INVESTIGATING NEXT-OF-KIN'S CAPACITY TO GIVE VALID CONSENT FOR RESEARCH ON BEHALF OF A CRITICALLY UNWELL LOVED-ONE IN THE CONTEXT OF THE INTENSIVE CARE UNIT

by

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LIST OF ABBREVIATIONS

APACHE II SCORE: ACUTE PHYSIOLOGY AND CHRONIC HEALTH EVALUATION II

BSL: BRITISH SIGN LANGUAGE

FB: FALSE BELIEF

ICU: INTENSIVE CARE UNIT

IRAS: INTEGRATED RESEARCH APPLICATION SYSTEM

NRES: NATIONAL RESEARCH ETHICS SERVICE

QEHB: QUEEN ELIZABETH HOSPITAL BIRMINGHAM

RN: RESEARCH NURSE

SAE: SIGN ASSISTED ENGLISH

SDM: SUBSTITUTE DECISION MAKER

SE: SPOKEN ENGLISH

TOM: THEORY OF MIND

UBACC: UNIVERSITY OF CALIFORNIA, SAN DIEGO BRIEF ASSESSMENT OF CAPACITY TO CONSENT

PREFACE

A META-INTRODUCTION TO THE THREE PLACEMENT PROJECTS

This thesis grows out of my research experience in three placements carried out during my one year MRes Clinical Psychology course at the University of Birmingham. It takes the title from my main research project, and its aim is to introduce the reader with a description of the placement projects using a chronological framework (from the Autumn placement to the Summer term placement). The thesis includes three chapters with a common structure, in that each of them is introduced by a practical summary of its contents, followed by a brief placement overview, the description of the core part of my research and a final part focusing on my reflections on learning.

Chapter 1. Placement 1: *'Are substitute decision makers able to provide valid consent/assent for their critically ill loved one?'* This chapter focuses on my first placement, during which my supervisors and I designed a pilot mix methods study aiming to investigate next-of-kin's capacity to consent for research involving a critically ill close relative. The chapter is all about a very applied piece of research, which arose from a need manifested by clinicians and aimed to provide practical recommendations. The report discusses the main aspects of the research protocol that I wrote under attentive supervision, and the main ethical issues taken into consideration during research planning and ethical approval seeking.

Chapter 2. Placement 2: *'Developing an appropriate battery of ToM tasks for deaf children (false belief paradigms)'.* This chapter outlines the main stages of my second

project, which aimed to provide more appropriate measures of False Belief (FB) reasoning in deaf children. In particular, I and my supervisor developed a new battery of FB tasks presented in video format: I filmed the stories creating *Lego* stop-motion animations. Three versions of the videos were produced in order to allow deaf children to use their preferred method of communication. The chapter provides preliminary results regarding the usability of the instrument, and shows how this battery, once appropriately improved, could overcome some methodological issues faced by previous studies. The chapter includes the slides of my *Prezi* presentation of the project, combined with a further commentary.

Chapter 3. Placement 3: '*Are substitute decision makers able to consent/assent for their loved one in the context of the Intensive Care Unit?' From the research protocol to its implementation.* This chapter focuses on my main research project, which involved the implementation of the mixed methods research designed in the Autumn term. Using a concise introduction summary, given that the theoretical background of the study was described in Chapter 1, the report takes the reader through the research paradigm used and the analytic procedure carried out with both quantitative and qualitative data. Results are presented following the order of the research questions.

Each chapter was written at the end of each project and afterwards joined together to compose this thesis; however, the three chapters should not be seen as separate elements, rather as different phases of a single challenging research adventure. Each chapter shows my learning process and my growing attention to methodological and ethical details, and also a gradually more confident use of quantitative and qualitative research methods as well as reflexivity exercise. Throughout the three projects, I had the possibility to learn directly from

experience what conducting a good piece of research means. I have learned that it is the researcher's responsibility to describe, in a honest and exhaustive way, the methodology used and the limitations of their study in order to achieve credibility standards, and allow generalisability of results and the possibility for other researchers to reproduce them. In addition, the use of supervision helped me develop critical thinking skills such as flexible and synthesis-thinking. I hope the reader will see throughout the chapters not only my determination and effort to present well-thought pieces of research but also the enthusiasm placed in each project.

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CHAPTER 1

PLACEMENT 1

Are substitute decision makers able to provide valid consent/assent for their critically ill loved one?

The following report will provide the reader with:

- A description of the aims of my first placement, highlighting my main objectives and role in the project (section 1);
- A reflection on the research context, that is the Intensive Care Unit (ICU) of a hospital in Birmingham. In particular, I will provide a reflection on the ICU environment and research practice (section 2);
- An introduction to the research rationale, design, methods, outcomes, and ethical issues (section 3);
- A reflection on learning (section 4).

1. Placement overview

The initial aim of my research project was to design a pilot study able to investigate with a qualitative design the experience of next-of-kin, friends, and representatives who have been asked, as substitute decision makers (SDMs), to consider providing consent for research on behalf of a critically unwell loved-one, unable to decide for him/herself. Through discussions with the wider research group, this initial idea evolved into a more articulated project combining qualitative and quantitative methods. In particular, the goal of the research group became focused on designing a research study able to tell us whether SDMs are able to provide valid consent/assent for research on behalf of their ill loved one in the context of the

ICU (the main research question), and what factors influence their capacity to do this. See Box 1.1 (page 7) for clarification about the difference between assent and consent.

In addition, it was proposed that the use of qualitative data could add exploration of the experience of the participants involved in being approached to give consent for their relative to take part in research (the latter answering a secondary research question).

I joined a multidisciplinary research team, including Dr J. Oyebode, Dr I. Mackenzie, Prof F. Oyebode and Prof J. Bion, and had the opportunity to be involved in the preliminary discussions and decisions concerning project design and methodology. Initially, my role involved searching MEDLINE and psycINFO for relevant literature about surrogate decision making in critical care research, and identifying whether there was an appropriate existing instrument for the assessment of the capacity to consent for research, adapting it, as necessary, to our subject of investigation. A further key objective was to become familiar with the ICU environment and research practice there. To fulfil this goal and explore the feasibility of the project, I visited the ICU – Queen Elisabeth Hospital Birmingham (QEHB) and had meetings with ICU research nurses.

The next objectives involved applying for the study ethical approval, through the Integrated Research Application System (IRAS), and writing a detailed research protocol. Last but not least, aware of the complexity involved in the study and in the assessment of mental capacity, I undertook the “Adult Lacking Capacity” NRES (National Research Ethics Service) online training and studied the Mental Capacity Act 2005 and the legislation relating to informed consent. Regular weekly meetings with Dr J. Oyebode and monthly meetings with the entire research group ensured continuous supervision.

2. Reflection on the ICU environment and research procedures

"Nowhere in medicine do health care providers, patients and families interact with such intensity and ambiguity, often for protracted periods of time."
Burns, Zubrinich, Marshall, & Cook, 2009, p. 1656.

In line with one of the main objectives of my research placement, I developed the following reflection about the ICU environment and research practice. Its aim is to provide a clearer picture of the research context, and to consider the influence that the environment and research procedures have in the SDM's experience of the ICU, using a mainly ethnographic approach where informal conversation with key informants, ICU research nurses and ICU consultants, were combined with direct observation.

To begin with environmental aspects, the first impact of the QEHB is that it is unlike a conventional hospital. The huge main hall is quite welcoming and grand, and the high ceiling prevents the claustrophobic sensation that hospitals often cause. The ICU is on the second floor. For each of the four ICU areas, a robust door with reflective mirrors prevents visitors from seeing through. A sign informs visitors about the visiting hours: 11 am – 8 pm. To access the unit the visitors need to ring the doorbell and wait for an unpredictable time. As an ICU consultant explains, outside visiting hours the waiting time can be considerable. Thus, often visitors simply follow the staff accessing the unit to get inside. Inside the unit, most of the beds are in an open space. In some areas, there is little natural light, making it difficult to judge whether it is day or night. The majority of the patients are unconscious or sedated and next to each bed a big red line demarcates the space for each patient, and his or her relatives. There is no privacy. Moreover, for each patient a board beside the bed displays details about their clinical records. I was told by staff that details regarding contacts

between staff and patients' families should be also recorded. This is not always the case, as a consultant explains. Is this factor an indicator of a gap in the abovementioned relationship? Is this linked to a medical culture that does not give enough importance to emotional and relational aspects of care? It is difficult to say and also is not the core subject of this reflection. However, timely/regular clinician-family communication may be extremely important as literature suggests that they can support the next-of-kin's confidence in the role of surrogate. As an observer, I noticed that the ICU staff seemed very attentive to families' needs for information.

A "key room" is the consultation room. Here, clinicians talk privately to the family. This makes this room potentially the 'bad news room'. Interestingly, in the same room, researchers connected with a range of clinical studies approach the next-of-kin about critical care research. We could thus hypothesise that disturbing emotions potentially evoked by the environment could influence the next-of-kin's decision-making process. All these physical and environmental aspects might have an important influence on the families' experience of the ICU, as well as their perception of the care received by their loved ones.

Let us now consider research practice in the ICU. In the ICU highly trained staff, sophisticated equipment and medication guarantee high-level care for life-threatening conditions and high-level research practice. The main areas of research are respiratory management, epidemiology and pulmonary trauma. ICU research studies commonly focus on potentially life saving interventions, and so require enrolment at the earliest opportunity. This means the next-of-kin often have to be approached soon after the patient's admission. When they have only just arrived, next-of-kin do not know what is going on and what will happen to their close relative. In this delicate time, when they may have already received

complex medical information, the next-of-kin is asked to make a decision about whether or not their loved one, unable to decide for him/herself, should take part in research.

The time pressure is not easy to manage for either the researcher or the SDM. Every day the researcher has to gather the information necessary to decide about the eligibility of newly admitted patients for a specific study. Once an eligible patient has been identified, the researcher has to approach the family as soon as possible, given the short window for enrolment characterising these medical studies. In general, enrolled patients are those who stay for 4-5 days or more (on average patients stay at the ICU for 2-3 days). The number of SDMs approached is limited, usually no more than 3 (i.e., for 3 eligible patients) a day. In other words, the vast majority of next-of-kin are not approached. The main reasons for this low recruitment rate lie in the innate features of research practice in the ICU: a limited number of patients meeting the research eligibility criteria, the short enrolment window, and problems in contacting the SDM. The SDMs generally have one hour or so to decide whether to consent for their relative to be included in a study. As some staff suggested, this can make the decision tougher than it would otherwise be. The time pressure on the SDM may cause additional distress, and may influence the decision making process and the final decision. The SDM's decision is also influenced by a number of other factors including misconceptions about the patient's recovery and unrealistic expectations of research outcomes. Therefore, researchers should always be aware of the risk that proxies will misunderstand the primary purpose of the trial as therapeutic.

In summary, the literature suggests that asking next-of-kin to decide about research on behalf of a critically ill loved one causes additional stress to the burden they are already experiencing. My observations and informal conversations also suggested that physical and

research procedural aspects influence the next-of-kin's perception of the ICU clinical and research activity, potentially affecting the burden experienced and the confidence in making the decision about research. Certainly my reflections, developed after a few visits to the ICU, cannot fully reveal the ICU environment. However, I hope I have highlighted some aspects that can provide a better picture of the context of my research project.

3. The research

3.1. Introduction

Clinical research is an important means of improving the care of severely ill patients. However, critically ill patients are unable, most of the time, to give consent to research due to their health condition or treatment; therefore researchers have turned to SDMs to gain consent for patients' participation in research (Luce et al., 2004; Scales et al., 2009). See Box 1.1 for details about the legal framework underpinning the SDM's role.

Box 1.1: Legal framework

Substitute decision making. In the UK, when research involves adults lacking capacity to consent, it is a legal requirement that someone close to the patient is consulted prior enrollment into research. For trails involving the study of investigational medicinal product (CTIMP research), the Medicines for Human Use (Clinical Trials) Regulations 2004 requires that a Legal Representative provides or declines consent on behalf of the participant lacking capacity. On the other hand, non-CTIMP research involving adults unable to consent is regulated by the Mental Capacity Act 2005 (MCA 2005), which states that the researcher must seek a consultee, who does not consent on behalf of the participant and instead is invited to provide informed assent, comfirming the patient's enrolment into research. The MCA 2005 specifies that the SDM's decision for the person lacking capacity must be made "in his best interest" considering, at the same time, "the person's past and present wishes and feelings" (4.6a), "the beliefs and values that would be likely to influence his decision if he had the capacity" (4.6b) and "other factors that he would be likely to consider if he were able to do so" (4.6c).

Since many ICU research studies require enrolment soon after the patient's admission, the time of approach about research may be particularly delicate for the SDMs, as they may be worried, sleep-deprived, and already dealing with complex medical information (Mehta et al., 2012). Therefore, the complex nature of the information about medical trials combined with the emotional burden already experienced and the burden inherent in making decisions for a critically ill relative 'renders the integrity of the decision-making process uncertain' (Burns, Zubrinich, Marshall, & Cook, 2009, p. 1656). Evidence suggests that next-of-kin do not adequately understand the nature of research about which they are approached. A French study involving the families of ICU patients showed that half failed to comprehend the diagnosis, prognosis, or treatment of the patient, due to various patient-related (e.g. age, reason for ICU admission), family-related (e.g. pour knowledge of French) and physician-related factors (first meeting with SDM <10 mins, no information brochure for the SDM) (Azoulay et al., 2000). Chappuy et al. (2010) described similarly poor understanding in parents who were asked to consent for their child to participate in a therapeutic leukaemia trial: half could not explain the aim of the research nor its potential benefit.

In addition, deciding about the patient's enrolment into research may place an additional burden on them (Pochard et al., 2001). A high prevalence of symptoms of anxiety (>60%) and depression (>30%) has been found among the majority of family members visiting ICU patients (Barrett et al., 2012; Pochard et al., 2005), underlining that anxiety and depression may have a major impact on SDMs' capacity to make decisions (Pochard et al., 2001). This means that SDMs may make decisions about their loved one participating in research based

on their own emotional state rathen than in line with the patient's wishes (Mehta et al., 2012; Menon et al., 2012; Pochard et al., 2001).

Although most SDMs seem to be willing to participate in research decision making (Barrett et al., 2009; Chenaud, Merlani, Verdon, & Ricou, 2009; Perner, Ibsen, & Bonde, 2010), there is some evidence that some family members do not want to make decisions about their critically ill close relatives' care: in Azoulay and colleagues' study (2004) nearly one-half of families preferred to let the doctors make the decision. Moreover, Barrett and colleagues (2012) found that although many SDMs were comfortable with being involved in the decision process (50%), the number decreased when risk of harm involved in an hypothetical research scenario was higher (34%) or enrolment window was shorter (41%). It seems also that SDMs prefer to share decision making with the medical team (Majesko, Hong, Weissfeld, & White, 2012; Heyland et al., 2003). This could alleviate some emotional strain associated with the decision-making, although concerns about therapeutic misconception and social desirability arise (Barrett & Scales, 2012).

Therapeutic misconsception refers to misunderstanding the primary purpose of the clinical trial as therapeutic (Henderson et al., 2007). This could be explained by an actual lack of understanding of the difference between the goals of clinical care and the goals of the research, or alternatively it may result from the ICU next-of-kins' desperation to help their loved one, predisposing them to see the research proposal as a potential new redeeming treatment (Barrett & Scales, 2012; Mehta et al., 2012).

This research demonstrates that although the SDM model for informed consent can protect the patient, it is not perfect (Barrett & Scales, 2012), and additional safeguards for research involving greater-than-minimal risk should be used to protect both the patients and

the SDMs (Ciroldi et al., 2004). As Silverman and colleagues (2004) have suggested, an independent person should assess SDMs' capacity and understanding about the benefits and risks of research studies.

In this study we aimed to investigate SDMs' capacity to give valid consent for research on behalf of a critically unwell loved-one and to find out which factors influence their ability to do this. These factors are presented in Box 1.2 and will be further described in Chapter 3. In particular, this study aimed to develop understanding of whether the SDMs' capacity to consent for research was influenced by the complexity of the study (see page 11). Complexity was included as a factor as a result of previous research (Rosenstein, 2004) showing that a given individual at a given time may be capable of consenting for themselves to participate in a simple research study, but may be incapable of consenting to a more complex research study. In other words, the more complex the study, the harder it may be for the decision-maker to understand factors such as its purpose, design, risks and benefits. For this reason, we considered two types of studies, observational and interventional, where the former usually involves limited complexity and limited risks, while the latter involves the use of drugs or procedure associated with a higher risk.

Therefore, this study tried to better understand these issues applied to the context of substitute consent for research in ICU.

Box 1.2: Factors related to SDMs' capacity to provide valid consent for research

Even though literature has highlighted numerous factors potentially related to the SDMs' capacity to provide valid consent for research, in this pilot study it will be possible to analyse only some of these factors. In order to decide which to include in the investigation, the research team discussed each in turn and ranked them for importance. Below are reported the top eleven ranked factors:

SDM-related factors: time of day of approach for research, time elapsed between notification of patient's admission and approach, SDM's health status/level of distress, proximity of the relationship with the patient, SDM's perception of additional risk, SDM's age and education, previous conversation with the loved one about preferences regarding clinical trials.

Study-related factors: enrolment window and type of study, i.e. observational vs. interventional. *Observational research* is based on the collection of data which is then correlated with patient outcome (limited risks associated); *Interventional research* involves the use of a drug, piece of equipment, system, or procedure. The risks involved can vary substantially depending on the novelty and specific characteristics of the experimental intervention.

Patient-related factors: Severity of patient's illness, age of patient.

We also wished to explore with those who were actually asked to consider providing surrogate consent, what their personal experience of doing this was like. The implications of such a study are important in terms of understanding whether the current model of surrogate consent works well for the next-of-kin of ICU patients, or whether there is a need to find better ways of protecting patients and their next-of-kin, and fostering valid consent, whilst permitting the advancement of medical care through research.

Principal research question:

- Are SDMs in the context of critical care research able to give valid consent for their loved ones?

Secondary research questions:

1. Is the SDM's capacity to consent for their loved one influenced by whether the study is observational or interventional?
2. What factors affect capacity at the point when SDMs are asked to make the decision for their loved one?
3. What is it like for SDMs to be involved in the decision-making process for critical care research?

3.2. Design and method

This is a descriptive and correlational study which combines quantitative and qualitative data. The study has been designed to include two stages:

Stage 1: Assessment of capacity to consent. This stage addressed the main research question. In particular, through a quantitative design, the proportion of SDMs able to consent/assent for their loved one was investigated. I assessed the SDM's capacity to consent for research using our adapted version of an existing test (Appendix 1).

Stage 2: Investigation of factors influencing capacity and the experience of SDMs. Following the assessment of capacity, I conducted a structured interview to investigate factors influencing SDMs' capacity to consent. The same interview included also a less structured section to allow for qualitative exploration of the experience of being involved in this decision (Appendix 1).

3.2.1. PARTICIPANTS AND RECRUITMENT

The study was conducted at the ICU QEHB. The participants were the SDMs of patients meeting ICU research eligibility criteria and previously approached to consider giving consent for their loved one to participate in clinical research studies ongoing in the ICU (including both those who gave and those who declined consent) (Figure 1.1). Eligibility criteria (Table 1.1) were assessed on the day the SDM is approached to take part.

Table 1.1. Inclusion/exclusion criteria

Inclusion criteria
• Patient meeting ICU research eligibility criteria • Patient with unplanned ICU admission • Patient with identifiable SDM
Exclusion criteria
• Substitute decision-maker under 18 years old • Substitute decision-maker who does not speak English • Patient unlikely to survive 24 hours (as this would place too much burden on the SDM)

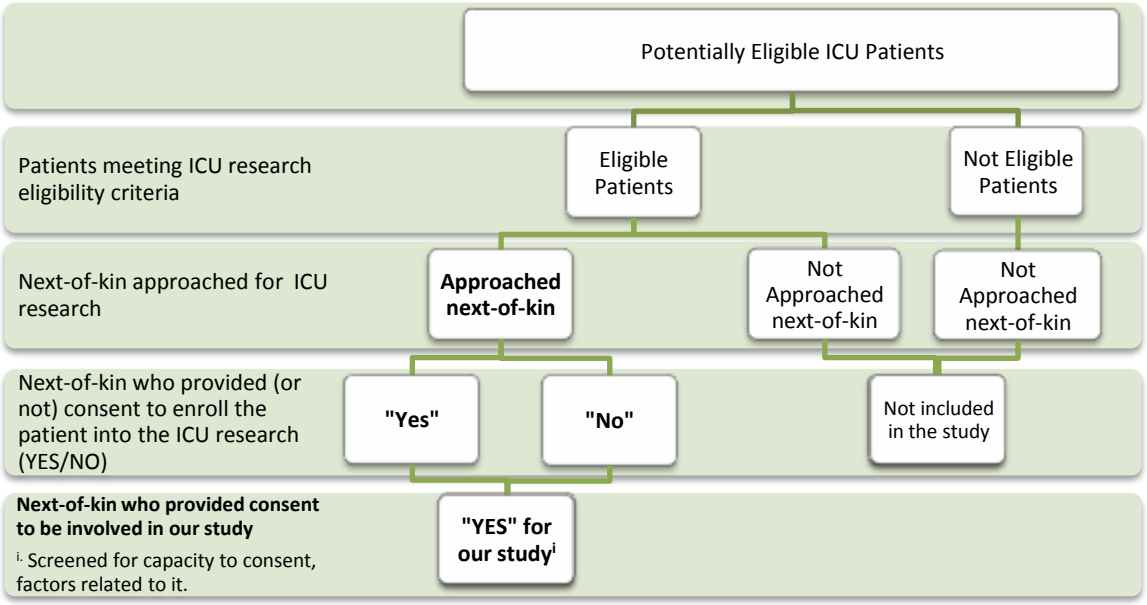


Figure 1.1. Participant identification

Figure 1.2 shows the recruitment procedure, which was designed to be as unintrusive as possible and was discussed and agreed with the ICU research team and a member of a panel of service users. See Chapter 3 for details of how it was conducted.

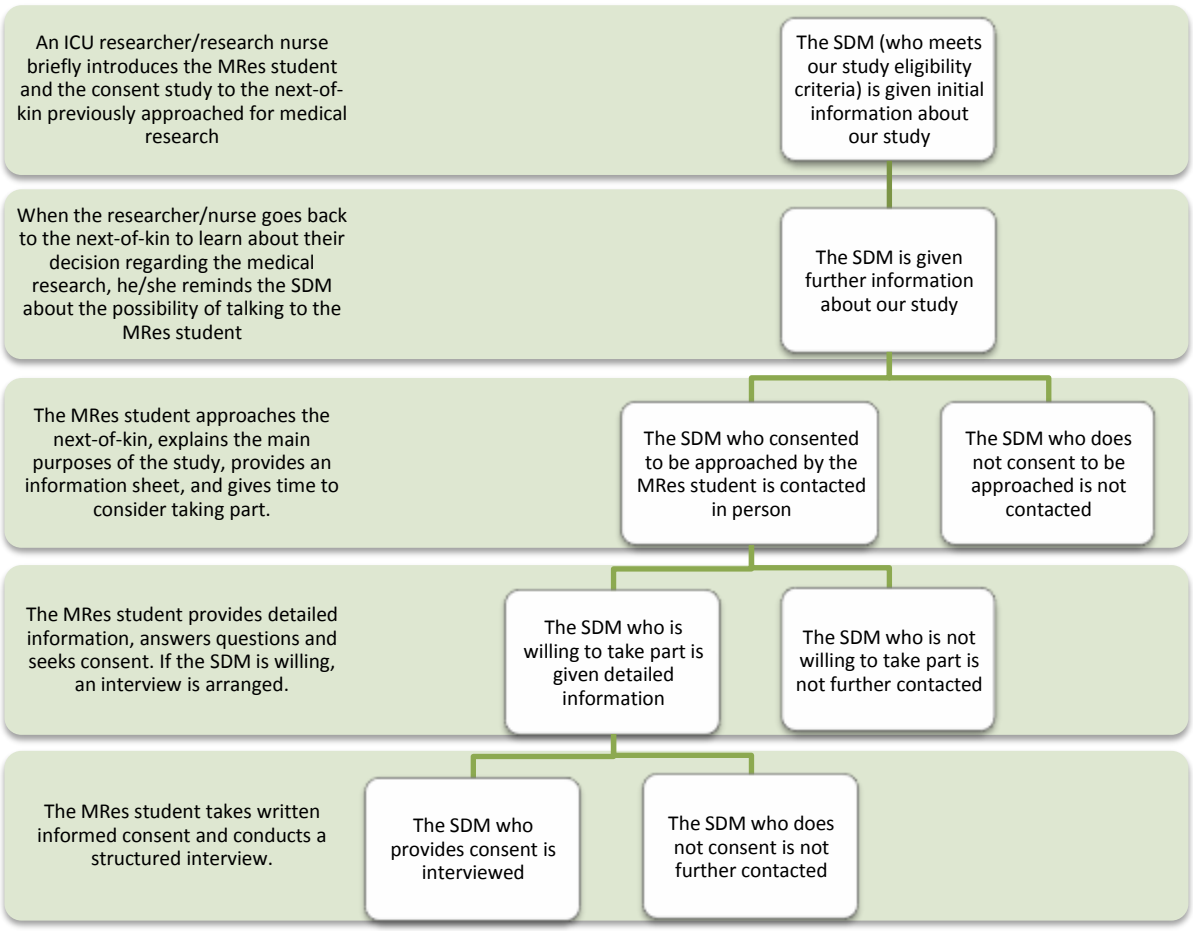


Figure 1.2. Recruitment procedure

3.3. Instruments

A combination of qualitative and and quantitative methods was considered to be the most appropriate approach to data collection, enabling us to gain a rounded understanding of the substitute consent process. The use of a quantitative measure of

capacity would allow clear conclusions to be drawn about the proportion of SDMs demonstrating capacity, whilst qualitative methods allowed us to contextualise the quantitative data within the personal experience of the SDMs, and uncover the range of meanings the participants themselves attributed to this experience.

3.3.1. MEASURES TO ASSESS MENTAL CAPACITY TO CONSENT FOR RESEARCH

Mental capacity is the ability to make a decision. According to the Mental Capacity Act (MCA) 2005 (section 3(1)), "a person is unable to make a decision for himself if he is unable— (a) to understand the information relevant to the decision, (b) to retain that information, (c) to use or weigh that information as part of the process of making the decision, or (d) to communicate his decision (whether by talking, using sign language or any other means)". Mental capacity is also time-specific and decision- specific. This means that capacity to consent for research needs to be assessed in relation to the specific decision which needs to be made and the particular time at which the decision is made. The above-mentioned areas of competence from the MCA 2005 are generally identified in the literature as four components of decisional capacity: understanding information relevant to the decision; appreciating the information; using the information in reasoning; and expressing a choice. I searched MEDLINE and PsycINFO for an appropriate measure to assess capacity to consent for research. Two main measures were identified: the MacArthur Competence Assessment Tool for Clinical Research (MacCAT-CR, Appelbaum & Grisso, 2001) and the University of California, San Diego Brief Assessment of Capacity to Consent – UBACC (Jeste et al., 2007).

Of these two, the MacCAT-CR is the most comprehensive, assessing all four domains of capacity through a semistructured interview, and able to be tailored to the details of a specific research protocol. It is also the most widely used measure to date, and recommended by Dunn et al. (2006) in their review of instruments. However, it also has limitations including the need for substantial training to administer the interview, and a long administration time of up to 30minutes (Dunn et al., 2006). These factors made us conclude that this tool was not appropriate for this study.

The UBACC was developed by Jeste and colleagues (2007) and is a 10-item scale, which assesses the first three components of decisional capacity and requires 5 minutes to complete. It has also good internal consistency, interrater reliability, high sensitivity, and acceptable specificity. The UBACC was validated against the MacCAT-CR (Appelbaum & Grisso, 2001), showing high correlation with its scores. Due to its reasonably comprehensive focus and brevity, it was deemed to be appropriate for the purpose of this study.

3.3.2. THE ADAPTED UBACC

Stage 1 of the interview included the UBACC (Jeste et al., 2007), which was minimally adapted in order to address the questions to the SDMs instead of directly to the patients. For example, the original item “If you participate in this study, what are some of the things that you will be asked to do?” was modified as follows: “If your loved one were to participate in the study, what are some of the things that will happen to him/her or they will need to do?”. See Appendix 1.

In line with the MCA (2005) assessment requirements, the items concern: understanding of the purpose of the ICU study (item1 and 3); the capacity to weigh the received

information about the study (item 2); appreciation of the possibility of dissent (item 4), understanding that the patient will get regular treatment in case of refusal (item 5), the appreciation of what the research implies for the patient (item 6); and understanding of potential risks (item 7) and benefits (item 8 and 9). In addition, we removed item 10 from the original version of the UBACC, which focuses on compensation in the event of injury as a result of participation in research, as not relevant to our investigation; instead, we added an item addressing the capacity to express a decision, which assesses the fourth domain of decisional capacity as required by the MCA 2005.

3.3.3. THE INTERVIEW

The interview included also closed-ended questions addressing the factors influencing SDMs' competence to make this decision (see Appendix 1). The items included were derived from existing questionnaires (Barrett et al., 2012; Mehta et al., 2012) and explored factors highlighted in the related literature. Furthermore, the inclusion of open questions aimed to explore, with a qualitative method, what the experience of being involved in this decision is like for the SDMs. This second part of the interview was designed to be more flexible and less structured than the initial part. The ICU research team and a member of a panel of service users were asked to determine the readability of the interview and the need to include other relevant items.

3.4. Analysis

3.4.1. QUANTITATIVE ANALYSIS

We planned to use descriptive statistics to describe the proportion of SDMs with capacity to consent for their loved one. Given that the main goal of our study was to obtain descriptive data, we did not perform a sample size calculation, and anticipated that 30-40 participants (with 30 as a minimum number) would have provided a reasonably representative sample, (recruiting 3 to 4 participants per week over an 11 week period). We planned to include the same participants in stages 1 and 2 of the research. In order to understand the correlation between SDMs' capacity to consent for their loved one and the factors influencing capacity, a logistic regression was considered the most appropriate analysis, with capacity to consent (two conditions: CAPABLE vs. UNCAPABLE group) as the criterion variable and the factors as predictor variables. We also planned to look at the association between capacity to give consent and complexity of the study for which the SDM was approached, using the Kruksall-Wallis test. Observational studies were categorised as low risk and interventional as high risk.

3.4.2. QUALITATIVE ANALYSIS

The 6 phases of thematic analysis proposed by Braun & Clarke (2006) were deemed to be the most appropriate framework for analysis of our qualitative data (see Chapter 3 for details). This approach is consistent with the researchers' theoretical framework, which is phenomenological and contextual constructionist. Mixing qualitative with quantitative data was considered to be the most appropriate approach to provide a greater understanding of

the substitute consent process, allowing the researcher to contextualise the quantitative data within the personal experience of the SDMs.

3.5. Outcomes

The primary outcome is the proportion of SDMs capable and incapable of giving consent for their loved one to take part in a critical care research study. Secondary outcome measures concern how SDMs' capacity to consent for their loved one relates to the complexity of the decision task and other factors (see Chapter 3 for further discussion).

3.6. Main ethical issues

The following section highlights the main ethical aspects taken into account during research planning and after consultation with the ICU research team (Box 1.3).

Box 1.3: Description of ethical issues as included in the IRAS form

Consent. We have no reason to think that the participants in our study are not able to consent for themselves. That being stated, distress about their ill relative added to the potentially burdensome effect of the SDM's task might make participants vulnerable, cognitively and emotionally. For this reason, several measures will be implemented to minimise additional risks or burdens. Firstly, to make sure that valid informed consent is taken, simple and understandable language will be used to communicate with the participant, explaining relevant information in an appropriate way for that person and checking his/her understanding. The participants will be informed about risks and benefits of their participation and will receive information about appropriate support. In order to reduce the burden of making simultaneous decisions (and not influence their decision about the medical research), the SDMs will be asked to consider participating in our study only after they have made a decision about the medical research. Although consideration has been given to timing and location, in our study it will not be practical and appropriate to allow the participants 24 hours to consider whether to take part. It will be important for us to assess the SDM's capacity to consent for their loved one soon after they have been approached by an ICU researcher, since our assessment is concerned with the real decision that has to be made.
Despite our short enrolment window, if necessary and feasible, we will give the opportunity to delay this decision. My studies in informed consent and the (National Institute for Health Research) Good Clinical Practice course should give me enough expertise to seek informed consent. The use of weekly to fortnightly supervision combined with my background in emergency psychology and psychosocial support in critical care will be useful in my work at the ICU.

Risks and benefits. Potential risks and benefits will be properly explained to the participant. We are aware that taking part could cause additional distress and discomfort. For this reason, we will try to minimise potential risks by offering the possibility of support from appropriate services within the hospital. It will be made clear to the participants that the goal of the research is not therapeutic. However, I will provide all participants with a sheet including useful local numbers (e.g., Local support associations). If a participant shows signs of distress, the interview can be stopped or delayed to a more suitable time for them.

Box 3: Description of ethical issues as included in the IRAS form - continued

Confidentiality. All personal data used will be carefully handled and stored. Only necessary personal information will be accessed, with related written consent. Specific consent will be asked for access to the patients' medical data for the admission diagnosis and information on clinical status, in order that we can give an appropriate description of our sample. Consent will also be sought for possible publication of direct but anonymised quotations from responders. Audio recording devices will be used during the interview. After analysis of the data the audio recording will be destroyed and the transcriptions will be kept securely and anonymously for five years at the University of Birmingham. All personal data will be encrypted. Manual files containing personal data will be safely stored under lock and key in a filing cabinet at the University of Birmingham. Personal

4. Reflections on learning

What did I learn during this placement? One of the most important lessons learned is that there are not necessarily definitive right or wrong answers to our several methodological questions, but well-pondered decisions that need to be justified by the researcher. In line with this, I appreciated that the integration of continuous reflection on methodological aspects with good use of supervision is essential to develop a good piece of research; even more considering that sometimes the researcher's work is solitary.

Even though I am at the beginning of this formative adventure, I recognise that I am developing critical thinking, and a sense of responsibility towards participants and the scientific audience, through the methodological and ethical decisions made in relation to the research discussed here.

I also realised that time management is essential, alongside enthusiasm and determination which help in dealing with the frustration generated by bureaucratic issues

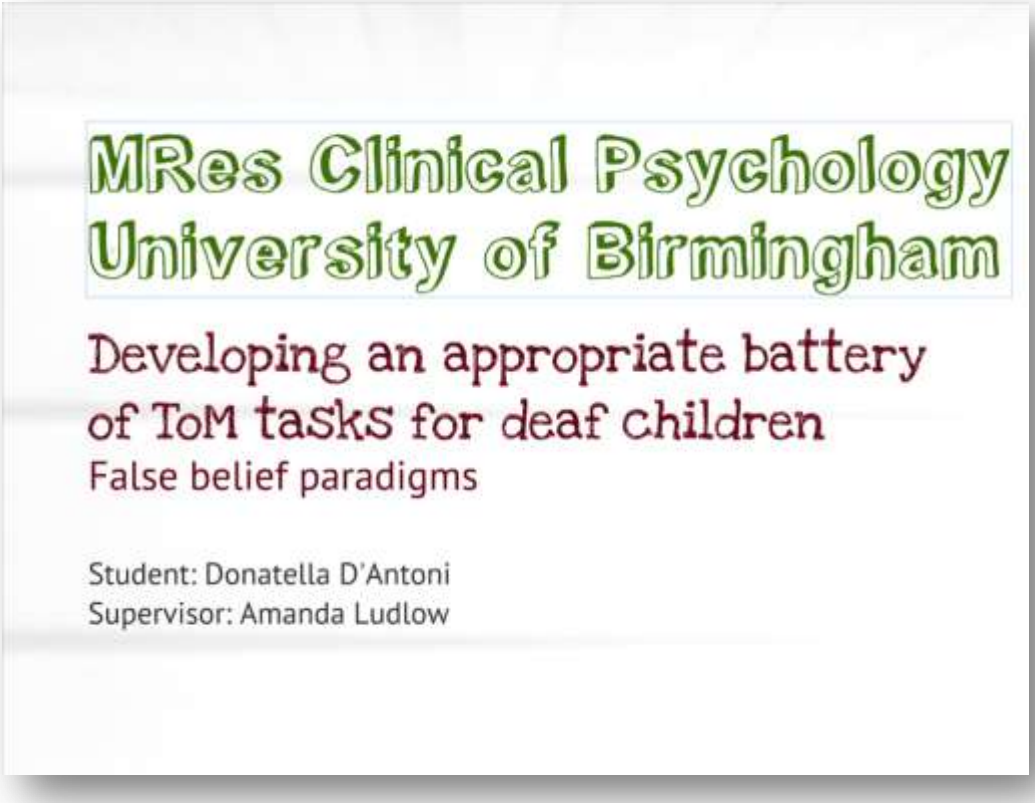
and delays (e.g. in the process of ethical approval). My enthusiasm for this research project made me decide to complete this study (data collection and analysis, and dissemination of results) as part of my main MRes project in the Summer term.

What could I have done differently? I could have tried to take a more distanced perspective on my work, to be able to avoid crystallised thoughts that prevent new insights and to help to generate more appropriate solutions to issues. Discussing my research more widely might have helped me consider issues from a different perspective, and also facilitate the identification of limitations. For this reason, the researcher's network or even a buddy willing to read what you write or how you communicate your research is important.

CHAPTER 2

PLACEMENT 2

Developing an appropriate battery of ToM tasks for deaf children. False belief paradigms

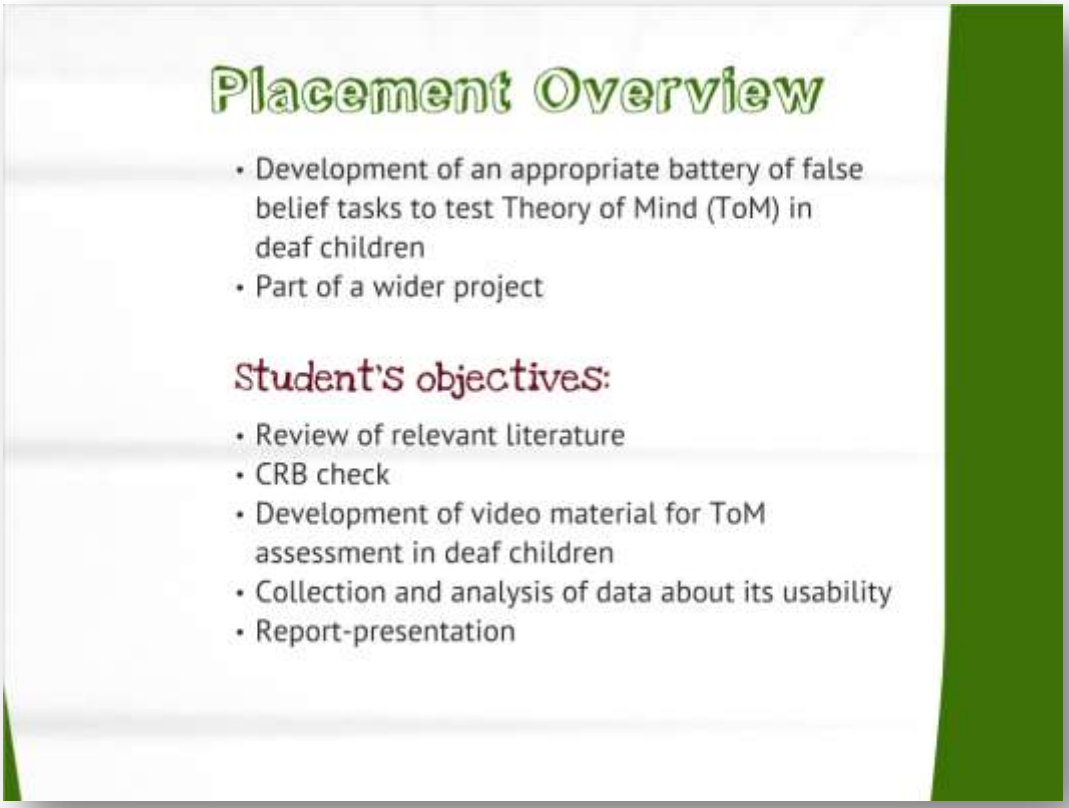


The following report will provide the reader with:

- A brief description of the aims of my second placement and my role in the project;
- A summary of the current literature about Theory of Mind in deaf children and, in particular, False Belief tasks paradigm.
- A discussion of the rationale of the study, method, preliminary results, conclusions and implications.
- A reflection on learning.

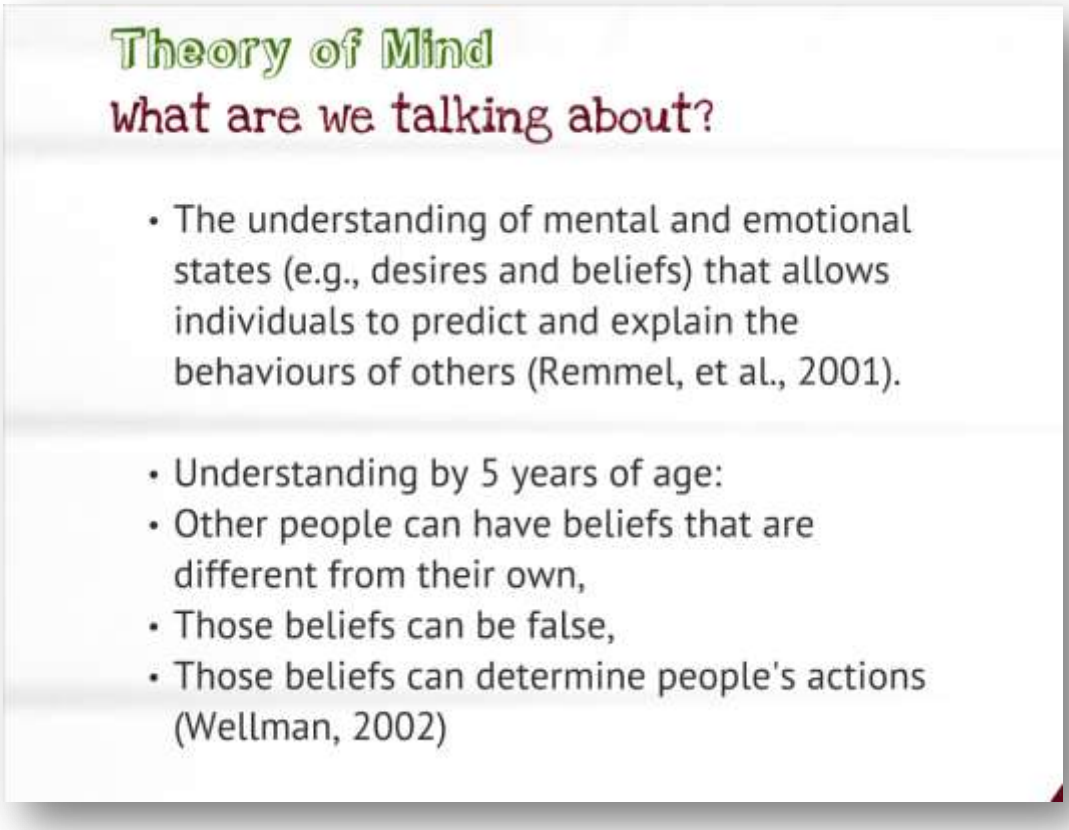
In this chapter are presented the slides from my oral *Prezi* presentation of my second placement. The slides are also accompanied by a further commentary (reported below each slide) to help the reader grasp the tone of the presentation.

1. Placement overview



The aim of this project was to develop an appropriate battery of Theory of Mind (ToM) tasks for deaf children. My role involved undertaking a literature review and further developing a new battery of false belief (FB) tasks that could overcome the several methodological limitations currently existing in assessing ToM in deaf children. I also collected and analysed data about the usability of this new battery. This project is part of a wider project, led by Dr Ludlow and focused on assessing ToM in deaf children using this new tool.

2. Theory of Mind



ToM refers to the understanding of mental and emotional states (desires, beliefs, intentions) which allows individuals to predict and explain the behaviours of others (Remmel, et al., 2001). ToM seems to enable children to make inferences, use deception, develop social and conversational competence, use sophisticate joint play and interpersonal sensitivity (Watson et al., 1999; Cutting & Dunn, 1999; Jenkins & Astington, 2000). Research shows that by the age of 5, children are able to understand that others can have beliefs that are different from their own, that those beliefs can be false, and that those false beliefs can determine a person's behaviours (Wellman, 2002). This ability to attribute FB is considered a milestone in ToM development.

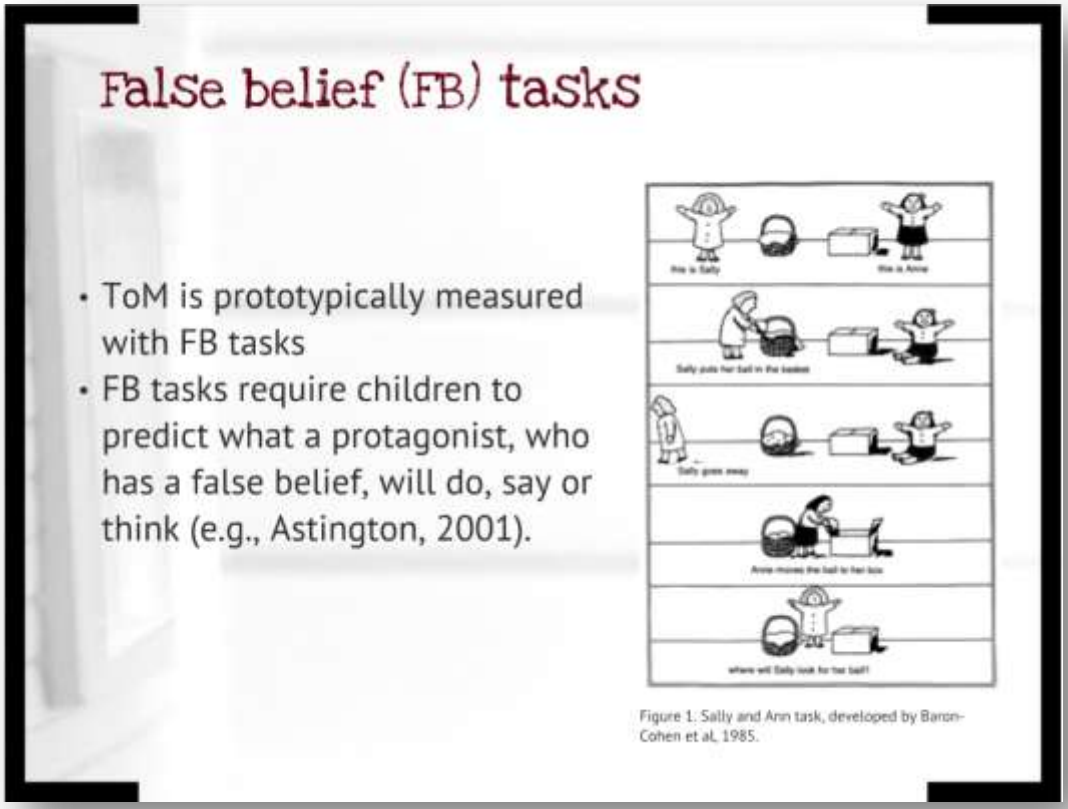


Figure 1. Sally and Ann task, developed by Baron-Cohen et al, 1985.

Numerous FB paradigms have been developed, which require children to predict what a protagonist, who has a false belief, will do, say or think (e.g., Astington, 2001).

For instance, in the 'Sally-Ann' task (Baron-Cohen et al., 1985) version of the original 'FB task' proposed by Wimmer & Perner in 1983) a child is presented with the story of Sally, who has a basket, and Ann, who has a box. Sally hides a marble in her basket and leaves the scene. While she is not there, Ann moves the marble and puts it into her box. When Sally returns, the child is asked: 'Where will Sally look for the marble?' followed by two control questions: 'Where is the marble now?', 'Where did Sally put the marble in the beginning?' To pass the task, the child has to correctly answer all the questions.

3. Theory of Mind in deaf children

Research has concluded that oral and late-signing deaf children of hearing parents show serious delays in their development of ToM, and in particular in FB tests, compared to typically developing children and to deaf children of deaf parents (e.g., de Villiers & de Villiers, 2000; Lundy, 2002). In contrast with the latter two groups, deaf children of hearing parents often experience a delayed access to a shared language with the environment, due to late identification of the hearing problem and poor acoustic and linguistic input (Peters, et al., 2009). It has been hypothesised that deficits in ToM in deaf children could be partially explained with language delays (de Villiers &de Villiers, 2000; Jackson, 2001), and limited

exposure to 'rich mental state discourse' with their mothers (Moeller & Schick, 2006) and the environment (Peterson & Siegal, 2000).

4. Why a new battery of tests?

Methodological considerations

It could be argued that the previous findings, showing a delay in ToM in deaf children, may simply reflect a lack of understanding of what is being asked of them due to the verbal nature of FB tasks (Bloom & German, 2000) and other methodological issues (Schick et al., 2007). In particular, these findings could be the result of the use of less-than fluent examiners; and even when the interpreter is fluent, a loss of information is likely. Moreover,

these tasks require the child to watch simultaneously the interpreter and the material presented, requiring visual coordination (Schick et al., 2007).

In addition, to date the majority of research has presented FB tasks in either Spoken English (SE) or British Sign Language (BSL), not taking into appropriate consideration the fact that many deaf children may struggle to learn the complex BSL and prefer communicate in Sign Assisted English (SAE), i.e., an easier manually coded language. To overcome these methodological limitations and give a fair representation of deaf children's overall ability to reason about other people's mind and mistaken beliefs, we present here a series of FB tasks in three different versions SE, BSL and SAE. In so doing we are developing tasks that are

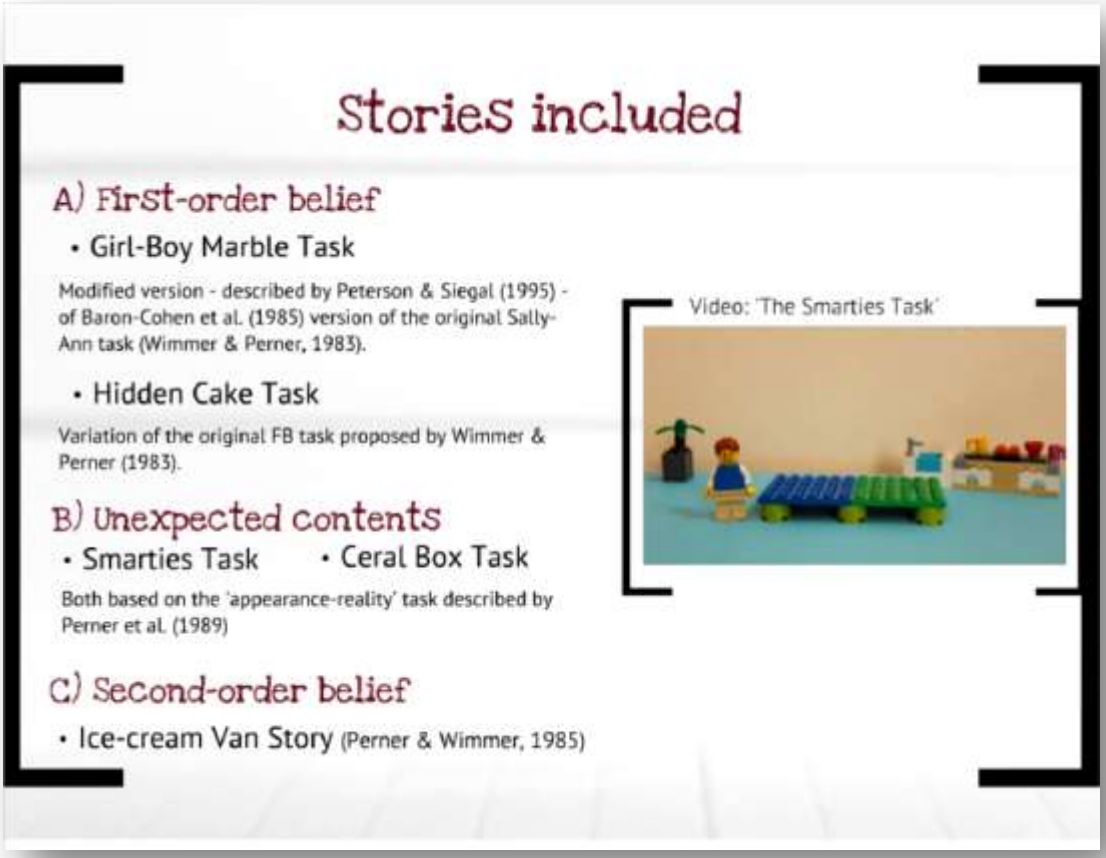
appropriately matched to deaf children's preferred language of communication, reducing the likelihood of lack of understanding.

5. Method

The battery of FB tasks

The battery is designed to test FB reasoning in younger and older children, and includes two standardised first-order FB tasks (see next slide), where the protagonist in the story has beliefs about situations that are different from the beliefs of the participant; two unexpected contents tasks, where misleading containers are used; and a second-order task, which involves a protagonist who mistakenly believes that another character believes

something (Perner & Wimmer, 1985). See Appendix 4 for the description and scripts of the stories.

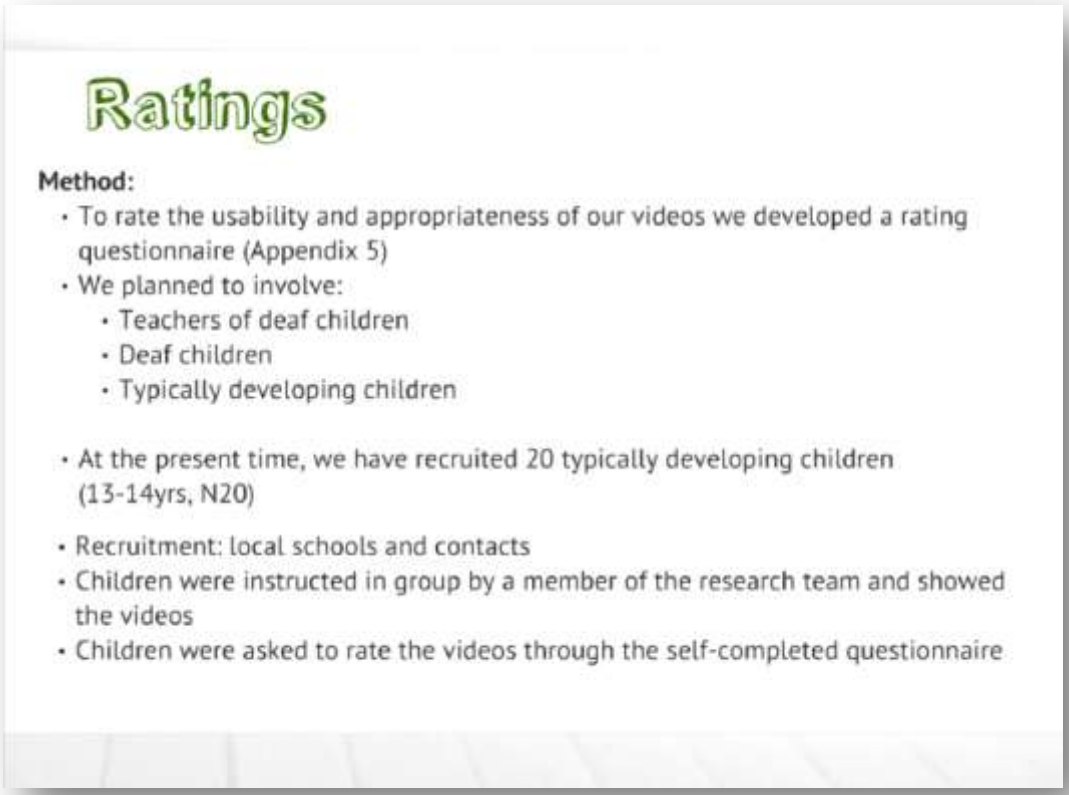


The tasks were designed to be accessible for deaf children, using a simple vocabulary, a storyline easy to comprehend and video animations to assure visual salience. The stories are presented in video format. The child is asked to watch the videos which show a narrator giving instructions and presenting the animated stories (the instructions and animations are not simultaneously presented). The narrator is a deaf person who is native signer proficient in BSL, SAE and SE. These measures make sure that both deaf children and their controls can be presented the tasks in the language they communicate with, ensuring their

comprehension. To pass the tasks the child needs to answer correctly both the test and control questions.

The children's responses are recorded in written format by the examiner.

6. Ratings



To rate the usability and appropriateness of our video material we developed a rating questionnaire (Appendix 5) containing 5-point Likert items and a few open questions. The questionnaire investigated aspects such as clarity of the storyline, the instructions and the animated story, checking also the understanding of the story with the use of probe questions. We planned to recruit, from local schools and contacts, deaf children, typically

developing children and teachers of deaf children to rate the videos. However, data collection is not complete and only preliminary results are available. At the present time, we have recruited 20 typically developing children of a local mainstream school (the control group, age 13-14 yrs). Children were instructed in group by a member of the research team who explained to them how to rate the videos using our rating questionnaire. They were showed the oral version of the videos containing the battery of FB tasks, and were asked to fill in the self-completion questionnaire on their own; however, it is possible that they influenced each in the completion as they were sitting close to one another. For technical reasons the Hidden Cake task was not presented to the children.

Informed consent was obtained.

Other schools have been contacted to rate the BSL and SAE versions of the videos.

7. Preliminary results

The slides below (pp. 34-35) show our preliminary results for all the four tasks.

1. **Girl-Boy task:** twenty children answered the questions related to this first task. The majority of them (*n*=13, 65%) reported that the storyline was not clear, and that the instructions presented were not understandable (*n*=8, 40%). However, 90% (*n*=18) of the responders passed the task, showing some evidence of at least a basic understanding. The animated story was rated as clear (*n*=12, 60%).

2. **Ice-cream task:** the majority of the twenty respondents (*n*=14, 70%) affirmed that the instructions in this task were not easy to understand, and the overall task not easy to

complete (*n*=18, 80%). Only half of them answered the probe questions, and 70% (*n*=14) of the responders answered incorrectly to two questions out of five.

3. Cereal Box task: Fourteen children completed the items concerning this task. Fifty per cent of them (*n*=7) rated the animated story as clear; 29% (*n*=4) gave a neutral answer. The opinion was divided between those who reported that the information received to complete the task was insufficient (42%) and those who thought the opposite (37%). All respondents passed the task.

4. Smarties task: Fifteen children rated this task. The majority of them (*n*=8, 54%) reported that the animated story was clear. Those who answered the probe questions (77%) also passed the task.

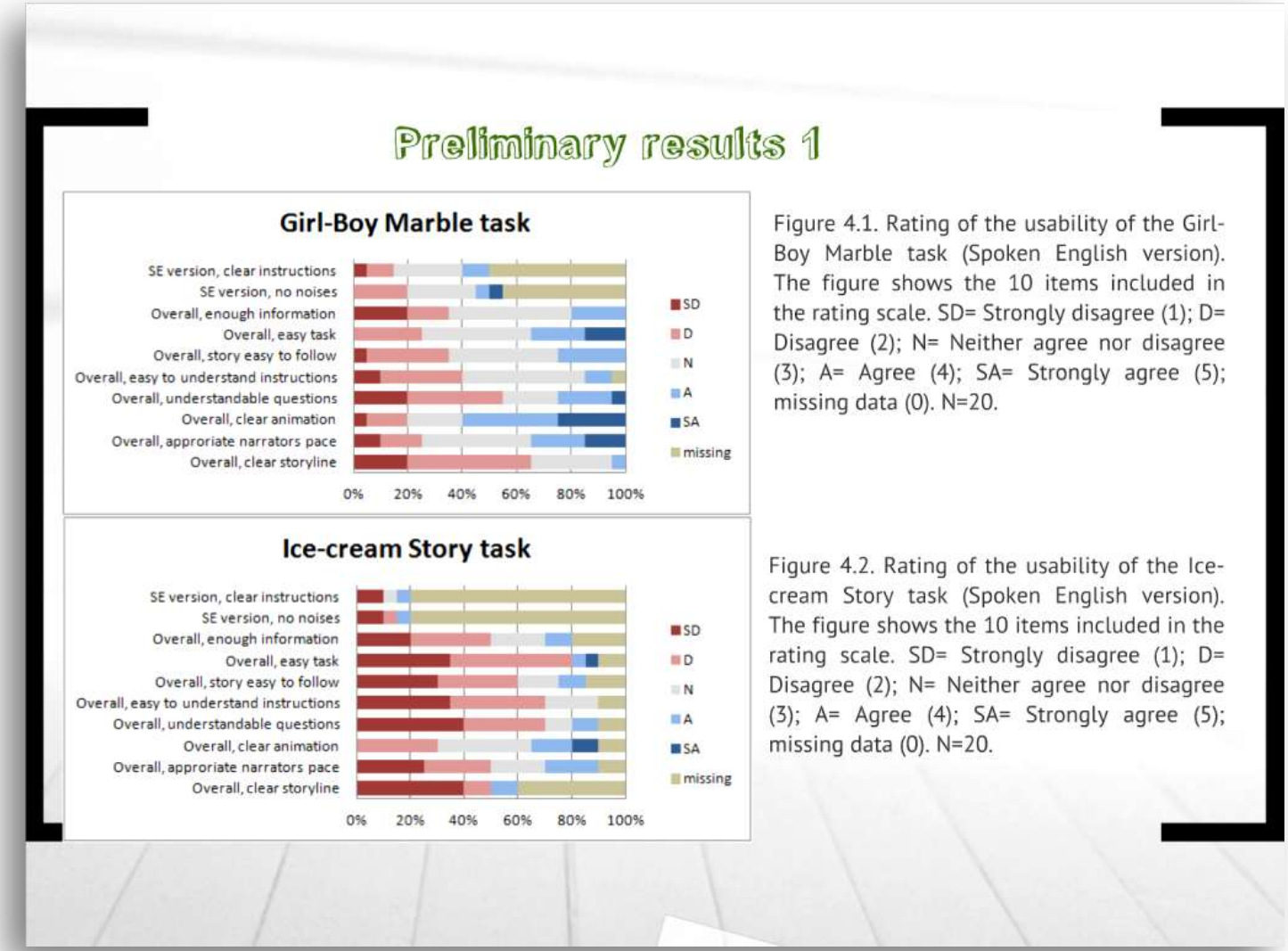


Figure 4.1. Rating of the usability of the Girl-Boy Marble task (Spoken English version). The figure shows the 10 items included in the rating scale. SD= Strongly disagree (1); D= Disagree (2); N= Neither agree nor disagree (3); A= Agree (4); SA= Strongly agree (5); missing data (0). N=20.

Figure 4.2. Rating of the usability of the Ice-cream Story task (Spoken English version). The figure shows the 10 items included in the rating scale. SD= Strongly disagree (1); D= Disagree (2); N= Neither agree nor disagree (3); A= Agree (4); SA= Strongly agree (5); missing data (0). N=20.

Preliminary results 2

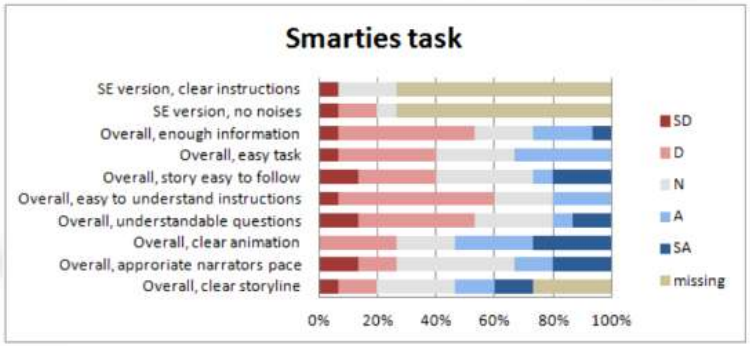


Figure 4.3. Rating of the usability of the Smarties task (Spoken English version). The figure shows the 10 items included in the rating scale. SD= Strongly disagree (1); D= Disagree (2); N= Neither agree nor disagree (3); A= Agree (4); SA= Strongly agree (5); missing data (0). N=14. Six questionnaires were substantially incomplete or difficult to interpret in the section related to this task.

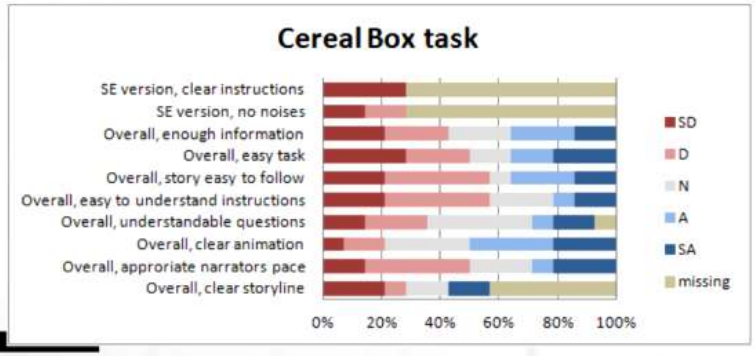
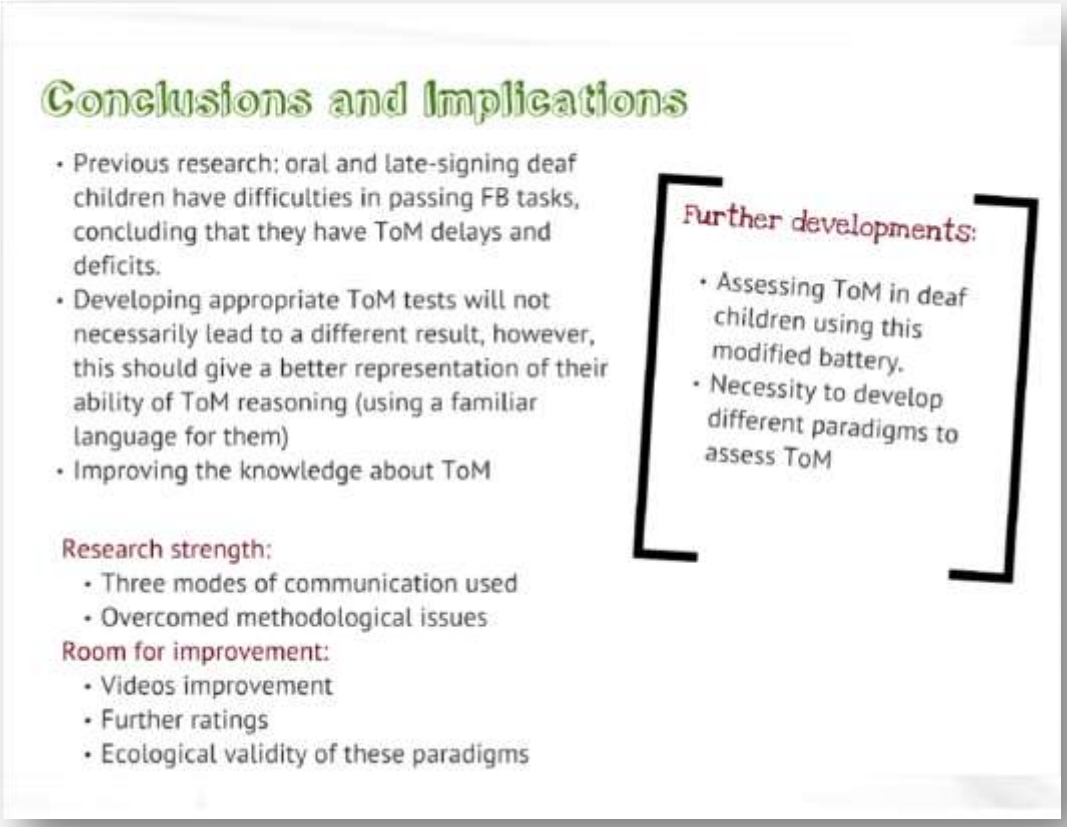


Figure 4.4. Rating of the usability of the Girl-Boy Marble task (Spoken English version). The figure shows the 10 items included in the rating scale. SD= Strongly disagree (1); D= Disagree (2); N= Neither agree nor disagree (3); A= Agree (4); SA= Strongly agree (5); missing data (0). N=15. Five questionnaires were substantially incomplete or difficult to interpret in the section related to this task.

8. Conclusions and implications



Based on the fact that deaf children of hearing parents struggle in passing FB tasks, previous studies have concluded that these children have delays in developing ToM. However, it is not clear whether these findings simply reflect a lack of understanding of the FB tasks due to their complex verbal nature and methodological issues such as the use of either spoken English or BSL, without considering that many deaf children may prefer communicate in SAE. The aim of this study was to provide more appropriate measures of deaf children's FB reasoning, improving existing ToM measures. Our battery of FB tasks is designed to test deaf children using the language they find more comfortable (including SE, BSL, and SAE version). Due to recruitment delays, it was possible to study the usability of the

oral version only: preliminary results showed that although the instructions for task completion were rated as not understandable by the majority of hearing children, the animated stories were rated clear and all the tasks, except for the second-order belief task, were passed. This means that even when children found that the instructions were unclear, they showed to have a sufficient understanding to pass the tasks. The oral instructions were provided by our deaf interpreter and this may have affected the participants' understanding.

The majority of children failed to pass the second-order belief task. This may be explained by the complexity of these tasks: as Bloom and German (2000) clarify, a good performance on FB tasks implies the ability to use multiple representations and is connected with how children understand the questions presented. Alternatively, this may have resulted from the children's lack of understanding of what they have been asked to do, due to the artificial nature of FB tasks. The fact that the stories had little relevance to the participants was confirmed by their comments about the abstract and unfamiliar feature of the tasks. It is also possible that, if the children had been provided with the scripts, their understanding of the tasks might have been improved. It may be worthwhile to test this hypothesis in next stages of the study.

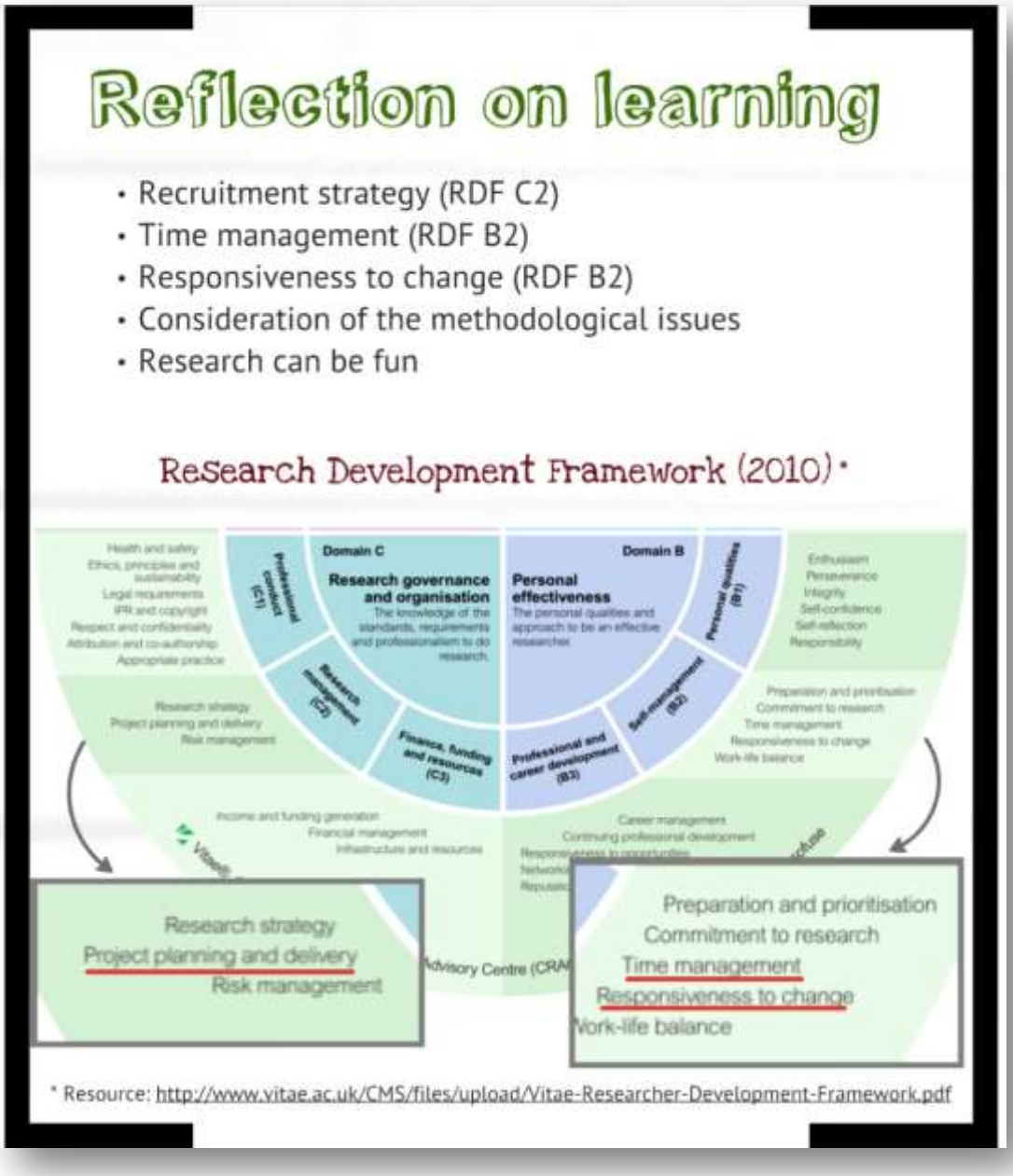
High rates of missing data, in particular for the last three tasks, may be due to respondents' lack of motivation, fatigue, and lack of understanding of how to rate the videos. We acknowledge that collecting data in group rather than on an individual basis may be problematic: given that children may have different levels of understanding, they may have benefitted from being shown the tasks at a different pace.

Our study has several limitations. Any conclusions should be regarded with caution, since our sample size was small and only composed by hearing children, and our results may be biased due to the amount of missing data. To draw solid conclusions about the usability of the battery, ratings about the BSL and SAE versions are needed.

Our findings offer important indications: the second-order belief task was perceived by the hearing participants as an unfamiliar, abstract, and lacking sense activity. We suggested that this may explain the participants' difficulty in passing the task. It is important to consider this, as deaf children may also show similar issues: as Marschark et al. (2000) concluded, deaf children who have ToM, as showed by their use of FB explanations in their spontaneous spoken narrative, have difficulty in demonstrating it in standardised FB paradigms due to syntactically complex questions and fairy artificial stories used.

Implications: our battery of FB tasks, once appropriately improved, could support the research in FB and overcome methodological issues faced by previous studies. Further research should examine the ecological validity of FB tasks and perhaps consider alternative and less artificial paradigms.

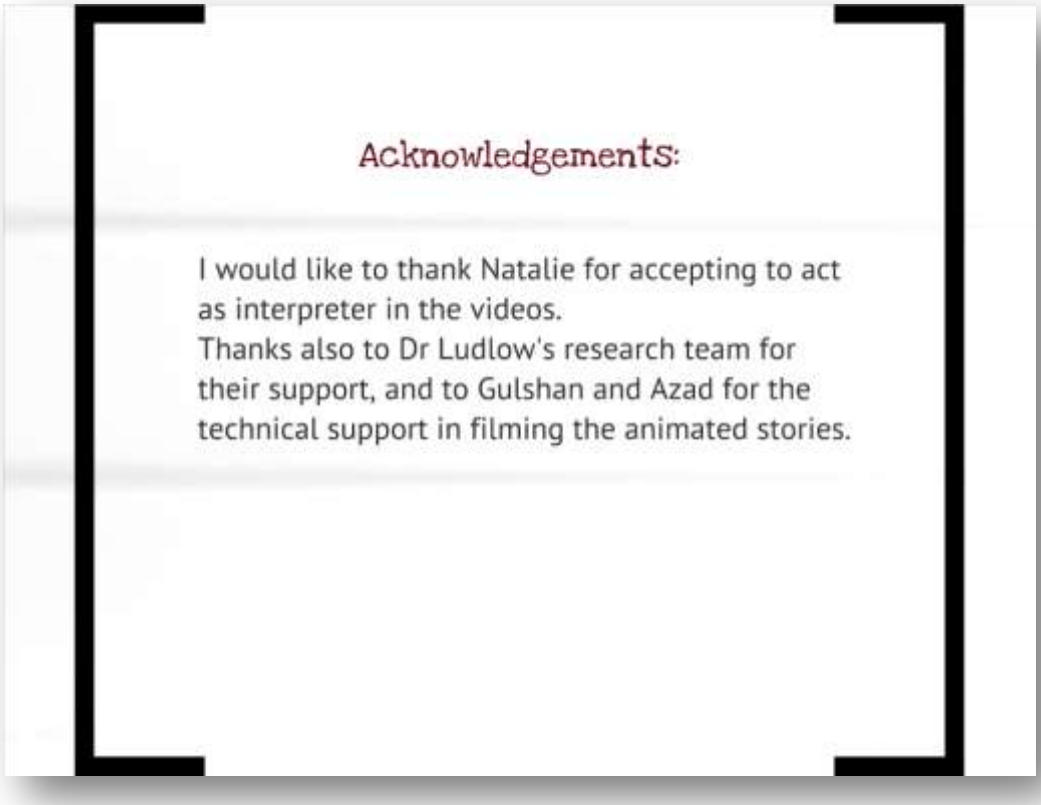
9. Reflections on learning



This placement allowed me to develop and improve some of the personal and professional areas highlighted by the Researcher Development Framework (RDF, 2010).

An important lesson learned is concerned with the application of an effective project management through the setting of research goals, intermediate milestones (RDF Domain C2 Research management), with particular attention to the recruitment strategy. For instance, delays with recruitment can disrupt the timetable of a research project and cause a limited sample size, as happened in this study. Therefore, a more effective research plan, able to take into consideration the possibility of dealing with the unexpected, could have reduced this kind of problem. Furthermore, I believe this project helped me improve two subdomains of the Self-management Domain (RDF B2), which are time management and responsiveness to change. The first refers to the ability to manage your own time effectively to complete research project, in line with your project plan and the necessity of the team work; the latter refers to the ability to adapt to changes, responding flexibly and learning to seek advice and reassurance when necessary, using supervision. Moreover, I developed awareness about the limitations of testing methods and methodological issues involved in assessing ToM in deaf children.


Thank you
for your
attention


Acknowledgements:
I would like to thank Natalie for accepting to act as interpreter in the videos.
Thanks also to Dr Ludlow's research team for their support, and to Gulshan and Azad for the technical support in filming the animated stories.

Below are reported the references used in the slides. For further references see Reference section at the end of the thesis.

References

- Astington, J. (2001). The future of theory of mind research: Understanding motivational states, the role of language and real world consequences. Child Dvelopment, 72, 685-687.
- Baron-Cohen, S., Leslie, A.M., & Frith, U. (1985). Does the autistic child have a theory of mind? Cognition, 21, 37-46.
- Bloom, P. & German, T. (2000). Two reasons to abandon the false belief task as a test of theory of mind. Cognition, 77, B25-B31.
- de Villiers, J.G., de Villiers, P.A. (2000). Linguistic determinism and the understanding of false beliefs. In P. Mitchell & K.J. Riggs (Eds.). Children's reasoning and mind, Hove, UK: Psychology Press, 191-228.
- Marschark, M., Green, V., Hindmarsh, G. & Walker, S. (2000). Understanding theory of mind in children who are deaf. Journal of Child Psychology and Psychiatry, 41, 1067-1073.
- Moeller, MP., & Schick, B. (2006). Relations between maternal input and theory of mind unerstanding in deaf children. Child Development, 77, 751-766.
- Perner, J., Frith, U., Leslie, A., & Leekam, S. (1989). Exploration of the autistic child's theory of mind: Knowledge belief and communication. Child Development, 60 , 689-700.
- Peterson, C.C., Siegal, M. (1995). Deafness, conversation and theory of mind. Journal of Child Psychology and Psychiatry, 36, 459-474.
- Peterson, C.C., Siegal, M. (2000). Insights into theory of mind from deafness and autism. Mind and Language, 15, 123-145.
- Remmel, E., Bettger, J.G., & Weinberg, A.M. (2001). Theory of mind development in deaf children. In M.D. Clark, M. Marschark, & M. Karchmer (Eds.). Context, cognition, and deafness, Washington, DC: Gallaudet University Press, 113-134.
- Russel, P.A., Hosle, J.A., Gray, C.D., Scott, C., Hunter, N., Banks, J.S., & Macaulay, M.C. (1998). The development of theory of mind in deaf children. Journal of Child Psychology and Psychiatry, 39, 903-910.
- Schick, B., de Villiers, P., de Villiers, J., & Hoffmeister, R. (2007). Language and theory of mind: A study of deaf children, Child Development, 78, 376-396.
- Wellman, H.M., (2002). Understanding the psychological world: Developing a theory of mind. In U. Goswami (Ed.). Blackwell handbook of childhood cognitive development, Malden, MA: Blackwell, 167-187.
- Wimmer, H., & Perner, J. (1983). Beliefs about beliefs: representation and the containing function of wrong beliefs in young children's understanding of deception. Cognition, 13, 103-128

CHAPTER 3

PLACEMENT 3

Are substitute decision makers able to consent/assent for their loved one in the context of the Intensive Care Unit? From the research protocol to its implementation

The following report will provide the reader with:

- A brief description of my third placement and my role in the project (section 1);
- A summary of the introduction of the study (extensively reported in Report 1), followed the presentation of methods, results, and conclusions (section 2).
- A reflection on learning (section 3).

The Appendices include our adapted test assessing mental capacity to consent for research and the interview used. Furthermore, extracts from the interviews and other documents showing the methodology used to carry out the qualitative analysis are provided.

1. Placement overview

The aim of the Summer term placement was to implement the research designed during placement 1. The general aim of this mixed methods study was to investigate the next-of-kin's capacity to give valid consent/assent for research on behalf of a critically unwell loved-one and to find out which factors influence their ability to do this (quantitative design). We also tried to find out from those who have been asked to act as SDMs, what their personal experience of doing this was like (qualitative design).

I was responsible for all the phases of the research implementation, analysis and dissemination of results. After an induction week, at the beginning of May 2013, I started collecting data through conducting interviews with next-of-kin of ICU patients. My

supervisors and I aimed to involve 30-40 SDMs over a period of 11 weeks at the ICU of the QEHB. However, the recruitment of only 14 people made it impossible to fulfil our second aim, which had been to analyse the association between factors potentially influencing the SDMs' capacity to consent and their ability to do this (such factors will not be further discussed in the body of the text, but are presented in Appendix 6 as descriptive statistics in Table 3.5). For this reason, this placement report focuses on the primary research question and the first and last of the secondary questions (page 46).

Once data collection was completed, my next objective was to perform the analysis of qualitative data, followed by the analysis of quantitative data. This order was planned to reduce the risk of influencing the qualitative analysis by knowing in advance the participants' performance on the mental capacity test.

During all the stages of the project, research progress was discussed with my supervisors, with particular emphasis on the recruitment process. Problem-solving support was available when necessary. In particular, I had direct support from one of my supervisors at the ICU. Supervision meetings also provided 'space' for discussion of the emotional impact of the placement on me as a research student and person.

My final objective was to write up the present scientific report.

2. The research

ABSTRACT

Background: Because ICU patients are frequently unable to decide for themselves, researchers have turned to substitute decision makers (SDMs) to obtain consent/assent before the patients are enrolled in medical research. However, the complex nature of the information about medical trials combined with the burden inherent in making a surrogate decision may make it hard, if not impossible, for them to make a valid decision (e.g., Burns et al., 2009). Our mixed methods study investigated the proportion of SDMs capable of providing valid consent/assent for research involving ICU patients. We also explored whether their capacity to do this was influenced by the complexity of the research, and what their personal experience of doing this was like (the latter two representing secondary research questions).

Methods: Using our adapted version of the University of California, San Diego Brief Assessment of Capacity to Consent (Jeste et al. 2007), 14 SDMs were assessed for their capacity to provide valid consent for the specific research study they were asked to consider by an ICU research nurse between May and July 2013. The participants were also interviewed to investigate the main aspects involved in the decision-making process. Both these investigations were addressed through a quantitative design. A second part of the interview explored how the participants made sense of their experience as SDMs (for this part a qualitative design was used).

Results: We found that 21% of the SDMs were not capable of providing valid consent for the specific research study about which they were approached. In particular, they did not understand the aim of the medical research nor appreciate the risks and benefits involved. Using thematic analysis, six themes emerged.

Conclusions: This study shows that approximately 20% of the SDMs assessed provided invalid consent for research. Our results lead to the recommendation for clinical researchers to check carefully whether potential participants are capable of providing valid consent.

Keywords: Surrogate decision makers; Informed consent; Research; Research ethics; Critical care; Intensive Care; Capacity to consent; Mental capacity

2.1. Summary introduction

Critically ill patients are often unable to make decisions for themselves as a consequence of either the severity of their illness or received treatment. In this situation, it is a legal requirement for someone close to the patient to be consulted before they are enrolled in

medical research (Medicines for Human Use (Clinical Trials) Regulations 2004; Mental Capacity Act 2005). However, previous literature suggests that this surrogate decision task causes additional stress to the burden already experienced by the next-of-kin (Pochard et al., 2001). In addition it may be difficult under these circumstances to understand what the research is about, making it hard, if not impossible, to make a valid decision (Azoulay et al., 2000; Burns, Zubrinich, Marshall, & Cook, 2009; Ciroldi et al., 2007).

In this study we investigated SDMs' capacity to give valid consent/assent for research on behalf of a critically unwell loved-one and explored whether their capacity to do this was influenced by the complexity of the research (interventional research being categorised as high complexity and observational research as low complexity). We also explored the personal experience of SDMs who had been asked to consider providing surrogate consent/assent.

Principal research question:

- Are SDMs in the context of critical care research able to give valid consent/assent for their loved ones? (quantitaive design)

Secondary research questions:

1. Is the SDM's capacity to consent for their loved one influenced by whether the study is observational or interventional? (quantitative design)
2. What factors affect their capacity at the point when SDMs are asked to make the decision for their loved one? (Question not addressed due to recruitment issues).
3. What is it like for SDMs to be involved in the decision-making process for critical care research? (qualitative design)

The implications of this study are important in terms of understanding whether there is a need to find better ways of protecting patients and their next-of-kin, and fostering valid consent, whilst permitting research.

2.2. Methodology

This section outlines the research paradigm and methods used (see also Box 3.1).

2.3. PROCEDURE

Induction week

Joining the research team, as an observer, I learned about the way the approach about research was carried out by researchers or research nurses in the context of the ICU and the information given to the SDMs. I also piloted the interview with the family of an ICU patient. As a result, some questions in the interview were rephrased and one question eliminated due to being repetitive (Appendix 7).

Participant recruitment

Every day the ICU research team was contacted to check whether they were planning to approach an SDM in order to seek consent/assent about their loved one's participation in medical research. Once a potential participant who met the eligibility criteria had been identified, a first contact with the person was established. I was introduced to the SDM by a research nurse in tandem with their own first approach. The SDM was then asked for permission to allow me to observe their interaction. At the end of their discussion, after the SDMs had made a decision about their relative's enrolment in research, I checked whether

they were willing to know more about my study. If so, I provided them with an information leaflet and gave more details (see Appendix 2). Where other family members were joining the discussion with the research nurse and were taking part in the decision about medical research, I took their permission to also include them in the qualitative aspects of the study; however, as each patient only had one formal SDM, these other family members were not asked to fill out the questionnaires, since they did not have to take responsibility for giving proxy consent. Their account was only considered in the qualitative analysis.

Given that the topic of the research interviews (mental capacity to consent for research) is time-sensitive (see Box 1.1 Chapter 1), potential participants needed to be contacted as soon as possible after the SDM had been approached for ICU research. To provide the best compromise between the need for timely interviews and the need not to overload the SDMs, my supervisors and I decided to allow 24 hours to consider participation. The interviews were carried out after taking formal written consent.

Box 3.1: Methods summary

Inclusion criteria for participants:
- Patient meeting ICU research eligibility criteria
- Patient with unplanned ICU admission
- Patient with identifiable SDM

Exclusion criteria
- Participant under 18 years old
- Patient unlikely to survive 24 hours
- Participant not available to be interviewed within 24 hours from consent/refusal of the medical research.

For details about the instruments used see instrument section Chapter 1.

Data recorded for each SDM
SDM: Age, gender, ethnicity, relationship to the patient, education, type of medical study that the participant had been asked to consider (observational or interventional).
Patient: Age, length of stay at time of interview, APACHE II score (the Acute Physiology and Chronic Health Evaluation is a severity of disease score and mortality estimation tool).

Characteristics of the ICU
The ICU at the QEHB has 100 beds divided between four adjacently situated areas on level 2 of the hospital. Each area is staffed by a specific group of nurses and ancillary staff, whereas the medical staff rotate between the areas. The nurse: patient ratio varies from 1:1 for the sickest or most disturbed patients, to 1:2 for others.

Conducting the interview

The interviews were conducted face-to-face in consultation or seminar rooms, where privacy and confidentiality was ensured. One interview was conducted in a waiting corridor due to lack of availability of consultation rooms. Considering the delicate time experienced by the next-of-kin, in order not to aggravate their distress and allow them to go back to their loved ones' bed as soon as possible, the length of the interviews was maintained reasonably limited. The interviews lasted 18 minutes on average.

A few participants spontaneously reported a positive impact of our discussion, as they had 'space' and time to give voice to emotions and organise thoughts. The interviews (both stage 1 and 2, see below) were audio recorded. Later, reflections on contextual aspects, regarding the consent process for medical research and the interview itself, were annotated in a research diary. The interview comprised the following stages:

Stage 1. Using our adapted version of the UBACC (Appendix 7), the participants were asked to answer ten questions concerning the main aspects involved in medical research for which they were approached by an ICU research nurse, within the last 24 hours. This test was used to assess the SDMs' capacity to provide valid consent/assent for that specific research study. The questions were rephrased when necessary, and probes were used when the participants' answer was uncertain or unclear, as suggested by Jeste et al. (2007). This stage lasted on average 5 minutes. The participants' answers were not scored up until the conclusion of the overall data collection (see Box 3.2: Ethical considerations).

Box 3.2: Ethical considerations

The UBACC was scored at the end of data collection and once the qualitative analysis was concluded. This was to keep the researcher blind to the outcome of the participant's performance on the mental capacity test, as this would have influenced the researcher's qualitative analysis.

This meant also that we did not find out whether an SDM was judged by the UBACC as being unable to provide valid consent for the research they had already consented to until some while after their interview. Whilst this issue represented a dilemma for our research team, we concluded that it would not have been appropriate to interfere with the judgements of capacity and informed consent made by the ICU research team, given that their own procedures already had ethical approval, and the research nurses were deemed to have adequate training to take consent.

If during the interview I had doubts about the SDMs' understanding of the medical research study for which they had consented as SDM, I advised them to get in touch with the research team and seek further clarification. In the long-run, we aimed to use our overall results to learn about how processes could be improved. The Ethics Committee gave us a positive opinion.

Stage 2. This second part involved conducting an interview which gathered both quantitative and qualitative data. The first part was designed to investigate the main aspects potentially implicated in the SDMs' capacity to consent/assent for research. Using a quantitative design, the answers were presented in either multiple-choice format or in five point Likert scales. The second part of the interview was designed to inform a qualitative investigation; open-ended questions were included to allow the participants to share their personal experience of being consulted about their loved one's participation in research. This latter part was design to provide further understanding of this relatively unexplored area.

2.3.1. METHODS OF ANALYSIS

UBACC (adapted version): scoring

SDMs who participated had been approached to take part in one of a range of medical research studies. The questionnaire was scored against criteria specific to the research protocol about which each SDM had been approached, as advised by Jeste et al (2007). To create the list of correct answers we consulted the information leaflets for each study, and the verbal information given to the SDM in person by the research nurse who approached them. Main aspects of the verbal information were noted by me, such as randomisation, benefits and risks of the study and the possibility of no benefit arising. The range for the score for each question was 0-2. Two was given for a full and correct answer, 0.5/1 was given when even after further probing the answer was still partial. Clearly wrong aswers were scored 0. Jeste et al. (2007) suggested that a cut off score of 14.5 out of a maximum of 20 indicates capacity. In other words, any participant showing a total score of >14.5 was deemed able to provide valid consent. This cut-off score was selected by Jeste et al. (2007) based on the receiver operating characteristic curve analysis of the UBACC total score against an experienced psychiatrist's judgment about capacity to consent of a sample of middle-aged and older outpatients with schizophrenia or schizoaffective disorder and healthy comparison participants, using a research simulated protocol. With the cut score of 14.5, the UBACC showed a sensitivity of 89% and specificity of 100%.

Data preparation

Each interview was transcribed and checked to ensure accuracy. Each participant was given a reference number to guarantee anonymity.

All identifiable names of participants and research nurses were replaced by fictitious names.

Quantitative analysis: analytic procedure

The intended sample size was based on information provided by ICU key informants about annual average approaches rates. It was reported that research nurses approached 3 eligible patients or SDMs each day with a view to recruiting them into studies. We therefore conservatively anticipated recuiting 30-40 people in total (3 to 4 participants per week over an 11 weeks period). Unfortunately this expectation was not met due to unusually low recruitment rates to medical research in Critical Care during the period of my study. It proved possible to recruit only 14 SDMs during the 11 week time period available. As a result, it was not possible to fully perform the intended quantitative analysis to identify links between capacity and factors that might be associated with it. However, descriptive statistics were run to describe the proportion of SDMs with capacity to consent for their loved one, and illustrate the characteristics of the decision-making process. I also looked at the association between capacity to give consent and complexity of the study (observational/interventional) for which the SDM was approached: for this comparison the non-parametric Kruskal Wallis Test was used, and a p value of <0.05 was considered to be statistically significant. However, any conclusions from this need to be treated with great caution since our small sample size, and the distribution across cells, affected the power of the Chi-Square tests to detect any statistically significant difference between the two groups of study. In particular, in this case the data was not suitable for use of Yates' correction (see Howell, 2002).

In the results section below, categorical variables are expressed as frequencies, while continuous variables as mean and standard deviation. IBM SPSS Statistics version 21 was used (IBM Corp., 2012).

Qualitative data: analytic procedure

The qualitative analysis involved transcripts of the whole interview, comprising free comments and remarks made by the participants, as they expanded on the questions included in the adapted UBACC, as well as their responses to open-ended questions. Three of the 14 interviews involved two family members, i.e. the SDM officially identified by an ICU research nurse and another member of the family (see 'Participant recruitment' p. 47). Therefore, the qualitative analysis, unlike the quantitative analysis, involved three additional next-of-kin. The aim of this approach was to try to capture a richer and more realistic account of the experience of substitute consent in ICU from all family members who had been in the discussion about the substitute consent with the research nurse.

The data were analysed using inductive (data-driven) thematic analysis and, in particular, the six phases described by Braun and Clarke (2006). Therefore, after familiarising with the data (phase 1) by transcribing and reading the interviews, initial codes were identified in a systematic line-by-line analysis (phase 2). An 'empathic' approach was used for the interpretation of the data, characterised by the attempt to engage with the interview transcripts while trying to keep out theoretical concepts from the outside and getting 'as close to the research participants' experience as possible' (Willig, 2013). Using also contextual data recorded in my research diary, the initial codes were further developed to potential themes, including collated codes (phase 3). The codes were informed by the focus

of the research question, and the researchers' theoretical framework (phenomenological and contextual constructionist). These potential themes were then reviewed (phase 4) and wrote down on post-it notes to let the data assume a systematic and consistent structure, as suggested by Braun and Clarke (2006). Through a continuous review of superordinate themes and subthemes (phase 5), the emerging story was further refined. In writing-up the results (phase 6) the data were related back to the research question (see Appendix 8).

Particular attention was paid to personal and epistemological reflexivity issues thanks to weekly face-to-face or Skype supervision meetings. Measures were also taken to ensure credibility of the study. Triangulation of researchers was assured by the use of supervision meetings, with the clinical supervisor, to consider the consistency, plausibility and completeness of my analysis and also its roots in the data; moreover, to explore the salience of the main themes, these were presented to all the research group as it includes consultants working daily in the ICU and familiar with the consent process for critical care research. In the results section, the use of direct quotations of both participants and some example of the researcher's questions and probes serve to show how the themes are grounded in the participants' accounts.

Epistemological position

Thematic analysis is a method compatible with different epistemological positions and does not predetermine theoretical and epistemological framework, which is described as 'theoretical flexibility' by Willig (2013). In this research, a phenomenological framework was used, focusing on the meanings the participants attributed to their experience as SDMs in

the ICU. In line with a phenomenological orientation, the researcher and subject of research are both considered beings interpreting and acting within networks of cultural meaning (Giorgi, 1995). Therefore, it is acknowledged that there cannot be such a thing as 'relationshipfree' interview (King, 2004, p. 11), as the researcher inevitably influences data collection. In addition, themes never just 'emerge' from data, rather it is the researcher who actively constructs and interprets them (Braun & Clarke, 2006). Using Madill at al.'s (2000) classification of epistemologies, the thematic analysis used in this study could be located somewhere close to the contextual constructionist position, in the continuum between naïve realist and radical relativist positions. This approach aims to provide a rich description of the phenomenon studied, recognising as well that every phenomenon observed is context-dependent.

Ethics

Ethical approval was granted by the NRES Committee West Midlands-Solihull (see Appendix 3).

2.4. Results

2.4.1. QUANTITATIVE DATA

In the period of 11 weeks from the 13th May 2013 out of a total of 25 SDMs initially identified by the ICU research team, 18 met eligibility criteria for the interview and 14 consented to participate. The reason for ineligibility was that for 7 SDMs the procedure of approach for ICU research was suspended because either the SDM was not available (n=3),

the patient regained capacity before the SDM was consulted (n=2) or the eligibility criteria for ICU research were not met anymore (n=2). Of the 18 SDMs initially meeting the eligibility criteria for the present study, 2 could not be contacted by me within 24 hours after their consent for medical research. Two SDMs refused consent: in one case, the SDM was too worried about her husband's condition; in the other, the SDM (who also refused consent for medical research) explained to be too emotional to be willing to share her personal experience.

Patient and SDM characteristics

The characteristics of the sample are shown in Table 3.1. Fourteen SDM were identified as eligible and interviewed. Interviews were conducted a median of 24 hours after the SDM's consent for medical research, and a median of 2 days after patient admission to the ICU. All the SDMs interviewed had consented to medical research. Three of the 14 interviews involved a further family member, who had contributed to the decision about medical research, in addition to the SDM. However, given that for each ICU patient only one person was officially identified as an SDM by the ICU research nurse, the questionnaire-based assessment of capacity and the quantitative analyses exclude the additional three family members and refer only to the SDMs.

One interview involved an interpreter as the participant preferred to be interviewed in her first language, rather than English.

Table 3.1 Patient and SDM characteristics

Patient characteristics*	
Age *M* (*SD*)	57.5 (18.3)
Apache II Score *M* (*SD*)**	14.5 (8-22)
Male gender *n* (%)	12 (85.7)
ICU length of stay at time of interview in days: *M* (IQR)	2.5 (2-5)
SDM characteristics***	55.1 (16.5)
Age *M* (*SD*)	3 (21.4)
Male gender *n* (%)	
Relationship to patient *n* (%)	3 (21.4)
Child	
Spouse	6 (42.9)
Parent	2 (14.3)
Sibling	3 (21.4)
Highest level of education *n* (%)	
Secondary school-to GCSE level/A level	7 (50%)
Graduate degree	5 (35.7)
No qualifications	1 (7.1)
Other: cannot remember	1 (7.1)
Ethnic Background *n* (%)	
African	1 (7.1)
Caucasian	13 (92.9)
English as first language *n* (%)	12 (85.7)

*N= 14.

** APACHE II (see Box 3.1). For instance, a score of 10-14 is associated with a death rate of 15%, a score of 15-19 with 25% death rate and 20-24 with 40% mortality rate. See Knaus, Draper, Wagner & Zimmerman (1985).

***N=14. These characteristics refer to the person who was officially identified as an SDM by an ICU research nurse, even though other relatives may have been involved in the decision.

SDMs' capacity to consent for research

Some SDMs (*n*=7) had been asked to consider their loved one's enrolment in an observational study, while others (*n*=7) had been asked to consider an interventional study. For those (*n*=2) asked to make decisions about more than one study at the same time, the test assessing capacity to consent focused only on the specific study for which the SDM was pointed out to me by the ICU research team and for which I consequently observed the

approach. In total 6 critical care studies were considered (3 interventional, and 3 observational). Table 3.2 shows the SDMs' performance on the modified UBACC. Three SDMs (21%) scored below the cut-off of 14.5, which is used to judge capacity, giving rise to important doubts about their understanding of the research they had consented to, and therefore also the overall validity of their consent. Two of these SDMs consented to an interventional study, and the other to an observational study. There was no significant difference between the distribution of scores across the observational (low-risk) and interventional (high-risk) studies, $X^2(1, N=14)=.39$, $p=.53$. Unfortunately, any conclusion about a lack of difference is suspect given that some cell sizes were <5, which does not allow the Chi-Square test enough power to detect relationships that may exist (Howell, 2002).

Table 3.2. SDMs' capacity to consent for research*

Total Score *n* (%) **	
7.5***	1 (7.1)
10.5***	1 (7.1)
11.5***	1 (7.1)
16	2 (14.3)
16.5	1 (7.1)
17	1 (7.1)
17.5	1 (7.1)
20	6 (42.9)

* *N*= 14.
**cut-off of 14.5. Min-max possible score= 0-20
*** SDMs unable to provide valid consent

Table 3.3 shows the SDMs' performance on each item. The majority of the SDMs interviewed were able to describe the main purpose of the medical study they consented to and were aware that refusing consent was not affecting their loved one's regular treatment. Even though the majority of them appreciated that the main purpose of the medical study was research, 3 described the main purpose as both research and therapy, one clearly

expressed a therapeutic misconception. The majority of them (*n*=9) correctly nominated at least two medical procedures involved in the ICU research. In addition, nine SDMs correctly cited the main potential risks involved, 4 SDMs had a vague or no notion of the risks involved. When asked about potential benefits of the ICU research, the majority of the SDMs (*n*=10) were able to answer correctly, while 4 produced very vague answers; furthermore, 3 SDMs did not appreciate the possibility of no benefits for their loved one as a result of their participation in research. Nine SDMs showed the ability to weigh the information received in order to make a decision, 3 produced a partially capable answer, and 2 did not show any clear reasoning behind their decision. All appreciated the possibility of withdrawing their relative from the study, and clearly expressed a decision.

Table 3.3. SDMs' performance on the adapted version of the UBACC

Adapted UBACC items *n* (%)	Clearly incapable response (score 0)	Incapable response (score 0.5)	Partially capable response (score1)	Capable response (score 2)
Item 1, Purpose of the study	3 (21.4)	-	-	11 (78.6)
Item 2, Understanding: research or treatment	1 (7.1)	-	3 (21.4)	10 (71.4)
Item 3, Can dissent	-	-	-	14 (100)
Item 4, Get regular treatment	1 (7.1)	-	-	13 (92.9)
Item 5, Things that will happen	1 (7.1)	1 (7.1)	3 (21.4)	9 (64.3)
Item 6, Risks	1 (7.1)	3 (21.4)	1 (7.1)	9 (64.3)
Item 7, Possible benefits	1 (7.1)	3 (21.4)	-	10 (71.4)
Item 8, No benefit possibility	3 (21.4)	-	1 (7.1)	10 (71.4)
Item 9, Weighing information	2 (14.3)	-	3 (21.4)	9 (64.3)
Item 10, Expressing decision	-	-	-	14 (100)

N= 14

Factors involved in the decision-making process

Table 3.4 shows the main factors involved in the decision-making process (Table 3.5 in Appendix 6 shows other factors investigated). Several SDMs perceived some degree of 'time pressure', as nearly half of them (*n*=6) perceived that only one hour or less was available to make a decision, and a few (*n*=3) only 5-10 minutes. The majority (*n*=9) reported deciding straight after the discussion with the research nurse. The majority of SDMs (*n*=10) made the decision without consulting the information leaflet provided, and also affirmed that there was no additional risk for their loved one as a result of research participation (*n*=9).

Table 3.4. Factors involved in the decision-making process

Characteristics	
Perceived level of stress *n* (%)	
Slightly stressed	2 (14.3)
Stressed	5 (35.7)
Unsure	2 (14.3)
Very stressed	4 (28.6)
Not stressed until the researcher arrived	1 (7.1)
Perception of time to decide *n* (%)	
As much as I wanted	5 (35.7)
5-10mins	3 (21.4)
Up to 1 hr	6 (42.9)
Actual time used to decide *n* (%)	
Straight away after the conversation with the research nurse	9 (64.3)
Up to 1hr 30 mins	5 (35.7)
People involved in the decision process *n* (%)	
Only the SDM	6 (42.9)
2	5 (35.7)
3 or more	3 (21.4)
Perception of patient's condition *n* (%)	
Serious but stable	7 (50)
Critical	7 (50)
Perception of additional risk as result of research participation *n* (%)	
No additional risk	9 (64.3)
A little additional risk	5 (35.7)
Prior conversation with patient about attitute towards research *n* (%)	2 (14.3)
Source of information to make the decision *n* (%)	
Information leaflet & verbal information from research nurse	4 (28.6)
Verbal information from research nurse	10 (71.4)
Verbal information from research nurse and clinical staff	0

N=14.

SDMs' attitudes towards their involvement

The majority of SDMs (*n*=13, 93%) across the two types of studies (observational and interventional) indicated willingness to be involved in the decision-making about research (Figure 3.1).

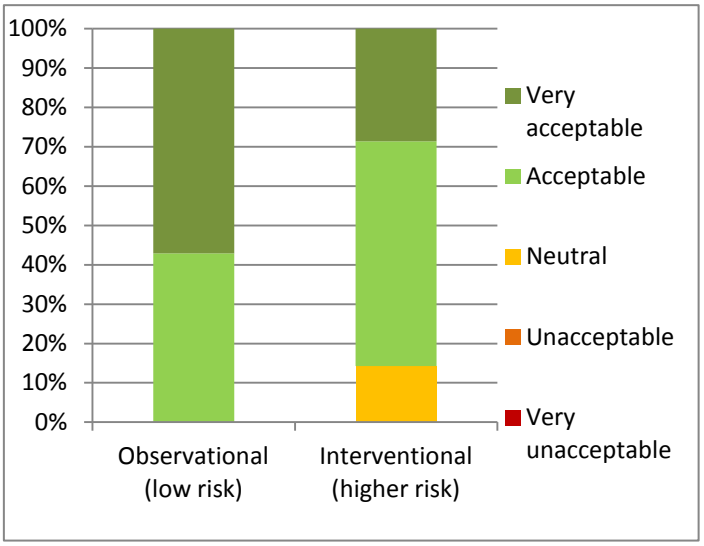


Figure 3.1. SDMs' rating of the acceptability of their involvement in research decision making across studies. Acceptability ratings were based on 5-point Likert scales from 1=Very acceptable to 5= Very unacceptable.

They also felt generally comfortable with making this decision: 50% declared feeling comfortable, 29% (*n*=4) very comfortable, 14% (*n*=2) neutral and 7% (*n*=1) very uncomfortable. Figure 3.2 shows the proportion across the two study types.

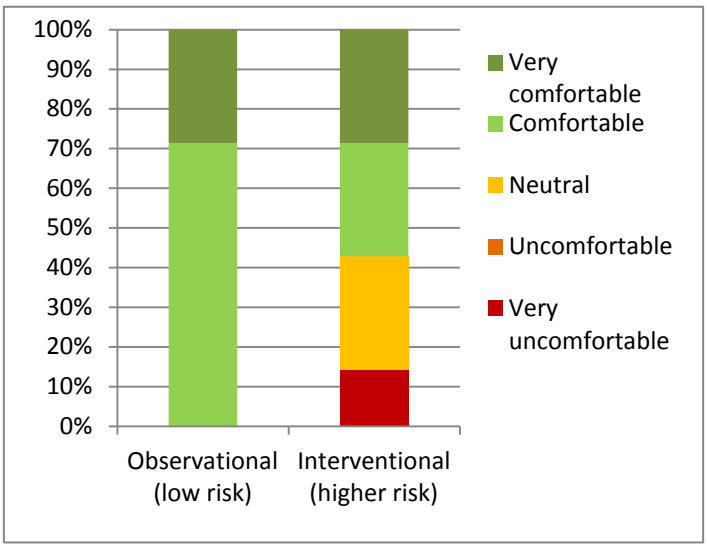


Figure 3.2. SDMs' rating of the comfort in making the decision about research across studies. Comfort ratings were based on 5-point Likert scales from 1=Very comfortable to 5= Very uncomfortable.

The majority of the SDMs perceived their involvement as a valuable opportunity (*n*=6, 43%) and an opportunity (43%); 14% (*n*=2) chose a neutral position (Figure 3.3).

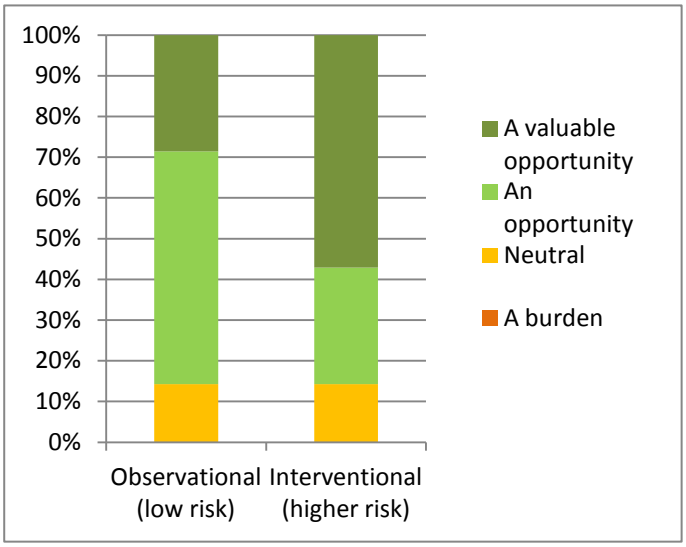


Figure 3.3. SDMs' perception of their involvement in making the decision about research across studies. The figure shows whether they perceived their involvement as either an opportunity or a burden.

When asked about who should have made the decision concerning their loved one's enrolment in research, the majority of SDMs (*n*=6, 43%) highlighted the necessity to equally share the decision with medical staff, followed by those who (*n*=4, 29%) answered 'medical staff with little advice from the SDM', and a few (*n*=3, 21%) who reply 'the SDM with little advice from medical staff'. One SDM declared a preference for the decision to involve only the medical staff. Figure 3.4 (below) shows these proportions across the studies. Even though we could not check for statistically significant differences, due to small sample size, in the proportion of responses across the two types of study, it is possible to identify a trend: 57% of the SDMs involved in the decision about an observational study showed a preference towards medical/research staff decision, against only 14% of those asked to consider an interventional study. For the latter group a decision equally shared (71%) seemed the preferred option.

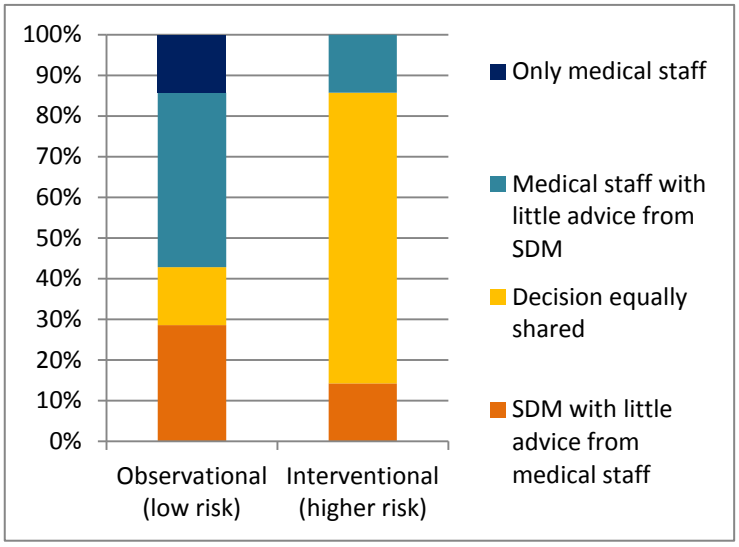


Figure 3.4. SDMs' rating of the preferred degree of involvement of medical staff vs. SDM in the decision-making about research, across studies.

2.3.2. QUALITATIVE DATA

Six themes are presented in this section. One of these, 'The time at the ICU: a draining roller-coaster' is related to the backdrop against which the approach about research took place. As such, material related to this peripheral theme is placed in the Appendix 9. The other five themes, which are more directly related to the experience of being asked to act as an SDM, are: 'Being approached for research: breaking a fragile homeostasis?'; 'Trust in medical staff's decision, with a proviso: being warned'; 'Understanding the research and its outcomes' (including three sub-themes: 'High expectations', 'Dealing with risk and probabilities: the illusion of certainty in dealing with uncertainty?', 'Dealing with verbal and written information'); 'Dealing with the decision-making: a straightforward decision vs. a delicate task'; 'Deciding for a loved one: the two sides of the coin'. Supporting quotations have been placed in text boxes to illustrate the place of each theme in the accounts of the

participants. As previously stated, three interviews involved two members of the same family, who decided jointly about their loved one's participation in research. The account of these additional next-of-kin was taken into consideration in the qualitative analysis (see p. 53: 'Qualitative analysis: analytic procedure'). Thus, the results reported below reflect the meaning attributed by multiple participants involved in the same interview, where more than one person was present. Except for one joint interview where there was initial disagreement about how each person understood the risks of the medical study (p. 72), the others showed substantial agreement between the family members. Where the interpreter was present his/her utterances are included.

'Being approached for research: breaking a fragile homeostasis?'

This theme focuses on the time when the next-of-kin are approached by a research nurse about one or more medical research studies. The approach itself seems to be a key time, which potentially evokes disturbing emotions and thoughts that may break the next-of-kin's fragile homeostasis. For some SDMs the approach did not cause additional distress to the burden already experienced:

Miss Smith: I was stressed not not because of you [she refers to the approach by the research team], because I had just been told he [her father] was going back for another scan, and I was concerned with him as person, not being approached for research.

For other SDMs, the approach not only resulted in a non-stressful experience but also caused a sense of relief that something else (i.e., some other drug) had been tried to improve their relative's condition:

I: So how stressed were you already when you were approached?
Mrs Green: I wouldn't say we were …
Miss Green (her daughter): We were stressed about dad, but I don't think
Mrs Green: The actual… [Overlapping voices]
Miss Green: Contributed to anything, probably a little bit of a relief to know that something else has been [overlapping voices] additional

However, the approach by the research nurse was described as intimidating by some SDMs. In particular, the very first impact seemed to be a delicate moment as the next-of-kin, already worried and unsure about what the researcher is going to say, may anticipate that they will be asked to make a potentially frightening decision about 'something serious':

Ms Jones: Yeah, I think I felt a little bit nervous because it's almost a new news to it:
"Oh my God! I've got to take responsibility here". […] at first I did feel a little bit like "Uh….!" [Laugh]
"Is this going to be something serious we've got to decide on?"

Another SDM portrayed the first approach about research as an experience of 'shock', as he received a phone call from the hospital which alarmed him and made him fear the arrival of bad news:

Mr Craine: […] I don't really know what I thought at that point 'cause it was such a shock when this phone rung and it was a member of hospital staff talking to me, so like I said I came here and I was like that literally.
I: Yes I also suppose it could be frightening when a research nurse approaches you and you don't know if you are going to talk about bad news or other things going on
Mr Craine: Yes it was… it was… coming through the door today, just erm "Please don't let me walk into bad news today"

The approach could also feel 'intimidating' when the location made the difference in terms of perceived 'formality': Miss Jones described how different in terms of formality and comfort were two contacts she had with a research nurse; where the first, in a corridor, resulted in a formal and intimidating approach:

Miss Jones: I think, I think probably I felt it more intimidating when I was asked [inaudible] yesterday [inaudible] [she is initially approached in a corridor in the ICU away from the bed space, while she is sitting on a chair waiting to go back to her love one's bed] . At least when I was next to the bed [the day after in a second approach] … with Paul it was it seemed a, a little less formal.

The adjective 'intimidating' came back again in another SDM's narrative, describing how a combination of a small room and several researchers made her feel uncomfortable:

Mrs Wane: […] with this room it's quite, with three of you sitting there it's quite intimidating…
[…] I think, as I said, it's a small room and I felt very crowded, erm I think to have three people come in was very difficult.

Furthermore, it seems that when the approach unexpectedly involved a large amount of information in a short and probably inconvenient time, it destroyed the person's attempt to hold things together. In particular, Mrs Wane told about her experience of distress as she was approached to discuss three different pieces of research (she was previously informed only about one), which resulted in an unexpectedly highly demanding task, the perception of time pressure and lack of staff empathy:

I: How stressed were you already when when actually all of us arrived?
Mrs Wane: I wasn't until you all arrived.
I: Yes
Mrs Wane: I was holding it together and I felt that it all came in after that.
[…] I was under the time pressure, I hadn't been told that I was going to spend, have that mean time, but I was asked to take part in one study [a consultant previously informed her about this study], where there were three of you who came in and I felt that it was just too much all at once [unexpectedly she was asked to consider three studies all at once].
[…] it wasn't the appropriate time to ask, people didn't, no one actually heard that I had another son to pick up. I was late picking him up.
[…] I: Erm yes so yes so you would have needed more time
Mrs Wane: Well, just some space, rather it all happening at the wrong time when I'd only got, I said I'd only got ten minutes anyway.

However, alongside the reflection below about the necessity for the researchers to tailor the time and approach to each case, participants also acknowledged that there are time constraints and other pressures for the research nurses as well as for the SDMs:

Mrs Wane: Erm, it would have been helpful, I think, to be given time to read the paperwork and then talk. Erm, I know on some of those bits they wanted it erm straight away erm because otherwise it wasn't any good, but that was their fault for not seeing me the previous day rather than leaving it to than right at the end there. So, erm…yes yo...you need to find out how someone wants: do they want to be talked to? Do they want the paperwork? There's different people do things in different ways.

In this theme therefore I aimed to capture participants' experiences around the initial approach to act as an SDM. The participants experienced a range of thoughts and emotions from those who did not find the approach added extra stress to those who found it quite intimidating, disturbing the fragile hold they had established over their fears for their relative's future.

'Trust in medical staff's decision, with a proviso: being warned'

This theme emerged consistently in all the interviews, and seemed to pervade the SDMs' experience at the ICU, where trust was placed in both the clinical and research staff, without always a clear appreciation of the distinction between the two. However, the trust in medical staff did not come alone, but was accompanied by another pressing wish: the necessity to be informed or better 'warned' about their loved one's enrolment in research.

Trust in medical staff could be extremely important in the SDMs' decision about research participation. For instance, the trust in the university hospital institution seemed a key factor in Mrs White's decision, being more relevant than any personal evaluation about additional risk involved in the proposed research:

I: So what factors did you take into consideration when reaching your decision?

Mrs White: I couldn't see any harm in it, you know, it seemed to be eh... also it is to do university research, that's for number one; two, I couldn't see any harm that would come to us or anybody else, so that's why I agreed really.

A total trust emerged also in the following extract, where another SDM explained how it is unlikely that the medical research involved substantial risk as if it were otherwise, the research team would not have proposed it to her:

Mrs Lee: […] If they thought it was risky, they wouldn't have proposed it.

Participants explained that the decision itself about research relied on the medical staff's knowledge and therefore was a 'decision of trust':

Ms Jones: […] we are trusting medical staff, you know. You know, it's not really equally shared because we are trusting them more to know what the […] the decision ultimately where wherever we go with the decision it's based on their knowledge

A particular circumstance was when the SDMs were asked to provide delayed consent, i.e., were asked to decide whether their loved one should continue being part of a research study after a consultant had previously decided to enrol them. Even in this case, Mrs & Miss Green trusted the medical staff's decision and justified it as necessary:

Mrs Green: […] I know the treatment was already started, but we actually erm, was asked if we wanted to carry on with it or whether we wanted [overlapping of voices] to stop [overlapping of voices].
Miss Green: They had to start it 'cause it has to [inaudible] in 6 hours

However, although trust was firmly placed in the medical staff, the proviso to be informed about any medical or research procedure remained essential for many SDMs:

I: Bearing in mind the risk involved in this study, erm, who do you think should have made this decision about research?

Miss Smith: Erm …well, I would probably say we need, you need to ask, so we need to be told what's going on, but I think it should be a mixture of all, you know, everybody stating what's happening. You know, as long as we are aware of what's going on and you've asked permission then we should be fine.

In line with this, two next-of-kin experienced an upsetting time when finding out that their relative was enrolled in a research study without them 'being warned' soon after the medical staff made the decision:

I: Could you please say more about how you felt when Nicole approached you about research?
[…] Mr Lewis: There was no warning was it?
Mrs Lewis: [Laughs] I just don't, you know, really know you gave anything like this [she refers to the unexpected news that her son was enrolled in medical research without the family been informed]
Mr Lewis: Erm, they should have come [inaudible] day, so we can do something about it in a couple of days time or whenever we come next, not just leave it on the spur of the moment.

The overall story emerging from this theme showed the centrality of the SDMs' trust in medical and research staff during the decision-making process. That being stated, the SDMs stressed the need to be informed about any research decision regarding their loved one.

'Understanding the research and its outcomes'

This overarching theme refers to how the SDMs make sense of the research aims and outcomes. Three subthemes are presented here:

<u>'High expectations'</u>

This subtheme focuses on the degree to which the SDMs perceived the research as progress and often attributed to it therapeutic characteristics, leading to high expectations about potential benefits for other people in the future and possibly for their loved ones in

the present. The majority of the SDMs offered a construction of the research as 'progress' and a 'good thing':

Mrs Berry: […] once I read I thought 'You know, it's got to be a good idea'
[…] You've got to think of the future, haven't you, and it's progress isn't it?

However, the SDMs sometimes misunderstood the main aim of the research as therapeutic, not infrequently believing that the research would directly benefit their critically ill relative. For instance, when asked to describe the potential benefit of the research, some SDMs produced answers which show this therapeutic misconception:

Mrs Nowak: [An interpreter translates her words] So she is saying she don't know, but basically she was explaining that if she wouldn't sign for it he wouldn't get this care. So she was explaining basically that she was not forced to do that, she just signed because she knows that without this signature he won't have his medication […] She is 100% sure it is gonna help him.

<u>'Dealing with risk and probabilities: the illusion of certainty dealing with the uncertainty?'</u>

This subtheme focuses on the SDMs' experience of having to deal with communication about risk and probabilities. Communication of risk was provided using 'probability language' instead of natural frequencies. Such communication could lead to misunderstanding 'the numbers' and overestimation of the benefits presented. On the other hand, when dealing with potential risks, the majority of SDMs seemed to underestimate the additional risks, even though the research study involved some degree of risk (interventional studies). Reading the following extracts where two SDMs are trying to refer (in terms of probabilities) to the communication received regarding the benefits of the research, it

appears that relative risk reduction in mortality is used as if it were absolute[1] (creating the illusion of amplified effects of potential benefits):

Mrs Green: […] it was actually, it was a treatment to actually, a study to give Andrew erm [inaudible] progesterone because it increases the risk…
Miss Green: Decreases
Mrs Green: Increases the risk of survival by 30%.
Miss Green: Decreases the risk!
Mrs Green: No, increases the risk of survival by 30%.

I: What are the benefits?
Miss Khan: The benefits are that barely 30% of people who have received the right, no the placebo, the actual… have survived, is that yes 30% less have died.

The same percentage appears several times in the narratives of those who received a similar communication about the potential benefits of the study: this shows how much the SDMs gripped onto these promising but misunderstood figures, probably being more receptive to them rather than to potential harm involved. In particular, the research was constructed as a not risky procedure where the benefits weighed more than the risks (Miss Khan) and where the understanding of the additional risk seemed to stem from a more physical and instinctual understanding rather than rational reasoning about figures (Ms & Miss Jones):

Miss Khan: […] I know about all the effects, and that's more beneficial than harmful

I: Could you please describe some of the potential risks or discomfort that he could experience…?
Ms Jones: […] she [the RN] didn't get me any slight sense of risk or concern that it might do anything negative to Paul

[1] Absolute risk reduction indicates the proportion of patients who die without treatment minus those who die with the treatment. For instance, Gigerenzer (2002) explains this reporting that pravastatin (a drug) reduces the number of deaths from 41 to 32 in 1000 (i.e., 9 in 1000 or 0.9%). On the other hand, relative risk reduction is the absolute risk divided by the number of patients who die without treatment (using the abovementioned data, it means 9 divided by 41, i.e. 22%). As it becomes clear relative risks are larger than absolute, leading to the appearence of higher benefit (Gigerenzer, 2002).

Where additional risk was acknowledged it was constructed as either a predictable risk (Miss Green), or at least a controllable risk, given the reassuring fact that the patient would receive critical attention if a problem or side effects emerged (Mrs White):

Miss Green: If he has a reaction, there is not many risk to it [overlapping voices], then you know how it is gonna be.

Mrs White: Oh! [Laughs] No, there was something erm…. [Silence] a kidney, they could affect the kidneys in some way, which I… yes, but they will do something about it as he is in this hospital in intensive care. So ain't no risks.

Another way to control the risks was through withdrawal:

I: Do you think there is any additional risk to your husband, as a result of him taking part in the research study?
Mrs White: I hope not, I don't, I hope not. Well, I thought, I thought there was no additional risk. She's got [inaudible], I guess I might say a little, okay a little, well I hope, but she [the RN] assured me that they will do something about it [inaudible], a little [inaudible], cause otherwise we can withdraw. Yeah.

However, despite these reassuring measures, a certain degree of worry about potential risks resonated from the narratives of some SDMs:

I: Do you think there are risks involved in the study?
Mrs Ellis: Well, in the leaflet it says, you know, that there may be risks erm, but I don't know exactly if there is a problem what you can do, apart from stopping that. There's nothing else you can do, is there? If there are side effects, something like that you know, you can stop it all together and then hopefully he'll go back to what he was.
I: Yes. Erm do you remember any of the potential risks?
Mrs Ellis: No, remind me! [Laughs] I don't. Oh! Hang on, [inaudible] okay, you tell me the potential risks.

Some research procedures themselves could evoke the phantom of uncertainty. For instance, medical decisions about research was seen as reassuring in that it was opposed to the uncertainty derived from the chance feature of the randomisation procedure, where a

machine or a coin makes the decision. In the following extracts an SDM described how she did not feel comfortable with signing for the second of two medical studies proposed, given that the procedure of randomisation was leading to a degree of uncertainty instead of leading to 'the best route', and where the lack of time did not give her chance to further explore this possibility:

Mrs Wane: […] I got something that was saying: "Well, this is the part one, and if we find that, we've done that, it will only be this, this and this, and well, monitor", and I was thinking "Well no, that effect you flip a coin and say which treatment he's going to have because you want three different proofs for your study, and it was too much without having been warned.
[…] The second study I wasn't quite as sure whether because I hadn't got time to read the notes and make enough enquiries whether actually they would flip the coin and go "Well, we don't know which is the best route so let's put him on one or the other", whereas I prefer them to make a judgement.

'Dealing with verbal and written information'

This subtheme is concerned with the SDMs' experience of dealing with the information received about research, where verbal information seemed to be easier to take in than written information. To understand how the SDMs dealt with new information, it is useful to consider the context of uncertainty and worry that they were experiencing, which it is likely to have limited the resources available to receive and process the information:

I: Can I ask you a little bit more about how did you feel when yesterday a research nurse approached you?
Miss Smith: Erm I think at the time, because we had just been told that he [her father] was gonna, you know, he could be poorly and he was going back for a scan, I was a bit, I wasn't fond of being approached, that's not a problem. Erm I was just panicking for him so my head wasn't completely with what was being said.

In such a context, the use of what is perceived as 'medical gobble de gook' language in the information leaflet appeared scary for some SDMs:

Mrs Wane: […] Some of the paper work, the top, the heading on the paper work, I haven't bought it with me, was medical gobble de gook, and anyone that saw that would perhaps run a mile.

For Mrs Lee it was difficult to even try to read the information leaflet:

I: What source of information did you use to make your decision?
Mrs Lee: Only the discussion with the RN. You don't really want to read things. It's okay to take verbal information but not to read things.

On the other hand, the discussion with a research nurse could help make the research understandable and the decision task more comfortable (Mrs & Miss Green), allowing the SDM or other relatives also to ask questions (Ms Jones):

I: How comfortable did you feel with making this decision?
Mrs Green: Very comfortable.
I: What do you think made you feel this way?
Miss Green: I think because Louise [the RN] has been spoken to us, she was so good in breaking down for us …I think partly that.

Ms Jones: For me, repeating, repeating what she [the RN] said, and you [Looking at the stepdaughter] asked a good solid question so just to make sure we haven't been, because I feel [inaudible] sometimes I've got to repeat it so that it settles for me; and to ask a couple of questions in cause I don't want to, cause it is [inaudible] just to say "Of course"

In this superordinate theme I tried to capture the SDMs' sense-making of the research aims and its outcomes. The SDMs interviewed constructed the research as 'progress' and 'a good thing' involving no risks for their loved ones but large benefits. Some SDMs perceived the research as a potentially redeeming treatment, also misunderstanding the

communication received about risks. In general, the participants' accounts highlighted their tendency to struggle with reading information leaflets.

'Dealing with the decision-making: a straightforward decision vs. a delicate task'

This theme addresses the degree to which the SDMs perceived the decision-making itself as being not a particularly challenging task, requiring limited time to make the decision, versus the belief that the task may be a delicate one, requiring resources and time. For decisions regarding observational studies, the limited time SDMs used to come to a decision appeared connected to the low complexity involved in the proposed study, as was the case for studies involving taking blood samples; therefore, making a decision straight after the discussion with the research nurse seemed plausible:

Mrs Best: We've just been told about it, but we didn't mind; when we had 2 or 3 hour to decide, it would be the same, so it doesn't matter.
[…] 'Cause it's about taking blood and it's gonna help [inaudible], like he [the RN] said, the quicker they do it the better, so out my mum to do a test. There's no harm in extra blood to come out.

However, the construction of a quite quick and straightforward decision emerged also from the narratives of SDMs asked to consider interventional studies. In particular, despite the fact that the proposed research was involving some degree of risk and limited time was available to decide, the decision still had the features of a not difficult or 'serious' decision. The reason for this seemed linked with the SDM's perception that the benefits clearly outweigh the risks given that no 'harmful drug' or invasive treatments were used:

I: How many hours did you have to decide?
Miss Khan: Not so much, but that's because the this treatment, as I'm sure compared to other treatments that can actually be both ways make it worse or make it better, these are harder to

decide on them, something like this. That's horrible that you already have, so it wasn't a very difficult decision. […] Compared to other bigger decision that could be like more harmful drug or things like this.

The construction of the decision-making task as not challenging could also be identified in the narratives of the majority of SDMs who were asked to provide delayed consent. In this case, their decision assumed the shape of a confirmation of medical staff's decision rather than a more challenging brand new decision (Mrs Wane), where the SDMs let the research staff guide them (Mr Craine):

Mrs Wane: Well, I understand the doctor did, the consultant has made the…, until they can see me on that first study, and I'm happy with that. Erm I think he [the RN] perhaps ought to have told me that that I was confirming his decision, rather than making it sound as if though it was my initial decision.

Mr Craine: […] as for risk, I would think that, that has been well assessed by the medical teams here and the ones above them, so you know that's, that's pretty well thought through before they go ahead […]. How I read this, is that they made this decision, more or less, and put two options to me, and acting on their advice and how John [the RN] presented it; and that was how [inaudible] the decision…[was made]

In addition, when having to make a decision about a critically ill loved one's enrolment in research represented 'another thing to think about', the attitude of some SDMs may have been to opt for an extemporaneous decision, even though additional time was available:

Ms Jones: […] at first I did feel a little bit like "Uh! [Laughs], is this going to be something serious we've got to decide on?" […] it's new again, it's another thing to thing about you know […]
I: How many hours did you have to decide?
Miss Jones: We went with it straight away

On the other hand, a few SDMs depicted the same decision-making process as a more complex task. For instance, Mrs Wane explained how she perceived the decision-making

process as a potentially delicate task, involving the risk of providing a pressured and invalid consent:

I: When you say that you felt pressure for that study, could you explain it a little further?
Mrs Wane: I think it was because the consultants had explained about one [research study] and I was quite happy for that, but the other one, because I didn't understand and I haven't got time to read erm because I was under time erm, and with three people in the room, I felt it was very difficult. I think if you had done that with someone that was a, a, someone that wasn't used to erm holding their own corner, they could have been pushed just into signing without really thinking about it. […] Might be better to ask people, it might be better to say "We might talk to you in a bit about, can we leave you some papers?", because then you can either take it or leave it.

This theme addressed the SDMs' construction of the decision-making task: the majority of SDMs perceived it as a not particularly challenging task and described a very rapid decisional process. A few participants' accounts highlighted the SDMs' awareness that the complexity of emotional and informational aspects involved in the decision-making may lead to little pondered decisions.

'Deciding for a loved one: the two sides of the coin'

This theme focuses on how the SDMs experienced their involvement in the consent process as either something which gave them the perception of exerting a certain degree of control or a burdensome experience due to the responsibility taken.

The exertion of control and power, as opposed to a sense of helplessness, is traceable in the SDMs' words where their decision about research participation meant for them increasing their loved one's chance of survival and contributing to their care:

Mrs Green: […] He has been through a terrible accident, and even if he has the placebo, it is not going to do any harm, if he has got the drug, there is a benefit. I rather give him every chance to actually survive.
I: So if you were to define your involvement from…? [Showing a 5 point Likert scale from a large burden to a valuable opportunity]
Mrs Green: A valuable opportunity cause we've been asked to take part in Andrew's care.
[…] He is in a critical condition, we want to do all we can to get through.

However, the other side of the coin seemed to be a certain degree of burden. In particular, the SDMs' main concern was whether or not they had put their loved ones in any danger (Mr Craine), and whether they had made a decision for which their relative would blame them in that it is against his/her wishes (Mrs White):

I: Could you describe how did you feel physically emotionally when John approached you?
Mr Craine: Yeah, well, well, I thought it was a little bit of a shock at first, and I asked "What I'm actually signing for?"; and if it was gonna put her in any danger at all

I: If you were asked now, would your decision about the research study be the same?
Mrs White: It would be the same, but I expected my husband came to, and he would be told about it, and he might say: "What have you done?! No, [inaudible] out of it! I am not going, why? I don't like this. Get me out of it […]"

In such a context again the withdrawal seemed to function as a reassuring proviso which eliminated the phantom of an irrevocable decision, with a consequent release of sense of responsibility (Ms Jones):

Ms Jones: […] and again she [the RN] gave us very good choice to come back. I did hear that a few times. If we want to withdraw, then we've got choice, if we get an… cause that does sometimes happen when you are sitting with yourself you think "Oh [inaudible]", and she said that a few times: "We can choose not to go ahead with it." So I liked that, cause that then takes the pressure off of what I've said now.

Furthermore, it seems that sharing the responsibility of making the decision with other family members enhanced comfort with the decision-making (Miss Jones):

Miss Jones: [Looking at the other member of the family] Because you were here I felt comfortable, but I think maybe, if I'd been on my own without…, I wouldn't have made the, the, the decision.

Nevertheless, a few SDMs were able to envisage a positive outcome where the loved one would soon get better and make his/her own decision about the medical research:

Mrs White: […] I'm sure we can get out of it if we see anything we don't like about it, and my husband hopefully will make the decision himself.

For many SDMs deciding for a critically ill loved one represented the possibility to be part of their family member's care. This allowed them to exert some degree of control over an uncontrollable situation. However, the other side of the coin was represented by a degree of burden as the SDMs felt the worry to have made a decision against their loved ones' wishes.

3. Discussion and conclusions

The SDM model for informed consent has been introduced in the UK to protect patients and their right to self-determination about participation in research. However, previous research has shown that deciding about a close relative's enrolment into ICU research may place an additional burden on the SDMs (Pochard et al., 2001), who may make decisions based on their own emotional state rather than in line with the patient's wishes (Mehta et al., 2012; Menon et al., 2012). In addition, evidence suggests that next-of-kin may not fully understand the nature of research about which they are approached (e.g., Azoulay et al.,

2000; Chappuy et al., 2010), which makes it difficult, if not impossible, for them to make a valid decision (e.g., Burns et al., 2009; Ciroldi et al., 2007).

This study investigated the proportion of SDMs capable of providing valid consent/assent for research involving a critically ill close relative in an ICU. Our results showed that 21% of the SDMs assessed were not capable of doing this. In particular, they did not understand the aim of the specific medical research about which they were approached or appreciate the risks and benefits involved. Unfortunately, the small sample size affected the significance test looking at the relationship between the capacity to provide valid consent and the complexity of the study, thus it was not possible to draw any solid conclusion about this.

Both the quantitative and qualitative investigation highlighted a trend in the SDMs' understanding of medical research, where the benefits were perceived as large while the risks as extremely low (for instance, two-thirds affirmed that there was no additional risk at all for their loved one). In addition, the thematic analysis showed that when risk communication was provided by research staff using probabilities instead of natural frequencies, the benefits of the studies were overestimated by the SDMs: their accounts showed a discursive construction of the benefits as unrealistically large benefits, as the relative risk reduction in mortality offered by the treatment was discussed as if it were absolute. In line with this, Gigerenzer (2002) suggests that relative risks are larger numbers than absolute risks and thus they give the illusion of higher benefits than really exist.

There are several potential explanations for this trend. Firstly, these results may stem from the next-of-kin's desperation to see the research as a potential new redeeming treatment (in line with what has been suggested by others, e.g., Mehta et al., 2012). Second, their incorrect evaluation may be explained by their limited understanding of risks and

probabilities. Third, these results may highlight the research staff's miscommunication of risks, which may be caused by their use of conditional probabilities in their communication with the SDMs. In line with this, Hoffrage & Gigerenzer (1998) found that even doctors may struggle when dealing with probabilities: in their study, only 10% of doctors correctly estimated the figures presented as probabilities, against 46% who correctly estimated information presented as natural frequencies.

In addition, the majority of SDMs (*n*=10; 71%) made their decisions without consulting the information leaflet provided, which means that their decision was based only on the discussion with the research staff. Thus, these results raise issues about what level and standard of information is required in order for SDMs to make a valid and informed decision about participation in research (see also, Antoniou et al., 2012).

The quantitative and qualitative results showed that a majority of SDMs (*n*=9, 64%) took a decision straight after the discussion with the research nurse, although more time was available (except for 3 SDMs (21%) who had only 5-10 minutes). This may highlight a tendency to arrive at extemporaneous decisions, potentially little pondered. Due to the small sample size, determination of statistically significant differences between the time taken to make a decision for low-risk and higher-risk studies was not possible. However, 57% of those asked to decide about greater-than-low risk studies were in fact asked to provide 'delayed consent', as a doctor had already made the decision about their loved one's enrolment. This may have simplified the decision-making for the SDMs, as they may have needed less time to 'confirm' a medical decision that had been already taken. Whilst this procedure may reduce the burden on the SDMs, it may also increase the SDMs' tendency to

agree with the medical staff, and make them mistakenly believe that the research has a primary therapeutic purpose.

Moreover, from my qualitative analysis it emerged that the participants experienced a range of thoughts and emotions connected with the approach about research, from those who did not find the approach added extra stress to those who found it quite intimidating (for instance, when it unexpectedly involved a large amount of information in a limited time, when the room was too small for the number of people in it, or when the content of the information was not clear from the beginning). This leads to the recommendation for researchers to place additional care in planning the approach, considering also physical aspects of the location and the way they introduce the purpose of their conversation with the next-of-kin.

Limitations. The main limitation concerns the small sample size, which limits any claim of generalisation of results, and also did not allow us to fulfil all our research aims. In particular, we were not able to analyse what factors influence the SDMs' capacity to consent for research. Furthermore, the SDMs may have perceived me as part of the hospital research team, given that I was introduced to them by the research nurses as their colleague, and that frequently the participants used the term "you" to refer to both the ICU research team and me. As a result, their answers may reflect social desirability bias, as it is possible that at a time of severe illness they tried to provide positive and desirable feedback. Other limitations were the use of leading questions in some cases, which may have directed the participants' answers, and the loss of audio in some recordings due to background noises, which affected the quality of the transcripts. Unfortunately, it was not possible to change the location where the interviews took place, and in this way control the background noises, in order to

mitigate the effects of these on the analysis and interpretation of the data collected. However, it may have been useful to ask the participants to confirm my interpretation of their accounts to enhance the plausibility and completeness of my analysis.

Strengths. The use of methodological pluralism, with which I refer to the use of a combination of quantitative and qualitative methods, allowed the adoption of different perspectives to achieve a holistic understanding of the experience of SDMs in the context of the ICU. From what we know, this is currently the only research trying to explore this issue using a mixed methods design, and also the only one to specifically investigate the SDMs' capacity to provide valid consent for research.

Implications. The implications of this study are important: its results stress the need to find better ways of safeguarding SDMs and fostering valid consent, whilst permitting the advancement of medical care through research. Showing that approximately 20% of the SDMs assessed had insufficient understanding of the medical research aims and the benefits/risks ratio, this study indicates that a not negligible number of people could provide invalid consent/assent for research. Consequently, it may be necessary to reconsider researchers' and research nurses' practice in critical care. We recommend researchers to carefully check potential participants' understanding of the proposed research and their capacity to provide valid consent (see also Silverman and colleagues, 2004). This could be done through the use of a screening test such as our adapted version of the UBACC, which could be quickly and easily conducted.

The results highlighted also that SDMs may misunderstand the figures about the risks and benefits involved in the medical studies, perceiving the benefits as large and the risks as minimal even for studies involving greater-than-minimal risks. For this reason, we suggest

training researchers and research nurses in risk communication. In particular, as Hoffrage & Gigerenzer (1998) suggest, using natural frequencies rather than probabilities may help the doctors to better communicate risks to patients, and patients to better understand these risks. Further research needs to understand, using a larger sample, the actual proportion of SDMs incapable of providing valid consent for research, as well as how to foster valid surrogate consent and whether there are alternative solutions to the current SDM model of consent.

4. Reflections on learning

This final placement represented the opportunity for me to conduct challenging research, where the complexity inherent in the topic of investigation and the delicate context and time of participants approach required consistent flexibility and reflexivity about methods and procedures used. I appreciated how demanding it may be to conduct a research about critical care research as the former needs to be adapted to the timing of the latter, and a good amount of energy is required to develop a synergic effort with the critical care research teams and staff, as key figures in the recruitment phase. I have also learned that researchers need to find appropriate compromises between their pressure to recruit as many people as possible, logistical constraints and the needs of participants and other researchers. In particular, this research has allowed me to recognise the need for researchers in the critical care context to put additional safeguarding measures for participants who may have been already approached by other researchers and may be overwhelmed by a large amount of information.

The time spent addressing reflexivity issues during the supervision made me understand how much my personal presuppositions may have influenced how the data was collected, analysed and synthesised, and my impact on the environment studied. The supervision helped me also to keep focused on the research questions, even though new aspects not originally considered, were emerging. Meetings with the ICU research staff improved my ability to present research in a concise and comprehensive way.

In addition, the combination of quantitative and qualitative methods of data collection and analysis allowed me to enhance my theoretical and practical knowledge about different research techniques and how they can be combined together in a pluralistic approach.

Appendix 2

INFORMATION SHEET AND CONSENT FORM

UNIVERSITY OF BIRMINGHAM

CONSENT FOR RESEARCH BY SUBSTITUTE DECISION MAKERS

Information sheet for next-of-kin

Version **1.1**, 23/04/2013

Invitation to join the research study

We would like to invite you to take part in our study which is trying to find ways of reducing the stress on the next-of-kin of patients admitted to intensive care (ICU). Before you decide we would like you to understand why the research is being done and what it would involve for you. My name is Donatella D'Antoni. I am an MRes Clinical Psychology student at the University of Birmingham. I am conducting this research, with supervision from Dr J Oyebode, Dr I Mackenzie, Dr F Oyebode and Professor Bion, as part of my Masters in Research. I will go through the information sheet with you and answer any questions you have. Please take time to read this information and feel free to talk to others about the research project if that would help you to decide whether you want to take part.

What is the purpose of the study?

The aim of this study is to explore the experience of people who have been asked to agree for their loved one taking part in research. We would like to know your personal point of view about this experience. We understand that this is a difficult time for you, and that you are worried about the health of your loved one. We also know that it is likely that you have recently received a lot of medical information that may not be easy for you to take in. In addition, we realise that asking you to decide if your loved one would have wanted to be involved in research can be stressful for you. For this reason, we want to understand what influences your ability to make decisions on their behalf and explore your views about this. The results of the study should help us to find the best way of carrying out medical research for the benefit of critically ill patients whilst minimising the stress and burden on you, friends and family members.

Why have I been invited?

You are being invited to take part in this study because you have been asked by an ICU researcher to enroll your loved one in a critical care research project.

Do I have to take part?

No. It is up to you to decide if you want to take part or not. If you decide not to, we would like to ask you if you would be willing to give us a short reason. If you agree to take part, we will then ask you to sign a consent form. You are free to withdraw at any time from the research. This will not affect the quality of care you and your loved one receive from the clinical team.

What would taking part involve for me?

If you decide to take part you will be interviewed by me. You will be asked some questions about your actual understanding of the proposed medical research. I will also ask you what it is like to be involved in making the decision about whether or not to enrol your loved one in an ICU research. Our interview should take about 20-30 minutes. If you agree to take part you will be involved in our study for no more than 1 day. We will need to record the interviews for later analysis and to use direct quotes in research reports. You will not be named in any report.

What are the possible benefits of taking part?

We think that taking part in our research could be of benefit for you, because you will have the chance to talk to someone about your experience of being involved in ICU research. This could also give you a chance to share your opinion, doubts, and emotions, and to find a source of support if needed.

We believe that the information we get from this study could improve the way next-of-kin are approached about research. Our findings could help to reduce any distress or anxiety that this experience could cause them.

What are the possible risks of taking part?

Because of the topics covered by our interview, taking part might cause you extra stress and discomfort.

What if there is a problem?

Any complaint or any harm should be reported to me or the named Contact Person at the foot of this sheet. In addition, if you feel very anxious and stressed, and you think you need help, please speak to me about the possibility of talking to an ICU psychology consultant. Attached to this sheet, you will find useful local numbers.

Will my taking part in the research study be kept confidential?

Yes. All information gathered will be kept according to the Good Clinical Practice Guidelines. It will be handled in confidence. All data will be kept very safely and securely, and will only be accessible to the research team. After the analysis of the recorded interview, the recording will be destroyed and the transcript will be kept securely and anonymously for five years.

What will happen if I don't want to carry on with the study?

You can withdraw at any time, but we would like to use the data collected up to your withdrawal. You can ask me to take your data out of the study at any time up to 2 weeks after the interview. If you ask for this, we will destroy your data and keep no record of it at all.

What will happen to the findings from our study?

Findings will be published in scientific journals, and may be used by the Trust to make practical recommendations. Nothing that could identify you will be included in any report or publication. If you wish, we would be pleased to provide study results to you when we finish the study.

Who is organizing and funding our study?

This study is being organized by a research group from the University of Birmingham (School of Psychology and University Dept Psychiatry) and the Queen Elizabeth Hospital Birmingham (Department of Anaesthesia & Intensive Care Medicine). It is sponsored by the University Hospitals Birmingham NHS Foundation Trust.

Who has reviewed the research study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interest. This study has been reviewed and given a favourable opinion by the NRES Committee West Midlands-Solihull.

What happens next?

If you decide to take part you will be asked to sign a consent form to confirm this. If you want to think about it for longer or discuss it with your friends or relatives, you can delay your decision and we will contact you later in the day to answer any questions or enroll you in the study.

Thank you for considering taking part and taking time to read this information sheet.

Sources of help

Patient Advice and Liaison Service (PALS) at QEHB

As a relative or carer PALS is available if you need someone to turn to for on-the-spot help, advice, and support. You can drop in to PALS during office hours (Monday to Friday, 10am to 4pm), leave them a telephone message (0121 371 3280), or send them an email (PALS@uhb.nhs.uk). PALS is on Level 0 and is signposted from the main entrance.

Your General Practitioner

It is important that your GP knows what is going on in the family, and many GPs can provide support and counselling. They can also provide help with problems such insomnia and depression.

ICU Steps

An organisation set up to provide help and support for patients, their relatives, and their friends. They have an excellent web site (www.icusteps.org) or can be contacted by mail (ICUsteps, 18 Fortescue Drive, Shenley Church End, Milton Keynes, MK5 6AU) or telephone (0870 471 5238).

British Association for Counselling and Psychotherapy

For details of counsellors and psychotherapists in your area.
Telephone 01455 883300.

Samaritans

An organisation that provides confidential, unbiased emotional support, 24 hours a day, for people who feel distressed, desperate, or suicidal.
Telephone 08457 909090.

UNIVERSITY OF BIRMINGHAM

Centre: Intensive Therapy Unit - QEHB
Study Number:
Participant Identification Number:

CONSENT FORM

Title of the Project: **Consent for research by substitute decision makers**

Chief Investigator: **J. Bion**

Please initial all boxes

1. I confirm that I have read and understand the information sheet dated 30/01/2013 (version 1.0) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

2. I understand that taking part is voluntary and that I am free to withdraw at any time up to 2 weeks after the interview without giving any reason, without my medical care or legal rights being affected. ☐

3. I give permission for the researcher to have access to my relative's medical record. ☐

4. I agree to take part in the above study. ☐

5. I consent for the researcher to use audio taping and to anonymously use quotation for publications and research reports. ☐

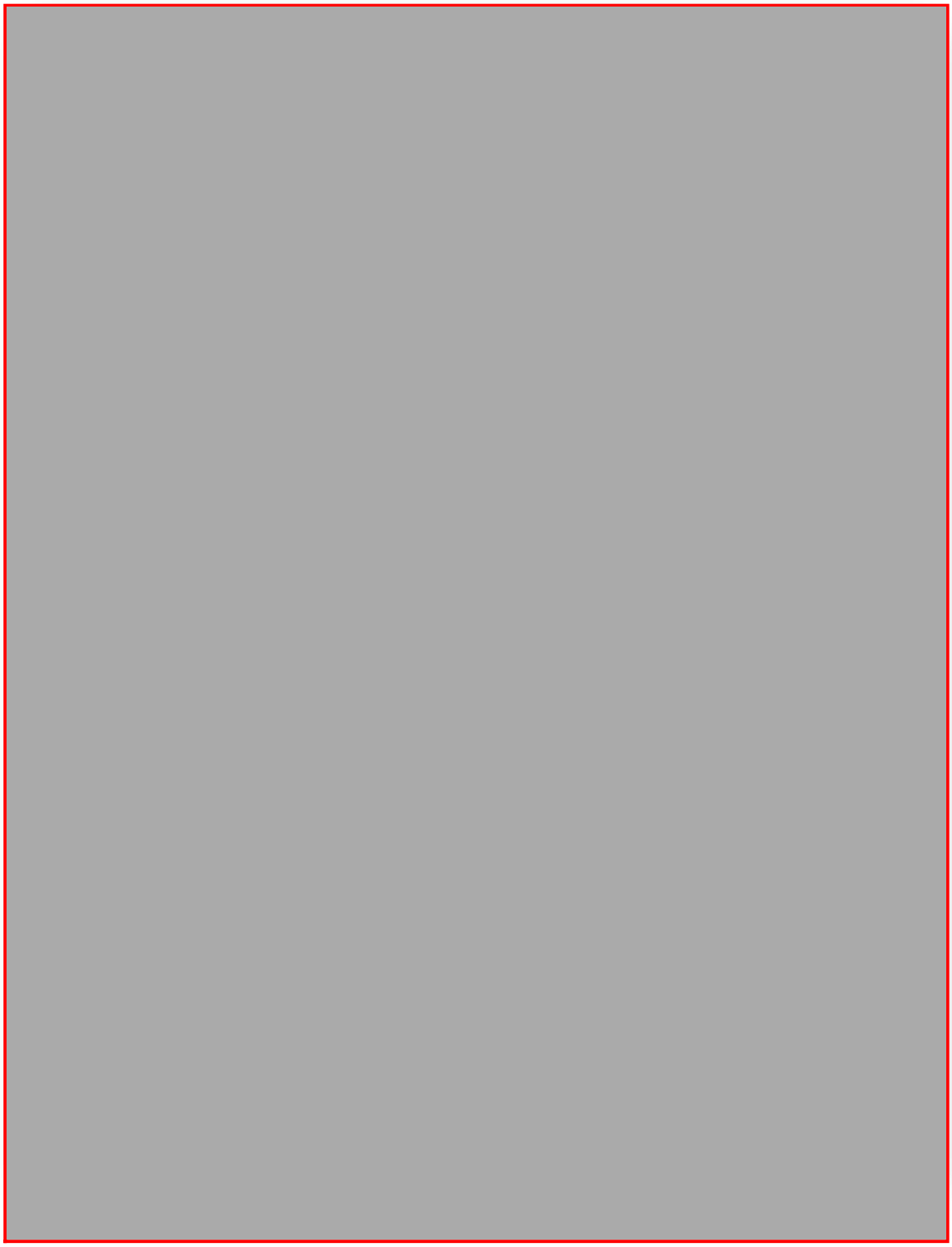
6. Tick here if you wish to receive a summary of the study results. ☐

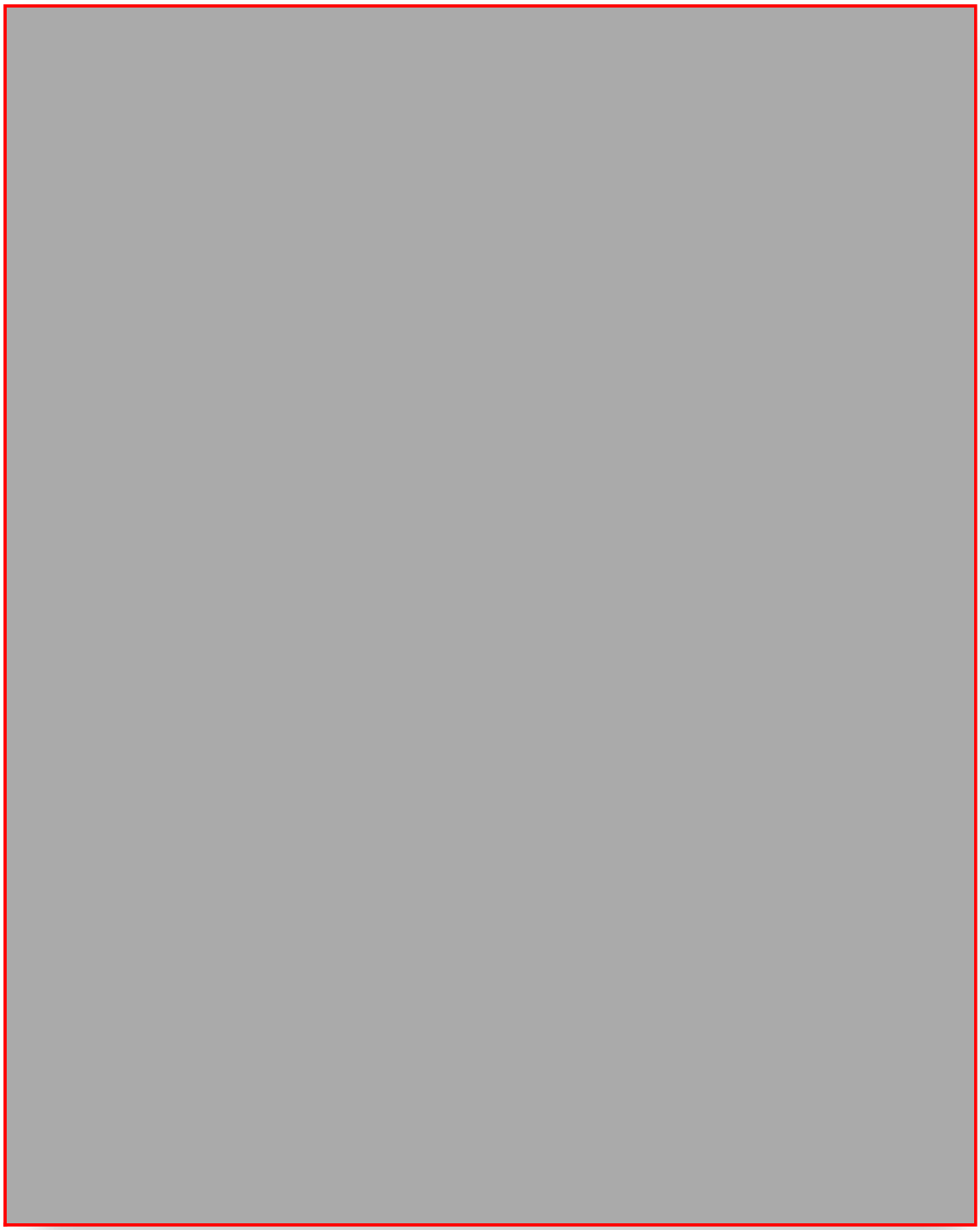
Name of Participant	Date	Signature
Name of Person taking consent	Date	Signature

This consent form is version 1.0 dated 30/01/2013 and refers to the project 'Consent for research by substitute decision makers'. The Chief Investigator for this project is Professor Julian Bion.

Appendix 3

RESEARCH ETHICS COMMITTEE APPROVAL LETTER





Appendix 4

THREE VERSIONS OF THE VIDEOS – ONE SPOKEN, ONE WITH SAE AND ONE WITH BSL.

The child is asked to watch the videos which show a narrator presenting animated stories. In order to pass the tasks the child needs to answer correctly to both the test and control questions. The children's responses are recorded in written format by the examiner.

FIRST-ORDER FALSE BELIEF TASK
Girl-Boy* (Sally-Ann) Marble Task

Description:
Slightly modified version - described by Peterson & Siegal (1995) - of Baron-Cohen et al. (1985) version of the original 'FB task' proposed by Wimmer & Perner (1983). The task involves a girl and a boy. The girl hides a marble in her basket and leaves the scene. Once she is away, the boy moves the marble and puts it in his box. When the girl returns, the child is asked: 'Where will the girl look for the marble?' followed by two control questions: 'Where is the marble now?', 'Where did the girl put the marble in the beginning?' To pass the task the child has to answer correctly to both questions.

Script:
Hello! My name is Natalie. I am going to tell you a story. Then you are going to answer some questions.

Here is the girl [*point at the Lego girl*]. Here is the boy [*point at the Lego boy*].
Which one is the girl? [*Pause*] **Which one is the boy?** [***Control/naming question***].
The girl has a basket [*show basket*]. The boy has a box [*show box*].
The girl has a marble [*show marble*]. She puts the marble in her basket to keep it safe and then goes out to play [*act out and remove the girl from view of camera*].
While the girl is away and cannot watch, the boy takes the marble out of the basket and puts it into his box [*act out*].
The girl come back [*the girl is back into the scene between the box and the basket*].
Where will the girl look for her marble? [*Belief question*].
Where is the marble now? [*Reality control question*].
Where did the girl put the marble in the beginning? [*Memory control question*].

** Girl and Boy exchanged for Sally and Anne to eliminate need for finger spelling names, highlighted by Peterson and Siegel (1995) as placing extra demands on attention span and memory of children.*

Hidden Cake Task

Description:

A boy places a tray of cakes in a green cupboard, and then goes out to play. A second character, a girl, moves the cakes and puts them in a blue cupboard. When the boy returns, the child is asked: 'Where will the boy look for the cakes?' followed by two control questions: 'Where are the cakes now?', 'Where did the boy put the cakes in the beginning?' To pass the task the child needs to answer correctly all the questions.

SECOND-ORDER FALSE BELIEF TASK
John and Mary (boy and girl) and ice-cream man

Description:

Four characters are involved (a girl, a boy, an ice-cream man, and the girl's mother) and 3 locations (a park, the girl's house, and the school). At the park, the girl and boy see an ice-cream van, but the girl does not have money for the ice-cream so she goes to her house to get some. The ice-cream man told her that he was going to stay at the park all day. Meanwhile, the ice-cream man decides to go to the school to sell ice-cream. The girl sees him and follows him to the school. Later the boy goes to the girl's house but her mother only tells the boy that she went to buy some ice-cream. To pass the task, the child should understand that the boy would mistakenly believe the girl went to the park (Perner & Wimmer (1985).

The narrator asks probe and control questions to ensure that the key elements are remembered and understood. Two open-ended questions: 'Where does the boy think that the girl went to buy the ice-cream? Why?' (Sullivan, et al. 1994).

Script:

John and Mary are playing together in the park. They see the ice-cream man coming. Mary really wants to buy an ice-cream cone but she doesn't have any money. She feels sad. The ice-cream man says to Mary: "Don't be sad, you can go home and get some money. I'll be here in the park all day long." So Mary goes home to get money to buy an ice-cream cone. See, there goes Mary to her house to get some money. John stays in the park and plays.

Probe Question 1. "Why did Mary go home?"
Probe Question 2. "What did the ice-cream man tell Mary?"

Now, John sees the ice-cream cart start to move away. John asks, "Hey, where are you going?" The ice-cream man says, "I'm going to the school to sell ice-cream. I can sell more ice-cream at the school." So the ice-cream man starts walking to the school to sell ice-cream there. See, there goes the ice-cream man to the school.

Probe Question 3. "What did the ice-cream man tell John?"

Now, John goes off to his house to have some lunch. Mary is at her house getting money for ice-cream. Mary walks outside her house and sees the ice-cream man going by. "Hey, where are you going?" asks Mary. "I'm going to the school to sell ice-cream," says the ice-cream man. Mary says, "Well, I'm so glad I know that. Now I have some money to buy an ice-cream cone, so I will follow you to the school." The ice-cream man and Mary go to the school.

(After they get to the school two opaque screens are placed in front of the park and school).

Remember the little boy John?

Probe Question 4. "Does John know the ice-cream man went to the school?"

Linguistic control question. "Does John know that the ice-cream man told Mary he was going to the school?"

Non-linguistic control question. "Does Mary know where the ice-cream cart is?"

[The questions are a balance of 'yes/no' responses to avoid preference of a response over the other. Feedback and correction should be given before continuing onto the next section of the story. The second-order ignorance question should be asked as the first test question and no feedback was given.]

Second-order ignorance question. "Does John know that Mary knows where the ice-cream cart is?"

Now, John has finished his lunch and he goes over to Mary's house to play with her. John knocks on the door. Mary's mother comes to the door. John asks her, "Where's Mary?" Mary's mother says, "Mary went to buy an ice-cream cone." So John goes off to find Mary. *[The scene finishes with John in front of Mary's house]*

Memory aid: Now, remember, John does not know that the ice-cream man told Mary where he was going.

Second-order false-belief question. "Where does John think Mary went to buy an ice-cream cone?"

Justification question. "Why?"

MISLEADING CONTAINER
Cereal Box/Smarties Task (2 trials)

Description:

Based on the 'appearance-reality' task described by Perner et al. (1989). Includes two trials.

First trial: the story presents a boy and his favourite sweets, Smarties. This tube of Smarties, however, contains a red crayon instead of sweets. Once the unexpected content is

revealed, the child is asked: 'What did the boy think was inside the tube before we looked inside?', 'What did you think was inside the tube before we looked inside?' Second trial: a cereal box is used, instead of a Smarties tube, which contains a pencil. The procedure and questions are the same as the first trial.

Script:

Here is a boy [*a Lego boy is shown in front of camera*]. He is very hungry.

Here is his favourite cereal/chocolate is called 'Cornflakes'/'Smarties' [th*e cereal or Smarties are shown on the table next to the boy*].

What do you think is inside the box/tube? [*Pause*].

Let's look and see what's inside [the boy *opens the box/tube, removes pencils/crayon and puts them on table*].

What was inside the box/tube? [*Pause*].

What did the boy think was inside the box/tube before we looked inside? [*Pause*].

What did you think was inside the box/tube before we looked inside? [*Pause*].

How would the boy feel when he found pencils/crayon inside the cereal box/smarties tube? Would he be happy or sad?

Appendix 5

RATING SCALE FOR ToM TASKS

Please watch the videos and respond to all the following items. Rate the usability of the videos: tick one option for each item. For items that are not applicable use N/A.
(SE= Spoken English; BSL= British Sign Language; SAE= Sign Assisted English)

1. THE GIRL-BOY MARBLE TASK:	Strongly disagree				Strongly agree	N/A
Overall, in the three versions (SE, BSL, SAE) the storyline was clear	1	2	3	4	5	
Overall, the narrator's pace was appropriate	1	2	3	4	5	
Overall, the animated story was clear	1	2	3	4	5	
Overall, the questions were understandable	1	2	3	4	5	
Overall, the instructions were easy to understand	1	2	3	4	5	
Overall, the story presented was easy to follow	1	2	3	4	5	
Overall, this task was easy	1	2	3	4	5	
Overall, the information provided is effective in helping me complete the task	1	2	3	4	5	
In the SE version, there were no disturbing noises	1	2	3	4	5	
In the SE version, the instructions were clear	1	2	3	4	5	
In the BSL version, the instructions were clear	1	2	3	4	5	
In the SAE version , the instructions were clear	1	2	3	4	5	

Control questions:

a) Where is the marble at the end of the story? _____

b) Where did the girl put the marble in the beginning? _____

c) Now, please read the script of the story and answer the following questions:

Having read the script, how accurately do you think the video reflects the script?

Not accurately at all 1 2 3 4 5 Very accurately

Why?

_____

_____

2. THE ICE-CREAM STORY TASK:	Strongly disagree				Strongly agree	N/A
Overall, in the three versions (SE, BSL, SAE) the storyline was clear	1	2	3	4	5	
Overall, the narrator's pace was appropriate	1	2	3	4	5	
Overall, the animated story was clear	1	2	3	4	5	
Overall, the questions were understandable	1	2	3	4	5	
Overall, the instructions were easy to understand	1	2	3	4	5	

Overall, the story presented was easy to follow	1	2	3	4	5	
Overall, this task was easy	1	2	3	4	5	
Overall, the information provided is effective in helping me complete the task	1	2	3	4	5	
In the SE version, there were no disturbing noises	1	2	3	4	5	
In the SE version, the instructions were clear	1	2	3	4	5	
In the BSL version, the instructions were clear	1	2	3	4	5	
In the SAE version , the instructions were clear	1	2	3	4	5	

Control questions:

a) Why did Mary go home? _____

b) What did the ice-cream man tell Mary? _____

c) What did the ice-cream man tell John? _____

d) Does John know that the ice-cream man went to school? Yes No

e) Does John know that the ice-cream man told Mary he was going to the school? Yes No

f) Now, please read the script of the story and answer the following questions:
Having read the script, how accurately do you think the video reflects the script?

Not accurately at all 1 2 3 4 5 Very accurately

Why?_____
_____

3. THE SMARTIES/CEREAL BOX TASK:	**Strongly disagree**				**Strongly agree**	**N/A**
Overall, in the three versions (SE, BSL, SAE) the storyline was clear	1	2	3	4	5	
Overall, the narrator's pace was appropriate	1	2	3	4	5	
Overall, the animated story was clear	1	2	3	4	5	
Overall, the questions were understandable	1	2	3	4	5	
Overall, the instructions were easy to understand	1	2	3	4	5	
Overall, the story presented was easy to follow	1	2	3	4	5	
Overall, this task was easy	1	2	3	4	5	
Overall, the information provided is effective in helping me complete the task	1	2	3	4	5	
In the SE version, there were no disturbing noises	1	2	3	4	5	
In the SE version, the instructions were clear	1	2	3	4	5	
In the BSL version, the instructions were clear	1	2	3	4	5	
In the SAE version , the instructions were clear	1	2	3	4	5	

Control questions:

a) What was inside the tube/box? _____

b) What did the boy think was inside the tube/box before we looked inside? _____

c) Now, please read the script of the story and answer the following questions:
Having read the script, how accurately do you think the video reflects the script?
Not accurately at all 1 2 3 4 5 Very accurately

Why?_____
_____

4. THE HIDDEN CAKES TASK:	Strongly disagree				Strongly agree	N/A
Overall, in the three versions (SE, BSL, SAE) the storyline was clear	1	2	3	4	5	
Overall, the narrator's pace was appropriate	1	2	3	4	5	
Overall, the animated story was clear	1	2	3	4	5	
Overall, the questions were understandable	1	2	3	4	5	
Overall, the instructions were easy to understand	1	2	3	4	5	
Overall, the story presented was easy to follow	1	2	3	4	5	
Overall, this task was easy	1	2	3	4	5	
Overall, the information provided is effective in helping me complete the task	1	2	3	4	5	
In the SE version, there were no disturbing noises	1	2	3	4	5	
In the SE version, the instructions were clear	1	2	3	4	5	
In the BSL version, the instructions were clear	1	2	3	4	5	
In the SAE version , the instructions were clear	1	2	3	4	5	

Control questions:

a) Where are the cakes at the end of the story? _____

b) Where did the boy put the cakes in the beginning? _____

c) Now, please read the script of the story and answer the following questions:
Having read the script, how accurately do you think the video reflects the script?

Not accurately at all 1 2 3 4 5 Very accurately

Why?_____
_____

OVERALL, list the most positive aspect(s) of the videos:

1.
2.
3.

List the most negative aspect(s) of the videos:

1.
2.
3.

How could we improve the videos?

_____
_____

Thank you for your collaboration

Appendix 6

TABLE 3.5. FACTORS INVOLVED IN THE DECISION-MAKING PROCESS - continued

Characteristics	
Prior research experience *n* (%)	4 (28.6)
Prior experience as SDM *n* (%)	1 (7.1)
Perceived physical health status *n* (%)	
Very poor	1 (7.1)
Poor	3 (21.4)
Moderate	4 (28.6)
Good	5 (35.7)
Very good	1 (7.1)
Perceived mental health status *n* (%)	
Very poor	1(7.1)
Poor	4 (28.6)
Moderate	3 (21.4)
Good	6 (42.9)
Time between patient's admission and research approach *n* (%)	
<24hr	4 (28.6)
24-48hr	3 (21.4)
>48hr	7 (50)
Clinical update form clinician before research approach *n* (%)	
Yes	11 (78.6)
No	3 (21.4)
Prior conversation with patient about attitute towards research *n* (%)	2 (14.3)

N=14.

Appendix 8

EXAMPLE FROM THE QUALITATIVE ANALYSIS:

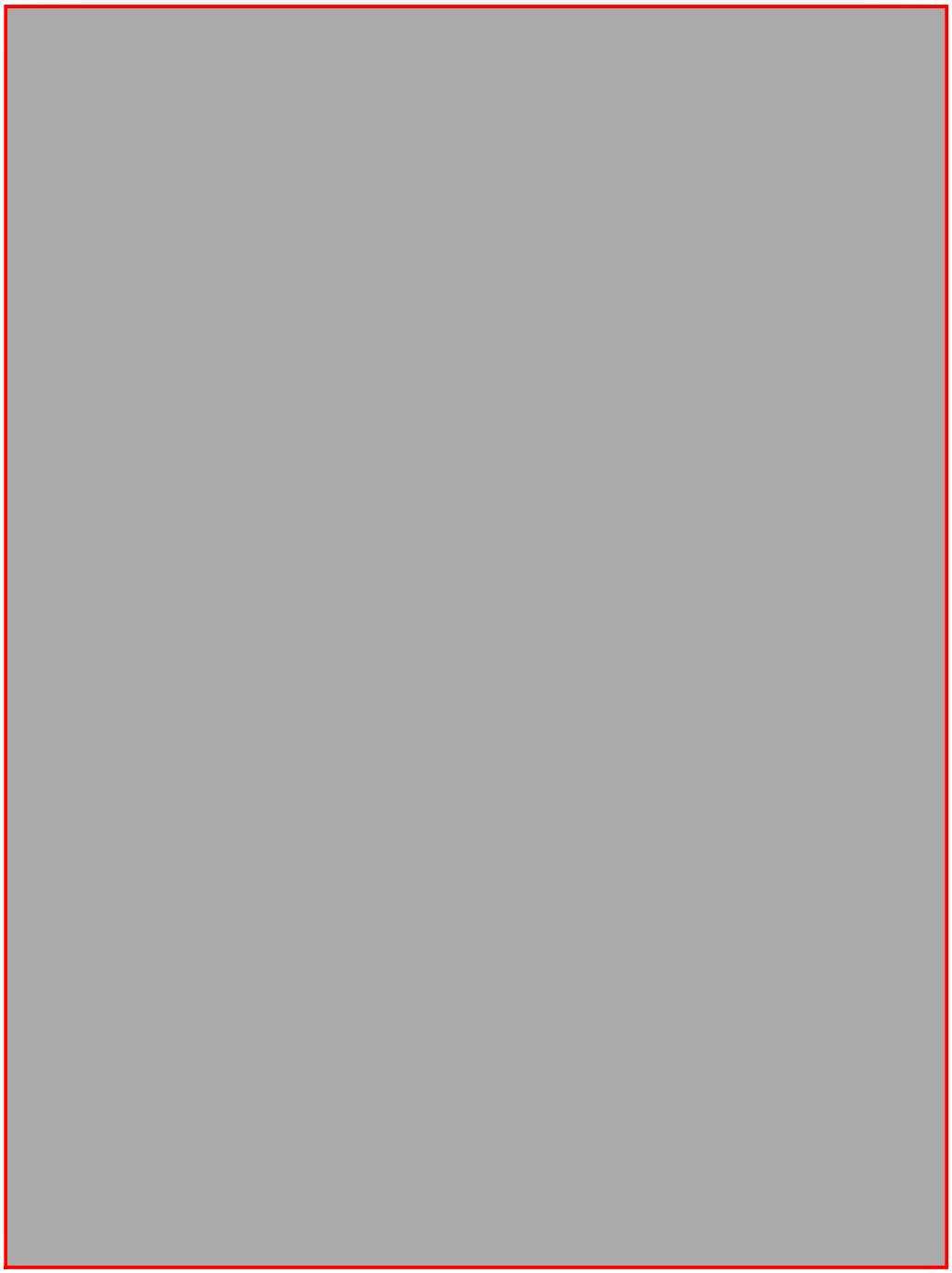


Figure 3.6. The Figure shows the initial stages of the qualitative analysis of one extract from an interview conducted with an SDM. In the inductive thematic analysis, the extracts were first read a number of times. Interesting or significant aspects were annotated using the right-hand margin. In the left margin were then reported the emerging theme titles.

Appendix 9

THEME: 'THE TIME AT THE ICU: A DRAINING ROLLERCOASTER'

For the next-of-kin, the time at the ICU seems characterised by a pervasive uncertainty about their loved one's condition:

> Mrs Best: […] we don't know we can't do anything for her for the way she is so we are sitting here without knowing anything …

A sense of helplessness seems also to characterise a waiting time, where the next-of-kin cannot really do anything else rather than wait and hope that the doctors, the machines, the patient and God will do something:

> Mrs Best: […] at the moment [inaudible] all the work the machines are helping her [inaudible] it's up to mum to pull through but we don't know if that is gonna happen we don't know please God it will I'm sure but it's very hard I mean [inaudible] 24 hours [inaudible] we think maybe 6 o'clock 8 o'clock tonight 24 hours things might be a little bit better but…

The SDMs appear also emotionally drained as they are going through an emotional rollercoaster. Such fluctuation of emotions seems to be connected with the nature itself of the ICU which exposes them to the consistent possibility of receiving stressful communications:

> Mrs Green: […] so the actual environment makes you drained anyway […] you know one minute you can be alright and relaxed and then you get to speak to a doctor that's [inaudible] stress but then the nurse comes back and you calm down a bit. […]
> Miss Green: It's a bit of up and down
>
> Miss Jones: I think it's just a constant stress so I don't know how it deviates from that so I feel like stressed

References

Antoniou, E. E., Draper, H., Reed, K., Burls, A., Southwood, T. R., Zeegers, M. P. (2011). An empirical study on the preferred size of the participant information sheet in research. *Journal of Medical Ethics*, 37,557-562.

Appelbaum, P. S., Grisso, T. (2001). *MacCAT-CR: MacArthur Competence Assessment Tool for Clinical Research.* Sarasota, FL: Professional Resource Press.

Azoulay, E., Chevret, S., Leleu, G., Pochard, F., Barboteu, M., Adrie, C., . . . Schlemmer, B. (2000). Half the families of intensive care unit patients experience inadequate communication with physicians. *Critical Care Medicine, 28*, 3044-3049.

Azoulay, E., Pochard, F., Chevret, S., Adrie, C., Annane, D., Bleichner, G., . . . Schlemmer, B. (2004). Half the family members of intensive care unit patients do not want to share in the decision-making process: a study in 78 French intensive care units. *Critical Care Medicine,* 32, 1832–1838.

Barrett, K. A., & Scales, D. C. (2012). Considering the vulnerabilities of surrogate decision-makers when obtaining consent for critical care research. *Intensive Care Medicine, 38*, 4-6.

Barrett, K. A., Ferguson, N. D., Athaide, V., Cook, D. J., Friedrich, J. O., McDonald, E., . . . Scales, D. C. (2012). Surrogate decision makers' attitudes towards research decision making for critically ill patients. *Intensive Care Medicine, 38*, 1616-1623.

Braun, V. & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, 3, 77-101.

Burns, K. E. A., Zubrinich, C., Marshall, J., & Cook, D. (2009). The 'consent to research' paradigm in critical care: Challenges and potential solutions. *Intensive Care Medicine, 35*, 1655-1658.

Chappuy, H., Baruchel, A., Leverger, G., Oudot, C., Brethon, B., Haouy, S., . . . Treluyer, J. M. (2010). Parental comprehension and satisfaction in informed consent in paediatric clinical trials: A prospective study on childhood leukaemia. *Archives of Disease in Childhood, 95*, 800-804.

Chenaud, C., Merlani, P., Verdon, M., & Ricou, B. (2009). Who should consent for research in adult intensive care? Preferences of patients and their proxies: A pilot study. *Journal of Medical Ethics, 35*, 709-712.

Ciroldi, M., Cariou, A., Adrie, C., Annane, D., Castelain, V., Cohen, Y., . . . Famirea study, group (2007). Ability of family members to predict patient's consent to critical care research. *Intensive Care Medicine, 33*, 807-813.

Cutting, A. L., & Dunn, J. (1999). Theory of mind, emotion understanding, language, and family background: Individual differences and interrelations. *Child Development*, 70, 853 – 865.

Dunn, L. B., Nowrangi, M. A., Palmer, B. W., Jeste, D. V., & Saks, E. R. (2006). Assessing Decisional Capacity for Clinical Research or Treatment: A Review of Instruments. *American Journal of Psychiatry*, 163, 1323-1334.

Gigerenzer, G. (2002). *Reckoning with Risk: Learning to Live with Uncertainty*. London: Allen Lane The Penguin Press.

Giorgi, A. (1995). Phenomenological psychology. In J.A. Smith, R. Haré & L. Van Langenhove (Eds.), *Rethinking Psychology*. (pp. 24-42). London: Sage.

Great Britain. Department for Constitutional Affairs. (2007). *Mental Capacity Act 2005: Code of Practice.* London: The Stationary Office

Henderson, G. E., Churchill, L. R., Davis, A. M., Easter, M. M., Grady, C., Joffe, S., . . . Zimmer, C. R. (2007, November 27). Clinical trials and medical care: Defining the therapeutic misconception. *PLoS Medicine, 4 Article* e324. Retrieved October 1, 2013, from http://www.plosmedicine.org/article/info%3Adoi%2F10.1371%2Fjournal.pmed.0040324

Heyland, D. K., Cook, D. J., Rocker, G. M., Dodek, P. M., Kutsogiannis, D. J., Peters, S., . . . O'Callaghan, C. J. (2003). Decision-making in the ICU: Perspectives of the substitute decision-maker. *Intensive Care Medicine,* 29, 75-82.

Hoffrage, U., Gigerenzer, G. (1998). Using natural frequencies to improve diagnostic inferences. *Academic Medicine*, 73,538-40.

Howell, D. C. (2002). *Statistical methods for psychology*. Fifth Edition. Belmont, CA: Wadsworth.

IBM Corp. Released 2012. IBM SPSS Statistics for Windows, Version 21.0. Armonk, NY: IBM Corp.

Jackson, A. L. (2001). Language facility and theory of mind development in deaf children. *Journal of Deaf Studies and Deaf Education*, 6, 161 – 176.

Jenkins, J. M., & Astington, J. W. (2000). Theory of mind and social behavior: Causal models tested in a longitudinal study. *Merrill-Palmer Quarterly-Journal of Developmental Psychology*, 46, 203–220.

Jeste, D. V., Palmer, B. W., Appelbaum, P. S., Golshan, S., Glorioso, D., Dunn, L. B., . . . Kraemer, H. C. (2007). A new brief instrument for assessing decisional capacity for clinical research. *Archives of General Psychiatry, 64*, 966-974.

King, N. (2004). Using interviews in qualitative research. In C.M. Cassell & G. Symon (Eds.), *Essential Guide to Qualitative Methods in Organizational Research*. (p. 11). London: Sage.

Knaus, W. A., Draper, E. A, Wagner, D. P. & Zimmerman, J. E. (1984). Apache II: A severity of disease classification system. *Critical Care Medicine*, 13, 818-829.

Luce, J. M., Cook, D. J., Martin, T. R., Angus, D. C., Boushey, H. A., Curtis, J. R., . . . American Thoracic, S. (2004). The ethical conduct of clinical research involving critically ill patients in the United States and Canada: Principles and recommendations. *American Journal of Respiratory & Critical Care Medicine, 170*, 1375-1384.

Lundy, J. (2002). Age and language skills of deaf children in relation to theory of mind development. *Journal of Deaf Studies and Deaf Education*, 7, 41 – 56.

Madill, A., Jordan, A. & Shirley, C. (2000). Objectivity and reliability in qualitative analysis: realist, contextualist, radical constructionist epistemologies. *British Journal of Psychology*, 91, 1-20.

Majesko, A., Hong, S. Y., Weissfeld, L., & White, D. B. (2012). Identifying family members who may struggle in the role of surrogate decision maker. *Critical Care Medicine, 40*, 2281-2286.

Mehta, S., Quittnat Pelletier, F., Brown, M., Ethier, C., Wells, D., Burry, L., & MacDonald, R. (2012). Why substitute decision makers provide or decline consent for ICU research studies: A questionnaire study. *Intensive Care Medicine, 38*, 47-54.

Menon, K., Ward, R. E., Gaboury, I., Thomas, M., Joffe, A., Burns, K., & Cook, D. (2012). Factors affecting consent in pediatric critical care research. *Intensive Care Medicine, 38*, 153-159.

Perner, A., Ibsen, M., & Bonde, J. (2010). Attitudes to drug trials among proxies of unconscious intensive care patients. *BMC Anesthesiology,* 10, 6.

Peters, K., Remmel, E., & Richards, D. (2009). Language, mental state vocabulary, and false belief understanding in children with cochlear implants. *Language, Speech & Hearing Services in Schools*, 40, 245- 255.

Peterson, C., & Siegal, M. (1999). Representing inner worlds: Theory of mind in autistic, deaf, and normal hearing children. *American Psychological Society*, 10, 126 – 129.

Pochard, F., Azoulay, E., Chevret, S., Lemaire, F., Hubert, P., Canoui, P., . . . French FAMIREA, G. (2001). Symptoms of anxiety and depression in family members of intensive care unit patients: Ethical hypothesis regarding decision-making capacity. *Critical Care Medicine, 29*, 1893-1897.

Rosenstein, D. L. (2004). Decision-Making Capacity and Disaster Research. *Journal of Traumatic Stress*. 17, 373-381.

Vitae®, Careers Research and Advisory Centre (CRAC) Limited (2010). Research Development Framework. Retrieved March 26, 2013, from http://www.vitae.ac.uk/CMS/files/upload/Vitae-Researcher-Development-Framework.pdf

Scales, D. C., Smith, O. M., Pinto, R., Barrett, K. A., Friedrich, J. O., Lazar, N. M., . . . Ferguson, N. D. (2009). Patients' preferences for enrolment into critical-care trials. *Intensive Care Medicine, 35*, 1703-1712.

Silverman, H.J., Druml, C., Lemaire, F., Nelson, R. (2004). The European Union directive and the protection of incapacitated subjects in research: an ethical analysis. I*ntensive Care Medicine, 35*, 1723–1729.

Smith, J. A. (1996). Beyond the divide between cognition and discourse: Using interpretative phenomenological analysis in health psychology. *Psychology & Health, 11*, 261-271.

Sullivan, K., Zaitchik, D. & Tager-Flusberg, H. (1994). Preschoolers can attribute second order beliefs. *Developmental Psychology*, 30, 395-402

Watson, A. C., Nixon, C. L., Wilson, A., & Capage, L. (1999). Social interaction skills and theory of mind in young children. *Developmental Psychology*, 35, 386-391.

Willig, C. (2013). *Introducing qualitative research in psychology*. Third Edition. Berkshire, UK: Open University Press/ McGraw Hill.