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Universal versus targeted newborn hearing screening in India: a decision-analytic cost-effectiveness model using secondary parameters

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Research Article

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Abstract

Background

Newborn hearing screening has been part of Rashtriya Bal Swasthya Karyakram since 2013 and the National Programme for Prevention and Control of Deafness now covers 587 districts, yet most Indian screening sites test only high-risk neonates. Whether universal screening justifies its additional cost over a high-risk-register approach was identified as unresolved in 2015 and has not since been answered. A 2024 Health Technology Assessment in India analysis compared screening devices but not screening strategies.

Methods

A decision tree compared universal two-stage screening (otoacoustic emissions followed by automated auditory brainstem response), targeted screening of neonates with Joint Committee on Infant Hearing risk indicators, and no organised screening, in a hypothetical cohort of 100,000 Indian live births from a health system perspective. Parameters were taken from published Indian and international sources and each was tagged as published, derived or assumed. Outcomes were cases of permanent congenital hearing loss (PCHL) confirmed and incremental cost per additional case detected. One-way, threshold and 10,000-iteration probabilistic sensitivity analyses were conducted.

Results

Modelled two-stage sensitivity (0.891) and specificity (0.998) closely reproduced published two-step performance (0.92 and 0.98), supporting internal validity. Universal screening confirmed 305.4 of 500 PCHL cases against 163.6 under targeted screening and 50.0 with no screening, at an incremental INR 43,228 per additional case detected versus targeted screening (probabilistic median 42,775; 95% interval 18,101–202,353). The result was highly sensitive to the share of cases occurring in the high-risk group: the ICER rose from INR 30,165 at 30% to INR 539,639 at 90%, and targeted screening became non-inferior beyond roughly 95%. Notably, 38.9% of all PCHL cases remained unconfirmed even under universal screening, driven by incomplete coverage and loss to follow-up rather than test performance.

Conclusions

Universal screening detects substantially more cases than a high-risk-register approach, but the magnitude of that advantage rests on a parameter never measured in India — the proportion of affected infants without risk factors. The finding that more than a third of cases are missed under universal screening because of coverage and attrition suggests that strengthening follow-up systems may yield more than the choice of screening strategy. This analysis uses published secondary parameters, not

primary costing, and reports cost per case detected rather than per QALY; it is therefore a structured exploration of the decision problem, not a definitive cost-effectiveness verdict.

1. Introduction

Permanent congenital hearing loss (PCHL) affects roughly 5 per 1000 live births in India [1, 2]. Hearing loss is recognised by the World Health Organization as a global public health priority for which early detection and intervention systems are a core recommendation [3]. Detection in the first months of life matters because language acquisition depends on early auditory input [4–6], and the Joint Committee on Infant Hearing benchmark of screening by one month, diagnosis by three and intervention by six months has become the international standard of programme performance [7].

India has the policy architecture for newborn hearing screening. Rashtriya Bal Swasthya Karyakram has included it since 2013 [8], and the National Programme for Prevention and Control of Deafness has expanded from a 25-district pilot to 587 districts across 36 states and union territories [9]. Implementation, however, has not followed the policy. A survey of 80 screening sites in Tamil Nadu found that 67.1% screened selectively — by high-risk criteria, physician referral, or neonatal intensive care admission — rather than universally, and that protocols varied site by site with no centralised tracking system [10].

Whether that matters economically is unresolved. Gupta and colleagues, reporting a universal screening programme at a tertiary centre in Delhi, concluded explicitly that “cost effectiveness of UNHS compared to high risk based screening needs to be determined” [11]. To our knowledge no subsequent Indian analysis has answered that question. A 2024 study by the Health Technology Assessment in India Regional Resource Hub did construct a decision tree for Indian neonatal hearing screening, but compared first-line devices — otoacoustic emissions (OAE) against portable automated auditory brainstem response — rather than the underlying choice between universal and targeted strategies [8]. The device question and the strategy question are distinct, and only the first has been addressed.

This analysis addresses the second. It asks what universal screening buys, relative to screening only neonates carrying recognised risk indicators, in additional cases of PCHL confirmed and at what incremental cost, using parameters drawn entirely from published sources.

2. Methods

2.1 Model structure and strategies

A decision tree was constructed for a hypothetical cohort of 100,000 Indian live births, matching the cohort size used in the preceding Indian analysis to permit comparison [8]. Three mutually exclusive strategies were compared (Fig. 1). Under universal screening, all live births are offered two-stage screening: OAE for every neonate, followed by automated auditory brainstem response (AABR) for those referred at stage one. Under targeted screening, the identical two-stage protocol is offered only to

neonates carrying Joint Committee on Infant Hearing risk indicators [7]. Under no organised screening, cases come to attention only through clinical suspicion. All three pathways converge on confirmatory diagnostic brainstem evoked response audiometry (BERA), and terminal states are confirmed PCHL, false positive, and undetected case.

The perspective is that of the health system, in line with the Indian Reference Case for economic evaluation [12]. The time horizon runs to confirmed diagnosis, within the first year of life, so no discounting was applied. Costs are in Indian rupees. The model was implemented in Python 3; code and parameter files accompany this manuscript. The analysis is reported in accordance with the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) 2022 checklist [13].

2.2 Parameters and their provenance

Every parameter carries an explicit provenance tag (Table 1). Published parameters are point estimates reported in named sources. Derived parameters are arithmetic transformations of published values, with the transformation stated. Assumed parameters are those for which no published Indian estimate could be located; each was varied across a wide plausible range in sensitivity analysis, and the assumed parameters are visually distinguished in Fig. 2 so that readers can see how much of the result rests on them.

Two parameters deserve comment. First, the proportion of PCHL cases occurring among infants with recognised risk indicators is taken as 0.50, following the United States Preventive Services Task Force systematic review finding that approximately half of infants with PCHL have no known risk factor [14, 15]. No Indian estimate exists, and the two available Indian cohorts disagree sharply: Gupta and colleagues found all five detected cases were high-risk [11], while Parida and colleagues reported detection of 22.78 per 1000 overall against 56 per 1000 among high-risk neonates, implying a substantial minority of cases outside the risk group [16]. This parameter is examined by threshold analysis in Section 3.3. Second, AABR sensitivity was reported as 100% in the source study [8]; a point estimate of exactly 1.0 from a finite sample is not credible for probabilistic analysis and was modelled at 0.99.

Table 1
Model parameters, plausible ranges and provenance

Parameter	Base case	Range	Provenance	Source / note
Prevalence of PCHL	5 per 1000	—	Published	Sahoo 2024 [8]
Share of births with JCIH risk indicators	0.10	0.05–0.35	Assumed	No Indian estimate located
Share of PCHL cases in the high-risk group	0.50	0.30–0.90	Published	USPSTF review [14, 15]
OAE sensitivity (stage 1)	0.90	0.69–0.98	Assumed	Sahoo 2024 reports 0.69 standalone [8]
OAE specificity (stage 1)	0.92	0.68–0.97	Assumed	Sahoo 2024 reports 0.68 [8]; AAP standard implies > 0.96 [24]
AABR sensitivity (stage 2)	0.99	0.92–1.00	Derived	Sahoo 2024 reports 1.00 [8]
AABR specificity (stage 2)	0.97	0.90–0.99	Published	Sahoo 2024 [8]
Screening coverage, universal	0.84	0.60–0.98	Published	Gupta 2015 [11]
Screening coverage, targeted	0.90	0.70–0.99	Assumed	Facility-based contact assumed higher
Loss to follow-up before confirmation	0.184	0.05–0.50	Published	Gupta 2015 [11]
PCHL clinically detected by 12 months, no screening	0.10	0.02–0.30	Assumed	No Indian estimate located
Unit cost, OAE (INR)	67	40–120	Published	Sahoo 2024 [8]
Unit cost, AABR (INR)	97	60–180	Published	Sahoo 2024 [8]
Unit cost, diagnostic BERA (INR)	1756	1000–3000	Published	Sahoo 2024 [8]

PCHL, permanent congenital hearing loss; JCIH, Joint Committee on Infant Hearing; OAE, otoacoustic emissions; AABR, automated auditory brainstem response; BERA, brainstem evoked response audiometry; AAP, American Academy of Pediatrics. Provenance: Published = point estimate reported in the named source; Derived = arithmetic transformation of a published value, stated in Section 2.2; *Assumed = no published Indian estimate located, varied across the stated range in sensitivity analysis.*

2.3 Outcomes and analysis

The primary outcome is the number of PCHL cases confirmed, and the primary economic measure is the incremental cost per additional case detected. Quality-adjusted life years were not used, because translating detection into lifetime health gain requires a downstream model of amplification, cochlear implantation and language outcomes that is beyond a short-horizon decision tree [17]. The implication of that choice is addressed in Section 4.

Internal validity was checked by comparing the model's emergent two-stage test performance against published two-step screening performance [14]. One-way sensitivity analysis varied each parameter across its stated range. Threshold analysis examined the share of cases in the high-risk group specifically. Probabilistic sensitivity analysis ran 10,000 Monte Carlo iterations with beta distributions for probabilities and gamma distributions for costs, following standard practice for decision-analytic models [18]; every sampled parameter was verified to reproduce its intended mean before results were generated.

3. Results

3.1 Internal validation

Combining the modelled stage-one and stage-two test characteristics yields a two-stage sensitivity of 0.891 and specificity of 0.998. The USPSTF systematic review reports 0.92 and 0.98 respectively for two-step OAE-then-ABR screening [14, 15]. The close correspondence, which was not imposed, supports the structural validity of the two-stage pathway.

3.2 Base-case results

Base-case results are given in Table 2. Of 500 true PCHL cases in the cohort, no organised screening confirms 50.0, targeted screening 163.6, and universal screening 305.4. Universal screening detects 141.8 more cases than targeted screening at an additional cost of INR 6,128,720, giving an incremental cost of INR 43,228 per additional case detected. Targeted screening relative to no organised screening costs INR 8,103 per additional case detected.

Table 2
Base-case results per 100,000 live births (500 true PCHL cases)

Strategy	Screened	Referred to BERA	Cases confirmed	Total cost (INR)	Cost per case (INR)	ICER (INR per additional case)
No organised screening	0	0	50.0	87,800	1,756	—
Targeted, high-risk only	9,000	222	163.6	1,008,173	6,163	8,103
Universal, two-stage	84,000	575	305.4	7,136,893	23,372	43,228

Strategies are ordered by total cost. ICER is calculated against the next least costly non-dominated strategy. Cases confirmed are true positives surviving loss to follow-up; fractional values arise from a deterministic cohort model.

A finding not anticipated by the strategy comparison is that 194.6 cases — 38.9% of all PCHL in the cohort — remain unconfirmed even under universal screening. This attrition is not a test-performance problem: it arises from 84% screening coverage compounded with 18.4% loss to follow-up between referral and confirmation.

3.3 Sensitivity analysis

One-way results are shown in Fig. 2 and Table 3. The dominant parameter by a wide margin is the share of PCHL cases occurring in the high-risk group. Threshold analysis (Fig. 3) shows the ICER rising from INR 30,165 when 30% of cases are high-risk to INR 539,639 at 90%, with universal screening ceasing to detect additional cases beyond approximately 95%. In other words, the economic case for universal screening is strong under the USPSTF estimate and collapses if Indian risk registers capture cases far more completely than international evidence suggests.

Table 3
Selected one-way sensitivity results (ICER, universal versus targeted)

Parameter varied	Low / high value	ICER at low (INR)	ICER at high (INR)	Provenance
Share of PCHL cases in high-risk group	0.30 / 0.90	30,165	539,639	Published
Unit cost, OAE	INR 40 / 120	28,945	71,265	Published
Screening coverage, universal	0.60 / 0.98	74,998	37,983	Published
Loss to follow-up	0.05 / 0.50	37,634	68,292	Published
OAE specificity	0.68 / 0.97	60,955	39,535	Assumed
OAE sensitivity	0.69 / 0.98	55,814	39,853	Assumed
Share of births high-risk	0.05 / 0.35	45,710	30,818	Assumed

Base-case ICER is INR 43,228 per additional case detected. Parameters are ordered by the width of the interval they produce.

Probabilistic analysis (Fig. 4) gave a median ICER of INR 42,775 (95% interval 18,101 to 202,353), closely matching the deterministic base case. Universal screening detected more cases than targeted screening in 98.4% of iterations. The proportion of cases never confirmed under universal screening had a median of 38.3% (95% interval 16.9% to 64.1%). Because no willingness-to-pay threshold exists in India for a case of PCHL detected, the acceptability curve is presented as a decision aid rather than as a verdict: the probability that universal screening is cost-effective reaches 0.49 at INR 43,000 per case, 0.81 at INR 75,000, and 0.89 at INR 100,000.

4. Discussion

Universal newborn hearing screening detects roughly 1.9 times as many cases of permanent congenital hearing loss as screening confined to a high-risk register, at an incremental cost of about INR 43,000 per additional case detected. That comparison, rather than the choice of screening device [8], is the decision most Indian districts actually face, and it has not previously been modelled [11].

The more useful finding may be the one that cuts across all three strategies. Even under universal screening, more than a third of cases are never confirmed, and the loss comes almost entirely from incomplete coverage and from families not returning between referral and diagnostic testing. Attrition of this kind is the dominant operational failure of newborn hearing screening in resource-limited settings [19], and comparable losses have been documented in programmes in Thailand [20] and Nicaragua [21]. Loss to follow-up alone moved the ICER from INR 37,634 to INR 68,292 across its plausible range. A programme that improved follow-up completion without changing its screening strategy at all would therefore recover a substantial share of the cases that universal screening is being asked to find. For

programme managers weighing where to place the next rupee, that is a competing claim on resources that a strategy comparison alone would not surface.

The central caveat is that the answer depends on a quantity India has never measured. The proportion of affected infants who carry no recognised risk indicator determines almost the entire result, and the two Indian cohorts that bear on it point in opposite directions from a very small number of cases [11, 16]. Indian data on congenital cytomegalovirus infection, a leading non-genetic cause of PCHL that frequently occurs without conventional risk indicators, further suggest that risk registers are unlikely to capture cases as completely as the higher end of the range implies [22]. Under the international estimate of 50% [14], universal screening is clearly worth its cost; if Indian risk registers were to capture 90% of cases, the incremental cost per case would exceed INR 500,000 and the argument would reverse. Establishing this parameter from Indian data — which requires only that existing universal screening programmes report detected cases stratified by risk status — would be an inexpensive and high-value addition to the evidence base, and would do more to resolve the policy question than any refinement of the model presented here.

4.1 Limitations

Several limitations bound what this analysis can support. It reports cost per case detected, not per quality-adjusted life year; no cost-effectiveness threshold exists for that outcome in India, so the analysis cannot pronounce a strategy cost-effective in the sense a formal HTA would require [12, 17]. Detection is a surrogate: the health gain from early detection accrues through language and educational outcomes that a short-horizon tree cannot capture [4, 5], and extension to a lifetime model of amplification and cochlear implantation, of the kind recently applied to hearing aid provision in India [23], is the necessary next step.

Parameters come from published secondary sources rather than primary costing, and several rest on single studies. Coverage and loss-to-follow-up values derive from one tertiary centre in Delhi [11] and may not describe district hospitals or community settings. Unit costs come from a single Indian analysis [8] and were not micro-costed across facility levels. Four parameters, including two governing OAE performance, are assumptions rather than published values; they are flagged as such throughout and varied widely, but they remain assumptions.

The published Indian device analysis reports OAE specificity of 0.68 [8], which implies that roughly a third of normally hearing neonates would be referred — far outside the American Academy of Pediatrics quality standard of under 4% referral [24]. This analysis used a more conservative 0.92 as the base case with 0.68 retained as the lower bound; readers should note that the published Indian estimate, if taken at face value, describes a programme that would not meet international quality benchmarks.

Finally, the model assumes screening coverage is non-differential with respect to risk status. In practice high-risk neonates, being more likely to have prolonged facility contact, are probably screened more

completely than well babies, which would flatter targeted screening relative to the estimates presented here.

5. Conclusion

Universal two-stage newborn hearing screening confirms substantially more cases of permanent congenital hearing loss than a high-risk-register strategy, at an incremental cost of approximately INR 43,000 per additional case detected under current published parameters. The strength of that conclusion is limited less by the model than by a single unmeasured Indian quantity — the share of affected infants without risk indicators — and by the finding that incomplete coverage and loss to follow-up remove more than a third of cases regardless of which strategy is chosen. Measuring the first and addressing the second are the two priorities this analysis identifies for the national programme.

Declarations

Ethics approval: Not required. The study used published aggregate data only, with no human participants and no identifiable information.

Data availability: All parameters and their sources are given in Table 1. The model code and output files are provided as supplementary material.

Competing interests: The authors declare that they have no competing financial interests

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Figures

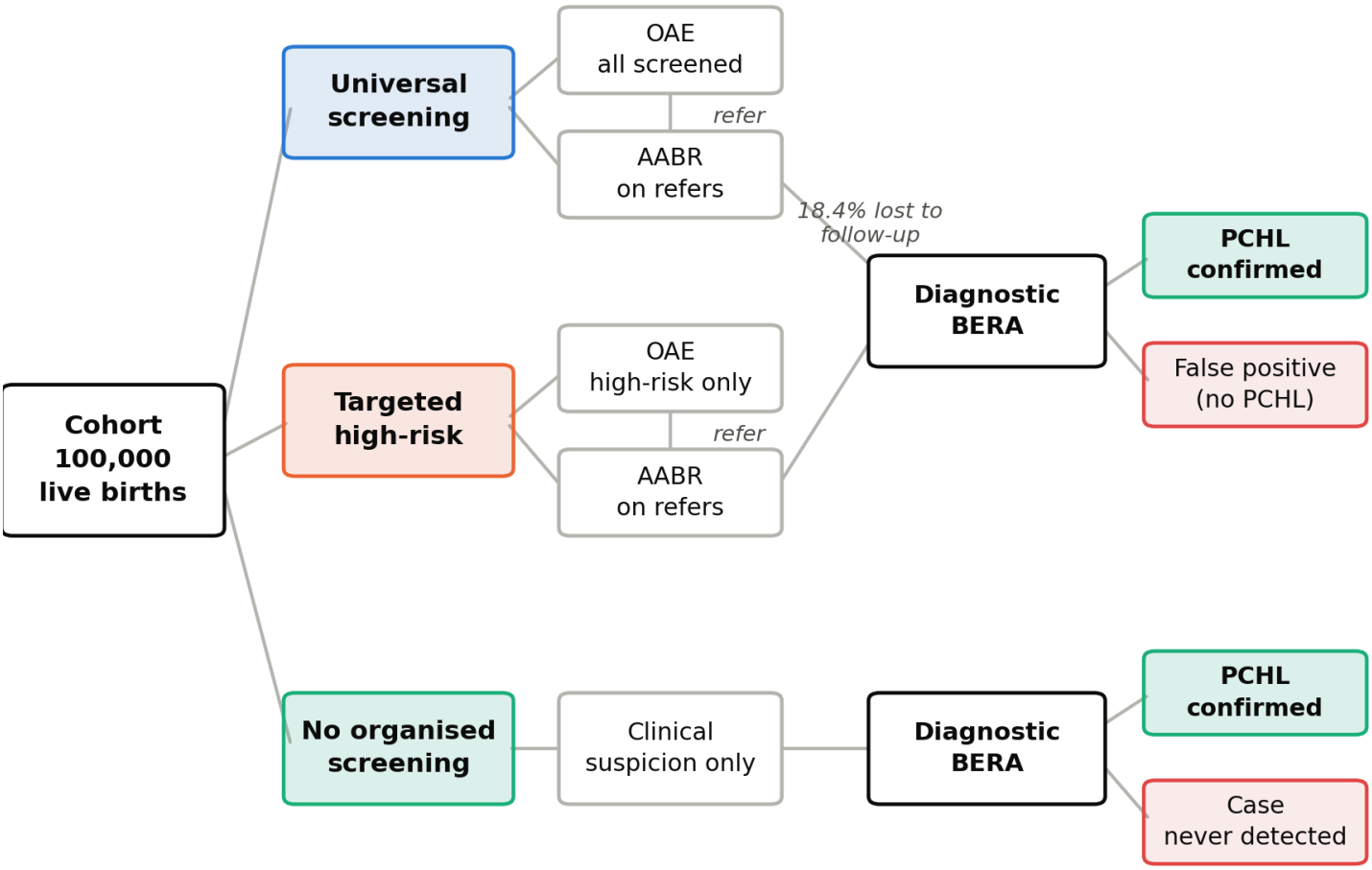


Figure 1

Decision-tree structure. Three mutually exclusive screening strategies applied to a hypothetical cohort of 100,000 live births. All branches converge on confirmatory BERA; terminal states are confirmed PCHL, false positive, and undetected case.

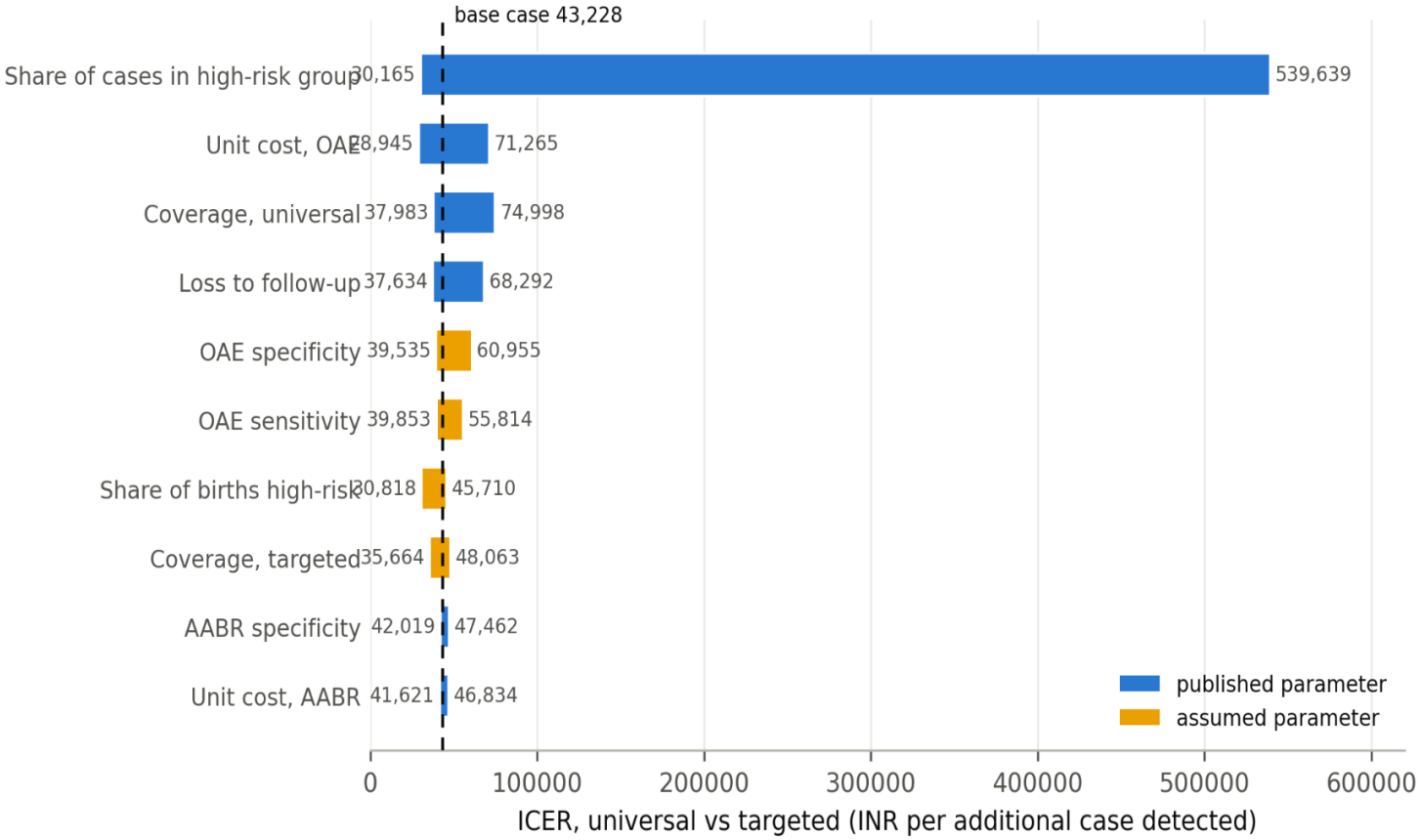


Figure 2

One-way sensitivity analysis of the ICER for universal versus targeted screening. Bars span the ICER across each parameter's plausible range; the dashed line marks the base case. Colour distinguishes published from assumed parameters.

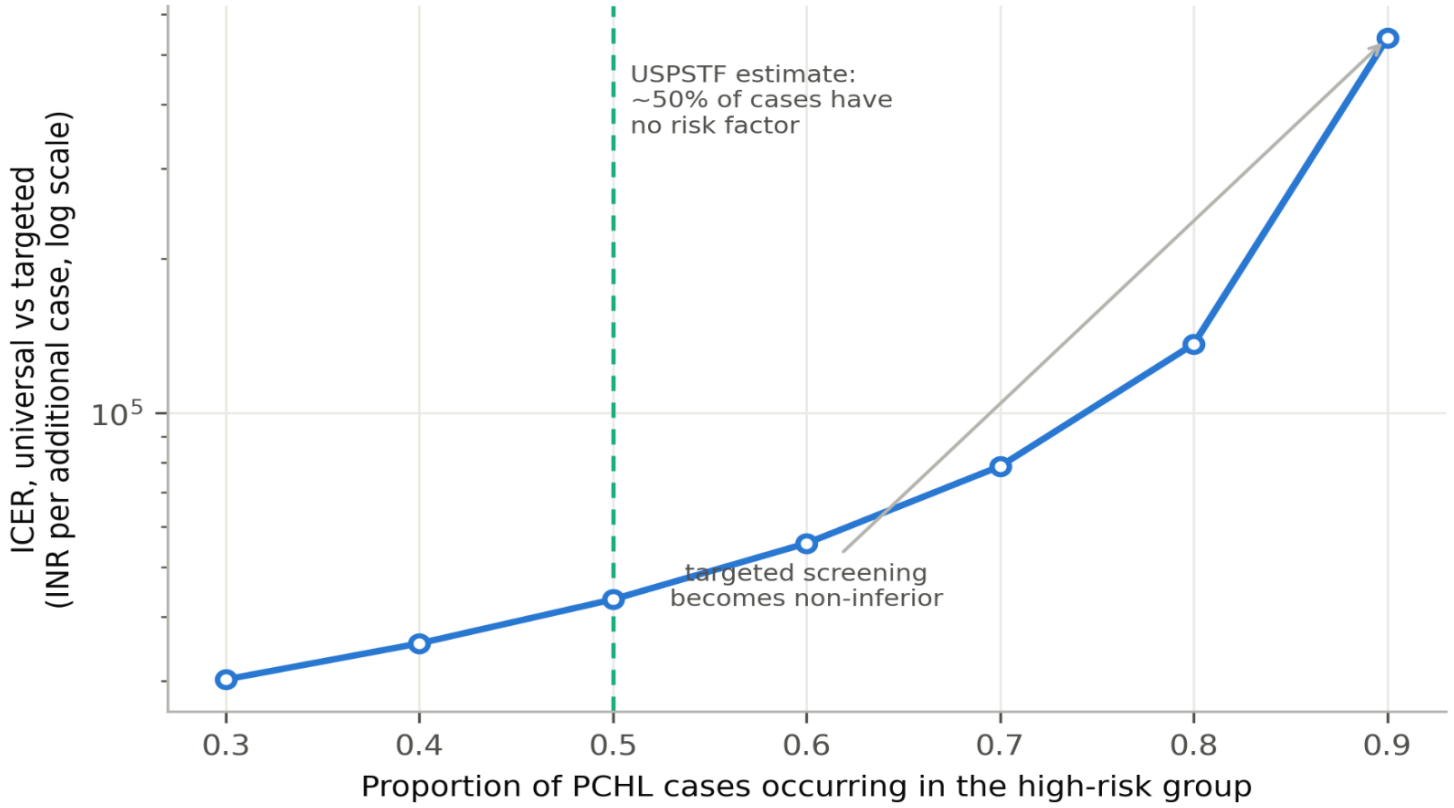


Figure 3

Threshold analysis on the proportion of PCHL cases occurring in the high-risk group — the parameter that dominates the result. The dashed line marks the USPSTF estimate of approximately 50%. Beyond roughly 95%, universal screening detects no additional cases.

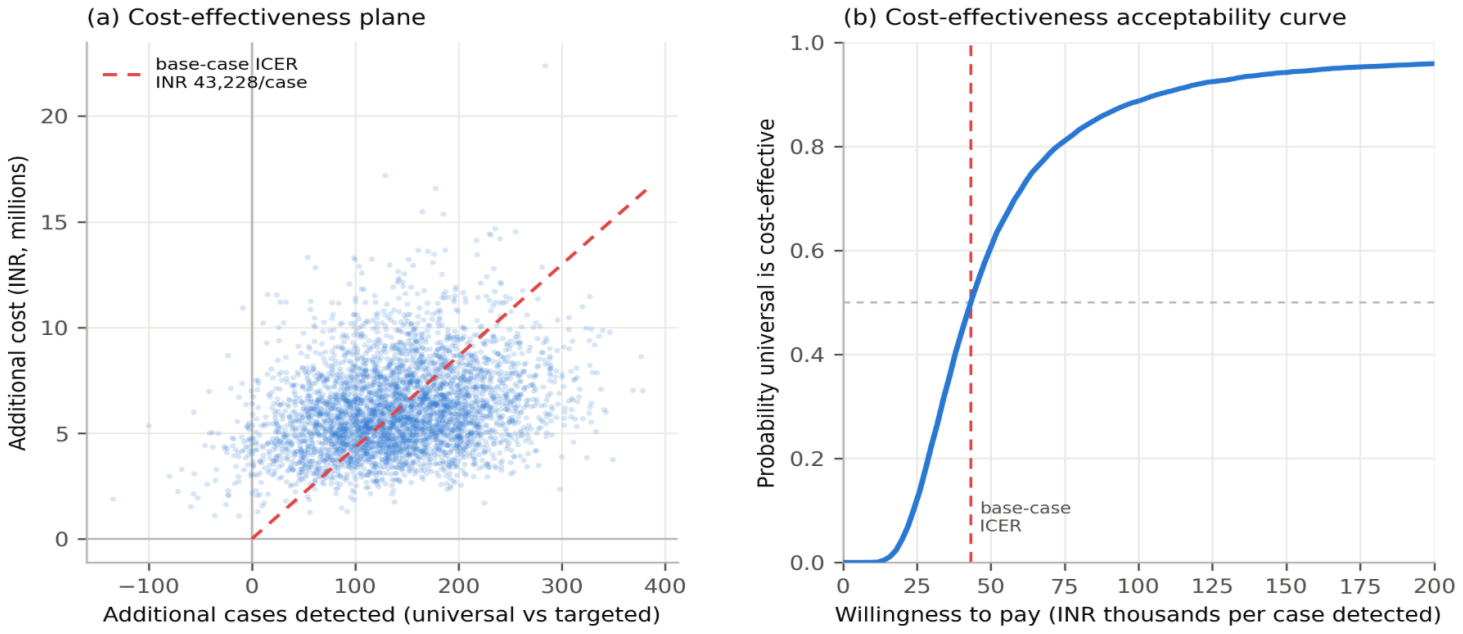


Figure 4

Probabilistic sensitivity analysis, 10,000 iterations. (a) Cost-effectiveness plane, universal versus targeted screening; the dashed line is the base-case ICER. (b) Cost-effectiveness acceptability curve. No Indian willingness-to-pay threshold exists per case detected, so (b) is presented as a decision aid rather than a verdict.