



# Lifetime cost of meningococcal disease in France: Scenarios of severe meningitis and septicemia with purpura fulminans

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## KEYWORDS

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**Summary** Invasive meningococcal disease (IMD) is life-threatening and can result in severe sequelae. In France, no data have been published on the costs of severe IMD cases. Two realistic scenarios were developed with national experts (clinicians and social workers): a 6-year-old child with *purpura fulminans* with amputation of both legs below the knee (case A) and a 3-year-old with meningitis and severe neurological sequelae (case B). Additional scenarios included other typical sequelae of IMD such as chronic kidney disease (CKD), profound deafness and epilepsy. Data on healthcare, disability, educational and other resource use were obtained from experts and families of patients with similar sequelae. Unit costs (2013) were mainly obtained from the literature and the National Health Insurance (NHI). Time horizon was based on life expectancies of patients (77 and 55 years, respectively). A 4% discount rate decreasing to 2% after 30 years was applied. Costs are presented from the perspective of the NHI, publicly funded organizations and patients' families or their private health insurances. *purpura fulminans* with amputations is associated with a lifelong discounted cost of €768,875. Adding CKD doubles the amount (€1,480,545). Meningitis with severe neuro-cognitive sequelae results in a lifelong discounted cost of €1,924,475. Adding profound deafness and epilepsy slightly increases the total cost (€2,267,251). The first year is the most expensive in both scenarios (€166,890

**Abbreviations:** CKD, chronic kidney disease; IMD, invasive meningococcal disease; MRF, Meningitis Research Foundation; NHI, National Health Insurance; PICU, Pediatric Intensive Care Unit.

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and €160,647 respectively). The main cost drivers for each scenario are prostheses and child/adult stays in healthcare facilities, respectively. Overall, patients' families or his private insurance had to pay around 13% of total cost (101,833€ and 258,817€, respectively). This study fills a gap in the body of knowledge on IMD sequelae care and lifetime costs in France. The potentially high economic burden of IMD, in addition to its physical, psychological and social burden, reinforces the need for prevention.

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## Introduction

Invasive meningococcal disease (IMD) is a severe bacterial infection caused by *Neisseria meningitidis* or meningococcus, the most disease-associated serogroups being A, B, C, W and Y. Young non-immunized children are the most at risk of contracting the disease. IMD can present as meningococcal septicemia, which can evolve to *purpura fulminans*, very serious septic shock or meningitis [1]. The disease can result in severe sequelae and is fatal in 50–80% of cases if not treated [2–4]. Even when treated, 5–10% of the patients die 24–48 h after the onset of symptoms [2]. This rate is even higher for patients with *purpura fulminans* (15–30%) [5,6]. Sequelae include cerebral lesions, hearing loss, learning difficulties in 10–20% of survivors, deafness in 3–15% and severe cognitive deficits, cerebral palsy or epilepsy in rare cases (3–4%) [7,8]. Of the initial survivors, 10–30% present with skin necrosis and limb ischemia requiring orthopedic surgical management, such as debridement, skin grafting, muscular flap coverage for limb salvage and sometimes even limb amputation [6].

The introduction of meningococcal conjugate vaccines in the last 20 years has resulted in a reduction in the incidence of IMD across Europe [5].

In France, the incidence of IMD has been stable for the last 25 years and is approximately 1 case per 100,000 inhabitants. There is a high predominance of serogroup B (58%) and C (26%) [9], and the incidence of IMD in France remains relatively high compared to other European countries such as Italy or Germany. The very first vaccine to protect against different strains of serogroup B meningococcal bacteria, Bexsero®, became a licensed product in January 2013 and is indicated for children 2 months of age in Europe. More recently in the USA, the FDA granted a license to Bexsero® and to another vaccine Trumenba®, both of which are indicated for those 10–25 years of age. At the time of writing, the UK is the only European country [10] to have started vaccinating infants

against serogroup B disease as part of children's routine immunization schedule.

To date, very little data have been published concerning the financial impact of IMD, and no data are available for France, particularly in regards to the different payer perspectives throughout a patient's lifetime. Therefore, we specifically designed this study to estimate the cost of exhaustive resource consumption of two severe and realistic scenarios of IMD cases and their sequelae in France, from the onset of symptoms to the end of life, from all payers' perspectives.

## Materials and methods

### Scenario development

We developed two scenarios of severe IMD cases: a 6-year-old boy with *purpura fulminans* resulting in amputation of both legs below the knee (case A) and a 3-year-old girl with meningitis resulting in severe neurological sequelae (case B). Additional scenarios were created to include other common sequelae of IMD. Scenarios were initially based on those previously selected in a UK study [11] and were then adapted to France with the help of national experts. Various healthcare specialists (pediatricians, orthopedic surgeons, neurosurgeons, physical therapists, nephrologists, otolaryngologists, prosthesis specialists, social workers, occupational therapists, psychomotor specialists, and independent living specialist) and families of patients with sequelae similar to those of patients A and B were interviewed to describe and evaluate patient management. In total, 19 healthcare professionals, 5 patient's families and one patient group agreed to participate in face-to-face or telephone meetings. Lifelong patient management was described in the interviews and all resource use associated with each step of management was collected. For each family, hypothetical revenues, type of home and distance from home to healthcare professionals were determined. The time horizon was based

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