ORIGINAL RESEARCH ARTICLE

# Counting the Cost of Meningococcal Disease

Scenarios of Severe Meningitis and Septicemia

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Published online: 16 January 2013 © Springer International Publishing Switzerland 2013

### Abstract

*Background* Meningococcal disease can result in severe disabling sequelae, but there is no published information about the lifelong rehabilitation costs of patients with severe outcome in the UK. As cost-effectiveness studies play a crucial role in determining whether immunization programmes will be implemented, it is important to identify these costs.

Objective The aim of the study was to estimate lifelong rehabilitation costs associated with severe cases of meningococcal disease and to present these costs in a format appropriate for use in a cost-effectiveness analysis. Methods Two severe scenarios of meningococcal disease with major sequelae were developed; one that presented acutely as meningitis and the other as septicemia. Scenarios were based on systematic reviews of the literature describing the sequelae of meningococcal disease, dialogue with Meningitis Research Foundation members who have experience of the disease, and discussions with pediatricians who have been responsible for managing children with this disease over many years. The two scenarios were devised to represent cases typical of the severe end of the spectrum. To obtain a comprehensive list of the health, educational and other resources used by survivors during and since their acute illness, families of individuals with

**Electronic supplementary material** The online version of this article (doi:10.1007/s40272-012-0006-0) contains supplementary material, which is available to authorized users.

sequelae similar to those in each of our scenarios were interviewed. Relevant academics and professionals in health, social care and education were consulted in order to ensure that our scenarios accurately represented the treatment and support that individuals with such sequelae might realistically receive from the National Health Service (NHS), the local authority and Personal Social Services (PSS). The majority of costs were derived from English Department of Health reference costs and unit costs of health and social care reflecting values for the financial year 2008-2009 indexed to 2010-2011. Costs were based on a life expectancy of 70 years in each scenario and are presented at a discount rate of 3.5 % for the first 30 years and 3 % thereafter, as recommended by the UK treasury (non-discounted costs are also presented for comparison). Costs are presented from both an NHS/PSS perspective and a government perspective.

*Results* This study has revealed that severe cases of disease that result in long-term sequelae can result in costs to the NHS/PSS of around £160,000–£200,000 in the first year alone. Over a lifetime of 70 years, discounted costs from an NHS/PSS perspective ranged from approximately £590,000 to £1,090,000 (£1,250,000–£3,320,000 undiscounted) and discounted costs from a government perspective ranged from £1,360,000 to £1,720,000 (£3,030,000–£4,470,000 undiscounted).

*Conclusion* This study fills a gap in the UK literature and produces estimates that can be used in cost-effectiveness analysis to better represent the cost of illness at the severe end of the spectrum. Costs from a government perspective highlight the wider impacts of this disease, which is important for clinical decision makers, and budgetary and service planners to be aware of when making decisions about the benefits of implementing public health interventions such as immunization programmes.

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#### Key points for decision makers

There are currently few UK data on the rehabilitation costs of cases of meningococcal disease with lifelong sequelae

Lifetime rehabilitation costs are substantial for survivors of severe cases of disease and are particularly high during the early years after recovery

Lifelong costs associated with severe cases of meningococcal disease are presented in a format appropriate for use in cost-effectiveness analysis from a National Health Service/Personal Social Services and government perspective

Results from this costing exercise may be used by economic analysts to represent the cost of severe illness when looking at the cost effectiveness of immunization programmes against meningococcal disease

# 1 Introduction

Meningococcal disease is the foremost cause of bacterial meningitis in the UK, where it is also a major infectious cause of death in early childhood [1]. Although the majority of people who survive appear to recover relatively unscathed, some are left with severe disabilities, and their long-term care imposes a considerable burden on healthcare resources. Meningococcal meningitis can result in major sequelae, such as mental disability, seizures, hearing loss, motor impairment, hydrocephalus and loss of vision [2], while limb loss, growth plate damage, renal failure and scarring can occur in cases of meningococcal septicemia [3, 4]. According to a recent systematic review of studies in children surviving meningococcal disease, 4 % have hearing loss, 7 % have neurological sequelae, 3 % have amputations, 3 % have other orthopedic problems, and 13 % have skin necrosis and scarring [1].

Economic evaluations such as cost-effectiveness analyses are becoming increasingly important in determining whether immunization programmes should be implemented [5]. Accurate cost-of-illness data is a fundamental component of such an analysis; however, a study of cost effectiveness of meningococcal serogroup C conjugate vaccination pointed to a lack of published information in the UK of costs associated with the treatment and rehabilitation of survivors of meningococcal disease with serious after effects [6].

As Meningitis Research Foundation (MRF) has unique contact with families affected and professionals involved in acute care and rehabilitation, we have attempted to fill this gap in the current UK literature through an estimation of the lifetime rehabilitation costs associated with severe cases of meningococcal disease. We present these costs in a format appropriate for use in cost-effectiveness analyses.

# 2 Methods

We developed two severe scenarios of meningococcal disease with major sequelae; one that presented acutely as septicemia (patient A) and the other that presented as meningitis (patient B). Scenarios were based on MRF members who have experience of meningococcal disease with severe outcome and systematic reviews of the literature describing the sequelae of meningococcal disease [1, 2, 7]. The compiled scenarios were refined through discussions with intensive care pediatricians, pediatric neurologists, pediatric orthopedic surgeons and pediatric infectious disease specialists responsible for managing children with meningococcal disease over many years. They agreed that our final scenarios were representative of cases at the severe end of the spectrum.

Once we had outlined the scenarios, we interviewed the parents of six children with sequelae similar to those of patient A, and the parents of six children with sequelae similar to those of patient B. This enabled us to compose a list of health, social service, educational, and other resources used by patients during and after their acute illness. From this we identified and consulted with relevant professionals and academics in health, social care, and education. Consultation with these professionals allowed us to estimate the amount of National Health Service (NHS), local authority and Personal Social Services (PSS) resources that these patients would use. Lifelong needs for continuing care and support were considered as the child grew, attended school, and made the transition to adulthood.

Initial consultations were carried out over the telephone or in person and the MRF members' experiences of resources were used as an initial template for discussion. Amendments to patient-reported resource use were made according to comments made by the consultees. A consultation document was then produced that detailed all of the assumptions made about the acute illness, immediate, long-term and late-onset sequelae, and consequent acute care, rehabilitation and long-term service provision. This document was sent to all of our professional consultees for validation and then amended according to the responses received. The revised document was then submitted to our consultees, who agreed that this was an accurate representation of resource use for such patients. Tables 1 and 2 summarize the NHS/PSS resources used by patients A and B, respectively. Full assumptions are provided in the appendix (Online Resource 1).

The resource use identified during the consultation period was costed following recent guidelines on costing of public services as part of economic evaluation [8]. 
 Table 1
 Patient A's National Health Service/Personal Social Services resource use

#### Outpatient appointments

Follow-up appointments with acute hospital pediatrician

- Quarterly appointments at disablement services centre for limb fittings
- Prosthetic physiotherapy and OT appointments associated with limb upgrades
- 6-monthly pediatric combined clinics with plastic surgeon and orthopedic surgeon
- Five appointments with adult plastic surgeon to deal with breakdown of skin grafts
- Community medicine
- Six-monthly to yearly appointments with community pediatrician
- Regular community physiotherapy and OT

Specialist equipment

Splints, pressure suits, manual and power wheelchair, walker, adapted crutches, therapy bench and standing frame

Prosthetic provision

- Non-articulating prosthetic legs until age 3 years (three pairs)
- SAKL prosthetic legs from age 3 to 5 years (two pairs)
- Junior articulated prosthetic legs from age 5 to 14 years (approximately one pair per year)
- Adult articulated prosthetic legs from age 14 years (initially two pairs, one pair renewed every 2 years, the other renewed every 5 years)
- Stubbies (replaced every 9 months as a child and every 4 years as an adult)
- Silicon gel liners for prosthetic/limb interface (renewed 6-monthly)
- Waterproof legs for access to pool and showering
- Cosmetic below-elbow prosthesis until age 15 years (replaced every 6 months)
- Child's mechanical functional below-elbow prosthetic arm until age 15 years (hand is replaced once every 2 years)
- Child's myoelectric functional below-elbow prosthesis (hand upgraded in size three times and serviced once every 2 years)
- Adult cosmetic below-elbow prosthesis from age 15 years (replaced every 5 years)
- Adult below-elbow prosthesis with split hook from age 15 years (limb replaced once every 10 years)
- Adult below-elbow myoelectric prosthesis (hand is serviced once every 2 years and exchanged once every 8 years)
- Sockets are replaced more frequently on child limbs compared with adult limbs
- Stump revisions and skin graft surgery
- Eight surgical procedures to correct contractures, breakdown of skin grafts and bony overgrowth within amputation stumps between ages 3 and 19 years

Behavioral problems

Appointments with a child psychologist

Ten sessions at a parenting group

40 mg/day immediate-release methylphenidate

Public health

Chemoprophylaxis for the family

# Table 1 continued

Contact tracing and information sharing

- Analysis of blood sample
- Personal Social Services direct payments
- Social care assessments, home visits and reviews
- Direct payments for homecare worker (7 hours per week)

Additional educational costs

Learning support assistant

School transport

SEN statement (issue and review)

School adaptations and equipment

Other government costs

Direct costs

Disabled facilities grant, government's specialized vehicles fund, disabled students allowance (university non-medical helper and equipment)

# Indirect costs

Lost tax revenue from both the mother's and the affected individual's unemployment

### Transfer payments

Carers allowance, child tax credits (additional due to disability), disabled living allowance (mobility and care), disabled students allowance (general allowance), working tax credits, housing benefit, council tax benefit, pension credit

Full assumptions are provided in the appendix (Online Resource 1) *OT occupational therapist, SAKL* self-articulating knee lock, *SEN* special educational needs

Currently, UK cost-effectiveness analyses (CEAs) are confined to an NHS/PSS perspective [8]. However, there is continuing debate about whether broader perspectives should be used in economic evaluation of medical innovations [9], so we have presented costs from both an NHS/ PSS and government perspective.

As the costs presented are distributed across a lifetime, discounting has been used to give less weight to future costs compared with current costs. In keeping with UK Treasury recommendations [10], all costs have been presented at a discount rate of 3.5 % for the first 30 years and at 3 % thereafter. Costs have been based on a lifespan of 70 years of age (7 and 12 years less than the respective average life expectancies for UK males and females [11]). Several pediatric specialists argued that the mobility limitations in these scenarios might give rise to cardiovascular problems that could shorten lifespan, and it has been reported that survivors of meningococcal septicemia report poorer general health than the population as a whole [12].

# 2.1 Unit Cost Data

All costs are expressed in pounds sterling. The costs of hospital stays, inpatient and outpatient appointments were derived from English Department of Health reference costs

# Table 2 Patient B's National Health Service/Personal Social Services resource use

#### Outpatient appointments

Follow-up appointments with acute hospital pediatrician

6-monthly appointments with pediatric neurologist (one per year is a multiprofessional appointment with a neuropsychologist)

Yearly appointments with neurosurgeon (regarding shunt)

Yearly appointments with orthopedic surgeon from age 6 to 20 years (monitoring for scoliosis)

Regular appointments with orthotist

Appointments with opthalmologist

Community medicine

6-monthly to yearly appointments with community pediatrician

Regular appointments with community physiotherapist and OT 12 sessions of hydrotherapy

Quarterly appointments with SALTs

Specialist equipment

Therapy bench, standing frame ( $\times$  2), specialist seating (Leckey Squiggles chair, Tumbleform feeder seat and wedge), walkers ( $\times$  4), specialist toys, lycra suit ( $\times$  6), sleep system ( $\times$  3), manual wheelchair, orthotic shoes (two per year until age 17 years, two every 5 years from then on), annual splint replacement, communication aid

#### Public health

Chemoprophylaxis for the family

Contact tracing and information sharing

Analysis of CSF

Cochlear implantation

Assessment, bilateral implant surgery, rehabilitation in year 1 (including ten appointments with SALT or ToD and eight tuning appointments with the audiologist in the clinic, rehabilitation in year 2 (including six appointments with SALT or ToD and two tuning appointments with the audiologist in the clinic)

Ongoing care and technical support for implants, including tuning, batteries, spares and upgrade of processors

General health problems

Paracetamol (acetaminophen) for pain management

Lactulose for constipation management

Four nappies per day provided by the NHS beyond the age of 3 years and four incontinence pants provided beyond the age of 7 years

#### Epilepsy

400 mg sodium valproate per day from ages 3 to 5 years

600 mg sodium valproate per day from age 6 years onwards Shunt revision surgery

Three elective shunt maintenance operations at ages 5, 28 and 48 years

Two non-elective operations due to shunt blockage at ages 11 and 33 years

Personal Social Services

Social care assessments, home visits and reviews

Direct payments for home help 3 hours a day, 5 days a week until age 16 years, and then 8 hours per month from age 16 years onwards One weekend in every 2 months in a specialist residential unit (short-break provision)

Full-time residential care from age 40 years onwards

Additional educational costs

Additional cost of attending an SEN nursery and then SEN school until age 19 years

Transport to and from school

SEN statement and annual reviews

Other government costs

Direct costs

Disabled facilities grant (for home adaptations) and government specialized vehicle fund

Indirect costs

Lost tax revenue from the mother and child's unemployment

Transfer payments

Carers allowance, child tax credits (for a disabled child) and disability living allowance (mobility and care)

Full assumptions are provided in the appendix (Online Resource 1) *CSF* cerebrospinal fluid, *NHS* national health service, *OT* occupational therapist, *SALTs* speech and language therapists, *SEN* special educational needs, *ToD* teacher of the deaf

2008–2009 [13]. Unit costs of community health and social services were largely calculated using unit costs of health and social care [14], although some costs such as shortbreak provision and educational services were based on independent research (Lam SC, personal communication) [15]. Medication costs were obtained from the British National Formulary [16, 17]. Prosthetic component costs were obtained from the NHS catalogue of prosthetic components at the NHS supply chain. All unit costs represent, or have been indexed to, 2010–2011 prices.

#### **3** Results

#### 3.1 Patient A

Patient A contracted meningococcal septicemia at 12 months of age, suffering severe septic shock, acute respiratory distress syndrome and renal failure. He also developed gangrene of the limbs due to purpura fulminans. He spent 31 days in the pediatric intensive care unit (PICU) during the acute episode. His respiratory and renal problems were resolved in the PICU, but both legs had to be amputated above the knee and one arm below the elbow. He also underwent various skin grafting operations during his initial hospital stay, which lasted a total of 6 months.

Once discharged from hospital, patient A became a lifelong outpatient of a disablement services centre for prosthetic limbs and attended regular appointments with the hospital pediatrician, orthopedic surgeon and plastic surgeon. Community therapists were involved in his rehabilitation at home and at school and he required specialist equipment to help him with his mobility. He experienced stump overgrowth and scar contractures on his residual limbs and as a result needed corrective operations throughout his childhood to trim his bones and alter the skin coverage of his amputation stumps.

At 4 years of age he started to display difficult behaviour. He saw a psychiatrist throughout his school years and took medication to help his concentration.

Patient A attended a mainstream maintained nursery and school. He had a special educational needs statement because of his physical and behavioural problems. During his early years he had a learning support assistant to help him with his mobility and daily physiotherapy exercises. He had free transport to and from school, and the school also underwent adaptations to accommodate his needs.

Patient A was one of two children in a two-parent family. After becoming ill, one parent gave up work to help care for him, which put a financial strain on the family. The child's disability and the reduced family earnings entitled the family to a range of government grants and benefit payments.

Patient A went on to further education at age 18 years and at age 21 years he secured a part-time job and moved from his parent's house into rented accommodation. Whilst he lived independently, he received direct payments which allowed him to employ a homecare worker for 7 hours a week to help with certain household tasks such as shopping. He worked 16 hours a week until he retired at the age of 65 years.

# 3.2 Patient B

Patient B contracted meningococcal meningitis at 3 years of age. She was hospitalized with raised intracranial pressure and intractable seizures, and required ventilation and intubation. Acute hydrocephalus was immediately treated by insertion of an external ventricular drain and later by insertion of a shunt. She required prolonged airways management for neurological complications, including repeated seizures. She spent 26 days in the PICU. Once stable she was transferred to a pediatric ward where she spent time recovering before moving to a neuro-rehabilitation unit for 5 months.

Patient B was left with severe neurological damage, including severe cognitive deficits, epilepsy, severe hemiplegia, homonymous hemianopsia, communication problems, and profound deafness. Although she eventually learned to walk, this was only for very short distances. She was predominantly a wheelchair user and had problems with her posture.

Once discharged from hospital, she had regular neurological outpatient appointments. She also had regular visits from community therapists and was provided with specialist equipment to help with her mobility and posture. She had bilateral cochlear implants and became a lifelong patient of the cochlear implant centre.

Patient B attended a special needs nursery and a maintained special needs primary and secondary school. Throughout her childhood and into adulthood she spent the occasional weekend in a residential unit so that her parents had a break from caring. Her parents also received direct payments so that they could employ home help for a few hours each week.

One parent gave up work to help care for Patient B shortly after she became ill, which put financial strain on the family. The child's disability and the reduced family earnings entitled the family to government grants and benefit payments. Patient B lived with her parents until the age of 40 years, when they became too frail to care for her, and then moved into full-time residential care.

# 3.3 Costs

The lifetime government and NHS/PSS costs associated with both patients are outlined in Table 3. Prosthetic provision was the primary cost driver for patient A, followed by the hospital length of stay. The most costly aspect of care for patient B was the residential care home. Cochlear implantation was the most costly medical treatment that she received over her lifetime.

The average undiscounted costs per year for patients A and B are shown in Tables 4 and 5, respectively. In both cases, considerable costs were accrued during the first year after disease onset. This is largely due to the prolonged stay in the PICU and the subsequent long spell in hospital undergoing multiple surgical procedures (in the case of patient A) and rehabilitation (in the case of patient B).

Yearly undiscounted costs to the NHS/PSS tend to be higher in the early years following the acute illness in both cases. For patient A, this is due to increased prosthetic provision in childhood, when limbs and their interfaces need to be changed more regularly to keep up with the growth of the child and due to skin stump fragility in the first years after healing, along with increased physiotherapy and occupational therapy soon after the amputation and throughout childhood. Increased costs for patient B were partly due to more intensive habilitation after cochlear implantation in the first 3 years post-implantation. Yearly costs for patient B increased again in later life because of the high cost of residential care, which was required from the age of 40 years onwards.

# 3.4 Univariate Sensitivity Analysis

To account for uncertainty associated with both costs and resource use in our scenarios, we adjusted cost category

Table 3	National	Health	Service/Persona	al Social	Services	and	government	costs	(discounted	and	undiscounted)	according t	.o c	category	fo
patients .	A and B	over a li	ifespan of 70 ye	ars											

Category	Perspec	tive	Patient A		Patient B		
	NHS/ PSS	Government	Discounted (£)	Undiscounted (£)	Discounted (£)	Undiscounted (£)	
Acute costs	•	•	157,101	157,101	136,401	136,401	
Outpatient appointments	•	•	32,591	57,151	20,226	35,086	
Community medicine	•	•	13,563	17,412	19,572	23,392	
Specialist equipment provision	•	•	14,464	27,630	29,967	59,528	
Prosthetic provision	•	•	259,288	654,784	NA	NA	
Stump revisions and skin graft surgery	•	•	21,793	30,809	NA	NA	
Behavioural problems	•	•	8,382	11,241	NA	NA	
Public health response	•	•	154	154	265	265	
Cochlear implantation	•	•	NA	NA	175,481	361,897	
General health problems	•	•	NA	NA	43,372	100,775	
Epilepsy	•	•	NA	NA	3,739	9,515	
Shunt revision surgery	•	•	NA	NA	18,610	33,269	
Social care assessment, home visits and reviews	•	•	2,390	8,188	16,238	24,501	
Direct payments	•	•	83,009	288,259	131,231	185,352	
Residential overnight provision	•	•	NA	NA	61,917	116,622	
Residential care home	•	•	NA	NA	430,413	2,235,844	
Maintained special needs school		•	NA	NA	147,318	189,868	
Learning assistant		•	100,629	127,242	NA	NA	
Transport to and from school		•	41,612	58,214	49,831	67,170	
Special educational needs statement		•	4,647	6,326	5,358	7,023	
School adaptations		•	30,634	38,717	NA	NA	
Direct government costs		•	127,389	256,602	70,748	99,839	
Indirect government costs (lost tax revenue)		•	105,688	252,480	139,125	351,105	
Transfer payments		•	365,121	1,046,410	221,956	436,975	
Total							
NHS/PSS costs			592,734	1,252,729	1,087,432	3,322,446	
Government costs			1,368,454	3,038,719	1,721,768	4,474,427	

NA not applicable, NHS/PSS National Health Service/Personal Social Services, • indicates that the cost category is included in this cost perspective

 Table 4
 Average yearly undiscounted cost for patient A (a severe case of meningococcal septicemia)

Perspective	No. of years post-acute illness							
	0	1–15	16–69					
National Health Service and Personal Social Services (£)	160,529	16,910	15,529					
Government (£)	166,474	51,615	38,852					

totals by 25 %. Acute care is a significant contributor to overall costs so we also examined how varying the length of stay in the PICU and length of stay in hospital by 25 % affected the lifelong costs. Sensitivity analyses are presented in Figs. 1 and 2.

# **4** Discussion

# 4.1 Main Findings

Severe cases of meningococcal disease which require 5- to 6month stays in hospital can result in costs to the NHS/PSS of around £160,000–£200,000 in the first year alone. These costs have not been taken into account in a recent UK costeffectiveness model for meningococcal B vaccines because acute care costs in this model have been based on average hospital inpatient day costs for meningococcal infection alone (which do not include costs associated with surgical procedures such as amputations, skin grafting, cochlear implantation, neurosurgery, or other inpatient rehabilitation, which may take place in complicated cases) [18]. Table 5Average yearlyundiscounted cost for patient B(a severe case of meningococcalmeningitis)

Perspective	No. of year	No. of years post-acute illness								
	0	1–2	3-12	13–36	37–69					
National Health Service and Personal Social Services (£)	196,243	31,948	26,666	13,982	79,357					
Government (£)	205,073	87,646	60,746	32,328	83,400					

This study demonstrates that resource use (and therefore costs) in severe cases tend to be higher in the early years after recovery from acute illness. Discounting scales down future events so that the further into the future they occur, the less important they are to a decision maker today, and the discount rate applied in a CEA can dramatically affect the resulting cost-effectiveness ratio [19]. It may be important to the outcome of CEA that these higher costs in the early years post-illness are fully represented by staggering the costs accordingly.

This study estimates costs over a lifetime of survivors of meningococcal disease with major sequelae approached from two different perspectives: NHS/PSS and government. A government perspective on costs gives a broader picture of the cost to society as a whole, and includes additional costs associated with the education of children with major sequelae. The government perspective also includes benefit payments such as disability living allowance, carer's allowance and disabled facilities grants which acknowledge (but do not fully compensate for) the increased cost to the family of bringing up a disabled child. In these scenarios, discounted costs from a government perspective are 1.5- to 2.3-fold higher than NHS/PSS costs alone.

We did not attempt to cost these cases from a full societal perspective as costs to individual families are more difficult to quantify. Previous studies have reported that the day-to-day costs to a family with a disabled child are three times those for a family of a non-disabled child [20], and one study found that the minimum essential budget required to bring up a disabled child was £7,355 per year compared with £2,100 for other children [21]. Reasons for these additional costs to the family may include increased heating bills due to the lack of mobility of the child, increased washing because of incontinence, and the need for specialist equipment to help with day-to-day activities and mobility. These higher day-to-day living costs in addition to a lower income (due to one parent perhaps needing to stop work to care for their child) mean that families with disabled children are 4-fold more likely to live in poverty [22].

# 4.2 Strengths and Limitations

A limitation of this study is that relying on experts with experience and interest in the sequelae of meningococcal disease to estimate resource input may produce estimates representing good practice care rather than actual practice. However, we have been careful to be realistic about the medical treatment given and included only such care as our experts considered would be available on the NHS for a child in these scenarios. For example, some children are able to get running legs by applying to their Primary Care Trust, but such expensive specialist equipment was not included in our scenario. Additionally, patient B was cared for by her parents until she reached 40 years of age rather than being cared for in a residential care home from an earlier age at a considerably higher cost.

Drug costs have been obtained from the British National Formulary, which may overestimate costs because NHS Trusts often have separate contracts for suppliers. Additionally, this could be the case for certain equipment costs where the only available price was a direct quote from suppliers; however, as these costs only make up a small proportion of the total, they are unlikely to significantly alter the overall amounts.

Although we describe severe examples of disease, these scenarios do not represent the most complex cases. For example, the extent of injury to the circulatory system in severe septicemia leaves a proportion of children with permanent renal failure, and adrenal or other endocrine damage in addition to amputations. Our scenarios are also conservative in that they have not taken into account additional costs to the NHS due to ill health of other members of the family. Previous studies have indicated that ill health is more prevalent in families with disabled children compared with other families as members of such families are particularly prone to stress-related illnesses such as depression and anxiety. It has also been reported that well siblings of children with chronic illness or disability are 1.6- to 2.0-fold more likely to experience behavioural and emotional problems [23].

# **5** Conclusions and Implications

At the time of writing, the first ever vaccine to protect against different strains of serogroup B meningococcal bacteria has been given a positive opinion by the European Medicines Agency and will soon be a licensed product in Europe. Another vaccine is in late stage development. CEA Fig. 1 Tornado plot showing results of univariate sensitivity analysis of various cost categories for patient A. Parameters were adjusted by  $\pm 25 \%$ . *NHS/PSS* National Health Service/Personal Social Services, *SEN* special educational needs



Fig. 2 Tornado plot showing results of univariate sensitivity analysis of various cost categories for patient B. Parameters were adjusted by  $\pm 25$  %. *NHS/PSS* National Health Service/Personal Social Services, *SEN* special educational needs

is used as a tool to decide whether licensed vaccines should be introduced into the national schedule.

Economic analysts who create cost-effectiveness models are often faced with estimating costs for meningococcal disease survivors with sequelae with limited information. Until now, there has been no UK literature detailing the costs of caring for and treating survivors of meningococcal disease with major sequelae. This study has identified the high costs associated with the acute treatment of severe cases of meningococcal disease, which to date have been unaccounted for in CEA of vaccination for meningococcal disease. We have also identified cochlear implantation and prosthetic limb provision to be amongst the most costly aspects of caring for survivors of meningococcal disease with severe sequelae. One recent study suggests that around 1.3 % of serogroup B meningococcal disease survivors will be left with disabling amputations and another 2 % will survive with profound bilateral hearing loss, although the proportion of survivors with severe brain damage is more likely to be less than 1 % [24]. Consideration of these long-term rehabilitation costs within CEA would certainly increase the cost effectiveness of a vaccine that protects against serogroup B meningococcal disease. It remains to be seen, however, whether their inclusion would be enough to reduce the cost of a particular vaccine to within the NICE £20,000–£30,000/quality-adjusted lifeyear (QALY) cost-effectiveness threshold.

In addition to the cost/QALY from an NHS/PSS perspective, it is imperative that decision makers and budgetary and service planners recognize the overall economic and human impact of such diseases when considering whether to implement vaccines that protect against them. As our study identifies costs from a wider government perspective, it highlights some of these wider impacts.

Acknowledgments Numerous professionals were consulted to complete this study. The authors would like to thank the orthopedic surgeons, plastic surgeons, specialist pediatricians, neurologists, consultants in rehabilitation medicine, prosthetists, psychiatrists, occupational therapists, physiotherapists, educational professionals and academics who provided assistance during the study. We would also like to thank the many individuals at the NHS Information Centre and Connecting for Health, and the families of children affected by meningococcal disease who took the time to share their experiences with us.

**Funding** Unrestricted educational grants from Novartis, Pfizer and GlaxoSmithKline provided funding for the study to take place. A knowledge transfer partnership grant from the Department of Trade and Industry funded interviews and work leading up to interviews with the families affected. No funding body influenced the study design, data collection, data analysis, data interpretation, writing of the report, or the decision to submit the paper for publication.

Outside of the submitted work, the MRF has received unrestricted educational grants and conference sponsorship from Novartis, Pfizer, GlaxoSmithKline, Baxter and Sanofi Pasteur. Claire Wright and Linda Glennie have also been paid for providing consultancy for Novartis.

**Contributors** Linda Glennie was responsible for study concept. Linda Glennie and Claire Wright were responsible for study design. All authors took part in interviews with members and health professionals to produce the patient scenarios. Claire Wright and Rebecca Wordsworth undertook the data collection for costs. Claire Wright produced the consultation document that was sent to health professionals and academics for verification. She also wrote the first draft of the report, which was critically revised by Linda Glennie and Rebecca Wordsworth. All authors take responsibility for the integrity of the data.

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