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Cost-Effectiveness of Decompression Strategies in Pediatric Chiari I Malformation: A Decision Analysis

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Abstract

Purpose: Comparing cost-effectiveness of bone-only posterior fossa decompression (PFD) versus decompression with duraplasty (PFDD) for surgical management of pediatric Chiari malformation type I (CM-I).

Methods: A decision analytic model was created comparing PFD versus PFDD and simulated a cohort of pediatric CM-I patients with exertional headaches. Four outcome definitions were evaluated, reflecting risk tolerances: permissive (any headache improvement), revision-averse (improvement without revision), complication-averse (improvement without complications), and stringent (improvement without complications or revision). Incremental cost-effectiveness ratios (ICERs) at a willingness-to-pay threshold of $20,000 per negative outcome avoided. One-way and multi-way sensitivity analyses examined parameter influence on cost-effectiveness. Threshold analysis identified cost/ variable margins at which strategy preference changes.

Results: Base case costs were $18,480 for PFD and $19,052 for PFDD (incremental cost $572). PFDD demonstrated reduced revision rates (8.3% vs 17.9%) but increased complications (23.4% vs 13.7%). In the complication-averse scenario, PFD dominated (68% vs 67% success at lower cost). In the revision-averse scenario, PFDD demonstrated robust cost-effectiveness (ICER $3,615 per negative outcome avoided; effectiveness advantage 82% vs 66%) with stability across sensitivity analyses and cost threshold margin of $2,868. Permissive and stringent scenarios showed moderate parameter sensitivity with ICERs of $7,919 and $11,562, respectively (both below the $20,000 threshold). Three-way sensitivity analysis of the stringent scenario revealed PFDD's cost-effectiveness depending on maintaining complication rate differences at or below current levels.

Conclusions: PFD is superior when minimizing complications is paramount; PFDD is cost-effective when avoiding revision is prioritized. These findings support individualized, family-centered surgical planning regarding complication-revision trade-offs.

INTRODUCTION

Chiari malformation type I (CM-I), characterized by caudal displacement of the cerebellar tonsils below the foramen magnum, is a common diagnosis in up to 4% of pediatric patients undergoing brain or cervical spine MRIs for any reason [10, 22]. Surgical management is indicated in patients with commonly associated symptoms, including posterior exertional headaches, cervical or cervicothoracic syringes, central sleep apnea, swallowing difficulties, or progressive atypical scoliosis [9, 19]. The most common symptom, posterior exertional headache, is thought to result from transient increases in intracranial pressure with Valsalva maneuvers resulting in further compromise to spinal fluid flow [20]. Due to this pathophysiology, suboccipital decompression to create more space and restore physiologic CSF flow at the foramen magnum is widely accepted as the treatment for symptomatic CM-I [2, 6, 15, 20].

Two major surgical strategies exist to achieve this decompression. A bone-only posterior fossa decompression (PFD) involves a suboccipital craniectomy with or without C1 laminectomy [8, 20]. A posterior fossa decompression with duraplasty (PFDD) takes PFD further by opening the dura and, at a minimum, sewing in a redundant dural graft. This dural opening also provides the opportunity to lyse arachnoid adhesions, cauterize or resect the cerebellar tonsils, and explore the obex [1, 4, 21]. While there are variabilities in which of these are performed based on surgeon preference and intraoperative findings, these techniques allow for maximal cerebrospinal fluid (CSF) flow through the bottom of the skull [3, 6, 7].

Both techniques are acceptable treatment options for patients with symptomatic CM-I; however, controversy exists over which is the more favorable option in pediatric patients. PFD is a less invasive procedure with a lower complication rate and hospital stay; however, there may be a lower success rate in terms of symptomatic improvement, which may necessitate further surgical intervention [8, 15]. Alternatively, PFDD may have a higher success rate but is more invasive with higher risk and severity of complications including CSF leak, postoperative hydrocephalus, and bacterial meningitis [1, 6, 10, 15–19].

First line treatment practices vary by institution and by patient presentation, with patients who present with a syrinx more likely to be offered PFDD upfront [19, 23]. Some studies suggest that PFD may be cost effective as a first treatment, particularly in the absence of syringomyelia, due to low complication rates, surgical time, and length of stay [13]. However, the potential differences in effectiveness, need for further surgery, and distinct complication profiles of these strategies were not integrated into this cost study.

These controversies in the literature make it challenging to determine the superior strategy without a large-scale multicenter efficacy trial. Cost-effectiveness analysis techniques allow for the exploration of the cost and comparative effectiveness of different treatment strategies simultaneously without the resource intensiveness of a large-scale trial. Here we provide a cost effectiveness decision model based on the literature comparing PFD and PFDD for the treatment of posterior exertional headaches to better understand differences in overall cost and if there is clinical equipoise between the two techniques.

METHODS

This evaluation of the cost effectiveness of PFD and PFDD for the treatment of exertional headache follows the reporting guidelines recommended by the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) [11]. CHEERS aims to standardize best practices in reporting economic evaluations in healthcare. Since no largescale clinical trial data are available, published reviews, meta-analyses, large observational studies, and cost analyses informed model inputs [1, 5, 12, 13].

Decision Model

Given unclear differential clinical effectiveness, complication rate, and repeat surgery rate between PFD and PFDD [2, 6, 8, 13–15], a model that includes a range of probabilities and costs of clinical outcomes

associated with these procedures is important to better understand the relative cost effectiveness. A decision model was developed that simulates the risks, benefits, and costs associated with choosing PFD or PFDD in a patient population whose primary CM-I symptom was exertional headaches. TreeAge Pro software was used to construct the model (version 2025 arch64, TreeAge Software, Williamstown MA). Probabilities of surgical complications within 6 months, revision surgery, and improvement of headache were based on the observational data of both PFD and PFDD from the Park-Reeves Syringomyelia Research Consortium study, which included 692 pediatric patients, the largest multi-center pediatric cohort to date [1]. The base case is an average patient with CM-I with syringomyelia and posterior exertional headaches who undergoes a decompressive surgery. Due to the lack of detailed long-term data, the time horizon for this analysis is one year after the index surgery and thus no discounting was applied for the analysis.

Because there are no easily accessible utility data for pediatric CM-I patients, binary effectiveness assignments were used, with 1.0 denoting a positive outcome and 0 a negative outcome, calculating the proportion of the modeled cohort with a positive outcome. To assess cost effectiveness changes based on individual risk tolerance, the analysis was run with four progressively rigid positive outcome definitions: 1. Permissive (headache improvement regardless of complication or revision), 2. Revision averse (headache improvement without revision surgery), 3. Complication averse (headache improvement without complications), 4. Stringent (headache improvement without revision or complication).

Model Inputs: Clinical Variables and Costs

Inputs and data sources for index procedures are summarized in Table 1. It was assumed that revision surgery was a PFDD in all cases, and that the complication risk was consistent with a primary PFDD procedure. Acute perioperative surgical complications were considered in this model including pseudomeningocele, CSF leak, wound revision in or out of the operating room, postoperative hydrocephalus, meningitis, and cervical instability. To accommodate variability in probabilities, wide sensitivity analyses were performed for each strategy as this was not available in prior meta-analyses nor large observational studies.

Table 1
Clinical parameters and input sources

Surgical Outcome Parameters	PFD Base Case	PFDD Base Case	Source
Complication risk	13.7% (1–25%)	23.4% (10–40%)	Akbari et al., 2022[1]
Improvement in headache	80.8% (70–90%)	89.6% (80–95%)	Akbari et al., 2022[1]
Revision surgery	17.9% (10–25%)	8.3% (0–25%)	Akbari et al., 2022[1]
Surgical cost parameters			
Surgical cost	$14,465 ($11,000-$18,000)	$16,096 ($15,000-$20,000)	Litvack et al., 2013[13] Lam et al., 2016[12]
Complication cost	$6,281	$6,281	Lam et al., 2016[12]

Base case values taken from largest available literature sources noted above. Sensitivity analysis values were determined by smaller studies discussed in the narrative above.

Surgery costs were obtained from the Lam et al. 2016 [12] study comparing pediatric CM-I treatment in different hospital settings using data from the Kid's Inpatient Database (KID). KID is the largest nationally available all-payer pediatric inpatient administrative database and was developed for the Healthcare Cost and Utilization Project, sponsored by the Agency for Healthcare Research and Quality. Costs were calculated via hospital specific application programming interface (API) cost to charge ratios (CCR) or group weighted average CCR. The costs were then adjusted by regional hospital wage level. This study estimated a baseline index surgery cost of $16,096 and an average surgical complication cost at $6,281 [12]. While this study did not stratify costs by procedure performed, a prior study estimated a 10% lower cost in PFD [13]. As such, $14,465 was used as the base cost for PFD. Also given cost potential variability, wide ranges were applied to these as well to accommodate differences in cost based on hospital type ($11,000, $20,000). A healthcare perspective was used for this analysis, and as such only direct medical costs were included.

To facilitate clinically meaningful interpretation of the results, variable values for the cost of index surgery and probabilities of complication, need for revision, and symptomatic improvement for PFDD were defined as a function of the PFD variable based on the absolute percentage point differences between the two identified in the literature.

Analysis

A cost-effectiveness analysis was performed to calculate cost per positive outcome gained (or equivalently, per negative outcome avoided) for each positive outcome definition. PFD and PFDD were compared using incremental cost effectiveness ratios (ICER) at each positive outcome definition. The ICER is the difference in treatment cost divided by the difference in probability of positive outcome for each strategy. The willingness-to-pay threshold was conservatively set to $20,000 per negative outcome avoided to account for an average surgical cost to mitigate a negative outcome.

One-way sensitivity analyses were performed for each positive outcome by varying model parameters over ranges listed in Table 1, generally based on reports from observational literature [1, 2, 5, 6, 8, 12–15, 18]. This allowed evaluation of each variable and its effect on the preferred treatment strategy for each positive outcome definition. For parameters whose variation could potentially affect the favorability of treatment strategies, a threshold analysis was performed to determine at what probability the change in favorability occurred. To further assess the effect on favorability between the two treatments, clinical parameters were jointly varied through two-way sensitivity analyses using the differences between revision probability, complication probability, and improvement probability for each outcome definition.

RESULTS

Base Case Results

The decision tree model (Fig. 1) compared costs and outcomes for PFD versus PFDD across the four outcome definitions. Base case costs were $18,480 for PFD and $19,052 for PFDD, an incremental cost of $572. Effectiveness varied substantially by outcome definition (Table 2). Under the permissive definition (any headache improvement), PFDD achieved 90% success versus 82% for PFD. However, when complications were weighted as failures (complication-averse definition), PFD achieved 68% success versus 67% for PFDD.

Cost-effectiveness ratios reflected these effectiveness differences (Table 2). PFDD was more favorable in the revision-averse scenario with an ICER of $3,615 per additional success (or per negative outcome avoided) and the stringent scenario wherein neither complication nor revision were tolerated with an ICER of $11,562 per negative outcome avoided. Both ICERs fall well below the average cost of a complication and revision surgery at $20,000. Conversely, in the complication-averse scenario, PFD dominated, i.e., PFD both cost less and was more effective. The permissive scenario (ICER $7,919) represented an intermediate position where PFDD remained cost-effective but less compelling than the revision-averse scenario.

Table 2
Base case cost effectiveness analyses

Outcome Definition	PFD (effectiveness, %)	PFDD (effectiveness, %)	ICER
Permissive	82	90	$7,919
Revision Averse	66	82	$3,615
Complication Averse	68	67	PFD Dominant*
Stringent	57	62	$11,562
ICER = incremental cost effectiveness ratio			
* PFD is less costly and more effective than PFDD at this outcome definition			

Outcome definitions reflect likelihood of a positive outcome at several risk tolerance levels with Permissive suggesting a positive outcome with symptom improvement regardless of need for more surgery or complications and stringent defining a positive outcome only if symptoms improve without complication or need for revision. Cost of PFD and PFDD for all outcome definitions was $18,480 and $19,052, respectively with an incremental cost of $572.

Sensitivity Analyses

In one-way sensitivity analyses, variations of multiple parameters led to changes in strategy favorability, indicating that effectiveness differences between techniques are sufficiently small that plausible parameter variations can reverse treatment preference. For each outcome, variables and thresholds that caused a change in favorable treatment were identified (Table 3). These variables were chosen for the subsequent multi-variable sensitivity analyses. Notably, the most influential variables in the one-way sensitivity analyses were those that represented the differences of cost, complication rate, improvement rate, and revision rate between the two treatments. Smaller differences in the incremental probability of improvement between the two surgeries favored PFD, whereas smaller differences in complication rate and cost favored PFDD. A greater likelihood of revision in PFD (i.e. more negative incremental value between PFDD and PFD), favored PFDD. Variables chosen for two-way sensitivity analyses were based on which incremental probability variables affected favorability at each outcome definition (Table 3). Figure 2 shows representative multi-way sensitivity analyses of outcome definitions.

Table 3
One-way sensitivity analyses. Parameters whose variation changed treatment favorability at a $20,000 per negative outcome avoided threshold.

Outcome and Parameter*			Favored	
	Base Value	Threshold Value	Below Threshold	Above Threshold
Permissive				
Diff. in Probability of Improvement	8.8%	3.5%	PFD	PFDD
Diff. in Probability of Revision	-9.6%	-4.5%	PFDD	PFD
Diff. in Index Surgical Cost	$1,631	$2,596	PFDD	PFD
Stringent				
Diff. in Probability of Improvement	8.8%	5.8%	PFD	PFDD
Diff. in Probability of Revision	-9.6%	-8.2%	PFDD	PFD
Diff. in Probability of Complication	10.6%	12.5%	PFDD	PFD
Diff. in Index Surgery Cost	$1,631	$2,093	PFDD	PFD
Revision Averse				
Diff. in Probability of Improvement	8.8%	-5.3%	PFD	PFDD
Diff. in Probability of Revision	-9.6%	-2.2%	PFDD	PFD
Diff. in Index Surgical Cost	$1,631	$4,499	PFDD	PFD
Complication Averse				
Diff. in Probability of Revision	-9.6%	-13.4%	PFDD	PFD
Diff. in Probability of Complication	10.6%	6.8%	PFDD	PFD

* Differences (Diff.) are calculated as the parameter value with PFDD minus the parameter value with PFD as listed in Table 1

For the permissive outcome definition (Fig. 2A), the base case is positioned within the PFDD-favorable region. However, PFD occupies a substantial portion of the parameter space, particularly when PFDD's improvement advantage or revision benefit approaches zero. This decision boundary indicates that

PFDD's cost effectiveness in this scenario depends on maintaining both its revision reduction and superior improvement rates, if either benefit is substantially reduced, PFD would be favored.

In varying differences in revision rate and complications for the complication averse scenario (not shown), PFD is clearly favored in the base case. PFDD is favored if there is at least a 7.8% lower revision rate and a lower than 12% difference in complication rate. Conversely, PFDD is clearly favored in the base case revision averse scenario when varying differences in revision rate and improvement (Fig. 2A). The PFDD must have at least a 0.7% *greater* rate of revision and at least a 14.6% *lower* improvement rate for PFD to be favored in this case.

In the stringent scenario, a three-way sensitivity analysis of the three most influential variables, absolute difference in the probability of revision, improvement, and complication was performed (Fig. 2B). In the base case at about a 10% absolute difference in complication risk, PFDD is favored but very close to the decision threshold line. This suggests that this favorability is fragile and sensitive to even small alterations of any of these three variables.

DISCUSSION

The adequacy of PFD for CM-I decompression has long been debated, with many surgeons favoring duraplasty based on understanding of the pathophysiology and higher revision rates observed in bone-only approaches [19]. This perspective may not adequately account for perioperative morbidity associated with dural opening. Recent work has suggested that PFD has good symptomatic relief in exertional headaches [2, 3, 5, 8]. The tradeoff between lower complication rate and cost, but possibly lower effectiveness, makes the PFD vs PFDD debate an ideal topic to explore using decision analytic modeling. This analysis suggests that neither PFD nor PFDD is universal. Rather, optimal strategy selection depends critically on patient-specific priorities regarding competing clinical risks. Our findings reveal changes in favorability based on how complications and revisions are weighted when defining a positive outcome.

Despite different risk profiles, PFD and PFDD have comparable cost effectiveness that slightly favors PFDD at a willingness to pay threshold of $20,000. PFDD has a reduced revision surgery rate (17.9% vs 8.3%) but has an equal increase in complications (23.4% vs 13.7%), while costing an additional $572. When complications were weighed as treatment failures, PFD demonstrates clear superiority, dominating by costing less ($18,480 vs $19,052) while achieving marginally better outcomes (68% vs 67% success). Conversely, when revisions were weighed as failures, PFDD demonstrated robust cost-effectiveness with an ICER of $3,615 per negative outcome avoided and a 16 percentage point effectiveness advantage (82% vs 66%). Threshold analysis reveals substantial margins: PFDD maintained cost effectiveness even if surgical costs increased by $2,868 above the current difference. Two-way sensitivity analysis demonstrates that PFDD remains preferred across wide parameter variations, requiring simultaneous reduction in both improvement advantage and revision benefit before PFD becomes preferred.

Favorability was less clear in the sensitivity analyses of the permissive (any improvement) and stringent (complications and revisions seen as failures) outcome definitions. PFDD was cost effective in the base case with ICERs of $7,919 and $11,562, respectively, both well below our $20,000 willingness-to-pay threshold. However, its favorability was moderately fragile in two-way sensitivity analyses, with decision boundaries positioned near base case estimates particularly in the stringent scenario. This suggests that in the stringent scenario, the optimal strategy may depend on more precise institutional performance and patient characteristics.

This pattern reflects a fundamental clinical trade-off between the revision rate, symptomatic improvement, and complication rate. PFDD reduces revisions by 9.6 percentage points (17.9% to 8.3%) and improves symptomatic outcomes by 8.8% points (80.8% vs 89.6%), but has a 10.6 percentage point increased complication rate (13.7% vs 23.4%) while costing $1,631 more for the index procedure. How one prioritizes these competing factors will determine the optimal strategy and should be individualized to patients and families. These threshold-crossing behaviors suggest that there is genuine uncertainty about the optimal strategy. Moreover, the difference in revision rate between PFD and PFDD effected threshold crossing behavior in each scenario, suggesting that revision rate has a very strong effect on the model for all definitions.

This fragile balance in the sensitivity analysis supports individualized surgical planning that considers patient-specific risk tolerance rather than defaulting to one technique as both options have distinct advantages. This may influence how surgeons frame the consenting of patients PFD is preferable when families prioritize minimizing perioperative complications, when patients have standard anatomic presentations without high-risk features, and when staged approaches are acceptable. PFDD is preferable when families prioritize single-surgery definitive treatment, when high anesthetic risk makes reoperation undesirable, when geographic barriers limit access to follow-up, or when high risk anatomic features such as a large syrinx, severe tonsillar herniation, or a medullary kink necessitate maximal decompression. For patients without strong preferences or high-risk features, shared decision-making is essential.

During the informed consent, surgeons might explain that there are two well-established approaches. The bone-only approach has lower complication rates but about 18% need a second surgery. The duraplasty approach reduces need for reoperation to 8% but has a higher upfront complication risk. Both achieve good symptom improvement for exertional headache. The choice depends on family priorities.

While the primary data used for this study includes patients with syringomyelia, the symptomatic outcome explored here is exertional headache improvement [1]. Recent studies have also suggested that there may be improvement or lack of progression of syringomyelia with PFD alone [8, 13, 14]. These findings may indicate that PFD may be a suitable procedure for an even larger patient population. Further large-scale studies are needed to better characterize this; however, a similar model could be used to assess syringomyelia as a primary outcome measure.

This study has several limitations. Our model uses observational data for all inputs, and as such is at the mercy of the data available in the literature. Since cost and probability data are not well defined across multiple large studies, we chose to vary all parameters widely in the sensitivity analysis, which may have affected our results. Moreover, institutional bias in observational data may exist as some may favor PFD over PFDD and vice versa. Our binary utility assessments do not capture the differences in complication severity or account for important patient-centered factors such as the psychological burden of revision surgery or initial complications. A utility assessment would better reflect patient specific preferences, but data is not available for CM-I pediatric patients. Third, available cost data were derived from studies at least nine years old and do not account for institutional variation or economic changes during this time period. We performed wide variability in costs to partially compensate for this. Our model aggregated all complications despite substantial variation in severity and management complexity. This was accounted for by a wide range in the complication cost variable. The threshold-crossing behavior in our sensitivity analyses reflects genuine parameter uncertainty rather than model instability. This uncertainty underscores the need for additional high quality comparative data.

CONCLUSION

This decision analysis, which compares PFD vs PFDD over multiple different outcome risk tolerance definitions, demonstrates that optimal surgical strategy for pediatric CM-I is context dependent rather than universal. PFD is superior when avoiding complications is paramount, dominating by costing less while achieving better outcomes than PFDD. Conversely, PFDD is superior when avoiding revision surgery is the primary goal. For stringent and permissive outcome definitions, both strategies are reasonable. This suggests that the optimal choice depends on patient specific priorities regarding the complication revision tradeoff. These findings support individualized, family-centered surgical planning where technique selection aligns with family priorities, suggesting the need for further qualitative work and the development of decision support tools surrounding CM-I surgery in children. Future work should also include systematic review and meta-analysis of PFD outcomes and further models exploring other outcome parameters such as syrinx improvement.

DECLARATIONS

AUTHORS’ CONTRIBUTIONS

Study conception and design were performed by Kenneth J. Smith and Taylor J. Abel. Data collection and analysis, decision model creation, and figure creation were performed by Martin G. Piazza. The first draft of the manuscript was written by Martin G. Piazza. Figure editing was performed by Kenneth J. Smith. All authors contributed to manuscript revision and editing, and supervision was provided by Kenneth J. Smith and Taylor J. Abel. All authors read and approved the final manuscript.

Funding and Competing interests:

The authors have no relevant financial or non-financial interests to disclose.

Ethics approval:

This retrospective chart review study involving human participants was in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. The Human Investigation Committee (IRB) of the University of Pittsburgh approved this study.

Data Availability

The data that support the findings of this study are not openly available due to reasons of sensitivity and are available from the corresponding author upon reasonable request. Data are located in controlled access data storage at the University of Pittsburgh.

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Figures

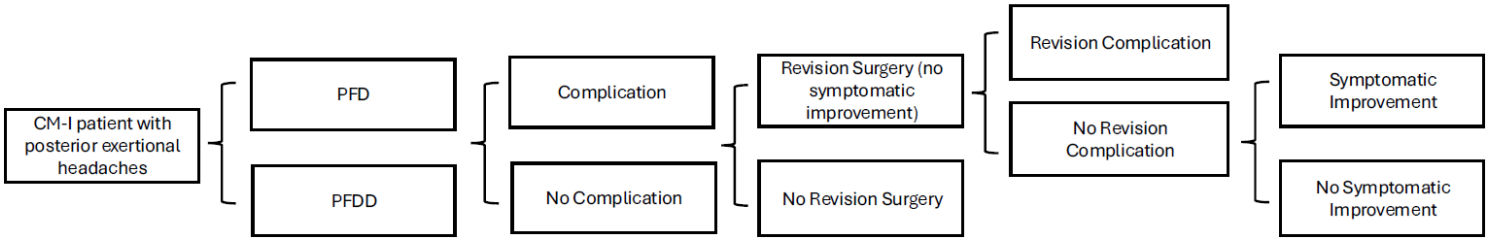


Figure 1

Decision analysis model. Schematic depiction of the decision analysis models used to simulate benefits, risks, and costs of PFD vs PFDD. Definitions of positive and negative outcomes were varied based on risk tolerance options.

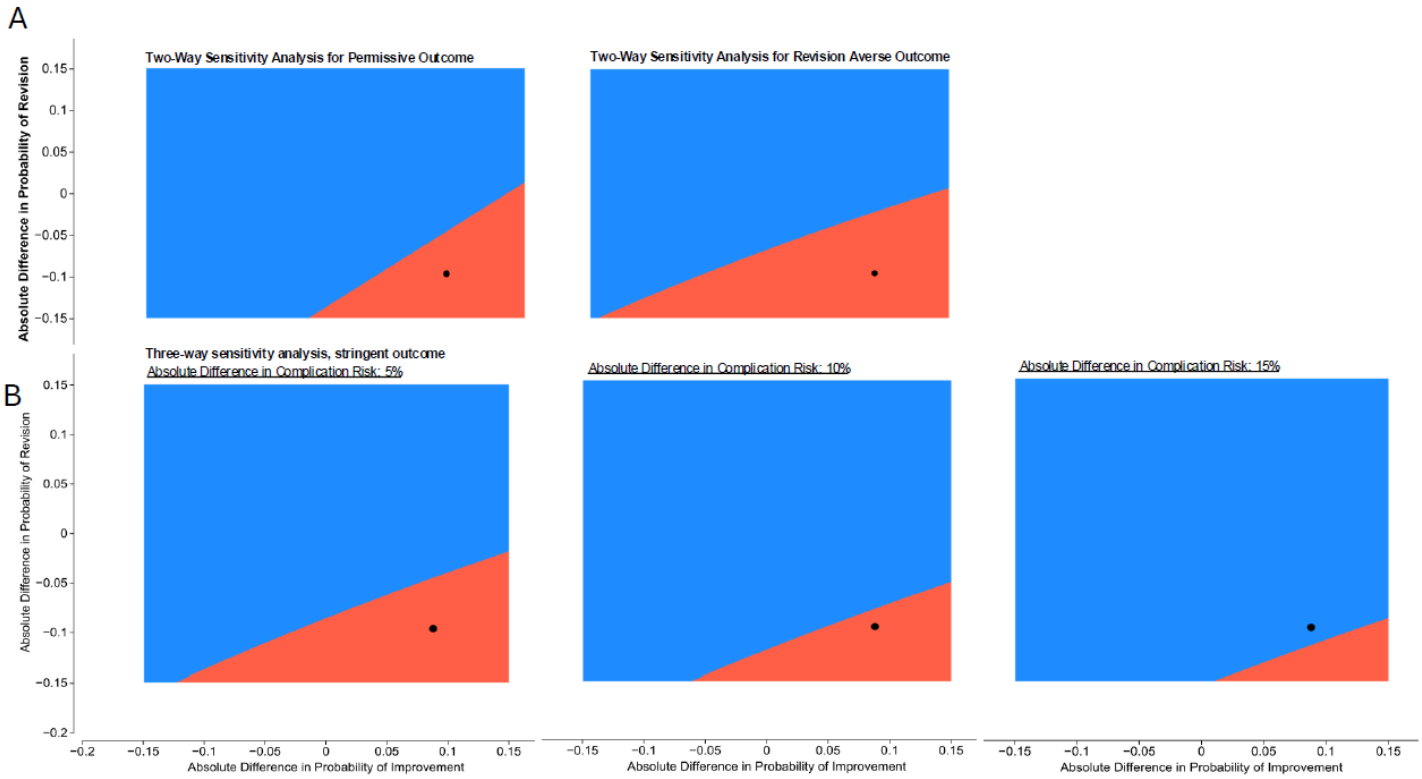


Figure 2

Representative sensitivity analyses.

(A) Two-way sensitivity analyses of clinically relevant variables for permissive and revision averse outcomes. Individual panels depict areas where strategies are favored (PFD in blue, PFDD in red) at a $20,000 per negative outcome avoided threshold with joint variation of differences in improvement and revision The black dot depicts base case values.

(B) Three-way sensitivity analysis, stringent outcome, varying differences in improvement, revision, and complications. Joint variation of differences in improvement and revision probabilities at complication risk differences of 5%, 10%, or 15%. The red area disappears when the complication risk difference between strategies is ≥23% (base case 10.6%).