Supplement. A Qualitative Assessment of Brain Injury, Cerebral Palsy and Stroke Patient, Caregiver and Clinician Values of Outcomes

Introduction

In conjunction with the evidence report on hyperbaric oxygen therapy (HBOT), we conducted a qualitative research project involving focus groups to guide our evaluation of the outcomes assessed in the evidence. Qualitative research can be a valuable adjunct to evidence reports by broadening the scope of evidence-based medicine and "can help bridge the gap between scientific evidence and clinical practice."[1] Qualitative methods also offer "insight into the factors that shape lay and clinical behaviour."[2]

Our goal was to examine the relative importance of different outcomes to patients, caregivers, and clinicians, and how experience and attitudes influence the value they place on different outcomes. Specifically, we sought to answer the following questions:

1. What outcomes are most important to patients, caregivers, and clinicians?
2. Are there differences among groups of patients and caregivers and clinicians in the value placed on each outcome? If so, are these differences related to the degree and duration of improvement?
3. How much effort, discomfort, or risk would participants be willing to accept to achieve a short-term improvement?

Methods

A Multiple Perspectives model, represented in the Figure, provided a framework for the collection and analysis of the data.[3, 4] In the figure, two overlapping ovals depict groups of people who hold different perspectives. The oval on the left represents "hyperbaric oxygen therapy experience," and the one on the right represents "brain injury, cerebral palsy, or stroke experience." The darkened area where the ovals overlap represents experience with HBOT and with brain injury, cerebral palsy, or stroke. The groups of people viewing their experiences and expressing their perceptions of them include six categories, labelled A through F. The rays pointing from each category to an oval or to the overlapping section of the circles indicate that these groups are viewing the objects within the ovals from their own perspectives. The individuals interviewed for our project were selected because they fit into one of these relevant groups. The results are descriptions of the views of each group about hyperbaric oxygen therapy; treatments for and recovery from brain injury, cerebral palsy, or stroke; or the intersection of the two.

Focus groups were the primary method for collecting data for this study.[5] Focus groups are group interviews where "several members of an aggregate are gathered together for a facilitated discussion about phenomena of import to them as a group."[6] Recognizing that there might be scheduling difficulties, because travel would be necessary for some interviewees, the researchers conducted individual interviews to augment the focus group data. These topical oral history

interviews, tape-recorded narrative accounts of past events experienced by those being interviewed,[7] provided in-depth descriptions of HBOT treatments over time. The data from the interviews therefore enriched the focus groups.

Participant Selection

Focus group participants were selected based on either their experience with HBOT and/or their experience with one of the three disease states; brain injury, stroke or cerebral palsy. Participants who had experience with HBOT were divided into two categories: those with experience with *approved* uses of HBOT, such as clinicians working at hospitals that have hyperbaric chambers, and those who had experience with HBOT specifically for treatment of stroke, cerebral palsy, or brain injury. Participants who did not have experience with HBOT were chosen based on their experience with stroke, cerebral palsy, or brain injury. This group was divided into patients (or caregivers) and clinicians. The categories of participants and the number in each category are summarized in the Table. We recruited clinicians from a local freestanding chamber facility (Group B), nominations from technical experts (Group D), and Oregon Health & Science University (OHSU) faculty (Group F). We recruited patients and caregivers through OHSU clinics (Groups A, C) and from a local freestanding chamber facility (Group E).

Table. Groups of patients or caregivers and clinicians.

	Patients or Caregivers		Clinicians	
Medical condition	HBOT experience	No HBOT experience	HBOT experience	No HBOT experience
Brain injury, stroke, or cerebral palsy	N=7 (Group E)	N=4 (Group C)	N=3 (Group B)	N=2 (Group F)
Other conditions	N=1 (Group A)		N=4 (Group D)	

Participants were recruited and consent obtained following approved university human subjects protocols. Participants were paid honoraria and travel expenses, where applicable.

Data Collection

We held five separate focus groups for participants in Groups B, C, D, E, and F. We also conducted six individual interviews:

- A patient in Group A.
- A stroke patient and her caregiver in Group E.
- A caregiver of a cerebral palsy patient in Group E.
- A caregiver of a brain injury patient in Group E.
- A clinician in Group B
- A clinician in Group D.

The research team developed the initial questions, which were designed to stimulate discussion about participants' experiences with brain injury and with HBOT treatment. We then developed interview guides so that appropriate questions would be asked, depending on whether

patients or clinicians were subjects. Questions were sometimes modified from one focus group to the next as experience was gained, which is typical of focus groups and the iterative nature of qualitative research.[8] As illustrations, the interview guides for three different focus groups and one of the individual interviews are shown in the Appendix E Attachment.

The focus groups were conducted as semi-structured interviews because, although the research questions needed to be addressed, we did not want discussion limited to those issues. Instead, we wanted individuals to raise issues of importance to them.

The six focus groups and six interviews were tape recorded and transcribed. Four of the focus groups were held on the Oregon Health & Science University campus in Portland, one was held at a local freestanding hyperbaric oxygen chamber facility, and one was held at a hospital with a chamber outside of Portland. The moderator for all sessions was a qualitative methods researcher with extensive interviewing experience. A second researcher took handwritten notes of both observations and discussion during each focus group. Each focus group session was two hours. The focus group moderator also conducted the interviews, four by telephone and two in person.

Data Analysis

A grounded theory approach was used to analyze the content of the 12 transcripts. This means that the informants' own words were the starting point for the development of themes.[9, 10] The primary qualitative researcher read each line of each transcript, pulling out important words and phrases, which became codes that were written in the margins. As an example, one focus group generated 61 codes, such as "problems with insurance now," "functional measures are needed," and "have to balance risk and benefit." The total number of codes for all transcripts was approximately 500, and from these, the researcher developed lists of patterns and themes. Patterns are issues that occur frequently, and themes are groups of patterns that form a larger idea. For example, a pattern seen in transcripts of patient interviews was that mobility improvements are highly valued. This pattern, along with several other patterns such as vision and speech, ultimately became a theme, "physical improvements," in the Values of Patients section. The data (i.e., the codes) were entered into a spreadsheet so that the phrases could be easily grouped.

Several procedures served to assure that an accurate picture emerged from the data. A second qualitative methodologist who was not otherwise affiliated with the project reviewed all of the transcripts and did high-level coding and analysis paragraph by paragraph. During a series of meetings of the two qualitative researchers, final themes and sub-themes were developed. The other research team members, who often had been present during the interviews and focus groups, then reviewed these results. This review provided another means of ensuring that themes were accurately described. The expert panel also reviewed the results. Finally, the EPC Director, Dr. Helfand, reviewed the transcripts and the report. These various forms of critiques are all methods for assuring trustworthiness, the qualitative equivalent of validity.

Limitations

Limitations of the study included difficulty in eliciting value judgments about HBOT from participants who had never experienced it. Scheduling focus groups for which participants needed to travel long distances was difficult but alleviated somewhat by the ability to do

interviews by phone. The nature of the interviews was different from the focus groups, in that synergy with others was not possible, but they had an advantage in that more detailed longitudinal information could be obtained. Another limitation was that participants familiar with HBOT who were nominated by HBOT clinicians tended to be strong supporters of the therapy. Finally, the individuals within each focus group were not homogenous. For example, interviewees in the focus groups of parents with children with cerebral palsy differed a great deal because their children's conditions varied so much.

Results

Values of Patients and Caregivers

Benefits of HBOT

Brain injury and Stroke. In the brain injury and stroke focus groups, responses varied greatly, and patients mentioned a broad range of outcomes. Each of four participants identified a different problem as his or her major problem—pain, vision problems, seizures, and cognitive impairment. The outcomes valued by HBOT-treated and non-treated brain injury or stroke patients did not differ. Both groups valued frequently measured outcomes, such as cognitive improvements, speech, independence, physical improvements, less pain, short-term memory gains, ability to work, seizure reduction, smell and taste, dexterity, and psychological improvements. They also valued more subjective and less measurable attributes, such as feeling better, less pressure in the head, more positive attitude, easier to live with, decrease in drugs, more energy, greater awareness, and increased self-esteem.

Cerebral palsy. For cerebral palsy patients, the responses reflect that of parents and caregivers brain injury and stroke. The outcomes parents cited as most important were steps that indicated increased interaction with parents, such as saying "I love you" or smiling for the first time. Many commonly measured outcomes were also important to them, including cognitive abilities, vision, spasticity, eating, fewer medications, reduced seizures, speech, and walking. Less commonly measured outcomes were of similar importance: warmer hands and feet, smiling, improvements to the immune system, awareness, less autism, and bladder control. Parents said things such as "sucking from a straw was like a mountain," "putting more than two words together (in a sentence) was a big step," and she was "not as sick as she used to be," thanks to HBOT. Relaxing a hand and decreased drooling were also mentioned as meaningful improvements.

Parents of CP children treated with HBOT spontaneously raised the subject of single photon emission computed tomography (SPECT) scans in every interview or focus group, while parents of CP children not treated with HBOT did not mention it, and when asked, had not heard of SPECT scans. The CP parents with HBOT experience believed the technique provides real evidence of improvement. One stated, "you can see that area of the brain working" [on a SPECT scan]. Another said, "That's why the SPECT scan is so critical. The SPECT scan can document something that makes it undeniable. This is the before. This is the after." This person went on to say that SPECT scans provide the evidence needed to show that HBOT is a "treatment necessary to correct (a physiologic deficit)," and that insurance should therefore cover payment.

There were other differences between parents whose children had and had not experienced HBOT. When asked about temporary regression of symptoms after any therapy, the CP parents without HBOT experience said that improvement for a week may not be worth the effort of seeking treatments, but improvement for 6 months would make it worthwhile. The parents of CP children with HBOT experience would definitely seek more HBOT treatments for either short- or long-term gain or for any degree of improvement as long as they felt the treatments were at a safe pressure level.

Harms and Risks of HBOT

For parents who did not use HBOT, the possibility that their children might undergo risk without benefit was of the highest importance. This attitude seemed to be related to their previous experience with conventional and unconventional therapies.

When asked about the downside of HBOT, patients and caregivers who had HBOT experience did not speak of risks. They tended to talk about the time driving for therapy and other ancillary negatives. One parent had broken an eardrum during HBOT treatments for his son when he entered the chamber, but he did not mention this in the context of risk.

Patients and caregivers (as well as clinicians) feared that the sale of chambers to individuals, often over the Web, is dangerous. Parents described other parents with "chambers in their basements and they have no clue what to do with them." They said of one parent, "People drive to dive in her garage." The parent movement, they said, "has taken on a life of its own…we've got to make it affordable. We have to make it safe."

Values of Clinicians

Benefits of HBOT

Clinicians valued many of the same outcome cited by patients and caregivers, for example, quality of life, cognitive abilities, speech, and motor function. Clinicians pointed out that interactions and independence are highly valued. Clinicians emphasized measurable improvements and objective measures more than did patients or caregivers. They consistently pointed out the variation in the severity of illness among patients, ranging from those who need constant total care to those who are relatively independent.

Unlike some parents, who expressed a strong belief that SPECT scans are valuable indicators of improvements in their children's conditions, clinicians were not so sure. We were told that SPECT scans might not measure outcomes "relevant to function" or measure "a clinical correlate." One clinician who used HBOT said that he has seen "success that is observable, objective, measurable, you know, by a variety of different physical therapy test methods," and he did not use SPECT scans because "I'm not running a big research facility here and there's very little money for it."

Clinicians were able to accurately identify the values parents and patients would have, e.g., they understood how important a child's ability to walk with flat feet for the first time might be to the parent of a CP child. Even when clinicians cited measurable outcomes as being important, they always added that less measurable outcomes, such as the patient interacting more with caregivers, were also extremely important.

Harms and Risks of HBOT

Clinicians who used HBOT for brain injury, stroke, or cerebral palsy and those who did not viewed the harms and risks of HBOT differently. Clinicians who did not use HBOT for brain injury described numerous risks, including financial risks for patients and families. They warned against "people wishing to prey upon families and patients" and disliked the idea of "people taking advantage of patients." The physical risks they mentioned included problems with ears and sinuses, claustrophobia, oxygen toxicity, seizures, blood sugar, vision, and fire. They gave detailed descriptions of the risks along with probabilities of adverse events. Those who used HBOT for conventional indications did not allow family members into the chambers at all because of risks. These clinicians believed that "basement/garage chambers are a disaster waiting to happen.

The HBOT clinicians who treat brain injury, cerebral palsy, or stroke considered the risks to be minimal or did not mention them at all.

Patterns and Themes

There were four overarching themes that emerged from the data: 1) View of Medicine, 2) Attitudes toward Risk, 3) Hope, and 4) Politics.

Views of Conventional Medicine

Patients and caregivers were frustrated with conventional medicine, but they varied in the degree of frustration. Those who had not experienced HBOT had dedicated themselves to various types of therapies, such as physical, speech, and occupational therapies and often believed that "the outcomes are so miniscule." For the particular conditions at issue here, the trial and error approach seemed like a difficult and never-ending journey. The difficult part of the journey, we were told, is seeing your child working so hard and in pain. Botox treatments for the CP patients were cited as one of the worst aspects of their experience, with surgery a close second because it "is a life-consumptive event," engulfing the entire family. When asked about the value of a therapy such as HBOT to help their children, these parents replied that they would want to see real evidence for its value before they would submit their children to one more kind of therapy. One mother stated that the family had been through so much with different therapies that "now I would thoroughly check it out and if I knew it would help, I would dedicate any amount of time to it." One parent said that doctors "are from a different world," and only a parent of a similar child would truly understand what her life is like.

Patients who had sought out HBOT had much stronger feelings about conventional medicine than those who had not used HBOT. They believed they had exhausted the limits of conventional medicine and moved on to alternative therapies. Most patients were using several complementary and alternative medicine therapies at once, which made it difficult to judge whether HBOT or something else was making a difference. Some caregivers believed that conventional medicine is sabotaging HBOT, undermining efforts to prove its efficacy. These patients and caregivers were more willing to trust strongly worded testimonials and case studies than scientific evidence, partly because of a lack of trust in the medical establishment.

The names of a few physicians were consistently invoked as having gained the trust of these patients and caregivers. Parents of children who had been treated with HBOT believed that

SPECT scans provide evidence of improvement, even if that improvement is not correlated with a clinical improvement.

Patients who had sought HBOT tended to have a large medical vocabulary of terms relating to the relevant condition. The terminology was retained from information they gathered, but the meaning was sometimes lost. Often, the terms were used in new ways that seem to fit the belief systems of the caregiver. For example, one parent equated her CP child's brain to a Pepsi: "you put a body inside a hyperbaric chamber and submerge it in a hundred percent oxygen with that pressure, every cell in that body is saturated with that oxygen. Most importantly the spinal fluid that goes up the back around the brain and it bathes the brain. It immediately starts breaking that calcification down."

Clinicians differed in their view of conventional medicine as well. The clinicians who did not use HBOT for brain injury, stroke, or CP wanted to see clear evidence that it works and does no harm: they would carefully weigh the risks and benefits. They "love to see people get better but need real evidence." These clinicians pointed out that many therapies in general use have not been proven completely useful and are sometimes prescribed so that "parents feel more engaged." They stated that they would like to see good studies on HBOT and were aware of and valued the 2001 Canadian study on cerebral palsy and HBOT.[11]

The clinicians who used HBOT for brain injury, cerebral palsy, or stroke considered their own experience to be clear evidence, and they could tell stories of their own patients as testimony. Two focus groups produced 15 stories of dramatic improvements after HBOT treatments. These clinicians stated that they would like to see good studies done, but they did not talk about evidence as often as the other clinicians. They spoke about other alternative therapies more often than clinicians who did not use HBOT and consistently emphasized the value of combining HBOT with nutrition and other wellness approaches. They also held strong beliefs in the benefits of diet, vitamins, stress reduction, and other therapies, with HBOT just one of many modalities in a holistic approach to treatment.

Health Information Seeking

This theme encompasses a wide range of activities, from phoning a society to requesting a book about cerebral palsy, which becomes "my first lifeline" after a parent learned her child had CP, to participating in online communities of patients, caregivers and supporters of HBOT.

Patients and caregivers unanimously said that they had done their own research about their conditions. All but one was active in networking online. The parent who said that doctors are from another world also said she actively asked other parents of children like hers for their advice. The CP parents said that parental decision-making is "like playing God," and that any help they could get was wonderful. The HBOT-supporter parents had active online communities with a listserv, large organizations of parents, and newsletters.

Clinicians used online resources as well. The clinicians who used HBOT for brain injury and stroke had a clear presence on the Web, and they have used the media, including television, to disseminate information about HBOT.

Hope

Faith and prayer were mentioned by both clinicians and patients and caregivers as therapies in their own right for those with brain injury, stroke, or cerebral palsy. However, the HBOT

caregivers volunteered many stories about miracles. HBOT was referred to as "God's medicine," and one parent said, "God's put the chamber here for the kids to be helped." Miracle stories from both parents and clinicians were many. One example was about an aphasia patient who "started spontaneously talking again" after 20 HBOT treatments, and another was a story about a stroke survivor who "lost the cane 3 days after starting the treatments." The caregivers who had not experienced HBOT spoke about little steps, about the small bits of progress they hoped their children would see. For that hope, they lived family lifestyles full of activity involving one therapy after another.

Politics

This term is being used broadly to signify politics at the national level and its relation to HBOT, politics among and within societies related to HBOT, and the politics of insurance and getting treatment paid for. The clinicians and patients and caregivers who had no experience with HBOT were unaware of such politics. However, clinicians and patients and caregivers with HBOT experience were extremely vocal about it. Those with HBOT experience for approved conditions knew about efforts at the national level to gain funding for clinical trials for HBOT for brain injury, cerebral palsy, or stroke, about the efforts to have insurance pay for HBOT treatments, and about the online communities of HBOT supporters.

Patients and caregivers with HBOT experience for brain injury, stroke, or cerebral palsy were pleased that the present systematic review was being done, and they were constantly aware that what they said might influence funding. Many were involved in advocacy for funding for HBOT therapy for brain injury, cerebral palsy, or stroke, clinical trials, and attention to the safety of chambers. This group distrusted the medical establishment, however, and did not believe that unbiased clinical trials could be conducted. One parent said it "is sad that our country would deprive us of something and we would have to go to those extremes" [such as going to another country] to get HBOT therapy.

Conclusions

We undertook this study to ensure that our systematic review sought evidence about outcomes that patients and caregivers care about. We also wanted to test the hypothesis that differences in attitudes about short-lived or small responses to therapy might explain differences in views about the effectiveness of HBOT.

We found that commonly studied outcomes, such as motor and speech function, are important to patients and caregivers. However, we also found that many outcomes that are not routinely measured in studies are of equal, if not greater, importance to patients and caregivers.

Our results did not reveal any consistent differences among the groups of patients or clinicians in how they valued small or temporary improvements. Rather, the results suggest that what we might consider to be a small or temporary improvement in function can have a large impact on caregiver burden. Discussions with caregivers revealed the most dramatic differences between what studies measure and what participants valued. Functional differences that might seem small or insignificant on standardized functional scales have a large impact on caregivers. Put differently, even if the standard measurement instruments are sensitive to small changes in

function, they may be insensitive to the impact of these changes on caregivers and on the family as a whole.

All focus groups (clinicians and patients and caregivers with and without HBOT experience) agreed on the need for more evidence about the efficacy and safety of HBOT for brain injury, stroke, and cerebral palsy. For patients and caregivers with HBOT experience, however, this need is tempered by distrust of the medical establishment. Without trust, it is questionable whether or not they will believe that the research was conducted in good faith.

Patients and caregivers who had not used HBOT are skeptical of the benefits and concerned about the potential adverse effects. This skepticism arises from their experience with other treatments for which the expected benefits never materialized or were not worth the harms, inconvenience, cost or risk. They will continue to be skeptical unless better-quality studies are done.

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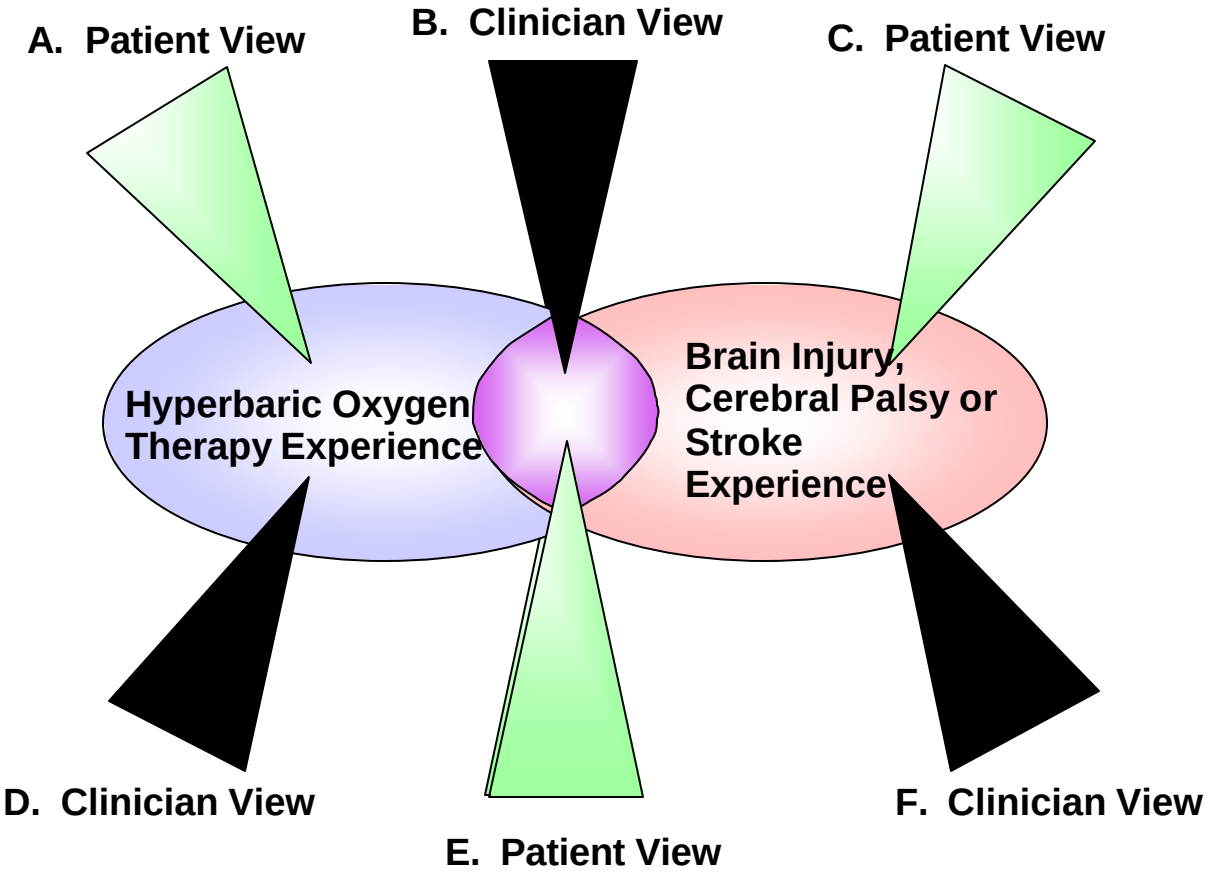
Supplement Figure



Supplement Appendix. Interview Guides

Guides for Focus Groups

Brain injury or stroke patients with no HBOT experience (February 23, 2002)

Introduction of study personnel
Purpose of focus group and importance of their roles
Signing the consent forms
Introductions: name, their injury (what it was, date, outcome)
Ground rules: all discussion voluntary, no criticism, confidentiality assured, try to represent peers

Exercise: write down the five worst things about their episode's aftermath; five positive things?
Go around and list them

List? What outcomes do they desire? Describe a really good outcome

Which are most important? Make up a scale and rate them (really important, moderately or less)

How important is duration of each outcome? For what outcomes would you be willing to accept only short-term outcomes?

What might your caregiver say?
Inconveniences and discomforts

Imagine that there is a therapy that can be delivered in a submarine that is perfectly safe. How would you feel about entering the submarine? On a daily or weeky basis?

Let me summarize what we've learned today: Have we missed anything?

Brain injury or stroke patients/caregivers with HBOT experience (March 11, 2002)

Signing the consent forms
Introduction of study personnel
Purpose of focus group and importance of their roles
Ground rules: all discussion voluntary, no criticism, confidentiality assured, try to represent peers

Introductions: name, their injury (what it was, date, outcome), therapies besides HBOT they've tried

Exercise: write down the five worst things about their situation; five positive things?
Go around and list them
List? What outcomes do they desire? Describe a really good outcome

What outcomes did HBOT produce?
Which are most important? Make up a scale and rate them (really important, moderately or less)

Outcome if short term Importance 1 to 3
Outcome if long term Importance 1 to 3

How important is duration of each outcome? For what outcomes would you be willing to accept only short-term outcomes?

What might your child say?
Inconveniences and discomforts

Let me summarize what we've learned today: Have we missed anything?

Clinicians with HBOT experience (March 15, 2002)
Signing the consent forms
Introduction of study personnel
Purpose of focus group and importance of their roles
Ground rules: all discussion voluntary, no criticism, confidentiality assured, try to represent peers
Introductions: name, their background that led to interest in HBOT
How did interest develop?

Upsides and downsides
The good and bad for each condition
Inconveniences and discomforts

What outcomes do they desire? Describe a really good outcome
What outcomes did HBOT produce? Listen for coordination, weakness, walking, cognition

Which are most important? Make up a scale and rate them (really important, moderately or less)
Outcome if short term Importance 1 to 3
Outcome if long term Importance 1 to 3

How important is duration of each outcome? For what outcomes would you or patients or caregivers be willing to accept only short term outcomes?

The SPECT scan issue

Focus on CP and stroke issues

Future research suggestions

Let me summarize what we've learned today: Have we missed anything?

Guide for Individual Interviews

(Note: This is a generic guide; these questions were always covered, but more specific questions and probing questions were also asked in addition to these.)

Introductions

Discuss the consent form

Purpose of project and interview

Can you start out by telling me what led up to your/his/her first HBO treatment?

Had you heard about HBO before? How did you hear about it?

What was the course of treatment like? How many? Kind of chamber? Did caregiver accompany? Feelings about it?

What difference did you notice and at what point in treatment? What was the most exciting breakthrough, if any?

Were there any discomforts or inconveniences?

What other therapies were you undergoing at the same time? Prior to HBO?

One of the things we're trying to find out is how people value the outcomes of HBO. What good does it do and what is it worth to you? To your caregiver or child?

Let me summarize what we've learned today: have we missed anything?