RESEARCH ARTICLE

Health and Economic Outcomes of Posterior Spinal Fusion for Children With Neuromuscular Scoliosis

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ABSTRACT

OBJECTIVES: Neuromuscular scoliosis (NMS) can result in severe disability. Nonoperative management minimally slows scoliosis progression, but operative management with posterior spinal fusion (PSF) carries high risks of morbidity and mortality. In this study, we compare health and economic outcomes of PSF to nonoperative management for children with NMS to identify opportunities to improve care.

METHODS: We performed a cost-effectiveness analysis. Our decision analytic model included patients aged 5 to 20 years with NMS and a Cobb angle ≥50°, with a base case of 15-year-old patients. We estimated costs, life expectancy, quality-adjusted life-years (QALYs), and incremental cost-effectiveness from published literature and conducted sensitivity analyses on all model inputs.

RESULTS: We estimated that PSF resulted in modestly decreased discounted life expectancy (10.8 years) but longer quality-adjusted life expectancy (4.84 QALYs) than nonoperative management (11.2 years; 3.21 QALYs). PSF costs \$75 400 per patient. Under base-case assumptions, PSF costs \$50 100 per QALY gained. Our findings were sensitive to quality of life (QoL) and life expectancy, with PSF favored if it significantly increased QoL.

CONCLUSIONS: In patients with NMS, whether PSF is cost-effective depends strongly on the degree to which QoL improved, with larger improvements when NMS is the primary cause of debility, but limited data on QoL and life expectancy preclude a definitive assessment. Improved patient-centered outcome assessments are essential to understanding the effectiveness of NMS treatment alternatives. Because the degree to which PSF influences QoL substantially impacts health outcomes and varies by patient, clinicians should consider shared decision-making during PSF-related consultations.

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Pediatric hospitalists frequently comanage the care of children with neuromuscular scoliosis (NMS). Postoperative comanagement has been associated with decreased length of stay.[1] Although less common, preoperative comanagement resulted in recommendations, including medication and nutrition changes and additional subspecialty involvement.[2] Identifying new opportunities to improve care for children with NMS can increase the value of care pediatric hospitalists provide in surgical comanagement.

NMS is the abnormal curvature of the spine secondary to an underlying neurologic condition and affects all nonambulatory children with cerebral palsy and 50% to 75% of children with mobility limitations.[3–5] Most patients with NMS have cerebral palsy, a disorder of spinal cord development, or muscular dystrophy, and children with NMS frequently have additional comorbidities with lifelong functional impairment.[6–8] Scoliosis progression extends beyond skeletal maturity in nonambulatory patients, with 50% of children with severe cerebral palsy progressing to moderate-to-severe scoliosis.[3,4] NMS progression often results in chronic pain, mobility impairment, and cardiorespiratory compromise.[3,9]

Treatment options for NMS include nonoperative and operative management. Nonoperative management, including bracing and physical therapy, frequently fails to halt curve progression.[10] Operative management is reserved for severe curvatures of a Cobb angle ≥50° or for patients with failed nonoperative therapy. The primary benefit of operative management is parent report of improved health-related quality of life (HRQOL) in pain control, mobility, and feeding.[11] However, objective assessments of physical function performed by physical therapists revealed no meaningful improvement in function above the preoperative baseline 12 months after surgery.[12] Although operative management has high costs and high complication rates, the frequency with which it is performed has rapidly increased over the past decade, likely influenced by the lack of alternative definitive treatment options.[7,8]

Posterior spinal fusion (PSF) is the most common operative management approach for NMS and uses implantable rods that mechanically straighten the affected portion of the spine, resulting in the subsequent fusion of the vertebrae over time. PSF in NMS frequently involves long portions of the spine, from the upper thoracic to the lower lumbar vertebrae or pelvis.[9] Although PSF produces marked improvements in the degree of spine curvature, its effects on quality of life (QoL) and health outcomes remain poorly understood.[13] Furthermore, children with NMS often have comorbidities that increase their risk of perioperative and postoperative complications (including hemorrhage, respiratory failure, surgical site infection, and death) and contribute to higher costs.[3,9,7,14]

Trade-offs between complication rates and cost with potential gains in QoL and health make the decision for PSF complicated to navigate. Among pediatric hospitalists in surgical comanagement programs, a greater understanding of the factors that affect outcomes for children with NMS can help identify opportunities to improve perioperative management of PSF.[15–18] Our objectives in this cost-effectiveness study were to assess the health and economic outcomes of operative and nonoperative management of children with NMS and to assess the clinical characteristics of children with NMS that could help guide treatment selection from the perspective of the health care system.

METHODS

Model Design, Treatments, Health States, and Target Population

We performed a cost-effectiveness analysis using a simple decision tree to compare 2 treatment strategies for patients with NMS: operative management with PSF and nonoperative management. The key health states and transitions are captured in Fig 1. In our base-case analysis, we assumed no crossover between treatment groups. A base-case analysis applies to the model with the most likely variable estimates and assumptions based on the literature.

Perioperative complications included major complications, such as intraoperative shock and hemorrhage and prolonged intubation.[19,20] Those who survived the perioperative period transitioned to 1 of 3 postoperative health states: no postoperative complications, a surgical site infection, or a noninfectious, implant-related complication requiring surgical site reoperation. Those with surgical site infection received treatment with long-term antibiotics alone or with reoperation. We excluded other complications, such as gastrointestinal complications, which were less well described. Patients could transition to death from each health state.

Our base-case analysis tracks a simulated cohort of 15-year-old patients with NMS and a Cobb angle of ≥50°. We excluded children who underwent combined anterior spinal fusion and PSF and those with muscular dystrophy because NMS progression in muscular dystrophy may be altered by corticosteroids, which are not viable treatment options for most children with NMS.[21]

Base-case parameters, ranges of values, and corresponding data sources are listed in Table 1. Parameter estimates were derived from literature about all populations of children undergoing NMS when available and otherwise from the subpopulation of children with NMS and cerebral palsy, the most prevalent comorbid condition associated with NMS.[6,8] We discounted costs and outcomes by 3% annually and adjusted all costs to 2018 prices using the Consumer Price Index Inflation Calculator.[22] We used a lifetime time horizon because NMS persists over time, and estimated values are from the health care system perspective. We calculated incremental costs per quality-adjusted life-year (QALY) (combines

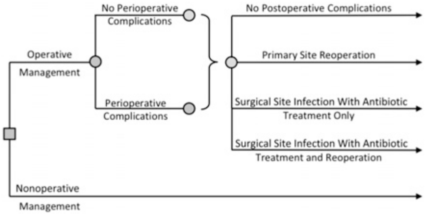

FIGURE 1 Health transition model. Death can occur from any state.

longevity and QoL into a single measure by multiplying the duration of time in 1 health state by indicating QoL on a scale of 0 [death] to 1 [perfect health]; eg, 1 year of life × QoL of 0.5 utility = 0.5 QALY; see Table 2 for definitions), reported as the incremental cost-effectiveness ratio (ICER) (additional cost to gain 1 additional QALY), and compared our findings against the traditional cost-effectiveness threshold of \$50 000 per QALY, with ICERs <\$50 000 per QALY considered more cost-effective.[23,24] Model construction and analyses were performed by using TreeAge Pro 2018 (TreeAge Software, Inc, Williamstown, MA) and Microsoft Excel. Selection of model inputs is detailed below.

Life Expectancy

We based the life expectancy of our base-case analysis on published literature on postoperative life expectancy in children with cerebral palsy and NMS after spinal fusion.[25] We estimated life expectancy to be 26.2 years. However, other sources in the literature had notably different projections of life expectancy, which we used in our sensitivity analyses.[26] In our base-case analysis, we assumed that uncomplicated PSF does not significantly change life expectancy because of inconclusive data on PSF's effect on preventing pneumonia and respiratory failure, the leading cause of death in children with neurologic impairment.[27–29]

HRQOL

To assess the effect of PSF on patients' QoL, we used utility-based estimates of QoL based on the EuroQol 5 dimensions, which combine measures of health on 5 dimensions (mobility, self-care, usual activities, pain and/or discomfort, and anxiety and/or depression) into a composite scale anchored at 0 (death) and 1 (full health).[30] For studies reporting HRQOL with alternative measures such as the Caregiver Priorities and Child Health Index of Life with Disabilities, we mapped results to the EuroQol 5 dimensions to calculate QoL for each health state.[31–35] Because of the markedly lower HRQOL for patients who had complications from operative management, we included these lower values in sensitivity analyses.[31] We assumed that HRQOL is altered by surgical intervention but does not otherwise significantly change over time because cerebral palsy, the most prevalent comorbidity associated with NMS, is a nonprogressive medical condition.

Adverse Events

Perioperative complications for the operative management group included intraoperative hemorrhage, pulmonary complications, and death.[20] Intraoperative hemorrhage included hemodynamic instability, defined as requiring multiple blood transfusions, hypotension, vasopressor support, and associated prolonged ICU monitoring beyond 24 hours postoperation.[20] Pulmonary complications included prolonged intubation beyond 24 hours, reintubation, the need for positive pressure support (above baseline) beyond 48 hours, and associated prolonged ICU monitoring.[19]

Postoperative complications for the operative management group included surgical site infections and implant-related complications.[20] Surgical site infections included prolonged antibiotic treatment and surgical site reoperation. Implant-related complications included malpositioning, malfunction, and other noninfectious, implant-related issues that required reoperation. We estimated transition probabilities for health states and decreases in life expectancy associated with each complication from published literature.[19,25,36] We assumed the risk of death during reoperation to be the same as for the initial operation.

Costs

We calculated the baseline annual medical costs of caring for a child with NMS in operative and nonoperative management groups from published estimates of annual care for children with cerebral palsy with an intellectual disability age of 14 to 17 years old from the MarketScan Multi-State Medicaid database.[37] Baseline costs included inpatient admissions, outpatient encounters, and medications. We selected this subgroup of children with cerebral palsy, which had the highest anticipated expenditures, because we felt they were reflective of the likely higher baseline costs of care for children with NMS who undergo PSF, who have a high prevalence of comorbid conditions: 95.6% have ≥1 comorbid condition, and 58.5% have ≥4 comorbid conditions.[7,37] Although we were unable to get annual baseline cost estimates for privately insured patients, the majority of children with NMS undergoing spinal fusion are covered exclusively or in part by Medicaid.[7] We assumed nonoperative management modalities, including bracing and physical therapy, were included in the baseline costs of care for this population. Because of the overall heterogeneity of health conditions in children with NMS, we included the annual cost estimates from children with cerebral palsy without intellectual disability in our sensitivity analyses.

We calculated the costs of operative management from weighted averages of published costs for PSF that included direct, indirect, variable, and fixed costs of individual encounters for NMS surgery from 2 distinct institutions.[14,15] We used 1 of these sources to estimate the costs of perioperative complications.[14] We estimated the costs for treatment of surgical site infections with antibiotics and reoperation from a cost-savings analysis of strategies to prevent surgical site infection in spinal fusion in adults with posterior spinal surgery involving multilevel fusions with hardware because pediatric sources were unavailable.[38] Costs were based on reimbursement received by the hospital and did not include costs of the new implant (if placed) or physician fees.

Sensitivity Analyses

Sensitivity analyses systematically evaluate the impact of uncertainty in estimates of model variables and of assumptions in the models.[24] When small changes in variable estimates or changes to assumptions result in large changes in the ICER, and thus cost-effectiveness of a treatment, they are important items to consider during decision-making. We tested all model variables with values across the full range of published values or ±15% of the

TABLE 1 Decision Tree Variables Examined in Base-Case and Sensitivity Analyses

Variable	Base-Case Value, Point Estimate (Range)[a]	Parameter Distribution[b]	Source(s)
PSF risks			
Relative risk, mean			
Perioperative death	0.003 (0–0.01)	β	Basques et al[51]; Tsirikos et al[25]; Asher et al[36]; Ersberg and Gerdhem[33]; Reames et al[8]
Perioperative complication	0.181 (0.04–0.28)	β	Ersberg and Gerdhem[33]; Sharma et al[19]; Reames et al[8]
Death after perioperative complication	0.0054 (0–0.01)	β	Reames et al[8]
Reoperation	0.072 (0–0.13)	β	McLeod et al[52]; Rappaport et al[15]; Asher et al[36]
Surgical site infection	0.089 (0.03–0.24)	β	Cahill et al[53]; Szöke et al[54]; Smith et al[55]; Sharma et al[19]; Ramo et al[56]; Reames et al[8]; Mackenzie et al[57]; Martin et al[58]
Reoperation for surgical site infection	0.580 (0–0.75)	β	McLeod et al[52]; Ramo et al[56]
Perioperative death with reoperation	0.003 (0–0.01)	β	Basques et al[51]; Tsirikos et al[25]
Costs			
Health care and nonhealth care costs, 2018 US dollars			
PSF	75 367 (46 980–107 970)	γ	Diefenbach et al[14]; Rappaport et al[15]; Berry et al[6]
Perioperative complication, additional cost	29 277 (0–58 455)	γ	Diefenbach et al[14]
Reoperation[c]	188 973 (58 574–555 163)	γ	Emohare et al[38]
Medical care, yearly (all groups)[d,e]	52 923 (16 721–172 229)	γ	Kancherla et al[37]
Utility wt and life expectancies			
PSF			
Utility wt	0.63 (0.43–0.79)	β	Ersberg and Gerdhem[33]; Bohtz et al[31]; Watanabe et al[34]; Sewell et al[32]; DiFazio et al[35]
Life expectancy, y	11.2 (10.5–11.9)	γ	Tsirikos et al[25]
PSF with reoperation			
Utility wt	0.59 (0.37–0.80)	β	Ersberg and Gerdhem[33]; Bohtz et al[31]; Watanabe et al[34]
Life expectancy, y	10.7 (10–11.3)	γ	Tsirikos et al[25]; Asher et al[36]
PSF with reoperation and surgical site infection			
Utility wt	0.49 (0.34–0.65)	β	Ersberg and Gerdhem[33]; Bohtz et al[31]; Watanabe et al[34]
Life expectancy, y	8.96 (8.4–9.5)	γ	Tsirikos et al[25]; Asher et al[36]
PSF with medically managed surgical site infection			
Utility wt	0.51 (0.36–0.68)	β	Ersberg and Gerdhem[33]; Bohtz et al[31]; Watanabe et al[34]
Life expectancy, y	9.33 (8.8–9.9)	γ	Tsirikos et al[25]; Asher et al[36]
Nonoperative scoliosis management			
Utility wt	0.40 (0.18–0.61)	β	Ersberg and Gerdhem[33]; Bohtz et al[31]; Watanabe et al[34]; Sewell[32]; DiFazio et al[35]
Life expectancy, y	11.2 (11.2–33.5)	γ	Tsirikos et al[25]; Strauss et al[26]

[a] Upper and lower bounds used in sensitivity analyses were based on the widest range of reported values unless otherwise noted.
[b] β is a continuous statistical distribution bounded between 0 and 1. γ is a continuous statistical distribution with a lower bound value of 0.
[c] Discounted 3% yearly on the basis of mean time to reoperation.
[d] Discounted 3% yearly on the basis of life expectancy.
[e] Value shown is based on the life expectancy of the nonoperative group.

base-case parameter when no wider range was available, as shown in Table 1. Our 2-way sensitivity analyses focused on evaluating the effects of HRQOL and life expectancy because of the uncertainty in our estimates of these values and the heterogeneity of comorbid conditions that can significantly affect a child's HRQOL and life expectancy. We also tested our assumptions that (1)

TABLE 2 Definitions of Relevant Health Economic Terms

Relevant Terms	Definition
Health utility wt	QoL rated on a scale: 0 (death) to 1 (perfect health)
QALY	Combines longevity and QoL into a single measure by multiplying the duration of time in 1 health state by a utility wt (eg, 1 y of life × QoL of 0.5 utility = 0.5 QALY)
ICER	Δ in costs between 2 treatments and Δ in QALYs between 2 treatments

HRQOL does not change over time, accounting for children who experience gradual clinical decline, and (2) there was no crossover between treatment groups because clinical practice suggests nonoperative management may delay but not replace operative management.

RESULTS

Base-Case Analysis

We estimated discounted life expectancy for the operative management group to be 10.9 years and for the nonoperative management group to be 11.2 years. The modestly shorter life expectancy was attributed to the additional mortality associated with the operation, intraoperative complications, and postoperative complications. On the basis of our review of the literature, we estimated the QoL to be 0.61 for the operative management group and 0.37 for the nonoperative management group, indicating that QoL is substantially improved with operative management (on a scale of 0 [death] to 1 [perfect health]). On the basis of discounted life expectancy and QoL, we estimated the QALYs to be 4.84 for the operative management group and 3.21 for the nonoperative management group. The longer quality-adjusted life expectancy with operative management reflects the substantial improvement in QoL relative to nonoperative management.

Costs were calculated on a lifetime time horizon. The main components of costs were cost of surgery, estimated as $75 400, and baseline care costs for NMS and comorbidities, estimated as $52 900 per year. We estimated discounted lifetime costs of care to be $507 750 for operative and $425 700 for nonoperative management.

We estimated the ICER as $50 100 per QALY gained for operative management when compared with nonoperative management. Table 3 summarizes these results. Figure 2 shows the cost-effectiveness frontier for the analysis, which plots costs against QALYs to demonstrate which treatment is associated with the lowest cost and which is associated with the highest QALYs.

Sensitivity Analysis

We conducted extensive sensitivity analyses, assessing the impact of uncertainty from all model inputs. The evidence about the effect of surgery on QoL is mixed, and therefore, we conducted sensitivity analyses on the improvement of QoL between operative and nonoperative management. The cost-effectiveness of operative management depends on the small differences in the QoL between nonoperative and operative management, as shown in Fig 3. In our base-case analysis, we estimated the QoL after operative management to be 0.61 and after nonoperative management to be 0.37, indicating an improvement in the QoL of 0.24, which is substantial. As the difference in the QoL between operative and nonoperative management decreases, the cost-effectiveness of operative management becomes markedly less favorable, as seen in Fig 3.

In 1-way sensitivity analyses, our findings proved to be robust to uncertainty among all probability variables at a willingness-to-pay threshold of $50 000 per QALY, except when probability of perioperative death exceeded 0.004, as seen in Supplemental Fig 4. Our findings were also sensitive to variations in annual baseline care costs, with operative management preferred when costs exceeded $53 192, as seen in Supplemental Fig 5, and variations in additional life expectancy, with operative management preferred when additional life expectancy exceeded 11.2 years, as seen in Supplemental Fig 6.

When evaluating the relationship between HRQOL and life expectancy, operative management is preferred for lower HRQOL but longer life expectancy, as seen in our 2-way sensitivity analysis for the nonoperative group in Supplemental Fig 7. Our model was not sensitive to the decline in HRQOL of 15% per year.

We also simulated crossover from nonoperative to operative management because clinical practice suggests that nonoperative management may delay but not replace operative management. We used a Markov model and assumed that 50% of patients undergo operative management immediately and patients crossover at 5% per year, with a cycle length of 1 month and a half-cycle correction. We estimated the life expectancy of the crossover group to be 11 years and QALYs to be 4.31, as seen in Table 3. Crossover management was more costly and less effective than operative management, as seen in Fig 2. Our findings were robust to 1-way sensitivity analyses of all variables, including the initial proportion of patients undergoing operative management and rate of crossover from nonoperative to operative management.

DISCUSSION

Children who are potential candidates for PSF often have medical complexity that may affect the length and QoL and make predictions about clinical trajectory

TABLE 3 Health and Economic Outcomes From NMS Treatment Approaches

Base-Case Analysis	Cost, $	QALY	LE
Operative management group	507 750	4.84	10.9
Nonoperative management group	425 682	3.21	11.2
Crossover group (sensitivity analysis)	515 280	4.31	11.0
ICER of operative versus nonoperative management	50 063 per QALY	—	—

LE, life expectancy; —, not applicable.

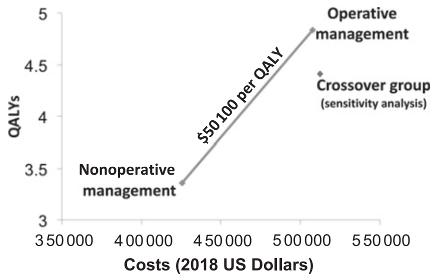

FIGURE 2 Cost-effectiveness frontier of PSF compared with nonoperative management for children with NMS (base case).

difficult.[39] Our results suggest (by using the traditional cost-effectiveness threshold of $50 000 per QALY gained) that from the health care system perspective, PSF is marginally not cost-effective. However, our estimates were highly sensitive to the degree to which PSF improves QoL, indicating that the cost-effectiveness of PSF depends on how children with NMS and their parents value the gains in QoL associated with PSF.

The decision to undergo PSF depends heavily on the balance between length of life and QoL. We observed that PSF appears more cost-effective for children with NMS when children have larger anticipated improvements in QoL and longer anticipated survival times, suggesting children without medical complexity or life-shortening conditions are better candidates for operative management. On the basis of our results, for children with medical complexity or with life-shortening conditions, PSF could trade length of life for QoL, but if NMS is the key factor in lowering QoL, PSF could result in marked improvements for the child's QoL and would be highly cost-effective. Although the expense of PSF surgery is a widely discussed consideration for PSF, we found that surgical costs minimally impact the cost-effectiveness of PSF.[6,40] The high annual baseline care costs of children with NMS have bigger effects on lifetime medical costs than PSF, likely limiting the effect of PSF costs on our results. Similarly, in sensitivity analyses, high surgical-complication rates minimally impacted the cost-effectiveness of PSF because their impact was short lived except for death. However, complications should be viewed from a safety perspective, which is not emphasized in cost-effectiveness.

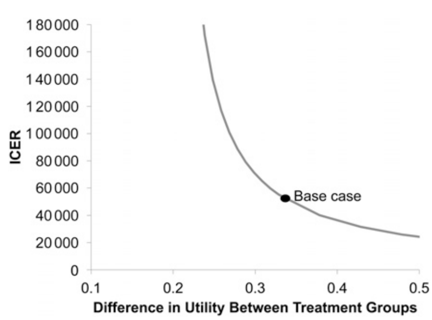

FIGURE 3 One-way sensitivity analysis of differences in HRQOL utility weight between operative and nonoperative management.

Our results identify additional opportunities for pediatric hospitalists in surgical comanagement roles to improve quality and health outcomes in PSF. Pediatric hospitalist comanagement is associated with timely attention to preoperative clearance needs and decreased length of stay.[1,2,15,17,41] Through preoperative comanagement, pediatric hospitalists can help support families in decision-making and identify comorbidities that may affect gains in HRQOL. On the basis of our results, the degree to which PSF influences HRQOL substantially impacts health outcomes and also varies by patient. Therefore, future work should include elucidating HRQOL for children with NMS and their families through shared decision-making, which helps align decisions with individual preferences and values when there is no clear best option.[42–44] Because of medical complexity, children with NMS may have increased risk of low-quality shared decision-making.[39,45] Hospitalists may also be well suited to proactively identify those children with NMS admitted for other conditions who may benefit from timely orthopedic consultation for PSF. For example, children with severe NMS may be repeatedly hospitalized for impaired lung function or pneumonia or be noted to have poor wheelchair fit during discharge planning. These symptoms could indicate that a timely referral to orthopedic surgery is needed.

The potential for improvement in HRQOL is a central consideration when discussing operative management of NMS. Gains in discounted HRQOL with operative management of ≥0.17 resulted in a more favorable cost-effectiveness ratio for operative management. This sizeable change in QoL is plausible and is analogous to a change in adults with osteoarthritis of the hip before versus after total hip arthroplasty.[46–48] The main contributors to QoL differences included parent and provider report of improved pain, mobility, and feeding tolerance from operative management.

If a child's HRQOL is primarily impacted by these contributors, PSF could significantly improve HRQOL. However, children with NMS may have overall low HRQOL because of multiple comorbidities. NMS surgery may not significantly improve HRQOL because it does not address all the comorbidities that contribute to low HRQOL. Decision-making for NMS treatment should include assessment of the determinants of HRQOL for the child and discussion about the degree to which PSF influences HRQOL. Future research that longitudinally and prospectively evaluates HRQOL in children with NMS who receive operative or nonoperative management could enable the development of clinical-decision support tools to help identify patients who may benefit the most from PSF.

Our study has several key limitations. Our analysis was limited by the quality of published data describing HRQOL and gains in HRQOL from operative management. Thus, we were unable to stratify our population on the basis of functional status. Furthermore, QoL assessments in this population are often indirect, provided by parents and caregivers, because developmental delay in many children with NMS precludes direct assessment. Indirect HRQOL assessments introduce additional uncertainty from possible caregiver bias. In previous studies comparing proxy QoL assessments by parents and children of the same developmental age as the child, parents provided more optimistic estimates of QoL than children.[46] Another limitation is insufficient published data on long-term outcomes of NMS, including life expectancy. Children with NMS frequently have comorbidities that impact their individual risk for PSF. However, our findings proved to be robust to variation in probabilities of surgical complications, suggesting that individual risk factors do not drive cost-effectiveness of operative management. Clinically, the long-term life trajectory of children with comorbid conditions is highly unpredictable, and even strong estimates of life expectancy in our model would not meaningfully inform counseling for NMS treatment selection.

CONCLUSIONS

Our analysis indicates that the effectiveness and cost-effectiveness of PSF depends strongly on the degree to which it improves QoL. If PSF results in substantial gains in QoL without substantial increased mortality, the procedure could provide important gains in quality-adjusted survival at a good value. However, gaps in empirical evidence about the effect of PSF on QoL and survival limit our ability to make a definitive assessment about its cost-effectiveness. Prospective and controlled evaluation with objective quantification of HRQOL and clinical outcomes is needed across a longer time span. In the face of this uncertainty about outcomes, a better understanding of patient and caregiver preferences and values may facilitate higher-quality, ethical decision-making and care concordant with their preferences and values.[49,50] Our analysis suggests these preferences and values should be a primary consideration in NMS treatment decisions.

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