The after effects and social consequences of encephalitis

Project Summary of Findings

Introduction to the Completed Project

Encephalitis is a life-threatening neurological illness which refers to inflammation of the brain parenchyma. The illness may result from a number of different causal agents including herpes simplex type 1 ('the cold sore virus'). Although, in more than half of the cases of viral encephalitis, the exact causal agent is never known (Booss and Esiri, 2003). The illness is also complex to diagnose, although application of the Liverpool diagnostic algorithm for viral encephalitis may aid the differential diagnosis (Solomon, Hart and Beeching, 2007).

After acute encephalitis, individuals can be left with an acquired brain injury. After herpes simplex encephalitis, almost two thirds of child and adult survivors of the illness experience significant post-encephalitic sequelae in spite of early administration of anti-viral therapy (Whitley and Kimberlin, 2005; McGrath, 1997). Neuropsychological sequelae are predominant, in particular anterograde amnesia, anomia, semantic memory deficit, retrograde amnesia and executive dysfunction (see Hokkanen and Launes, 2000). Significant morbidity can also result from encephalitis due to other aetiologies.

Hospital episodes of encephalitis have tended to go unreported (Davison et al. 2003), even though the illness has been a notifiable infection since 1918 under Public Health (Infectious Diseases) Regulations. Consequently, there are no accurate prevalence rates for the illness within the U.K. (A study by The Health Protection Agency into the aetiology of encephalitis in England is ongoing). Drawing on U.S. statistics, a U.K. figure has been estimated of 4,000 new cases of the illness per year (Easton, Atkin and Dowell, 2006). However, because of the likelihood of post-encephalitic brain injury, there remains a need for individuals and their families to have access to a variety of services for many years after the illness.

In 2008, the Department of Health Sciences at the University of York completed a research project into the after effects and social consequences of encephalitis. The project was conducted in collaboration with the Encephalitis Society, an organisation of international reputation which has become a seminal resource for support, information and research into encephalitis for individuals who have had the illness, their families, and for health and social care professionals. The Encephalitis Society has run a number of successful seminars for health and social care professionals in recent years, attracting key figures from neuropsychology and cognitive rehabilitation such as Professor Barbara Wilson who is also the Society's President.

The project focussed on post-encephalitic chronic pain and central pain states. The prevalence of chronic pain after traumatic brain injury has been estimated between 22% and 95% (see Ofek and Defrin, 2007). Central pain after traumatic brain injury can also occur, where there is damage to the central nervous system itself (see Tyrer and Davis, 2005; also Ofek and Defrin, 2007). Central pain states include allodynia (in which a painful response is triggered by a normally non-noxious stimulus) and hyperalgesia (where there might be an increased response to a stimulus which is not normally considered painful). With regard to encephalitis, both chronic pain and central pain have been reported in the literature in particular chronic headache (see Haglund and Günther, 2003).

The mixed methods project comprised a postal survey and interviews with individuals who had had encephalitis, their family members (carers) and also with different service practitioners. These professionals included neurologists, neuropsychologists, occupational therapists and social workers. Key aims of the project were: to investigate the range and frequency of self-reported post-encephalitic sequelae including chronic and central pain; to provide individual, family carer and practitioner perspectives on living with the social consequences of these sequelae; and to evaluate provision of and engagement with service support.

The University of York

The Encephalitis Society
action for support, awareness, and research
www.encephalitis.info

supported by
BIG LOTTERY FUND

Findings from the postal survey

A postal survey was sent to all adult members of the Encephalitis Society (n=1281). The survey was devised based on expert appraisal and a knowledge review of previous research. It comprised a number of question items including those on chronic and central pain. The questionnaire also included the SF-36 - version 2 (a health-related quality of life measure) and the Brief Inventory - Short Form. A high response rate of 59.4% was achieved (n=717 usable returns).

Respondent Profile

	Survey Sample		Men		Women	
Frequency	n=717		n=303 (42.5%)		n=410 (57.5%)	
	Mean		**Mean**		**Mean**	
Age at self-report	48.0 (SD=15.4)		48.1 (SD=15.3)		47.9 (SD=15.5)	
Age when had encephalitis	37.26 (SD=18.4)		37.25 (SD=18.8)		37.26 (SD=18.2)	
Years since the illness	10.6 (SD=11.1)		10.7 (SD=11.3)		10.5 (SD=11.0)	
	Percentage		**Frequency**			
Ethnicity	White British	90.5%	White British	277	White British	370
	White Irish	3.9%	White Irish	10	White Irish	18
	Other White	2.4%	Other White	6	Other White	11

- Most respondents had had encephalitis within the last 15 years
- 43.8% felt they could no longer do paid work due to encephalitis
- 73.5% felt their earning ability had been affected due to encephalitis
- 33.0% did not know the cause of their encephalitis; 19.6% were unsure
- 28.2% reported having post-encephalitic epilepsy
- 70% (or more) reported fatigue, concentration & memory problems
- 60 to 69% reported depressed feelings, personality change & impaired self-confidence
- 58.6% (n=420) attributed chronic and/or central pain to encephalitis

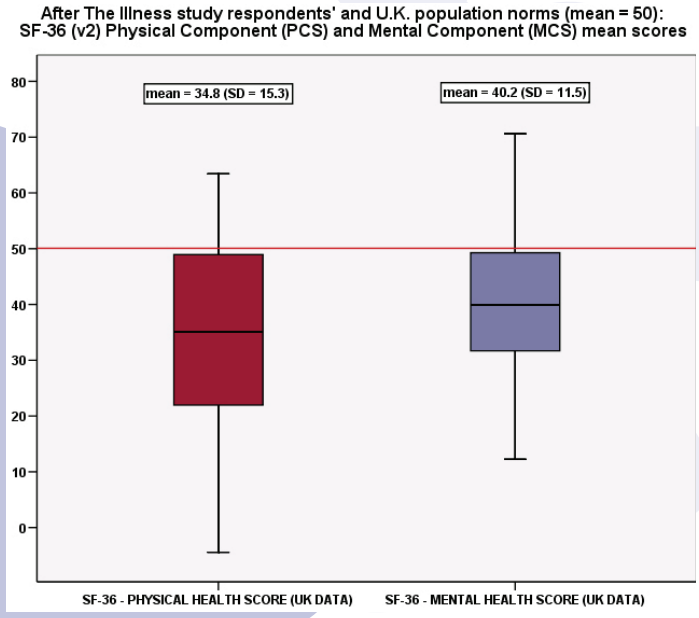
Post-Encephalitic Chronic Pain and Central Pain States (n=420)

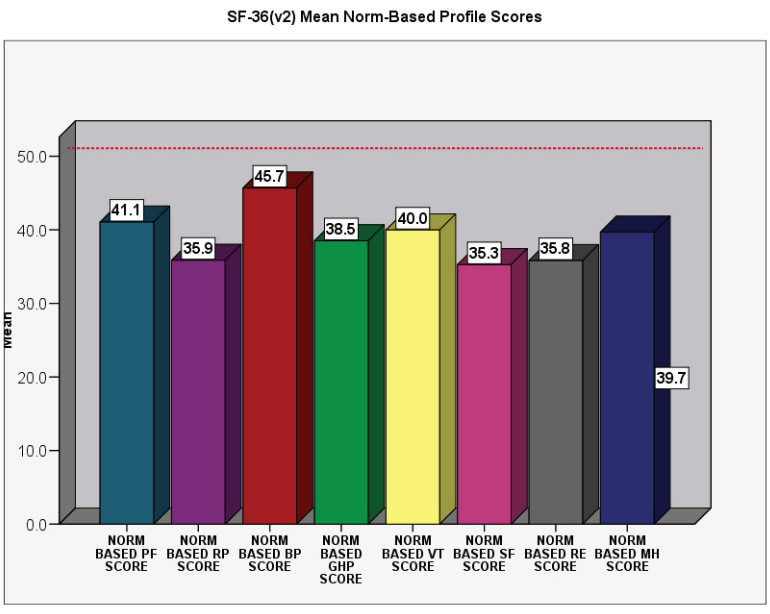
- **Only 9.4% had attended a pain management clinic**
- Preliminary evidence for central pain states:
  - thermal hypo- & hyperalgesia (48.2%); allodynia (51.8%)
  - central pain descriptors from interviews
- Locations for pain: head (56.9%); legs (51.9%); hands (50.2%)
  - head pain – unrelated to epilepsy or gender
- Head pain – reported up to 8 years after encephalitis by n=236 (32.9% of n=717)
  - service provider interviews: chronic headache after ABI is common & intractable
- Brief Pain Inventory indices and the SF-36(v2)
  - significant associations between chronic pain and health-related quality of life
    - Pain Severity Index: mean = 4.49 (SD=2.30)
    - significant negative correlation for SF-36 PCS (r= -0.53, p=0.000) & large effect size (r^2=0.28)
    - significant negative correlation for SF-36 MCS (r= -0.26, p=0.000) & small effect size (r^2=0.07)
    - Pain Interference Index: mean = 5.29 (SD=2.80)
    - significant negative correlation for SF-36 PCS (r= -0.63, p=0.000) & large effect size (r^2=0.39)
    - significant negative correlation for SF-36 MCS (r= -0.26, p=0.000) & medium effect size (r^2=0.15)

Summary of Key Findings:

- Post-encephalitic sequelae (including chronic/central pain) can persist for several years after the illness
- Chronic headache after encephalitis should be considered for inclusion in the International Headache Society's ICHD-2 classification
- SF-36 summary and profile scores were worse than U.K. population norms (and SF-36 data from studies into other acquired brain injuries)
  - respondent cluster (n=201)
    - worse SF-36 MCS & PCS and greater impact of pain
  - mean scores (n=201)
  - SF-36 MCS =33.1; SF-36 CS=24.1
  - BPI Pain Severity = 4.9
  - BPI Pain Interference = 6.1

SF-36 (version 2): Summary Physical and Mental Component Scores and Mean Norm-Based Profile Scores in a U.K. Post-Encephalitic Population





Findings from the Interviews with individuals who have had encephalitis

Twenty three interviews were conducted with individuals who had had encephalitis. These individuals had been selected from those who had volunteered to participate after completing the postal survey. Selection criteria (based on both purposive sampling and stratified purposeful sampling) included gender, chronic headache and years since the illness. Interviewees ranged in age from twenty nine to seventy four; fourteen women were interviewed and nine men. Thematic analyses of all three sets of interviews were facilitated by Atlas.ti 5.0 qualitative software. During their interviews, individuals were able to explicate the meaning of post-encephalitic sequelae within their everyday lives, suggesting the social consequences of encephalitis in terms of work, family relationships and their sense of identity. In addition interviewees discussed their engagement with services and in particular their experiences of service delivery beyond the post-acute illness phase.

Interview accounts suggested that people had difficulty understanding that long-term sequelae can result from acute illness. Reported sequelae included sensory overload, retrograde amnesia, anterograde amnesia, prosopagnosia and executive dysfunction. Sensory overload referred to the inability to 'filter' sensory input from the environment. Accounts of pain and fatigue were common and expressed in terms of uncertainty, whereby variable sequelae were negotiated within people's everyday lives. Head pain was often reported as constant 'background' pain, which at times could 'break through' or worsen. Pain and fatigue fluctuated with regard to 'breakthrough' pain and 'burn-out' respectively. In particular, when such sequelae were severe, these could intrude on work, social activities and social relationships. Notably, central pain descriptors were evident from interviewees' accounts. Exemplar descriptors included 'burning pain', 'dry rot' and 'very heavy pins and needles'.

Key concerns for many of the interviewees included reconciling loss and revising social roles and relationships. This was in terms of losing prior skills and activities but in particular related to a loss of self or becoming a changed person. Loss of self-confidence was apparent which sometimes also related to loss of work identity. Several interviewees reported changing their jobs or careers or having to give up work completely as a consequence of encephalitis, which entailed the loss of a key aspect of their identity. The impact of encephalitis on individuals' work opportunities meant a reduction in family income; for example, going from a salaried income to receipt of welfare benefits. However, when applying for welfare benefits such as Disability Living Allowance, interviewees' accounts suggested that welfare benefits agencies did not understand the complexities and implications of acquired brain injury, particularly in relation to cognitive impairment. There was often a continued fight for individuals to 'prove' their disability within a welfare benefits system which seemed set to disbelieve and stigmatise them. When welfare benefits and insurance claims were in dispute, this process was stressful and could mean severe financial hardship for individuals and their families in the interim.

Interviews suggested that individuals and their families were often managing the consequences of encephalitis in isolation from statutory services. Variability in service provision or a 'gap' between acute and follow-up services was supported by the interview accounts of service providers themselves. However, individuals reported needing access to services such as neuropsychology in order to validate their experience (post-encephalitic sequelae) and so inform their coping and reassert their sense of self. In addition, with regard to pain, engagement with services could be inhibited by individuals' difficulty in articulating pain, especially central pain states. Similarly, finding a language to describe the long-term effects of encephalitis was difficult. Post-encephalitic sequelae cannot be described as a syndrome or as a chronic condition. This may be why many individuals discussed encephalitis as an ongoing illness, even many years after the acute illness phase.

Case: John (aged 36; chronic head pain)

- Encephalitis in 2003 (unknown cause)
- 2 weeks in local hospital; home for 1 week; 3 weeks in regional hospital where diagnosed
- Memory problems and severe intractable head pain since the illness 4 years ago
- Morphine injections for 'breakthrough' head pain
- Rehabilitation for anger management
- Attended pain management clinic
- Unable to work due to head pain and memory problems
- Fight to secure insurance claim and welfare benefits
- Impact of head pain on family relationships

"My life's ruled by it basically. So everybody else is living by what I do, so their lives are ruled by headaches as well. So if I'm crabbier one morning, they'll know to leave me alone…It's consuming. It totally consumes you..everything…My pain is just constant and it's not as painful as when I was first diagnosed with the encephalitis. It borders on a 10 but it never goes over a 10. A 10 is pretty intolerable as far as I'm concerned. It's really bad. On an average day, it will be about a 7 to 8".

Interview Exemplars of Central Pain

- increased/decreased sensitivity to heat/cold
- sensitivity to having hair brushed
- sensitivity to shower water pressure
- "burning head pain"
- "maggots in my head"
- "a very heavy pins and needles"
- "like I've got a log tied to my leg"
- "leg aches like toothache"
- "just like dry rot"
- "like bedsores"
- "nerve pain like walking on cotton wool"
- "having your skin ripped off, being beaten with a baseball bat and then being put in a vat of vinegar"

Summary of Key Findings:

- Difficulty defining the long-term consequences of encephalitis after acute illness and difficulty in articulating pain
- Reconciling loss and revising social relationships after encephalitis
- Managing post-encephalitic sequelae in isolation from statutory services
- Misunderstanding of acquired brain injury by welfare benefits agencies

Findings from the Interviews with family members

"Well, I've often used the analogy that [on] (date of husband's illness) at just after 3 o'clock in the afternoon probably has had the same impact on me and the family unit as somebody that undergoes a bomb blast, an earthquake, a tsunami. It hits without warning and the devastation and the consequences, the fallout that comes from it can be never ending. And because nobody warns you, it's just basically survival tactics you're using initially on a day to day basis and to make sense of it. But somewhere along the line you have got to pick the pieces up and rebuild and it won't be the same as it was before".

Philip's wife: 5 ½ years after the illness

Eighteen interviews were conducted with the family members (carers) of those who had had encephalitis. Their participation was with the consent of their relatives who had also nominated them to take part. Most interviewees were the husbands or wives of those who had a post-encephalitic acquired brain injury.

Family members reported a number of different ways in which encephalitis and its sequelae had impacted on the family and family relationships. For example, family members discussed consequences of the illness such as redistributing domestic tasks or taking on responsibility for the family finances. Several accounts referred to family members observing their relatives' loss of self-confidence due to encephalitis or to their altered mode of self-expression; for example, no longer being the driving force behind the family or having problems with self-initialisation. Moreover, interviewees reported that coming to terms with their relatives' altered personality was particularly difficult. Certainly, across many of the family member interviews, rebuilding a relationship with a 'different' person was evident, either because the individual him- or herself had changed or because responsibilities within the family had shifted. Some family member accounts even suggested having to reconcile 'grief' for the person they had lost whilst at the same time learning to live with the person who had survived the illness. In this way, reconciling loss was a dominant theme across both the interviews with individuals who had had encephalitis and with members of their families.

With regard to their own situations and needs, family members were often reluctant to perceive themselves as 'carers'. This was in spite of performing a variety of different care and support roles such as personal care and prompting completion of everyday tasks. There was the suggestion that family members assumed a sense of emotional and moral responsibility for their relatives, with such responsibility continually renegotiated in terms of variable post-encephalitic sequelae over time and in relation to family members' ongoing acceptance and accommodation of such sequelae and their social consequences. This responsibility was another means by which family relationships were altered, in that the nature of family relationships became dissonant by challenging normative assumptions; for example, being 'carer and patient' rather than 'husband and wife'. Importantly, care-giving was seen as normative or legitimate within the context of expectations of family relationships. Family members' caring roles were subsumed within notions of family obligation, so that family members did not view their needs as separate from those of their relatives.

Family member accounts also suggested ongoing, difficult engagement with health and social care professionals in order to secure service support for their relatives. Family members discussed taking on responsibility for their relatives' rehabilitation following the acute illness. Similar to individuals themselves who wanted expert help to implement effective coping strategies, many carers wanted their rehabilitative care efforts validated in terms of information and guidance from health professionals. Certainly family members did not report health and social care professionals as focusing on their separate needs as carers. Information and service support for both individuals and their carers was often reported as variable. Furthermore, because their care roles were viewed as normative, 'carer as resource' (or perhaps 'carer as co-worker') was a role to which family members were vulnerable. The blurred boundary between normative and rehabilitative care may mean 'carer as resource' is perhaps a role to which family members are readily assigned during negotiations with health and social care agencies.

Summary of Key Findings:

- Reconciling loss due to encephalitis whilst rebuilding family life and relationships
- Care-giving as normative within expectations of family relationships
- Variation in service support for individuals and their family carers
- Health and social care professionals not addressing carers' needs separately from those of their brain-injured relatives
- Blurred boundary between 'normative' and 'rehabilitative' care with a positioning of 'carer as resource'

"If you know what the worst can be, you hope for something in the middle. But at least you're prepared for whatever happens. Whereas now, we're just sort of fumbling along in the dark…it just seemed as if you're ill, you're in hospital, you're discharged. Cheerio!"

Jenny's husband, 14 years since the illness

Findings from the Interviews with service providers

"I think it's absolutely haphazard and pot luck whether they get through to the right people or not. And that's partly if they've got the relatives asking the right questions or making a fuss. It's partly the medical people that they've seen first, if they're knowledgeable about rehab. But I'm sure many, many, many of them never get to see a neuropsychologist or get support that they need or that they could have. I don't think it's a good situation in general".

Professional 1: Clinical Psychologist

"The idea that everybody with a serious brain injury or for that matter any serious long-term condition should have a main person in the community that they can contact as the first point of call for a problem is a joke really. Unfortunately".

Professional 4: Clinical Consultant in Rehabilitation Medicine, discussing the NSF for Long-Term (Neurological) Conditions

Telephone interviews were conducted with eighteen service practitioners from the fields of acquired brain injury and pain management. These practitioners included neurologists, psychologists, occupational therapists, nurses, social workers and experts in chronic and central pain. Research governance was agreed with each individual NHS Trust concerned. Analyses focussed on service delivery in acquired brain injury whilst also enabling comparison with themes from the interviews with individuals and family members.

Service provider accounts suggested that acute encephalitis is a problematic diagnosis, particularly in cases of mild presentation where delayed or psychiatric misdiagnoses are possible. These accounts concurred with those of individuals who had had encephalitis, for whom a psychiatric misdiagnosis was difficult to reconcile with their experience of the illness and its sequelae. In addition, a delayed diagnosis might mean a delay in administration of anti-viral and other therapies and so the risk of poorer functional outcomes. Professionals identified a range of post-encephalitic sequelae but described cognitive impairments, personality and behaviour change as common post-encephalitic sequelae seen within their services. They also supported the idea that chronic head pain was a common, sometimes intractable sequela of acquired brain injury and one which some viewed as more likely to be due to central nervous system damage rather than to analgesic-rebound headache. Service provider accounts of 'cognitive fatigue' were concordant with individual interviewees' reports of 'sensory overload'. Together with chronic pain, practitioners supported the possibility of central (neuropathic) pain as a sequela of the illness, suggesting (as was apparent from the interviews with those who had had encephalitis) that both chronic and central pain may be difficult for individuals who have had the illness to articulate.

"It's not all uncommon with neuropathic pain for people to find it very difficult to describe their symptoms…very, very, very commonly with neuropathic pain, the description of the pain is weird. Some patients feel embarrassed to say what it feels like. And that may be a very good reason why. It's a bit like a distortion in your hi-fi system…They're often made to feel it's their fault. Some of them feel guilty because they can't explain it, because there's no easy answer, because there's nothing to see. And sometimes they perhaps just give up in despair because nobody really understands what they're talking about".

Professional 10: Consultant in Chronic Pain Management and Anaesthesia

As in the interviews with individuals and carers, managing issues of loss after acquired brain injury was a theme identified by practitioners as central to their practice for both individuals and family carers. Service provider interviewee accounts suggested some shift towards regarding 'carers as co-clients'. Lack of resources meant that service delivery for both individuals and particularly carers was sometimes precluded, so that effective implementation of the NSF for Long-Term Neurological Conditions was restricted, as was support for families in the longer term. Moreover, practitioners perceived service delivery as variable with 'pockets' of good services only. In particular, practitioner interviewees suggested that those with cognitive impairments ("the walking wounded") could find such sequelae were missed during the post-acute illness phase. In discussion of post-acute services following encephalitis, service provider interviewees described how cognitive rehabilitation might be excluded from practitioners' representations of what 'rehabilitation' encompassed. There may be insufficient focus on the cognitive, emotional and psychosocial sequelae with regard to acquired brain injuries. Therefore the role of cognitive rehabilitation and neuropsychology in rehabilitation should be emphasised and adequately resourced. Post-encephalitic care pathways are imperative.

Summary of Key Findings:

- Variability in service provision and a 'gap' between acute and follow-up care
- Excluding neuropsychology and cognitive rehabilitation from representations of 'rehabilitation'
- Poorer care for those with cognitive impairments ('the walking wounded')
- Lack of adequate resources to implement the NSF for Long-Term Neurological Conditions

Summary

- Both chronic and central pain can persist as post-encephalitic sequelae many years after the acute illness.
- Chronic headache is a common problem after acquired brain injury and is also prevalent after encephalitis. It is currently not recognised within the International Headache Society's ICHD-2 classification.
- Articulating chronic and central pain is difficult for individuals and this may complicate their engagement with service providers. Individuals sometimes experience difficulty in finding the words to describe what they are going through.
- Individuals who have chronic/central pain due to encephalitis would benefit from access to pain management services. Few report access to such provision.
- SF-36(v2) summary and profile scores in a U.K. post-encephalitic group are worse than SF-36 U.K. population norms. (They are also worse when compared with SF-36 data from populations with other acquired brain injuries).
- A significant group of individuals report poor outcomes after encephalitis, particularly in relation to chronic/central pain and health-related quality of life. This group represents a key target group for service support and intervention.
- Understanding that an acute illness can result in acquired brain injury may be difficult for individuals and their families. This tension may be complicated by the lack of language and linguistic strategies to define the after effects of encephalitis in the longer term.
- Individuals and families need to reconcile a sense of loss after encephalitis, particularly in terms of the individual's altered personality resulting from the illness.
- Family members may perceive the role of 'carer' as normative and legitimate within the context of family relationships and family obligations. (They do not see themselves as carers).
- Family members' lives are impacted by looking after someone following encephalitis. There may be financial implications such as those associated with loss of family income as well as the need to assume the practical tasks of caring for someone after an illness. Family members also assume emotional responsibility for their relatives.
- Access to and engagement with service provision may be variable after acquired brain injury.
- Cognitive rehabilitation and neuropsychology may be underrepresented as key aspects of service provision following encephalitis.
- Individuals who have sustained cognitive impairments as a result of encephalitis ('the walking wounded') may not get adequate service provision for their needs.
- Welfare benefits agencies may not understand the complexities and implications of cognitive impairment in acquired brain injury.
- The role of social care agencies in offering support seems almost non-existent.
- Individuals may feel stigmatised by the 'hidden disability' of post-encephalitic acquired brain injury.
- The Encephalitis Society needs pro-active engagement with individuals and families from diverse ethnic communities.

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