consequences of the illness education

(a) Has haemophilia affected your education or career choice?
(b) How did it affect it?
(c) Has Hepatitis C affected your education or career choice ....
(d) How did it affect it? ....
(e) Any achievements affected by haemophilia/hepatitisC/other?
(f) What qualifications do you have?...

19 What is the major source of income for you household?

a. Income from employment of case?
b. Income from employment of partner or parent?
c. Income from Pension? (please state), state pension, occupational pension, private pension?
d. Other state benefits (please state)Income Support, Mobility Allowance
e. How much is your usual weekly income? (*It would be helpful to us if you are able to provide this information. However, if you feel unwilling to do so please complete the remainder of the questionnaire.)*
f. up to £60
£61-100
£101-150
£151-200
£201-250
£251-300
£301-350
£351-400
£401-500
£501-600
£601-750
£751-

20. What proportion of your weekly expenditure is spent on
Food ....
Housing....................................
Transport.................................

21. How much additional expenditure per week do you consider you spend because of disabilities attributed to
(a) haemophilia / von Willebrand's disease.................£.............................
(b) hepatitis C...£..........................
(c) other blood borne illnesses....................................£.............................
(d) other illnesses £
(e) Have you had any other regular expenses
(f) If yes, do you associate these with haemophilia/hepatitis C/other?
(g) If yes, what were they?
(h) Have you had any large exceptional expenditure related to you illness?...
(i.). If yes that you associate with haemophilia/hepatitis C/other?
(j) If yes, what were they? ....
(j) How much did this cost?..

22 Consequences of the illness on housing
(a) Do you own, rent or have a mortgage on a house?
(b) Did you encounter any problems with accommodation because of illness?..........................
(c) If yes, was it associate with haemophilia/hepatitis C/other?

23. Consequences for life insurance and pension?
(a) Have you had any problems in obtaining life insurance?
(b) If yes, was that associate with haemophilia/hepatitis C/other?
(c) Have you had any problems in obtaining a pension?
(d) If yes, did you associate this with haemophilia/hepatitis C/other?

24. Leisure activities
(a) Has you illness affected your leisure activities? ....
If yes, how?...
(e.g. Family holidays, sport etc.)
(b) If yes, do you associate this with haemophilia/hepatitis C/other?

25. Consequences to family life
(a) Has the illness affected family life? Yes/ no ....
(b) If yes, in what ways?..
(c) If yes, do you associate this with haemophilia/hepatitis C/other?
(d) Is there anything you would like to say about the impact of the illness on your life and the life of your family ....
(e) If yes, do you associate this with haemophilia/hepatitis C/other?...

(f) Do you have any pressing anxieties or worries about hepatitis C?

....

....

(g) What would help most to deal with any anxiety you feel relating to hepatitis C?

....

(h) Anything else you would like to say about your illness?

Thank you so much for your help

Appendix F

It would appear that unless we understand the attitudes towards and knowledge of hepatitis C amongst those involved with treating cases we are unlikely to be able to change their practices. It is thus suggested that a parallel study of clinical staff be undertaken.
Some possible questions are appended.

Questionnaire that might be used to assess the knowledge and views of clinicians who treat those with haemophilia and hepatitis.

What is hepatitis C?
How is it transmitted?
What is the natural history of the illness?
What symptoms would you expect patients to experience during the course of the illness?
When do you test for hepatitis C?
What tests would you use?
What do you tell patients and their families about the disease?
Do you put them in touch with agencies able to offer support and information to cases?
Do you offer counselling?
Do you recommend treatment?
If yes, what treatment would you recommend?
Is this available in your area?
What treatment do you offer?
What success would you expect from such treatment?
What symptoms might the patients experience whilst on treatment?
Are any clinical governance or control assurance processes in place to assure the good management of hepatitis C?
If yes what are they?
If no what do you think they should contain and who do you think should be responsible?
Are there any lessons to be learnt from the management of hepatitis C that might inform any subsequent iatrogentic illnesses?