Newborns With Suspected Occult Spinal Dysraphism: A Cost-Effectiveness Analysis of Diagnostic Strategies

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ABSTRACT. *Objective.* **To assess the clinical and economic consequences of different diagnostic strategies in newborns with suspected occult spinal dysraphism.**

Methods. **A decision-analytic model was constructed to project the cost and health outcomes of magnetic resonance imaging (MRI), ultrasound (US), plain radiographs, and no imaging in newborns with suspected occult spinal dysraphism. Morbidity and mortality rates of early versus late diagnosis of dysraphism and the sensitivity and specificity of MRI, US, and plain radiographs were obtained from the literature. Cost estimates were obtained from a hospital cost accounting database and from the Medicaid fee schedule.**

Results. **We found that the choice of imaging strategy depends on the underlying risk of occult spinal dysraphism. In low-risk children with intergluteal dimple or newborns of diabetic mothers (pretest probability: 0.3%–0.34%), US was the most effective strategy with an incremental cost-effectiveness ratio of $55 100 per quality-adjusted life year gained. For children with lumbosacral dimples, who have a higher pretest probability of 3.8%, US was less costly and more effective than the other 3 strategies considered. In intermediate-risk newborns with low anorectal malformation (pretest probability: 27%), US was more effective and less costly than radiographs and no imaging. However, MRI was more effective than US at an incremental cost-effectiveness of $1000 per quality-adjusted life year gained. In the high-risk group that included high anorectal malformation, cloacal malformation, and exstrophy (pretest probability: 44%–46%), MRI was actually cost-saving when compared with the other diagnostic strategies. For the intermediate-risk group, we found our analysis to be sensitive to the costs and diagnostic performances (sensitivity and specificity) of MRI and US. Lower MRI cost or greater MRI diagnostic performance improved the cost-effectiveness of the MRI strategy, whereas lower US cost or greater US diagnostic performance worsened the cost-effectiveness of the MRI strategy. Therefore, individual or institutional expertise with a specific diagnostic modality (MRI versus US) may influence the optimal diagnostic strategy.**

Conclusion. **In newborns with suspected occult dysraphism, appropriate selection of patients and diagnostic strategy may increase quality-adjusted life expectancy and decrease cost of medical work-up.** *Pediatrics* **2001; 108(6). URL: http://www.pediatrics.org/cgi/content/full/108/6/e101; *cost-effectiveness analysis, occult spinal dysraphism, newborns, MRI, ultrasound.***

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ABBREVIATIONS. MRI, magnetic resonance imaging; QALY, quality-adjusted life year; US, ultrasound; LE, life expectancy.

Three percent of neonates have major central nervous system or systemic malformations.[1] Furthermore, 5% to 15% of pediatric neurology hospital admissions are related to cerebrospinal anomalies.[2] Occult spinal dysraphism (skin-covered lesions with no exposed neural tissue) is the most prevalent spinal axis malformation[3] and the most common indication for spinal imaging in children.[4] Occult spinal dysraphism encompasses the entire range of skin-covered spinal column and neuraxis anomalies resulting from the failure of primary or secondary neurulation.[5,6] Occult spinal dysraphic lesions include dorsal dermal sinus, tethered cord with spinal lipoma, lipomyelomeningocele, and diastematomyelia[4,6–8] and are commonly associated with urinary tract anomalies.[9]

The clinical spectrum of occult dysraphism is broad, ranging from skin stigmata such as a dimple, sinus tract, hair patch, or hemangioma to motor, bladder, or bowel dysfunction.[6–8] Approximately 50% to 80% of occult spinal dysraphic cases exhibit a dermal lesion.[10–13] However, 3% to 5% of all children have skin dimples.[14,15] The prevalence of occult dysraphism has a broad range from as low as 0.34% in children with pilonidal sinuses (intergluteal dimples) to as high as 46% in newborns with cloacal malformation.[9,16] Because of the broad range of occult dysraphism prevalences, controversy exists as to which risk group may or may not benefit from imaging.

Early detection and prompt neurosurgical correction of occult spinal dysraphism may prevent upper urinary tract deterioration, infection of dorsal dermal sinuses, or permanent neurologic damage.[17–22] Several studies have demonstrated that motor function, urologic symptoms, and urodynamic patterns may be improved by early surgical intervention in patients with occult spinal dysraphism.[23,24] The surgical outcome may be better if intervention occurs before the age of 3 years.[23–25] Spinal neuroimaging, therefore, has the important role of determining the presence or absence of an occult spinal dysraphic lesion so that appropriate surgical treatment can be instituted in a timely manner.

There is no consensus regarding the most appropriate neuroimaging modality to diagnose occult dysraphism. Some authors advocate spinal ultrasonography as the primary imaging modality for lower spine congenital anomalies because of its reasonable diagnostic performance at a low cost and without sedation.[15,26] Others emphasize the importance of the more expensive spinal magnetic resonance imaging (MRI) examination because of its better diagnostic performance, excellent soft tissue characterization, and importance in presurgical planning.[6–8,27–29] Although plain film imaging is inexpensive and widely available, few authors advocate its use as a diagnostic test because of its low specificity.[30] Conversely, some clinicians have emphasized the importance of close clinical follow-up with no imaging in low-risk children who present with simple intergluteal dimples, because the probability of finding an occult dysraphic lesion is only 0.34%.[16]

Because occult spinal dysraphism is a relatively common pediatric disorder and early diagnosis and treatment are important, the role and cost-effectiveness of the imaging modalities for different risk groups need to be determined. Accordingly, we constructed a decision-analytic model to compare the cost and health outcomes of various work-up strategies in children with suspected occult spinal dysraphism.

METHODS

The Model

Cost-effectiveness analysis is a method used to evaluate the health outcomes and costs of interventions designed to improve health.[31–34] Primary and secondary (medical publications) data are used to construct a comprehensive decision-analytic model. The results of an analysis are summarized in a series of cost-effectiveness ratios that show the cost of achieving 1 unit of health outcome (eg, the cost per quality-adjusted life year [QALY] gained) for different risk groups and strategies.[31–34]

We constructed a decision-analytic Markov model to compare the costs and health outcomes of 4 diagnostic strategies in children who are suspected of having occult spinal dysraphism: 1) MRI, 2) plain radiographs followed by MRI for positive results (radiographs), 3) ultrasound (US) followed by MRI for positive results (US), and 4) no imaging with close clinical follow-up. We analyzed 3 risk groups based on the prevalence (pretest probability) of disease: 1) low (intergluteal dimple and infants of diabetic mothers; prevalence: 0.3%), 2) intermediate (low and intermediate anorectal malformation; prevalence: 27%–33%), and 3) high risk (high anorectal and cloacal malformation; prevalence: 44%–46%). Figure 1 shows a simplified representation of the decision tree.

The cost-effectiveness analysis was performed from a societal perspective. Costs were expressed in 1999 US dollars. Costs and cost per QALY were discounted at 3% per year.[32] Incremental cost-effectiveness ratios were calculated as the additional cost of one strategy divided by the additional effectiveness compared with the next most effective strategy.[33] Strategies that were both less effective and more costly than the alternative were eliminated because of dominance.[33] Strategies that had a higher incremental cost-effectiveness ratio and a lower effectiveness than another were eliminated because of extended dominance.[33] Cost-effectiveness ratios were expressed as cost per QALY gained. The model was constructed and analyzed by using the computer program Decision Analysis (DATA; TreeAge Software, Williamstown, MA).

Data

Data sources included medical publications from 1962 to 1998 reporting on occult spinal dysraphism. Model baseline values and ranges are summarized in Table 1.

Risk Groups

Children with suspected occult dysraphism were divided into low-, intermediate-, and high-risk groups. Children in the low-risk group included those with simple skin dimples as the sole manifestation or newborns of diabetic mothers. The prevalence (pretest probability) of a dysraphic lesion among low-risk patients has been estimated at 0.3% to 3.8%. In the low range (0.3%) are children with low intergluteal dimples (pilonidal sinuses); children in the upper range (3.8%) have higher lumbosacral dimples.[9,15,16]

Children in the intermediate-risk group included those with complex skin stigmata and low and intermediate anorectal malformations. The prevalence (pretest probability) of a dysraphic lesion among intermediate-risk patients has been estimated at 27% to 36%. Children in the high-risk group included those with high anorectal malformations, cloacal malformation, and cloacal exstrophy. The prevalence (pretest probability) of a dysraphic lesion among high-risk patients has been estimated at 44% to 100%.

Diagnostic Tests

Several studies have shown that MRI and US have better diagnostic performances (ie, sensitivity and specificity) than plain radiographs[4,13,26,30]; these data are shown in Table 1. The sensitivity of spinal MRI and US has been estimated at 95.6% and 86.5%, respectively.[4,26] The specificity of spinal MRI and US has been estimated at 90.9% and 92.9%, respectively.[4,26] Conversely, the sensitivity and specificity of plain radiographs have been estimated at 80% and 18%, respectively.[13,30] Because there are no large series assessing the joint diagnostic performance of MRI and CT, we assumed conditional independence between these tests for the case in which a positive plain radiograph or US examination was followed by MRI (Fig 1).

Natural History and Surgical Intervention

Symptoms from occult spinal dysraphism often are not apparent until the child becomes older.[15] The most common clinical

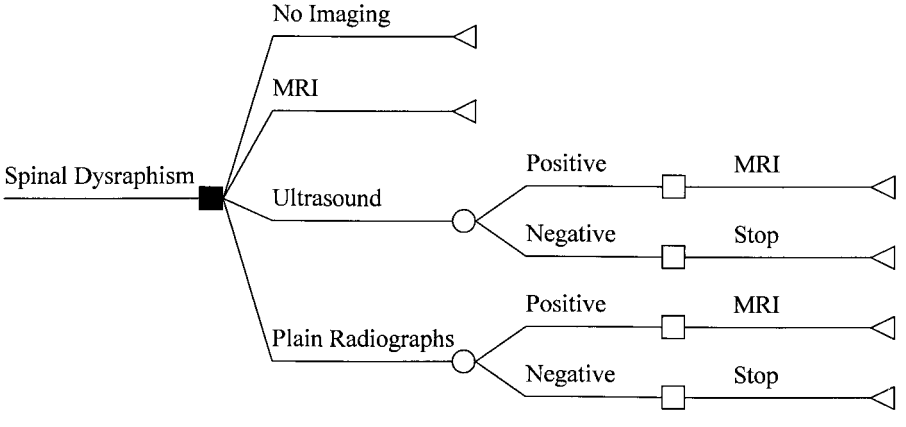

Fig 1. Summarized decision tree of diagnostic strategies in newborns with suspected spinal dysraphism.

TABLE 1. Summary of Data Used in the Analysis

Variable	Baseline Value	95% Confidence Interval*	Reference
Low-risk group			
Offspring of diabetic mothers	0.3%		20, 49, 50
Pilonidal sinuses (intergluteal dimples)	0.34%		16
Lumbosacral dimple	3.8%		15
Intermediate-risk group			
Low anorectal malformation	27%		51
Intermediate anorectal malformation	33%		51
Complex skin stigmata†	36%		15
High-risk group			
High anorectal malformation	44%		51
Cloacal malformation	46%		9
Cloacal exstrophy	100%		9
Diagnostic test performance			
Ultrasound			
Sensitivity	86.5%	75%–98%	26
Specificity	92.9%	84%–100%	26
MRI			
Sensitivity	95.6%	89.8%–99.7%	4
Specificity	90.9%	75.7%–98.1%	4
Plain Radiographs			
Sensitivity	80%	80%–100%	13, 30
Specificity	18%	11%–25%	30

* 95% confidence interval derived from the available literature.
† Hemangiomas, hairy patches, and subcutaneous mass.

presentations for occult dysraphic states later in infancy include delay in development of sphincter control, delay in walking, asymmetry of the legs or abnormalities of the feet (ie, pes cavus and pes equinovarus), and pain in the back or lower extremities.[12,13,18,24,35–39]

Several studies have demonstrated improvement of the multiple symptoms associated with occult dysraphism if surgical intervention is performed.[23–25] Newborns with spinal dysraphism were divided into early and delayed diagnosis. The early dysraphism diagnosis group consisted of patients whose dysraphism was diagnosed and surgically treated in the newborn period. The late dysraphism diagnosis group consisted of patients whose dysraphism was not diagnosed in the newborn period but rather diagnosed and surgically treated by or after age 3 years as the children became symptomatic.[25] Using surgical outcome data from the study by Satar et al,[25] in the early diagnosed and surgically treated patients, 60% became asymptomatic, 30% were unchanged, and 10% worsened. Conversely, the same study data for the late diagnosed and surgically treated patients (ie, surgery after 3 years of age) demonstrated that 27% became asymptomatic, 27% improved, 27% were unchanged, and 19% worsened.[25] To model these surgical outcomes, 4 health states were created ranging from asymptomatic (AS) to mild (S1), moderate (S2), and severe (S3) neuromuscular, urinary, and bowel symptoms.

Dysraphic patients with a central nervous system communicating dorsal dermal sinus (ie, 10% of all dysraphic patients) were considered to be at risk for infection.[13] The probability of becoming infected if diagnosed late was 50% over a 3-year period. A conservative mortality rate of 1% was used for meningitis.[40–44] Therefore, the total probability of any dysraphic child dying of infection was estimated at .0005.

In the model, only severely symptomatic patients with dysraphism (S3) were considered to be at risk of upper urinary tract deterioration.[45] In the S3 health state, only 15% had upper urinary tract deterioration[45] and of those with progressive renal damage, 7.5% were considered to develop end-stage renal disease over a 10-year period if undiagnosed.[45]

Long-Term Outcomes

The base-case patient was a newborn child without coexistent disease. The probability of dying of causes other than complications from occult spinal dysraphism was obtained from the US vital statistics data at the National Center for Health Statistics. To determine the effect of imaging on outcome, the following parameters were incorporated into the model: 1) the natural history of delayed diagnosis of occult dysraphism and associated renal abnormalities, 2) improved prognosis from early surgical intervention, 3) morbidity and mortality of undiagnosed occult spinal dysraphic lesions (ie, infected dermal sinus) and associated renal abnormalities (ie, end-stage renal disease[46,47]), 4) morbidity and mortality of imaging sedation, and 5) quality of life (Tables 1–3). We projected long-term outcomes such as life years and QALYs gained using these parameters.

The quality-of-life weight (utility) was obtained from the treating physician perspective (Table 3). A utility can vary from 0, representing death, to 1, representing perfect health. The quality weight (utility) for a year of life spent in a more favorable state (eg, mildly symptoms of dysraphism) versus a more unfavorable state (eg, severe symptoms of dysraphism) was incorporated into the Markov model.

Costs

Costs (not charges) of the radiologic and nonradiologic (ie, hospitalization, surgery, infection and complication treatment, laboratories, and pharmacy) procedures were estimated from the Cincinnati Children's Hospital Medical Center cost accounting system. Fixed, variable, and overhead costs were analyzed. Summary of cost data for the different diagnostic studies, symptomatic stage, neurosurgery, surgical complications, infection, and end-stage renal disease are shown in Table 2. The cost of the radiologic studies includes the technical and professional fee.

Sensitivity Analysis

Sensitivity analysis was performed for each parameter of the model over a clinically plausible range of values (Table 1–3). The effect of these changes on outcomes, costs, and cost-effectiveness ratios was analyzed.

RESULTS

Baseline Analysis (Table 4)

For low-risk children with intergluteal dimple or in offspring of diabetic mothers (pretest probability: 0.3%–0.34%), no imaging with close clinical follow-up cost less and improved health outcomes compared with the plain radiographs and MRI strategies. However, the US strategy offered a higher effectiveness at an incremental cost-effectiveness ratio of $55 100 per QALY gained. In low-risk children with a lumbosacral dimple (pretest probability: 3.8%), the

TABLE 2. Summary of Cost Data Used in the Analysis

Variable	Direct Cost	Total Cost*	Medicaid†	Sensitivity Analysis Range
Spine radiographs	$21	$37	$27	20–60
Ultrasound	$64	$110	$84	
MRI	$319	$550	$391	
Sedation	$70	$120	0#	0–150
Neurosurgery		$12 000**		
Surgical complication‡		$4000**		
Infection complication		$5500**		
End-stage renal disease		$40 000††		
Symptomatic 1§		$500††		
Symptomatic 2‖		$1400††		
Symptomatic 3¶		$2600††		

* Medical center cost estimates, includes direct (fixed and variable) and indirect (overhead) costs. Total costs were used for the case-base study.
† Medicaid reimbursement.
‡ ie, bleeding or suture dehiscence.
§ Mildly symptomatic.
‖ Moderately symptomatic.
¶ Severely symptomatic.
# Sedation by nonanesthesiologist is not reimbursed by medicaid.
** Mean cost per event (including surgery, hospitalization, pharmacy, laboratory studies, and follow-up visits).
†† Mean cost per year.

TABLE 3. Summary of Data Used in the Analysis

	Baseline	Range	Reference
Probability dermal sinus	10%	10%–35.6%	13, 24
Probability die from meningitis*	1%	1%–13%	40–44
Probability of upper urinary tract deterioration	15%		45
Probability end-stage renal disease†	7.5%		45
End-stage renal disease 5-y survival	95%		46
Probability of death from a reaction to MRI contrast material	0.25/1 000 000	0.2–0.5/1 000 000	52, 53
Probability of severe adverse reaction from MRI contrast material	0.25/100 000	0.2–0.3/100 000	52, 53
Probability of death from sedation	1/1 000 000	0.5–2/1 000 000	54, 55
Probability of systemic complication from sedation	1/1000		54, 55
Probability of death from neurosurgery‡	0.5%	0.1%–1%	56, 57
Probability of severe complications from neurosurgery‡	3%	0.5%–5%	56, 57
Utilities§			
Asymptomatic	1		
Mildly symptomatic	.95		
Moderately symptomatic	.9		
Severely symptomatic	.8		
Surgery no complications	.95		
Surgery short-term complications	.85		
Infection	.8		
End-stage renal disease	.7		

* Applies to patients who have dermal sinus and get meningitis.
† Applies only to patients who have severe dysraphic findings and progressive upper urinary tract deterioration.
‡ Data from institutional statistics and available neurosurgical literature.
§ As estimated by neurosurgeon.

US strategy cost less and was more effective compared with all other diagnostic strategies.

For children in the intermediate-risk group with pretest probability of 27% (ie, low anorectal malformation), the US strategy cost less and was more effective than the plain radiographs and no imaging strategies. However, the MRI strategy was more effective than US at a marginal cost-effectiveness ratio of $1000 per QALY gained. For high-risk children with high anorectal malformation (pretest probability: 44%), the MRI strategy cost less and was more effective than all other diagnostic strategies.

Sensitivity Analysis

For the intermediate-risk group, the incremental cost-effectiveness ratio of MRI decreased as the diagnostic performance (ie, sensitivity and specificity) of MRI increased (Table 5). At an MRI sensitivity and specificity at or above 100% and 93%, respectively, MRI became cost saving in comparison with all other strategies.

In the intermediate-risk group, the marginal cost-effectiveness ratio of MRI increased as the diagnostic performance (ie, sensitivity and specificity) of US

TABLE 4. Cost-Effectiveness in QALYS*

Strategy	Cost	QALY	Cost-Effectiveness Ratio ΔC/ΔQALY†
Low-risk group prior probability 0.3%			
No imaging	$150	29.088	
US	$312	29.091	$55 100
Plain	$1569	29.079	Dominated
MRI	$1833	29.077	Dominated
Low-risk group prior probability 3.8%			
US	$1186	29.064	
No imaging	$1899	29.013	Dominated
Plain	$2462	29.048	Dominated
MRI	$2514	29.058	Dominated
Intermediate-risk group prior probability 27%			
US	$6980	28.880	
MRI	$7026	28.926	$1000
Plain	$8382	28.844	Dominated
No imaging	$13 491	28.517	Dominated
High-risk group prior probability 44%			
MRI	$10 000	28.830	
US	$11 000	28.746	Dominated
Plain	$13 000	28.695	Dominated
No imaging	$22 000	28.153	Dominated

* Costs and effects are per person in the target population.
† Rounded in hundreds.
Δ Incremental, ie, cost or QALY gained.

decreased. At an US sensitivity and specificity at or below 85% and 89%, respectively, MRI became cost saving in comparison with all other strategies.

Figure 2 shows a 2-way sensitivity analysis of MRI cost and MRI sensitivity for the intermediate-risk group. The incremental cost-effectiveness ratio of MRI decreased as the cost of MRI decreased and MRI sensitivity increased. MRI became a cost-saving strategy when the following criteria were met: 1) MRI cost of $390 and MRI sensitivity of 83% or higher, 2) MRI cost of $500 and MRI sensitivity of 90% or higher, and 3) MRI cost of $670 and MRI sensitivity of 100%.

In the low-risk group (pretest probability: 0.3%), the increment cost-effectiveness ratio of US decreased as the sensitivity and specificity of US increased (Table 5 and Fig 3). Figure 3 shows a 2-way sensitivity analysis of US cost and US specificity. The incremental cost-effectiveness ratio of US decreased as the cost of US decreased and the US specificity increased. US became a cost-saving strategy when the US cost was $50 and the US specificity was 98.5% or higher.

Analysis of the data using as outcome life expectancy (LE) without quality adjustment (ie, no utilities) was done. In the low-risk group with a pretest probability of 0.3% (ie, intergluteal dimple or offspring of diabetic mother), no imaging was cost saving because it cost less and was more effective in LE gained compared with all other diagnostic strategies. In the low- and intermediate-risk groups with a pretest probability of 3.8% (ie, lumbosacral dimple) and 27%, respectively, US was cost saving because it cost less and was more effective in LE gained compared with all other diagnostic strategies. In the high-risk group (pretest probability: 44%), the MRI strategy cost less and was more effective in LE gained than the plain radiographs and no imaging strategies. However, the US strategy was more effective than MRI at a high marginal cost-effectiveness ratio of $870 000 per LE gained. In the high-risk group at a pretest probability of 50% or higher, MRI was the preferred strategy because it cost less and was more effective than all other diagnostic strategies.

DISCUSSION

Controversy exists in regard to the most appropriate neuroimaging modality to assess occult spinal dysraphism in the newborn. We used the methods of decision analysis to compare the long-term costs and health outcomes of various imaging strategies for children with suspected occult spinal dysraphism. We found that the choice of imaging strategy depends on the underlying risk of occult spinal dysraphism. In low-risk children with intergluteal dimple or in newborns of diabetic mothers (pretest probability: 0.3%–0.34%), the US strategy was the most effective with an incremental cost-effectiveness ratio of $55 100 per QALY gained. For children with lumbosacral dimples, who have a higher pretest probability of 3.8%, US was less costly and more effective than the other 3 strategies considered.

In intermediate-risk newborns with low and intermediate anorectal malformation or skin stigmata (pretest probability: 27%), US was cost saving when compared with radiographs and no imaging. However, MRI was more effective than US at an increment cost-effectiveness of $1000 per QALY gained. In the high-risk group that included high anorectal malformation, cloacal malformation, and exstrophy (pretest probability: 44%–46%), MRI was actually cost saving when compared with the other diagnostic strategies.

The cost-effectiveness ratios allow comparisons with alternative health care programs and may assist in resource allocation decisions.[33] The low-risk group US cost-effectiveness ratio at $55 100 per QALY saved compared favorably to other well-ac-

TABLE 5. Cost-Effectiveness Ratios of the Sensitivity Analysis

	MRI†	Ultrasound
Intermediate-risk patients with low anorectal malformation (pretest probability: 27%)		
MRI sensitivity		
100%	Cost-saving	Dominated
95.6%*	$1000	—
90%	$2500	—
80%	$6000	—
70%	$10 200	—
MRI specificity		
93%	Cost-saving	Dominated
90.9%*	$1000	—
80%	$28 000	—
70%	$80 000	—
Ultrasound sensitivity		
80%	Cost-saving	Dominated
85%	Cost-saving	Dominated
86.5%*	$1000	—
90%	$10 000	—
95%	$67 000	—
97%	$423 000	—
98%	Dominated	Cost-saving
99%	Dominated	Cost-saving
Ultrasound specificity		
89%	Cost-saving	Dominated
90%	$200	—
92.9%*	$1000	—
95%	$1600	—
99%	$2700	—
Probability ESRD		
1.5 × baseline	Cost-saving	Dominated
baseline*	$1000	—
0.75 × baseline	$2500	—
0.5 × baseline	$4200	—
Die from meningitis		
1.5 × baseline	$1000	—
baseline*	$1000	—
0.5 × baseline	$1000	—
Die from surgery		
1.5 × baseline	$1100	—
baseline*	$1100	—
0.75 × baseline	$900	—
0.5 × baseline	$800	—
Discount		
0%	Cost-saving	Dominated
3%*	$1000	—
5%	$15 700	—
Low-risk patients with pilonidal sinuses (pretest probability: 0.3%)		
Ultrasound sensitivity†		
90%	—	$51 300
86.5%*	—	$55 100
80%	—	$63 700
70%	—	$81 400
Discount†		
0%	—	$3700
3%*	—	$55 100
5%	—	$106 300

ESRD indicates end-stage renal disease.
* Base case.
† No imaging where indicated by † symbol.

cepted diagnostic strategies. For example, annual mammography and breast examination for women age 40 to 49 years[48] costs $62 000 per life year saved, annual cervical cancer screening for women beginning at 21 years of age costs $50 000 per life year saved,[48] and colonoscopy for colorectal cancer screening for people older than 40 years costs $90 000 per life year saved.[48]

For the intermediate-risk group, we found our analysis to be sensitive to the costs and diagnostic performances (sensitivity and specificity) of MRI and US. Lower MRI cost or greater MRI diagnostic performance improved the cost-effectiveness of the MRI strategy, whereas lower US cost or greater diagnostic performance of US worsened the cost-effectiveness of the MRI strategy. Therefore, individual or institutional expertise with a specific diagnostic modality (MRI versus US) may influence the optimal diagnostic strategy. For example, radiologists who are more proficient with MRI than US may perform the diag-

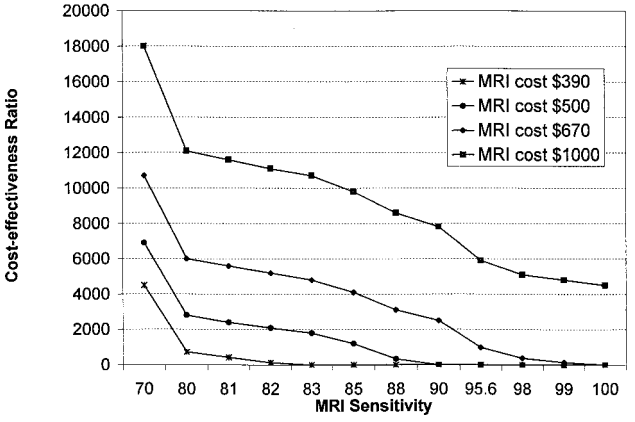

Fig 2. Two-way sensitivity analysis of MRI according to MRI cost and MRI sensitivity in intermediate-risk patients.

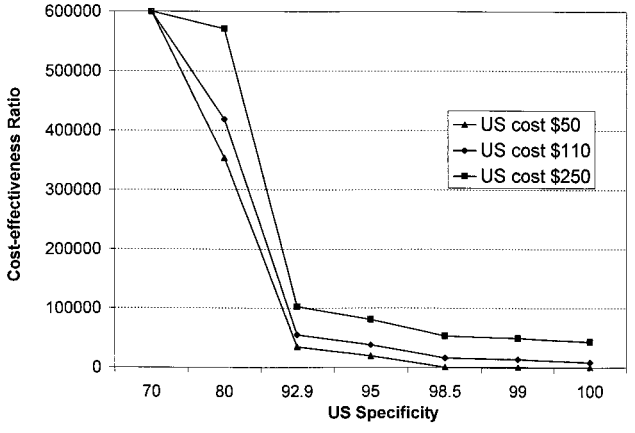

Fig 3. Two-way sensitivity analysis of US according to US cost and US specificity in low-risk patients (P = .3%).

nostic work-up of intermediate risk children with the former, whereas radiologists who are proficient with both may elect US because of the higher marginal cost-effectiveness ratio of the MRI strategy. Likewise, cost of the imaging strategies may affect the work-up strategy of choice. For example, a developing country with low US cost but high MRI cost may elect to recommend US to study children in the intermediate-risk group.

In the low-risk group (pretest probability: 0.3%), our analysis was sensitive to the cost and diagnostic performance (sensitivity and specificity) of US. Lower US cost or higher US diagnostic performance improved the cost-effectiveness of the US strategy. For example, in a developing country with well-trained radiologists performing low-cost ultrasonography, spinal US would be the strategy of choice for low-risk patients.

The results shown herein are summarized in Fig 4 as a suggested decision tree for use in newborns with suspected occult spinal dysraphism. The decision tree reinforces the primary importance of a careful acquisition of a medical history and performance of a thorough examination. For patients in the high-risk group, imaging of the spine with MRI is suggested. For patients in the intermediate-risk group, imaging of the spine with MRI or US is suggested, whereas in the low-risk group, the strategies of US or no imaging may be indicated. Selection between these 2 strategies per risk group may be based on individual and institutional diagnostic performance and cost per test. In newborns with suspected occult dysraphism, appropriate selection of patients for imaging based on these risk groups may maximize health outcomes for patients and improve health care resource allocation.

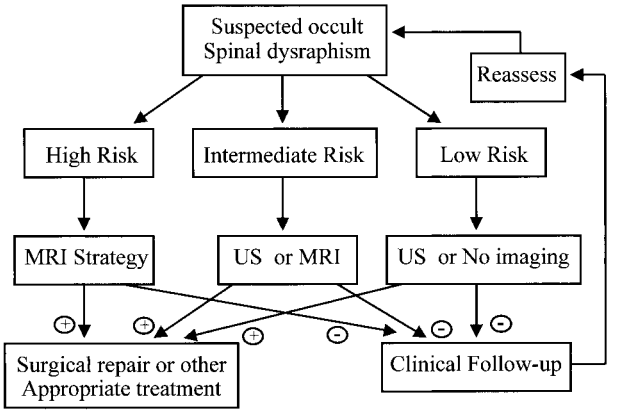

Fig 4. Suggested decision tree for use in newborns with suspected spinal dysraphism. For patients in the high-risk group, MRI is suggested. For patients in the intermediate-risk group, US or MRI is the strategy of choice; for the low-risk group, US or no imaging is recommended. For patients with negative imaging studies, close clinical follow-up with periodic reassessment is recommended.

Future studies clearly are needed. Because the spectrum of children with suspected occult dysraphism is so broad, other clinical risk groups need to be studied and their prevalence (prior probability) determined. Large prospective cohort studies in children with other well-defined stigmata or clinical findings of occult dysraphism are required for this purpose. Such studies might ultimately form the basis for valuable evidence-based practice guidelines for the large population of children evaluated for suspected occult dysraphism each year in the United States and other countries.

CONCLUSION

Our analysis suggested the MRI strategy in high-risk newborns (high anorectal malformation, cloacal malformation, or exstrophy) because it is cost saving. In the intermediate-risk newborns (low and intermediate anorectal malformation or skin stigmata), MRI or US is the preferred strategy. US or no imaging with close clinical follow-up is the preferred strategy in low-risk newborns. In the intermediate- and low-risk newborns, choice between the 2 preferred strategies is based on individual and institutional diagnostic performance and cost of the tests. In newborns with suspected occult dysraphism, appropriate selection of patients and diagnostic strategy may increase quality-adjusted LE and decrease cost of medical work-up.

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