Cost-effectiveness analysis of screening for congenital dislocation of the hip to help prioritise and plan a clinical trial

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Paper presented to the Health Economists' Study Group, 14-16 July 1999, Aberdeen.
Working document. Please do not quote.

INTRODUCTION

The **objectives** of this work are to:

- determine whether there is sufficient evidence to support or exclude particular screening options for congenital dislocation of the hip (CDH) within the UK setting without further primary research;
- identify key areas of uncertainty that it might be important to address in further primary research, particularly within the context of a randomised trial;
- explore the relation between the costs of a potential trial and the anticipated cost savings and other benefits that would be predicted to follow from policies which might result from such research.

Background

Current UK Policy

A national policy of universal neonatal CDH screening was formally introduced in the UK in 1969. Current recommendations, revised in 1986, advise clinical screening by the Ortolani-Barlow (O-B) test to detect hip instability on three occasions: within 24 hours of birth, at discharge from hospital of birth, and at 6 weeks of life. Infants in whom hip instability is subsequently confirmed are usually treated non-surgically with a splint appliance. Splinting is usually performed up to the age of 3 months. Thereafter, any primary treatment tends to be surgical. It is further recommended that infants with recognised risk factors for CDH, such as a positive family history or breech presentation, should be identified and followed until walking normally. Specific guidance on the use of ultrasound in the screening programme was not given.

Problems in evaluating effectiveness

Evaluation of the current policy is complicated by problems in case definition, uncertainty regarding the natural history of established dislocation, lack of a 'gold standard' with which to judge the performance of screening tests, uncertainty about the effectiveness of non-surgical treatment with a splint appliance, as well as a lack of agreed measures of process and outcome, and information systems to monitor them, within the current UK programme. The O-B test is associated with an appreciable false negative rate, which may reflect a test that performs poorly, lack of expertise on the part of the primary screener, or variability in the natural history of CDH in that not all cases are detectable before 6 weeks of age. The birth prevalence of CDH in *unscreened* populations is estimated to be 0.8 - 1.6 per 1000 Northern Europeans,

suggesting that 500 to 1000 children who will develop CDH are born each year in the UK. The positive predictive value of the O-B test is probably low, given that the prevalence of neonatal splinting in *screened* populations is from 4 to 25 times more common than that of established dislocation in *unscreened* populations. Non-surgical treatment by splint is not always successful, but evidence from randomised trials is lacking on effectiveness. Up to 5% of infants detected by screening and treated with a splint appliance will require subsequent surgery. In addition, treatment by splinting or surgery can cause iatrogenic avascular necrosis (AVN). AVN occurs when the blood supply to the head of the femur is disrupted. In severe cases it leads to loss of congruity between articulating surfaces in the hip joint and may result in hip pain and dysfunction in early adult life. Treatment of this complication is not always satisfactory.

Alternative strategies

The perceived failures of the current screening policy, together with implementation of universal primary ultrasound screening in some European countries, have generated interest in ultrasound imaging of the neonatal hip, used in addition to clinical examination, as a primary screening test in the UK. Ultrasound may be used for primary screening either on a selective basis for infants with recognised risk factors, or as a universal test. Non-randomised studies of selective use of ultrasound in infants at high risk of CDH, in the UK, suggest that this may not be an effective means of reducing the false negative rate associated with clinical screening. It is equally uncertain whether universal ultrasound screening reduces the prevalence of false negatives, as small numbers and a short period of follow up limit interpretation of published studies. Furthermore, as universal ultrasound screening is associated with higher rates of splinting and follow up than currently reported for clinical screening there may be considerable disbenefits to such a policy.

METHODS

Overview

A cost-effectiveness analysis was conducted using decision trees in Excel 97, and confirmed in DATA 3.5 by TreeAge, to represent the different screening strategies for CDH. The target population was newborn and young infants. Data on the pathway probabilities were obtained from a systematic review of published and unpublished data

sources, supplemented where required by information from collaborating experts. The strategies compared were:

- 'no screening'
- current policy of universal clinical screening (by the O-B test) and risk factor assessment
- selective use of ultrasound screening in infants with clinical hip instability and/or established risk factors (taken in the model as those with breech presentation and/or a positive family history of CDH). Thus, relative to current practice the selective strategy allows identification of 'high risk' infants in whom clinical examination has been deemed normal but ultrasound examination is abnormal.
- universal ultrasound screening

Characterisation of screening models

A multidisciplinary subgroup developed the decision trees (See Figures 1-4). The screening models have been characterised by considering the variety of policies and practices identified from the literature as well as from a recent MRC survey. Factors considered in the choice of models included practical feasibility within the NHS as well as acceptability to informed health care professionals. This information was supplemented by discussions with collaborating experts, a national survey of paediatric radiologists and visits by the study team to three UK centres, two currently or previously engaged in universal ultrasound screening (Coventry, Hinchingbrooke) and one using ultrasound selectively (Oxford). This information has been used to refine aspects of the screening scenarios defined in the models, particularly those relating to ultrasound screening.

Outcomes

The analytic horizon was taken to be until the end of adolescent growth to reflect the objectives of screening which are to ensure 'a functionally and developmentally normal hip joint at the end of the period of growth in childhood and adolescence'. As functional assessment of hip function throughout childhood is not thought to be a reliable predictor of developmental normality, emphasis was given to radiological assessment of the hips at skeletal maturity. A radiological Severin score of 1 to 3 at skeletal maturity was taken as a 'favourable' treatment outcome and one of 4 or worse to be an 'unfavourable' treatment outcome (see table 1).

Screening can lead to over treatment, thus the model was set up to identify those truly affected with CDH and those not. Favourable and unfavourable treatment outcomes could then be classified according to whether the child was truly affected with CDH. All children without CDH who were not treated were assumed to have a favourable outcome.

The model was also able to identify true negative, true positive, false negative and false positive screening outcomes, the non-surgical treatment rate, the Odds of CDH given a positive screen and the likelihood ratio.

The model was also run to identify the number of iatrogenic AVN cases expected with each strategy according to screening outcome. Of particular interest was AVN arising in false positives.

Costs

Unit costs were estimated for a clinical examination and risk factor assessment, the ultrasound screen for selective and universal use, follow-up ultrasound, non-surgical treatment by splinting and surgical treatment.

Clinical examination and risk factor assessment

A number of experts were consulted to determine the length of time taken to perform the O-B test and risk factor assessment. It was assumed to take two minutes of a senior house officer's time plus a midwife and administrative support for each test. The midpoints of the 1998 salary scales were used. Overheads were added at the rate of 40%.

The time taken to conduct the hip check at 6-week developmental check was costed for a GP performing the test and added to the cost of the clinical examination and risk factor assessment.

Under current practice, infants with a positive screening examination or risk factor assessment are brought back for re-examination at an outpatient visit. The cost of re-examination was estimated at the same cost as the first examination plus the cost of an outpatient visit.

Ultrasound screen

A questionnaire was sent to 77 members of the Paediatric Radiology Imaging Group (BPRI) who were identified as being responsible for ultrasound imaging of hips in their centre. The response rate was 75%. Information was sought on the personnel mainly responsible for performing and reporting hip ultrasounds and additional staff present throughout the examination plus the average time taken to perform and report ultrasound images. The questionnaire also asked for information on the type of ultrasound machine used and the proportion of time each machine is dedicated to the imaging of hips in the context of screening or managing suspected, or confirmed, cases of CDH. Those using ultrasound for screening were also asked the purchase and maintenance costs of their ultrasound machines.

Since practices were found to vary considerably in terms of the quantity of staff and machine time dedicated to CDH screening, scenarios of typical screening programme configurations were constructed on the basis of the data and costed. These scenarios were consistent with reported practices at the three centres visited where universal or selective ultrasound imaging is currently or has been performed. Capital costs were allocated per infant screened by assuming a provider unit screens 3500 children per annum. Overheads were added at a rate of 40% of the capital and labour costs.

Follow-up ultrasounds

Infants who have a positive screening result are assumed to have 1.5 additional ultrasounds before it is clear that they do not need treatment. The number of additional ultrasounds was based on data from the current MRC 'Hip' trial which is trial investigating the use of ultrasound in the management of CDH. The cost of a follow-up ultrasound was based on a scenario from the BPRI survey and added to the cost of an outpatient visit.

Non-surgical treatment

The cost of non-surgical treatment was based on all infants being splinted in a Pavlik harness with the unit cost per Pavlik harness. Manufacturers' prices were used to cost the Pavlik harness. It was assumed that each child splinted is followed-up to check the development of the hip. It was assumed that the response to treatment would be assessed by ultrasound on an average of four occasions as an outpatient. The number of ultrasound follow-up visits was based on data from the MRC Hip trial. Thus the total

cost of early treatment was estimated as the cost of splinting plus 4 follow-up ultrasounds at an outpatient visit.

Surgical treatment

On the basis of the literature review, it was assumed that children with CDH in whom splinting is unsuccessful will require one surgical procedure (closed reduction). Children with CDH who are not screened, miss screening, are false negatives or are screen positive but treated after 3 months were assumed to receive surgery in the following combinations: 56% have closed reduction alone, 14% have closed followed by open reduction and 30% have open reduction alone.

The cost of surgical treatment was based on the mean cost of open procedures for CDH (HRG h29 1998) provided by the national case-mix office. Data on closed procedures were not available. The cost of a closed reduction was estimated by assuming the same ratio of the cost of closed to open reduction reported in a Canadian study (Tredwell 1990).

To this cost was added an annual follow-up cost for a further 15 years, based on four outpatient visits a year for the first five years and then one outpatient visit a year for the remaining years. These costs were discounted at 6%.

The pathway probabilities

Data on pathway probabilities were obtained from review of published and unpublished sources. A computerised search of Medline (1966 to January 1998) and Embase (1974-January 98) was carried out. Papers of potential value to the cost-effectiveness analysis were identified using search strategies developed with input from an information specialist at the Cochrane Collaboration. Additional references were identified from reference lists of published articles and from literature identified by the review conducted by the American Academy of Pediatrics Sub-Committee on Early Detection of CDH as well as from a subsequent review conducted for the Canadian Task Force on Preventive Services. Unpublished data were obtained from Dr Edmund Hey (Northern Region), the Aberdeen Maternity and Neonatal data bank (Dr Marion Hall), Dr Becker (Hinchingbrooke), and Dr Lutz Altenhofen (Germany).

The mean value weighted by study size was taken as the baseline probability for parameters within the model. The exception was for the probability value associated with branch H ie. the probability of having CDH given a negative screening result, for which the median value was used. Heterogeneity of estimates for branch H suggested that a weighted mean would be an overestimate for current practice, due to larger studies coming a single centre with a high false negative rate, and for the universal use of ultrasound it would be an underestimate, due to a large study from a single centre with no false negatives.

In addition, it was not possible to get a direct estimate of the probabilities for branches K (ie. the probability of having CDH given splinting by 3 months following a positive screening result) and L (ie. the probability of not having CDH given splinting by 3 months following a positive screening result) as

- there is no diagnostic test and treatment is given to prevent development of definitive disease ie. treatment is given on the basis of risk of developing a condition (similar to risk factors for cardiovascular disease) rather than following confirmation of the presence of a condition (such as Down's or cystic fibrosis)

- risk estimates ie. the probability of developing CDH, are uncertain as all children with a given clinical finding or ultrasound appearance have been treated and there are no useful natural history studies

L, and therefore K, were estimated by assuming a constant prevalence of CDH across all strategies, given the values for H,C,J,E,D and F.

The base case probabilities are given in the appendix along with other assumptions used in the model.

Incremental cost-effectiveness ratios

Incremental cost-effectiveness ratios were estimated for the alternative screening strategies and presented as the additional cost per favourable treatment outcome in children with CDH.

Sensitivity analyses

A series of sensitivity analyses were conducted to examine the effect on the incremental cost-effectiveness ratios of varying assumptions. These were as follows:

i: Reduce the false negative rate (FNR) in current practice to the lowest reported in the literature ie. 0.34 per 1000 live births instead of 0.78 per 1000 live births.

ii. Increase the FNR in selective use of ultrasound to 0.81 per 1000 live births instead of 0.49 per 1000.

iii. Assume a splinting treatment rate in universal ultrasound screening of 44 per 1000 live births, as reported in Austria and Norway, instead of 5.18 per 1000 in the base case.

iv. Increased surgical treatment costs as a proxy for more complex surgery.

v. Halve the probability of an unfavourable treatment outcome.

vi. Double the probability of an unfavourable treatment outcome.

RESULTS

The costs used in the base case are shown in table 2. Table 3 ranks the screening strategies in order of effectiveness and gives the incremental cost-effectiveness ratios. Universal ultrasound is the most effective screening programme. No screening is least effective, followed by current practice and selective use of ultrasound. The total cost of the strategies increases with effectiveness, however, current practice is extendedly dominated by selective use of ultrasound ie. selective use of ultrasound achieves more favourable treatment outcomes at a lower additional cost than current practice. The cost-effectiveness ratios are depicted graphically in figure 5.

Without any screening programme, about 90 of the 120 affected children per 100,000 live births can be expected to have a favourable treatment outcome at skeletal maturity. Conversely, 30 will have an unfavourable treatment outcome. With current practice, about 94 of the 120 affected children can be expected to have a favourable treatment outcome, similarly with selective use of ultrasound 106 children can be expected to have a favourable treatment outcome and with universal ultrasound screening 110 children can be expected to have a favourable treatment outcome.

Table 4 shows the case of AVN that can be expected with each of the strategies. The other rates predicted from the analysis are shown in table 5.

Sensitivity analysis

The FNR in current practice was reduced from 0.78 per 1000 live births to 0.34 per 1000 live births, the lowest reported in the literature. The FNR =HxCxA. H (and therefore G) was re-estimated given CxA. In order to maintain a constant prevalence of 1.2 children affected by CDH per 1000 live births, L (and therefore K) was also re-estimated, given H,C,J,E,D and F. Current practice is then no longer extendedly dominated by the selective use of ultrasound (see Figure 6). The incremental cost-effectiveness ratio for current practice, compared to no screening, is £27,989 per additional favourable treatment outcome ie. lower than the base case. For selective use of ultrasound, compared to current practice, the incremental cost effectiveness ratio is £338,255 ie. higher than the base case. The incremental cost-effectiveness ratio for universal screening compared to current practice is £343,649 per additional favourable treatment outcome, which is less than the baseline value. The threshold value for the FNR in current practice at which current practice is no longer extendedly dominated by the selective use of ultrasound is 0.64 per 1000 live births.

If a FNR of 0.34 per 1000 live births in current practice could be obtained by improved training of primary screeners, the equivalent of £5.28 per child screened could be spent on training before current practice became extendedly dominated by the selective use of ultrasound. The incremental cost-effectiveness ratio for current practice compared to no screening would then be £64,595 per additional favourable treatment outcome ie. the same as selective use of ultrasound.

Assuming the base case FNR in current practice remains at 0.78 per 1000 live births, the FNR in the selective use of ultrasound needs to be increased to 0.81 per 1000 (from the baseline of 0.49 per 1000) for current practice to be no longer extendedly dominated by the selective use of ultrasound. The incremental cost-effectiveness ratio for selective use of ultrasound is now much higher than the base case at £137,402 per additional favourable treatment outcome and the ratio for universal use of ultrasound much lower than the base case at £138,343 per additional favourable treatment outcome.

To model the Austrian/Norwegian splint treatment rate for universal ultrasound screening meant re-estimating E and F and then re-estimating L, assuming a constant prevalence 1.2 children affected by CDH per 1000 live births. As expected universal

ultrasound screening is even less attractive. Moreover, it becomes dominated by current practice and selective use of ultrasound ie. it is less effective and more costly. Data are not shown.

The surgical costs need to be increased 5.818 times for no screening to be extendedly dominated by selective use of ultrasound. The incremental cost-effectiveness ratio for selective use of ultrasound compared to no screening becomes £19,296 per additional favourable treatment outcome and £381,226 per additional favourable treatment outcome for universal ultrasound screening. Both ratios are lower than the base case. Current practice is still extendedly dominated by the selective use of ultrasound (see Figure 7).

If the probability of an unfavourable treatment outcome is halved selective use of ultrasound no longer extendedly dominates current practice. The incremental cost effectiveness ratio for current practice compared to no screening becomes £11,984 per additional favourable treatment outcome ie. lower than the base case. The incremental cost effectiveness ratio for selective use of ultrasound compared to current practice becomes £81,174 per additional favourable outcome and for universal use of ultrasound compared to selective use it become £1,232,240 per additional favourable treatment outcome. Both of the latter ratios are higher than the base case.

If the probability of an unfavourable treatment outcome is doubled, current practice remains extendedly dominated by selective use of ultrasound. Re-estimation of the incremental cost-effectiveness ratios excluding current practice gives an incremental cost-effectiveness ratio of £32,411 for selective use of ultrasound compared to no screening and an incremental cost-effectiveness ratio of £66,907 for universal use of ultrasound compared to selective use. Both ratios are lower than the base case.

CONCLUSIONS

Despite very favourable assumptions regarding coverage and treatment rates for universal ultrasound screening and the fact that it is the most effective screening strategy, its high cost-effectiveness ratio is likely to suggest that it is not worthwhile implementing. Based on existing data, the cost-effectiveness analysis suggests that selective use of ultrasound may be best strategy but this is sensitive to assumptions

about the false negative rate achieved with current practice and the probability of an unfavourable treatment outcome.

It may be possible to reduce the current practice false negative rate by improving training and consistency of primary screeners and this is also relevant to the selective screening strategy.

The management of infants with breech or positive family history but without neonatal clinical hip instability will need addressing in either strategy.

If ultrasound is used selectively in these infants then treatment effectiveness in those who are clinically normal but have abnormal ultrasound appearances should be clarified.

In addition, the literature review has highlighted significant gaps in knowledge relating to:

1. long term outcome of splinting or surgery
2. effectiveness and safety of splinting and surgical treatment practices
3. parental views on splinting and compliance with treatment
4. association of acetabular dysplasia and hip pain and dysfunction in early adult life

POINTS FOR DISCUSSION

- Improving the sensitivity analysis
- Ways of addressing the third objective ie. to explore the relation between the costs of a potential trial and the anticipated cost savings and other benefits that would be predicted to follow from policies which might result from such research.

ACKNOWLEDGEMENTS

The work was funded by the Medical Research Council and has benefited from the comments made by members of the MRC Working Party on CDH. In particular the help provided by Alastair Gray, Rosemary Arthur, Nick Clarke, Edmund Hey and Diana Elbourne is acknowledged.

Key to figures non-surgical treatment = splinting; FO = favourable outcome; FTO = favourable treatment outcome; UFTO = unfavourable treatment outcome.

Figure 1: Decision tree for no screening

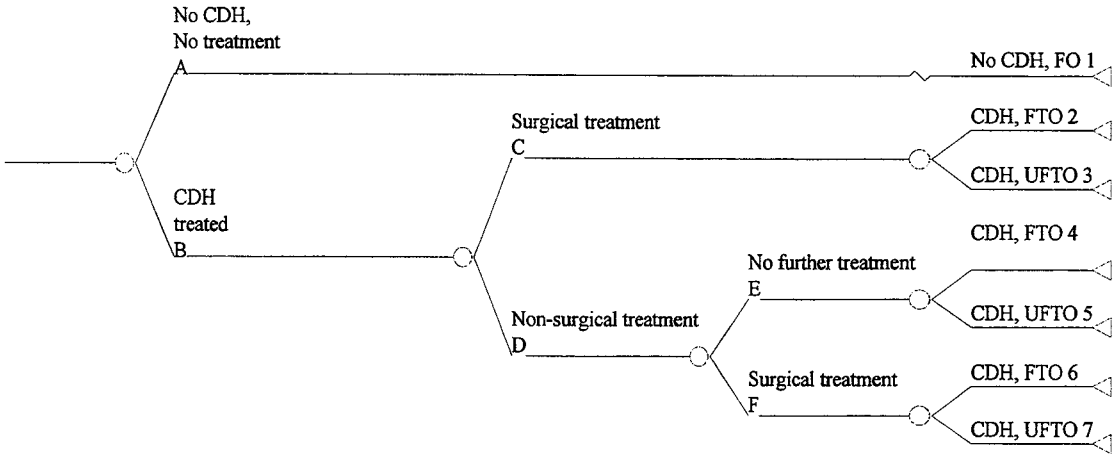


Figure 2: Decision tree for current practice

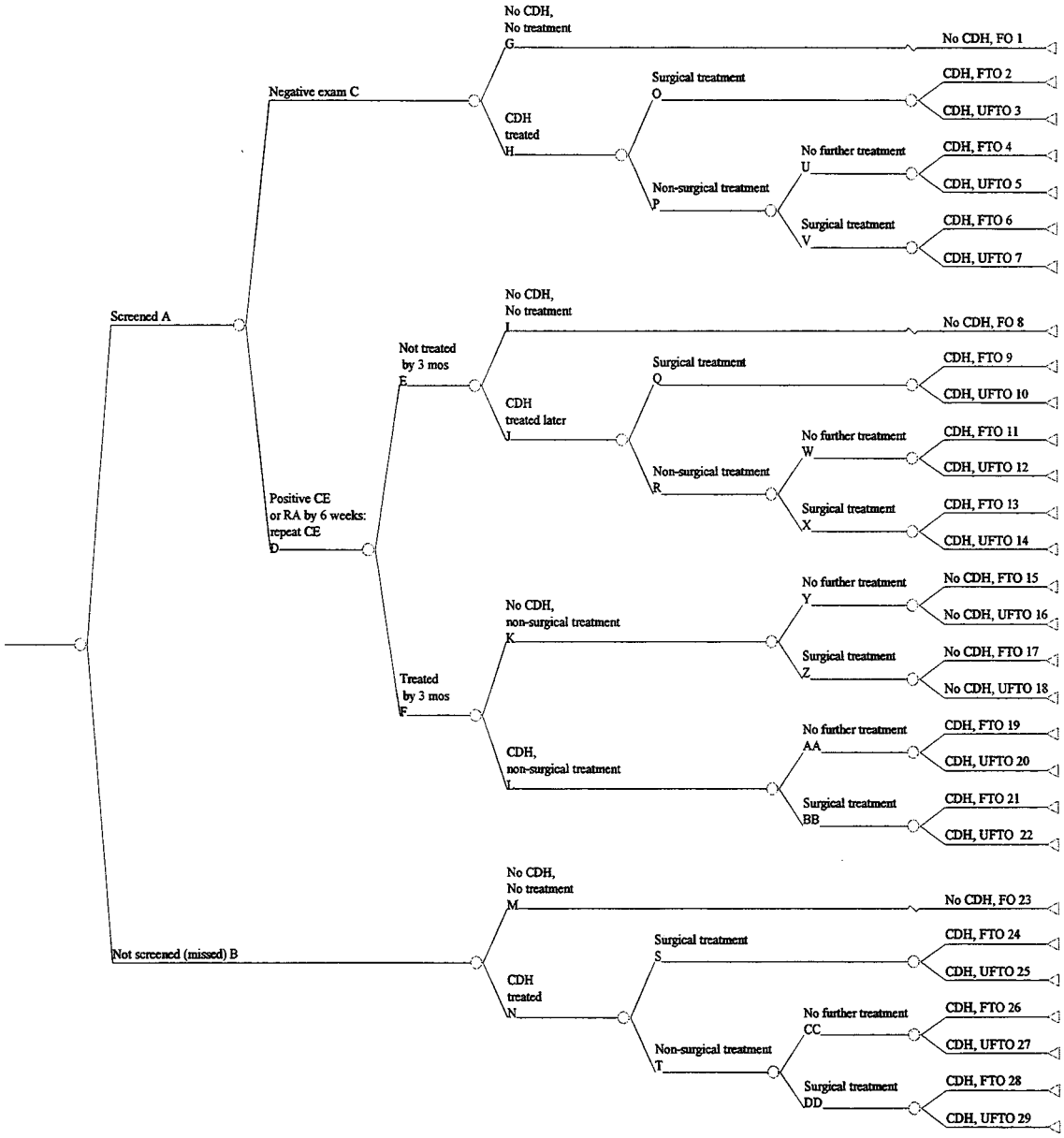


Figure 3: Decision tree for selective use of ultrasound

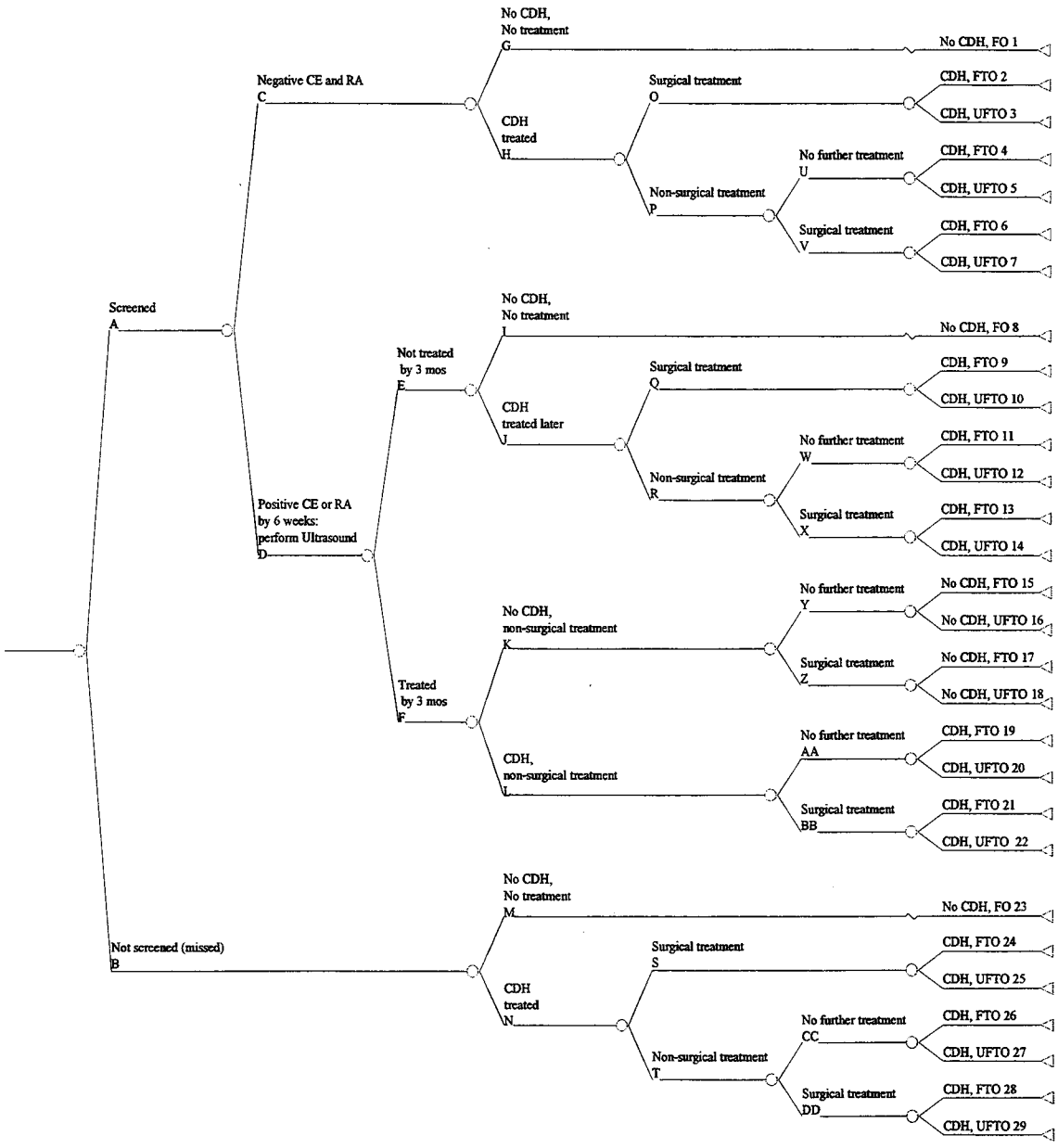


Figure 4: Decision tree for universal ultrasound screening

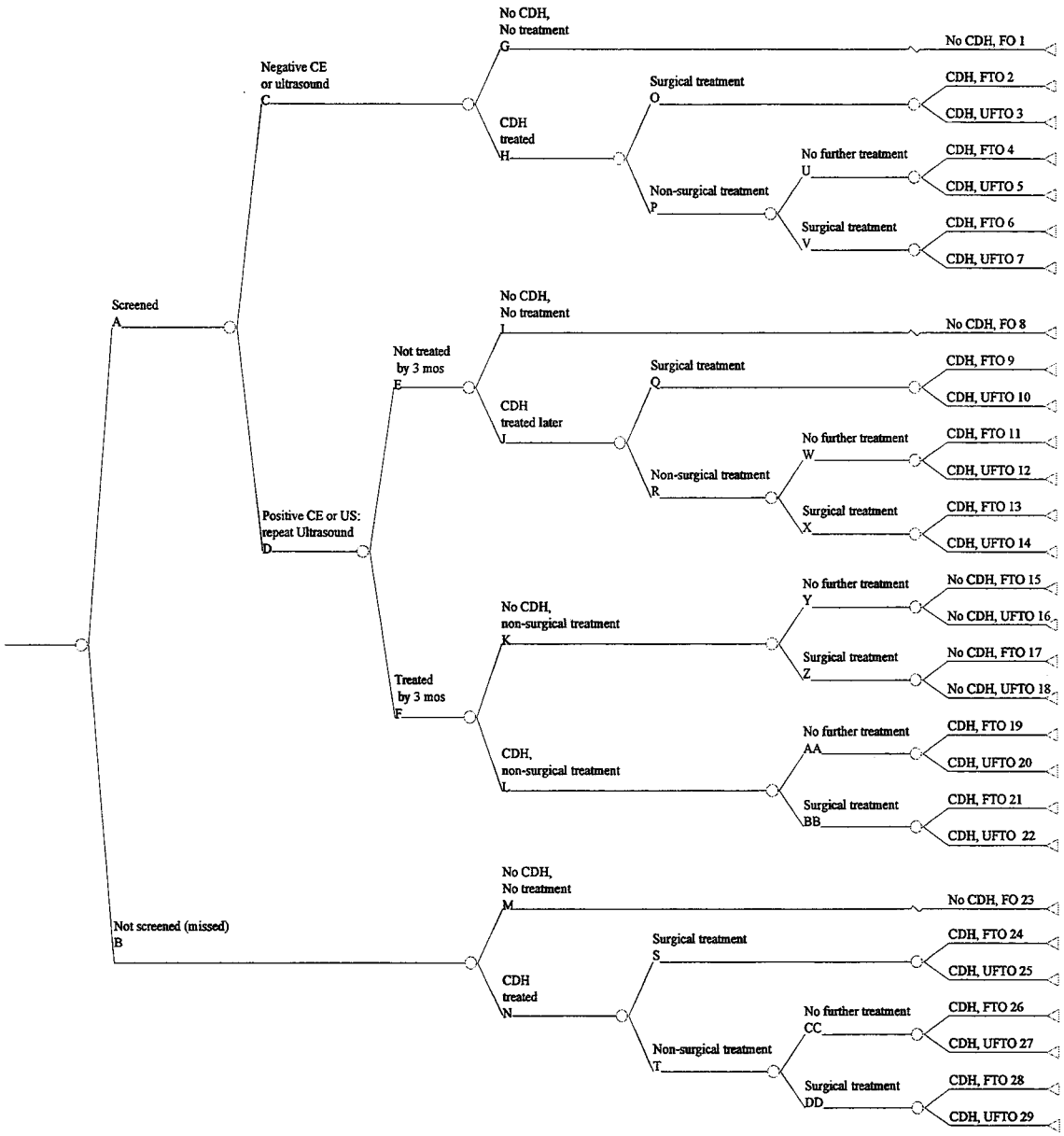


Fig 5

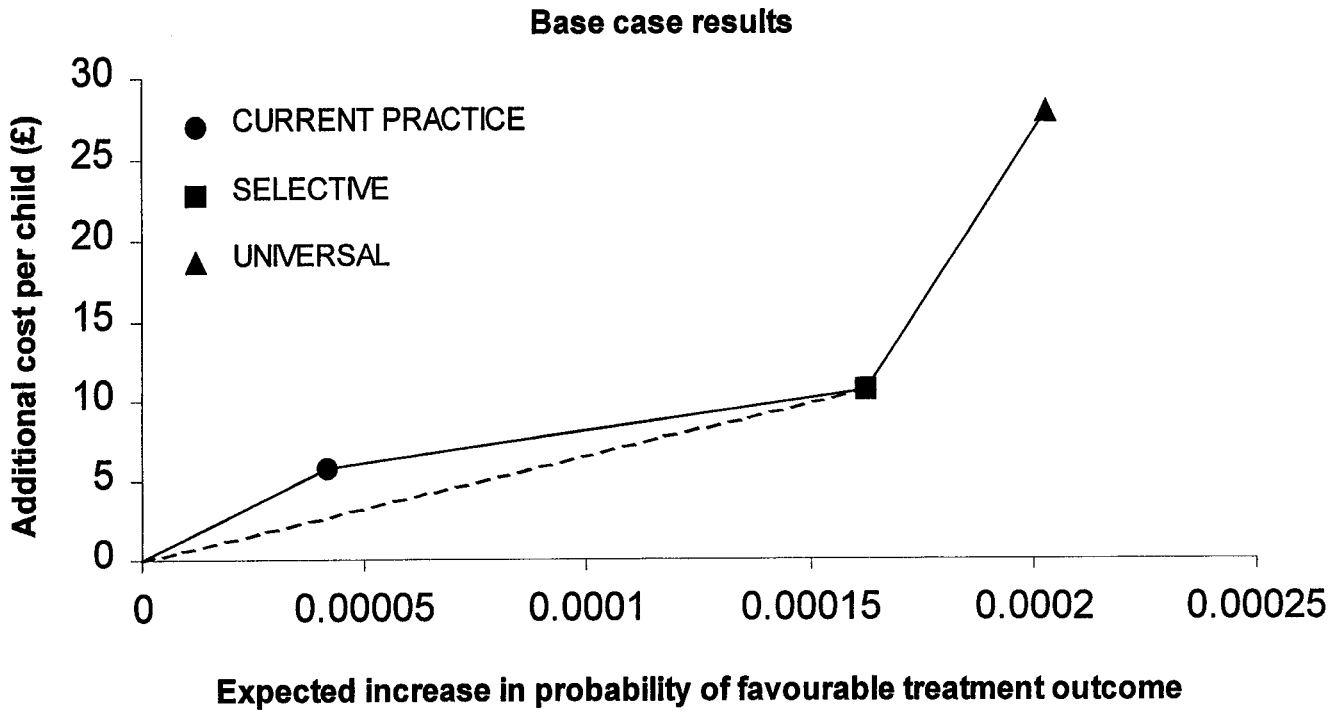
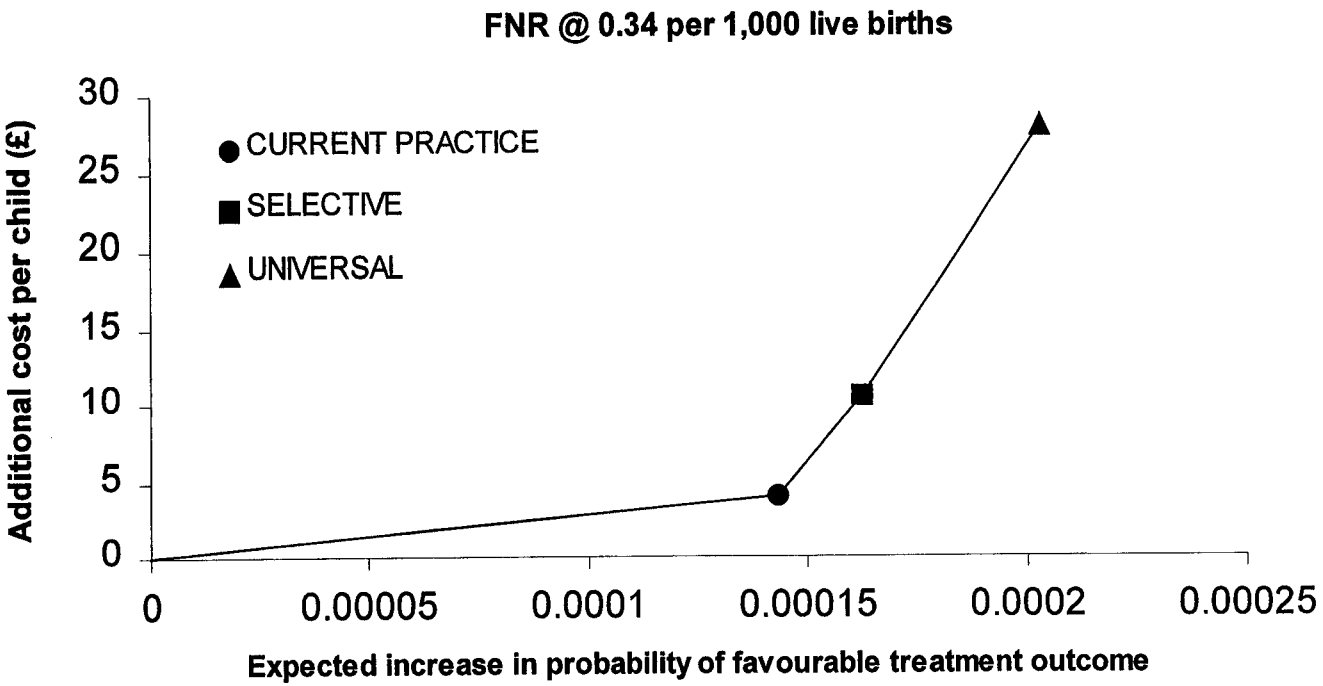

Base case results
Additional cost per child (£)
30
25
20
15
10
5
0
CURRENT PRACTICE
SELECTIVE
UNIVERSAL
0
0.00005
0.0001
0.00015
0.0002
0.00025
Expected increase in probability of favourable treatment outcome

Fig 6


FNR @ 0.34 per 1,000 live births
Additional cost per child (£)
30
25
20
15
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5
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CURRENT PRACTICE
SELECTIVE
UNIVERSAL
0
0.00005
0.0001
0.00015
0.0002
0.00025
Expected increase in probability of favourable treatment outcome

Fig 7

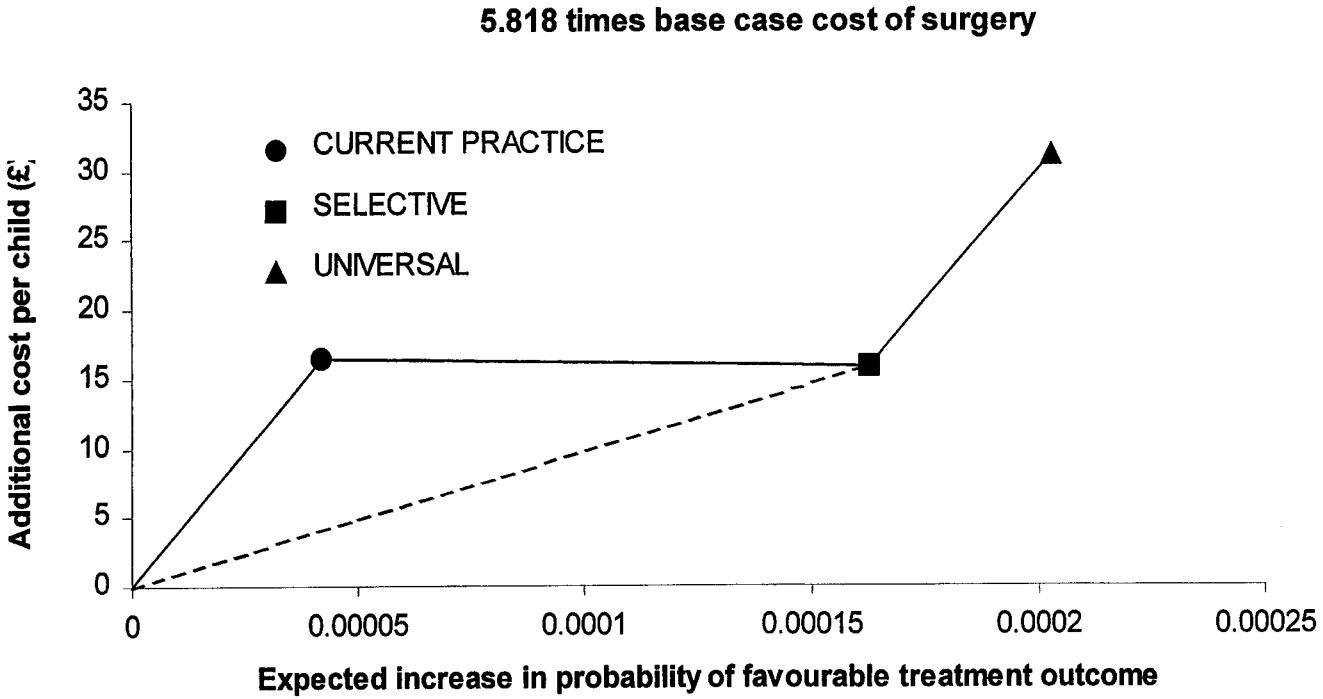
5.818 times base case cost of surgery
Additional cost per child (£)
35
30
25
20
15
10
5
0
CURRENT PRACTICE
SELECTIVE
UNIVERSAL
0
0.00005
0.0001
0.00015
0.0002
0.00025
Expected increase in probability of favourable treatment outcome

Table 1 Modified Severin Score

Severin Score	Appearance of hip	
1	Normal femoral head and neck	No subluxation
2	Deformed femoral head and/or neck	No subluxation
3	Dysplastic femoral head and/or acetabulum[1]	No subluxation
4	Subluxed femoral head[2]	
5	Femoral head articulates with false acetabulum	
6	Femoral head dislocated[3]	

Source: Gibson and Benson, 1982

[1] Shallow hip socket (acetabulum)
[2]Subluxation is a partial dislocation ie. the hip can move within the confines of the socket
[3]Dislocation implies the head of the hip is outside the socket

Table 2 Base case unit costs

	Unit cost (£'s 1998 prices)
Initial clinical exam	3.19
Re-examination clinically	56.49
Screen ultrasound:	
Universal	11.52
Selective	29.17
Follow-up ultrasound	77.31
Splint (Pavlik Harness)	37
Treatment*:	
Total OR costs	3684.15
Total CR costs	3340.53
Mixed CR/OR	3842.58

*OR = open reduction; CR = closed reduction

Table 3 Total costs, effects and incremental cost-effectiveness ratios per 100,000 live births

Strategy	Total cost (£'s) of programme per 100,000 live births	Total favourable treatment outcomes in CDH affected per 100,000 live births	Additional effects	Additional cost per additional effect
No screen-ing	461,000	89.90		
Current Practice	1,028,000	94.05	4.15	136,412
Selective ultrasound	1,515,000	106.15	12.10	40,251 (64,823*)
Universal ultrasound	3,252,000	110.16	4.01	433,399

Favourable treatment outcome = Severin 1, 2 or 3 at skeletal maturity
*Recalculated ICER dropping current practice, which was extendedly dominated

Table 4 Cases of AVN per 100,000 live births expected with each strategy

	No screening	Current practice	Selective ultrasound	Universal ultrasound
Total number treated: splint and surgery	120	523	759	547
Total AVN	18.27	19.56	14.25	9.44
AVN in false negatives	-	11.87	7.40	3.97
AVN in true positives - early	-	0.21	0.86	1.10
AVN in true positives- late	-	3.68	0	0
AVN in affected	*18.27*	*15.76*	*8.26*	*5.07*
AVN in false positives	-	3.78	5.98	4.01
AVN in missed screen	-	0	0	0.37

Table 5 Rates predicted from the decision trees (based on 100,000 live births)

	No screening	Current practice	Selective ultrasound	Universal ultrasound*
Positive screening test	-	2127	8125	7662
Non-surgical treatment rate	-	421	709	518
Surgical treatment rate	120	102	50	29
False negatives	-	78	49	26
True positives	-	18	71	93
False positives	-	403	637	427
Odds of CDH| Positive screen	-	1:118	1:114	1:84
Likelihood ratio**	-	7.1	7.31	9.92

*The false negatives and true positives do not sum to 120 because approximately 1 case of CDH is missed (i.e. not screened)

** The Likelihood Ratio (LR) is the likelihood that a given test result would be expected in a patient with the target disorder compared to the likelihood that that same result would be expected in a patient without the target disorder. It may be calculated by dividing the post-test probability of CDH by the pre-test probability of CDH. The LR is used to assess how good a diagnostic test is and to help in selecting an appropriate diagnostic test(s) or sequence of tests. It has advantages over sensitivity and specificity because it is less likely to change with the prevalence of the disorder and takes into account information from both sensitivity and specificity in the model.

Appendix: Base case probabilities

i. Pathways

No screening		Current practice		Universal ultrasound		Selective use of ultrasound	
Branch	**Probability**	**Branch**	**Probability**	**Branch**	**Probability**	**Branch**	**Probability**
Varying probabilities							
A	1-B	**A**	1	**A**	0.98	**A**	1
B	0.0012	**B**	0	**B**	0.02	**B**	0
C	1	**C**	0.978733	**C**	0.921821	**C**	0.918749
D	0	**D**	0.021267	**D**	0.078179	**D**	0.081521
E	1	**E**	0.802131	**E**	0.932432	**E**	0.912769
F	0	**F**	0.197869	**F**	0.067568	**F**	0.087231
		G	0.999203	**G**	0.999711	**G**	0.999471
		H	0.000797	**H**	0.000289	**H**	0.000529
		I	0.985824	**I**	1	**I**	1
		J	0.014176	**J**	0	**J**	0
		K	0.957422 093	**K**	0.824321	**K**	0.899257
		L	0.042577 907	**L**	0.175679	**L**	0.100743
		M	0.9988	**M**	0.9988	**M**	0.9988
		N	0.0012	**N**	0.0012	**N**	0.0012
Constant probabilities							
AA*	0.976247	**AA***	0.976247	**AA***	0.976247	**AA***	0.976247
BB	0.023753	**BB**	0.023753	**BB**	0.023753	**BB**	0.023753

***AA,BB:** probability of surgery following non-surgical treatment is assumed constant at all points in tree for each of the 3 screening strategies i.e. probabilities at U,W ,CC are equivalent to AA and probabilities at V,X,DD are equivalent to BB

Other assumptions:

- Prevalence of CDH in all trees is 0.0012 ie 1.2 per 1000 live births
- Coverage in current practice and selective use of ultrasound is 100%, but 98% for universal ultrasound
- Risk of CDH in those not screened (branch B) in a screening strategy is equivalent to that in an unscreened population ie 0.0012
- Non-surgical treatment rates for universal ultrasound have been based on the more conservative UK data
- All children detected by screening and treated by 3 months are given a trial of non surgical treatment
- False positives given non surgical treatment will never receive surgery, therefore probabilities at Y = 1 and Z = 0
- All treatment among those not screened, missed or not treated by 3 months following positive screen is surgical. Therefore probabilities at branches O,Q,S = 1 and P,R,S = 0

Appendix continued

ii. Outcomes

a. **Probability of a favourable Severin Score on pelvic Xray at 16 years**

Favourable outcome = Severin Grades 1, 2 or 3
Unfavourable outcome = Severin Grades 4, 5 or 6

b. Probability of avascular necrosis of any type (using Kalamchi & MacEwen, Salter or Bucholz where available, minimum 2 years follow up)

Favourable outcome = no avascular necrosis
Unfavourable outcome = avascular necrosis

Assumptions:

- Children with CDH who fail non surgical treatment receive a closed reduction only
- Children with CDH who are not screened, miss screening, are false negatives or are screen positive but treated after 6 months all receive surgery in the following combinations: 0.7 have closed reduction and in 20% of these this is followed by an open reduction, giving 0.56 probability of closed reduction alone and 0.14 probability of closed followed by open reduction; 0.3 have an open reduction alone.

No Screening

Branch	Probability of Severin 4 or worse	Probability of avascular necrosis
1	1	1
2	0.749161	0.84771
4/6*	1	1

**set to 1 as all cases receive surgery as primary treatment only*

Current practice, universal ultrasound or selective use of ultrasound

Branch	Probability of Severin 4 or worse	Probability of avascular necrosis
Unaffected Favourable outcome Branches 1/8/23	1	1
CDH False negatives Favourable treatment outcome Branches 2/9	0.749161	0.84771
CDH Non surgical treatment only Favourable treatment outcome Branches 4/11/19/26	0.98062	0.9906
CDH Surgery following non-surgery Favourable treatment outcome Branches 6/13/21	0.82	0.8772
False positives Non surgical treatment only Favourable treatment outcome Branch 15	1	0.9906
False positives Surgery following non-surgery * Favourable treatment outcome Branch 17	1	0.8772
Missed being screened Surgery Favourable treatment outcome Branch 24	0.749161	0.84771
Missed being screened Non surgical treatment only Favourable treatment outcome Branch 26	0.98062	0.9052
Missed being screened Surgery following non-surgery Favourable treatment outcome Branch 28	0.82	0.8772

**set to 1 as all false positive cases receive non surgical treatment only*

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