CONSERVATIVE MANAGEMENT OF ASYMPTOMATIC SPINAL LIPOMAS OF THE CONUS

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OBJECTIVE: The natural history of spinal lipomas of the conus (SLCs) has not been well studied. Because of disappointing long-term results with early surgical treatment of asymptomatic children with SLCs, we have followed a protocol of conservative management for these patients. The results are presented in this report.

METHODS: Since 1994, all asymptomatic children with SLCs who were examined at Necker-Enfants Malades Hospital were subject to a protocol of conservative management. The records for those patients were reviewed, to determine the incidence and timing of neurological deterioration. The findings were compared with those for a previously reported historical cohort of asymptomatic patients who underwent early surgery at our institution.

RESULTS: Fifty-three asymptomatic children (35 girls and 18 boys) with SLCs were monitored, with conservative management. During a mean follow-up period of 4.4 years (range, 12 mo to 9 yr), 13 patients (25%) exhibited neurological deterioration. At 9 years, the actuarial risks of deterioration, as determined with the Kaplan-Meier method, were 33% for the conservatively treated patients and 46% for the surgically treated patients. With a Cox proportional-hazards model, there was no significant difference in the risks of neurological deterioration for patients who were treated conservatively and those who underwent early surgery.

CONCLUSION: The incidences and patterns of neurological deterioration seemed to be very similar, regardless of whether early surgery was performed. These results suggest that conservative treatment of asymptomatic patients is a reasonable option. A more definitive randomized study will be required to clarify the relative efficacy of early surgery for SLCs among asymptomatic patients.

KEY WORDS: Lipoma, Natural history, Pediatric, Spinal dysraphism, Surgery

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Spinal lipomas of the conus (SLCs) are congenital lesions that usually present at birth with an obvious lumbosacral mass. The optimal treatment of asymptomatic patients with SLCs remains uncertain. Many articles and book chapters on this condition advocate early prophylactic surgery, on the basis of the idea that this offers improved outcomes, compared with the natural history (2, 3, 7–10, 12–14). However, two facts remain undeniable. First, several published series on the surgical treatment of SLCs have demonstrated that a substantial percentage of previously asymptomatic patients experience deterioration and develop symptoms with time, despite early surgery (4, 5, 10, 11, 15). Second, the true natural history of this condition is not known but has been estimated with indirect inferences (7, 9).

At Necker-Enfants Malades (NEM) Hospital, the standard approach for SLCs for more than 20 years was to offer surgery to all patients at presentation. However, a previously published review of the results of that management protocol, including early surgery for asymptomatic patients, provided some disconcerting results (11). In particular, patients who presented without neurological symptoms and underwent early surgery exhibited continued deterioration with time, with an actuarial risk of symptoms of nearly 60% at 12 years. Those results, combined with the lack of good evidence in the literature, prompted our group to begin using a prospective protocol in which all asymptomatic patients with SLCs underwent close clinical monitoring, rather than prophylactic surgery. The results of this management protocol are presented in this report.

PATIENTS AND METHODS

Patient Selection

Beginning in 1994, all pediatric neurosurgeons at NEM Hospital agreed to follow a revised protocol for the treatment

of patients with asymptomatic SLCs (see below for details of protocol). All asymptomatic patients who presented after the implementation of this revised protocol were prospectively recorded in a database. Inclusion criteria were the presence of a spinal lipoma with attachment to the conus, as demonstrated with magnetic resonance imaging (MRI), and no neurological symptoms or deficits. Exclusion criteria were a lipoma of the filum only and the presence of any neurological symptom or deficit (including clearly abnormal urodynamic features) at presentation.

Follow-up Monitoring and Outcome Measurements

In their initial assessments, patients underwent a clinical examination, a MRI study, a urodynamic evaluation, and, at the discretion of the attending surgeon, an electromyographic study. Patients were monitored with clinical assessments and urodynamic studies every 6 months until the age of 2 years and then with annual clinical assessments only. Routine urodynamic studies were not performed beyond 2 years of age unless there was a specific concern. Follow-up MRI studies were performed at the discretion of the attending surgeon. A functional neurological score, i.e., the modified NEM score, was recorded for each patient. The NEM score provides a rating of 4 (worse) to 18 (normal) by grading motor, sensory, bowel, and bladder functions (*Table 1*). This is a slightly modified version of a scale we previously described (11), in that the category of bowel function now includes four levels of function, rather than three. Therefore, a normal score on the modified NEM scale is 18, rather then 17. Although this scoring system is used in daily practice at our institution, its reliability and validity have not been formally assessed.

During the period of follow-up monitoring, patients were considered to have experienced deterioration if they developed any evidence of neurological symptoms or deficits, including 1) motor weakness or gait difficulty, 2) lower-limb deformities, 3) bowel or bladder dysfunction (based on clinical or urodynamic assessments), 4) sensory deficits, or 5) back or leg pain thought to be attributable to the lipoma. To ensure the accuracy of the data, the medical records of the patients were independently reviewed by a neurosurgeon (AVK) from another institution, who had not been involved in the medical care of any of the patients.

The primary outcome measure used in our analysis was the time (in months) to neurological deterioration. This was defined as the time from initial presentation to the time at which neurological deterioration was first noticed by a health care professional.

A comparison was also made with a historical cohort of asymptomatic patients with SLCs who had been treated at NEM Hospital during the previous 22-year period (1972–1994). The data for that series were previously published (11). That group of patients all underwent early prophylactic surgery at the time of presentation, with the goals being freeing of the spinal cord attachment, subtotal resection of the lipoma, and enlargement of the dural tube with graft material. The time to deterioration for those patients was

TABLE 1. Modified Necker-Enfants Malades functional score[a]

Score	Motor	Sensory	Bladder	Bowel
1				
Children/adults	Wheelchair	Skin ulceration or amputation	Day and night incontinence	Incontinence
Infants	Major deficit		Incontinence	Incontinence
2				
Children/adults	Major orthosis or two crutches	Pain	Night incontinence	Painful constipation requiring digital maneuvers
Infants		Pain	Retention	
3	Minor or distal orthosis	Painless deficit	Intermittent catheterization	Constipation
4				
Children/adults	Fatigue with walking	Normal	Dysuria, UTI, or stress incontinence	Normal
Infants		Normal	UTI or stress incontinence	Normal
5				
Children/adults	Normal		Normal	
Infants	Normal		Normal	

[a] UTI, urinary tract infection.

defined as the time (in months) from early surgery to the detection of neurological deterioration, including immediate postoperative deterioration.

Primary Analysis

Survival curves were calculated with the Kaplan-Meier method. Survival analysis of the time to neurological deterioration was performed with a multivariable Cox proportional-hazards model. This analysis was performed with the combined data set for the present series and the historical cohort. The following independent variables were tested: early surgery (yes/no) and patient age at presentation (in months). The proportional-hazards assumption was checked for each variable by plotting the partial residuals against time. The multivariable model was used to provide an adjusted estimate and 95% confidence interval of the hazard ratio for the variables. The Wald test was used for significance testing of the hazard ratios in the multivariable model, at a significance level of 0.05.

Secondary Analyses

Several secondary analyses were performed, as follows. 1) A comparison of the mean decrease in NEM scores (at the final follow-up assessments) between the conservatively treated patients and patients who underwent early surgery was performed with an independent-samples *t* test. 2) With the data for the current, conservatively treated cohort, the associations between certain patient characteristics and ultimate deterioration were examined. The following variables were studied with the χ^2 test: sacral agenesis (present versus absent), type of insertion of the lipoma on the cord (purely dorsal versus posterolateral), and spinal cord herniation outside the spinal canal on MRI scans (present versus absent). These were considered only secondary exploratory analyses; therefore, no specific significance level was set for the *P* value. All statistical analyses were performed with the SPSS advanced statistics software package (Version 8.0; SPSS, Inc., Chicago, IL).

RESULTS

Patient Characteristics

For the current series, there were a total of 54 eligible patients. Of those, one patient was assessed in an initial visit and was then lost to follow-up monitoring because the family moved to another country. The remaining 53 patients formed the basis of all further statistical analyses.

There were 35 female patients and 18 male patients. Forty-five patients presented at birth, whereas eight patients presented at ages ranging from 12 months to 19 years. The most common mode of presentation was detection of an obvious lumbosacral mass (52 patients). For one patient, the spinal lipoma was detected during the evaluation and treatment of perineal malformations (which did not constitute Currarino syndrome). Four patients had been diagnosed prenatally. Six patients presented with associated anomalies, i.e., three with partial sacral agenesis, two with perineal anomalies, and one with congenital cardiac anomalies.

Urodynamic Assessments

The initial urodynamic studies were available for review for 36 patients. For 29 patients, the findings were unequivocally normal. For seven patients (all infants, ranging in age from 3 to 19 mo), the results demonstrated sphincter spasticity, which was interpreted as being physiological, rather than pathological. In five of those cases, follow-up urodynamic assessments demonstrated completely normal results. In one case, repeated studies continued to demonstrate physiological sphincter spasticity for a completely normal child with no clinical evidence of urinary dysfunction. For one child, follow-up studies demonstrated deterioration with clearly abnormal urodynamic results, including incomplete bladder emptying. This occurred in association with clinical deterioration, consisting of loss of ankle reflexes, back pain, and urinary tract infections.

Imaging Characteristics

The initial MRI studies were available for review for 48 patients. The majority of lipomas were of the transitional type (n = 35), with a minority being dorsal (n = 7) or terminal (n = 6) types. The level of the conus termination ranged from L1–L2 to S3, with the vast majority (n = 45) terminating at the L3 level or below. The median number of levels over which the lipoma was attached to the conus was three (range, one to six). Most lipomas were attached purely to the dorsal aspect of the conus, but posterolateral attachment was noted in 16 cases (10 on the left and 6 on the right). Associated features included meningoceles (n = 7), evidence of spinal cord herniation outside the spinal canal (n = 7), and syringomyelia (n = 13). Cine MRI was performed for 29 patients and demonstrated spinal cord mobility in 5 cases and a fixed cord in the remainder.

Among the 40 patients who remained in clinically normal condition during the period of the study, 24 follow-up MRI studies (obtained a mean of 2.4 yr after the initial MRI studies; range, 3 mo to 6.4 yr) were available for review. The majority demonstrated no substantial changes from the initial findings. The following changes were noted, however: a decrease in the size of the lipoma (n = 4, with 2 having almost disappeared), a change in the size of the syrinx (2 bigger and 1 disappeared), and the development of a new syrinx (n = 4).

Patients with Deterioration

The patients were monitored for a mean of 4.4 years (range, 12 mo to 9 yr). During that period, 13 patients (25%) developed neurological deterioration, a mean of 24 months (range, 6 mo to 3.2 yr) after presentation. The following modes of deterioration were noted for 12 patients: bowel or bladder dysfunction (n = 8), foot or leg weakness (n = 6), foot or leg atrophy or deformity (n = 4), pain (n = 3), and numbness (n = 3). For one patient (mentioned above), physiological sphincter spasticity was demonstrated in the initial urodynamic

study; at the time of clinical deterioration, this spasticity worsened, with incomplete bladder emptying.

The deficits developed during a period of months for 11 patients and during a period of weeks for 2 patients. Eleven patients underwent surgery after they experienced deterioration. For the remaining two patients, neurological deterioration developed near the time of preparation of this manuscript and surgery was being planned but had not been performed. For the 11 patients who underwent surgery, the operation consisted of subtotal resection of the spinal lipoma, with untethering of the spinal cord in all cases and the use of duraplasty closure in 10 cases. No patient developed acute neurological worsening after surgery, although two patients required acute reoperation because of local wound complications (one cerebrospinal fluid leak and one pseudomeningocele with wound infection). Those 11 patients were monitored for an additional mean time of 3 years after the date of surgery (range, 6 mo to 4.6 yr). During that period, four patients exhibited improvement, six remained in stable condition, and one experienced further deterioration. The latter patient exhibited deterioration 4 years after the first surgical intervention (during which duraplasty had been performed with synthetic polyesterurethane graft material) and underwent a second untethering operation. At a follow-up time of 8 months after the second operation, the patient's neurological status was considered stabilized, compared with his status just before the second operation. Overall, the mean NEM score for this group of patients at the last follow-up examination was 16.1 (range, 11–18). The outcomes of specific symptoms after surgery were as follows: bowel or bladder dysfunction, three cases improved, two cases exhibited no change, two cases worsened, and one patient has not yet undergone surgery; foot or leg weakness, one case improved and five cases exhibited no change; foot or leg atrophy or deformity, two cases exhibited no change and two patients have not yet undergone surgery; pain, three cases improved; numbness, two cases improved and one case exhibited no change.

Imaging Results for Patients with Deterioration

The initial MRI scans for 10 patients who experienced deterioration were available for review. The majority of lipomas were of the transitional type (n = 9), with a minority being dorsal (n = 1) or terminal (n = 1) types. The level of the conus termination ranged from L2–L3 to S3. The median number of levels over which the lipoma was attached to the conus was three (range, two to six). Most lipomas were attached purely to the dorsal aspect of the conus, but posterolateral attachment was noted in three cases (all on the left). Associated features included meningoceles (n = 3), evidence of spinal cord herniation outside the spinal canal (n = 3), and syringomyelia (n = 4). Cine MRI was performed for six patients; no patients exhibited evidence of spinal cord mobility.

MRI studies performed at the time of deterioration, just before surgery, were available for review for 10 patients. Three of those studies demonstrated no changes. The remaining seven demonstrated only the following minor changes: increased syrinx size (n = 2), decreased syrinx size (n = 2), and development of a new syrinx (n = 3).

Postoperative MRI studies were available for review for eight patients. Removal of the lipoma appeared partial for seven patients and appeared subtotal (i.e., only a rim of fat remaining) for one patient. Cine MRI was performed for three patients and demonstrated no mobility of the cord for two and only slight mobility for one.

Prophylactic Surgery Historical Cohort

Data from the historical cohort of prophylactically surgically treated patients were available for 100 of the 109 original children in the series (11), including 56 female patients and 44 male patients. The mean age at the time of surgery was 22 months, and the mean follow-up period was 4.4 years. Twenty-two patients (22%) exhibited deterioration during the follow-up period.

Survival Analysis

The survival curves for both the current series and the historical, surgically treated cohort are presented in *Figure 1*. At 9 years, the actuarial risks of deterioration, as determined with the Kaplan-Meier method, were 33% for the conservatively treated patients and 46% for the surgically treated patients.

A Cox proportional-hazards model was used to assess the effects of age at presentation and early surgery on deterioration-free survival rates. Examination of partial residual plots suggested that both variables satisfied the proportional-hazards assumption. In a multivariable Cox proportional-hazards model, neither variable demonstrated a hazard ratio significantly different from 1. The hazard ratio for age was 0.99 (95% confidence interval, 0.97–1.0; $P = 0.2$) and that for early surgery was 0.89 (95% confidence interval, 0.42–1.88; $P = 0.8$).

Secondary Analyses

The mean decreases in the final NEM scores were 0.41 for the conservatively treated patients and 0.96 for the surgically

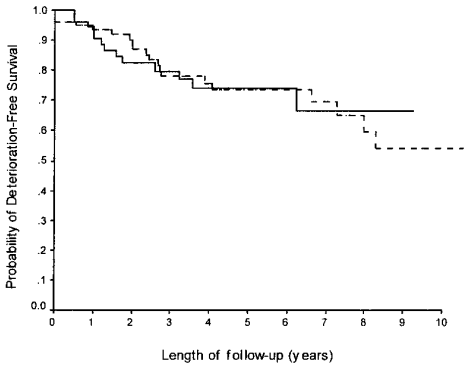

FIGURE 1. *Survival curve showing the deterioration-free survival of patients who were conservatively treated* (solid line) *and those who underwent early surgery* (dashed line).

treated patients ($P = 0.07$). There seemed to be little statistical difference in the outcomes of the various conservatively treated subgroups evaluated in the secondary analyses. The incidences of neurological deterioration were as follows: sacral agenesis: present, 33% deteriorated; absent, 24% deteriorated ($P = 0.7$); lipoma insertion on the cord: posterolateral, 19% deteriorated; dorsal, 25% deteriorated ($P = 0.6$); cord herniation outside the spinal canal: present, 43% deteriorated; absent, 20% deteriorated ($P = 0.3$).

DISCUSSION

The management of asymptomatic SLCs is still an unresolved matter, although many strongly advocate prophylactic surgery. The arguments made in favor of this approach usually rely on the idea that the outcomes after surgery are superior to the natural history. This argument has been put forth in different ways, each of which, we think, has methodological limitations.

One line of argument is based on what is considered to be a poor natural history for untreated patients. However, before this study, the literature contained virtually no data on the true natural history of asymptomatic SLCs. Some have inferred a grave natural history for this condition because the relative ratio of symptomatic to asymptomatic patients seems to increase with patient age, suggesting that the condition worsens as the patients become older (7, 9). The major weakness of this argument is clear; the true ratio of symptomatic to asymptomatic patients at any age is not known, because we cannot account for asymptomatic patients who never come to medical attention. Although findings are certainly not conclusive, autopsy studies suggest that the latter group of patients may be substantial (1). Therefore, the argument that prophylactic surgery offers improved outcomes, compared with the natural history, simply cannot be made, because the natural history is unknown.

Another line of argument is based on the observation that the outcomes of patients who were surgically treated before the development of symptoms (virtually all infants) were better than those of patients who were surgically treated only after the development of symptoms (mostly older children) (10, 15). However, this ignores the fact that these represent possibly completely different types of patients, whose natural history, regardless of early surgical intervention, might be very different, as highlighted in a recent report from London (6). In addition, asymptomatic older children, who never come to medical attention, were (once again) not accounted for in this analysis. Without knowledge of their numbers, little can be said regarding the relative efficacy of prophylactic surgery.

Given the limitations of the existing literature, the major contribution of this series is that it presents a sizable, modern, prospective series of conservatively treated patients with asymptomatic SLCs. This allows us, for the first time, to make some reasonable comments regarding the true natural history of this condition. We had the additional advantage of being able to compare our results with those of our earlier protocol of prophylactic surgery for the same types of patients. We observed that the course of deterioration with conservative management and that with early surgical management were remarkably similar. At 3 years, the actuarial risks of deterioration were 22% with early surgical management and 20% with conservative management. At 8 years, the risks were 40 and 33%, respectively. Neither prophylactic surgery nor patient age at presentation seemed to be associated with a statistically significant difference in the risk of neurological deterioration. The mean decreases in the NEM functional scores at the last follow-up examinations were not substantially different between the conservatively treated cohort and the surgically treated cohort. In addition, no other variables associated with neurological deterioration could be identified in the conservatively treated cohort. However, these secondary analyses were limited by small sample sizes for some of the comparison groups.

Several previous series of asymptomatic SLCs also demonstrated patterns of continued late deterioration of varying degrees, despite early surgery for all patients. Recent series from Chicago (10), Pittsburgh (5), and Vancouver (4) reported actuarial risks of deterioration that plateaued at approximately 20% (at 8 yr), 40% (at 8 yr), and 70% (at 9 yr), respectively. In addition, Xenos et al. (15), reporting on the Birmingham, England, experience, noted deterioration for 21% of 14 asymptomatic patients in 5 years of follow-up monitoring. In addition, early postoperative complications, most commonly wound-related complications, demonstrated incidences of approximately 11 to 24%, with incidences of acute neurological complications of 0 to 5% (2, 10, 11, 15).

Interpretation of Results

We think that this study makes it clear that early surgery for asymptomatic SLCs is a matter that is still open to debate. It seems, on the basis of our nonrandomized data, that the patterns of deterioration with or without surgery are similar, suggesting that either approach could be reasonable. Therefore, we have chosen to continue with the current protocol of conservative management. It is important to stress that the use of such a protocol requires careful follow-up monitoring of each child. It can sometimes be difficult to determine whether a newborn infant is in neurologically normal condition. This determination requires an experienced neurosurgeon who is familiar with the neurological examination of newborns and an experienced urologist who is familiar with the interpretation of sometimes-difficult infant urodynamic studies.

Another issue that arises during conservative management involves aesthetic concerns regarding the lipomatous mass as the child becomes older. Although in the past we have eschewed aesthetic surgery for the mass, with the change in our management we now recommend it as an option for parents who desire it. Such surgery can be performed via liposuction (when the anatomic features of the lesion on MRI scans exclude the possibility of spinal cord herniation or a meningocele in the subcutaneous space) or via an open superficial procedure. In our series, nine patients have undergone purely aesthetic surgery.

Pathophysiological Process of Neurological Deterioration

In addition to treatment recommendations, the current data force us to reexamine the proposed pathophysiological process of neurological deterioration with SLCs and the proposed mechanism by which surgery, even theoretically, addresses this pathophysiological process. On the basis of our earlier results demonstrating progressive neurological deterioration even after surgical untethering, we suggested that tethering might not be the only factor causing neurological worsening (11). These data seem to reinforce that view; the incidence and timing of patient deterioration were remarkably similar whether or not the patient had undergone early surgery. This certainly suggests that surgery did not ameliorate all of the relevant pathophysiological factors. The potential mechanical benefits of surgery, namely, untethering of the cord and possible relief of mass effect, do seem to be beneficial for at least a subset of patients, in particular, patients whose neurological deficits improve after the mechanical interventions of surgery. Would this group of patients, or perhaps another subset, benefit from early surgery? This question cannot yet be answered, and we currently seem to have no way to prospectively identify such patients.

Other postulated mechanisms of neurological deterioration include myelodysplasia and arachnoiditis (11). Arachnoiditis can develop with time among these patients, even those who have never been surgically treated. This has been put forth as another argument in favor of early surgery, i.e., surgery is technically easier when performed for infants, compared with later in childhood, when the appearance of scarring is more common. However, scarring also occurs after early surgical intervention and could possibly be more severe, resulting in late deterioration.

Limitations of the Study

Although our study represents the largest series of conservatively treated patients, an even larger sample size would have allowed more precise estimates of the survival curve. Similarly, a longer follow-up period would have allowed a more precise estimate of the end of the survival curve and more statistically powerful secondary analyses. We intend to continue monitoring this cohort of patients, especially as many of these infants and young children progress into adolescence.

Comparison with a historical cohort also presents certain limitations. For example, the period of accrual for the historical cohort was rather long and involved some minor variations in surgical techniques, including introduction of the ultrasonic aspirator, the contact laser, and intraoperative neuromonitoring. In addition, the follow-up period for the historical cohort was different from that for the series of patients. Although we attempted to account for this difference with the use of survival analyses, a longer follow-up period is still important.

CONCLUSIONS

This series of 53 children with asymptomatic SLCs who were treated conservatively, as part of a prospective protocol, demonstrated an incidence of neurological deterioration that was very similar to that observed for a similar cohort of patients treated with early surgery. Although we think that early surgery offers no improvement, compared with the natural history, we acknowledge that strong conclusions regarding the relative efficacy of surgery versus conservative management cannot be made without a randomized prospective study. However, it would obviously be preferable to avoid the added risk and inconvenience of surgery, if it was demonstrated to be of no added benefit in a more definitive study.

REFERENCES

1. Barson A: Symptomless intradural spinal lipomas in infancy. **J Pathol** 104: 141–144, 1971.
2. Byrne RW, Hayes EA, George TM, McLone DG: Operative resection of 100 spinal lipomas in infants less than 1 year of age. **Pediatr Neurosurg** 23:182–186, 1995.
3. Chapman PH, Davis KR: Surgical treatment of spinal lipomas in childhood. **Pediatr Neurosurg** 19:267–275, 1993.
4. Cochrane DD, Finley C, Kestle J, Steinbok P: The patterns of late deterioration in patients with transitional lipomyelomeningocele. **Eur J Pediatr Surg** 10[Suppl 1]:13–17, 2000.
5. Colak A, Pollack IF, Albright AL: Recurrent tethering: A common long-term problem after lipomyelomeningocele repair. **Pediatr Neurosurg** 29:184–190, 1998.
6. Dorward N, Scatliff J, Hayward R: Congenital lumbosacral lipomas: Pitfalls in analysing the results of prophylactic surgery. **Childs Nerv Syst** 18:326–332, 2002.
7. Hoffman HJ, Taecholarn C, Hendrick EB, Humphreys RP: Management of lipomyelomeningoceles: Experience at the Hospital for Sick Children, Toronto. **J Neurosurg** 62:1–8, 1985.
8. Iskandar B, Oakes W: Occult spinal dysraphism, in Albright AL, Pollack IF, Adelson PD (eds): *Principles and Practice of Pediatric Neurosurgery*. New York, Thieme Medical Publishers, Inc., 1999, pp 321–351.
9. Kanev PM, Bierbrauer KS: Reflections on the natural history of lipomyelomeningocele. **Pediatr Neurosurg** 22:137–140, 1995.
10. La Marca F, Grant JA, Tomita T, McLone DG: Spinal lipomas in children: Outcome of 270 procedures. **Pediatr Neurosurg** 26:8–16, 1997.
11. Pierre-Kahn A, Zerah M, Renier D, Cinalli G, Sainte-Rose C, Lellouch-Tubiana A, Brunelle F, Le Merrer M, Giudicelli Y, Pichon J, Kleinknecht B, Nataf F: Congenital lumbosacral lipomas. **Childs Nerv Syst** 13:298–334, 1997.
12. Satar N, Bauer SB, Scott RM, Shefner J, Kelly M, Darbey M: Late effects of early surgery on lipoma and lipomeningocele in children less than 1 year old. **J Urol** 157:1434–1437, 1997.
13. Sathi S, Madsen JR, Bauer S, Scott RM: Effect of surgical repair on the neurologic function in infants with lipomeningocele. **Pediatr Neurosurg** 19:256–259, 1993.
14. Wu HY, Kogan BA, Baskin LS, Edwards MS: Long-term benefits of early neurosurgery for lipomyelomeningocele. **J Urol** 160:511–514, 1998.
15. Xenos C, Sgouros S, Walsh R, Hockley A: Spinal lipomas in children. **Pediatr Neurosurg** 32:295–307, 2000.

COMMENTS

I consider this an important contribution to the literature on spinal lipomas. In a previous publication from the same center (4), the usefulness of prophylactic surgery was called into question. In fact, the neurosurgeons there considered the evidence sufficiently compelling to change their management protocol. The results of their more recent experience are presented here. Although the article title emphasizes the management of spinal lipomas, the article is directed toward the natural history. Dor-

ward et al. (3) previously called attention to the possibility that asymptomatic infants and symptomatic older children represented dissimilar groups that could not be directly compared in analyses of the potential benefits of prophylactic surgery. This study minimizes that bias. Eighty-five percent of the patients were diagnosed at birth. However, those who hoped that asymptomatic lipomas among infants might have a relatively benign prognosis will be disappointed by the authors' findings. If it is assumed that the earlier surgical interventions had no effect on the natural history of spinal lipomas and the data from the two series are combined, then there is a likelihood of neurological deterioration of 33 to 46% at 9 years and almost 60% at 12 years. Others who have advocated prophylactic surgery have made similar dire predictions regarding the natural history of these lesions. The mean follow-up period for this study was less than 5 years, and the longest follow-up period was only 9 years. Recognizing that this is a disorder that may cause progressive neurological deficits throughout the patient's lifetime, we must have some apprehension concerning the futures of these young children. We certainly hope that the authors and their successors will continue to provide us with follow-up data. We also look forward to current follow-up data on their patients who were previously treated with prophylactic surgery. It has been 6 years since their original publication, and any additional insights regarding that cohort would be welcomed.

Given the substantial likelihood of neurological deterioration, as indicated in this and other studies, it is not surprising that neurosurgeons have been eager to intervene in a constructive way. As early as 1950, Bassett (1) postulated that tethering was the mechanism by which lipomas caused neurological deficits and that surgical relief of traction on the conus was a mandatory part of successful surgery. This, along with debulking of the lipoma, has continued to be a primary goal of surgery. A problem with some of the reported series is that lipomas that are relatively straightforward surgically are grouped with more complex lesions. When this occurs, conclusions may be drawn that are misleading when applied to individual patients. Authors such as Cochrane et al. (2) have commendably addressed this issue. In this study, most of the lipomas were of the transitional type. This is relevant to the issue of prophylactic treatment, because such lipomas are the most difficult surgical lesions. In my experience, the extent and the intimate relationship between the lipoma and neural elements may preclude effective untethering, especially among older children and adults.

It must be asked why the authors found the results of prophylactic surgery to be so disappointing in their highly skilled hands. It cannot be assumed that the operations were technically inadequate. Given the large number of transitional lipomas, it is possible that complete untethering could not be achieved for an unknown fraction. Retethering, which is much more likely with extensive lesions, might also account for some of the poor outcomes. However, even if the majority of lipomas were incompletely untethered or were later retethered, we would still have to accept the authors' conclusions regarding management outcomes as valid. The authors raise the possibility that there is some other factor that is not amenable to surgical correction that is at least partly responsible for neurological deterioration. They mention arachnoiditis as one possibility. When lipomas among patients of all ages are treated, it is found that lesions among older individuals are often associated with more extensive fibrous tissue, especially within the lipoma itself, at the interface with neural tissue. We can only speculate regarding the effects of this process on the imbedded neurons and axons, as well as the microcirculation that nourishes them.

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1. Bassett RC: The neurologic deficit associated with lipomas of the cauda equina. **Ann Surg** 131:109–116, 1950.
2. Cochrane DD, Finley C, Kestle J, Steinbok P: The patterns of late deterioration in patients with transitional lipomyelomeningocele. **Eur J Pediatr Surg** 10[Suppl 1]:13–17, 2000.
3. Dorward N, Scatliff J, Hayward R: Congenital lumbosacral lipomas: Pitfalls in analysing the results of prophylactic surgery. **Childs Nerv Syst** 18:326–332, 2002.
4. Pierre-Kahn A, Zerah M, Renier D, Cinalli G, Sainte-Rose C, Lellouch-Tubiana A, Brunelle F, Le Merrer M, Giudicelli Y, Pichon J, Kleinknecht B, Nataf F: Congenital lumbosacral lipomas. **Childs Nerv Syst** 13:298–334, 1997.

The authors review a historical cohort of asymptomatic children with spinal lipomas of the conus (SLCs) who were treated with a protocol of conservative management. It is presently thought that the standard approach after identification of a spinal lipoma should be surgical treatment, in an attempt to decrease the potential long-term problems. Because of the lack of good evidence for surgical intervention for this condition, a review of conservative management raises the possibility that certain groups may benefit from a more conservative approach. In this review, the authors observed that children who were closely monitored and treated only after neurological deterioration demonstrated less risk of deterioration, compared with surgically treated patients. The authors also observed that there was no significant difference. These findings suggest that conservative management for asymptomatic children with SLCs is a reasonable option. This study involved a historical cohort and, although the statistical findings suggest conservative management as a reasonable option, definitive recommendations can be established only after a randomized controlled trial of early surgery versus conservative management. A trial of that type might also better describe patients who are likely to exhibit deterioration, with the potential for better early identification of such patients for surgical treatment. This report clearly indicates the potential for further study in this area.

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The treatment of children with SLCs is controversial. The entity represents a complex, skin-covered, neural tube defect, and conventional wisdom suggests that prophylactic surgery for asymptomatic patients is indicated to prevent delayed neurological deterioration. The natural history of this disorder has been

indirectly estimated, with studies suggesting that older patients are more likely to exhibit symptoms than younger patients, but no study has demonstrated the ratio of symptomatic to asymptomatic patients at any particular time. Therefore, the natural history of SLCs is really unknown, making it difficult to estimate the true effects of surgical intervention. Despite early surgery for asymptomatic patients, a certain percentage of patients eventually exhibit deterioration.

In this thoughtful analysis, Kulkarni et al. describe a cohort of 53 asymptomatic children with SLCs who were monitored prospectively, with nonsurgical treatment. The pattern of neurological deterioration was not different from that for a surgically treated historical cohort from the same institution. The authors conclude that early surgery does not change the natural history of asymptomatic SLC.

This is an intriguing conclusion, but one that must be taken in context. The surgically treated historical cohort spanned more than two decades, beginning in 1972, during which time there have been numerous changes in neuroimaging, surgical techniques, and intraoperative neurophysiological monitoring. Despite this criticism, this study suggests that surgery may not always be the answer for children with asymptomatic SLCs.

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There is debate regarding the treatment of infants and children with closed neural tube defects with lipomatous malformations, i.e., whether surgical intervention changes the natural history for patients who are asymptomatic at the time of diagnosis. For clarification of whether this subset of patients should undergo surgery despite being asymptomatic, the group from Necker-Enfants Malades (Paris, France) monitored 53 asymptomatic children with the aforementioned diagnosis. At a mean follow-up time of 4.4 years, 15 of 53 patients (25%) demonstrated neurological deterioration. At 9 years, 17 of 53 patients (33%) exhibited evidence of deterioration. In comparison, in a historical control group of 100 patients, 46 patients (46%) exhibited evidence of deterioration at 9 years. Therefore, at 9 years, there was no statistical difference between asymptomatic patients who had not been surgically treated and asymptomatic patients who had undergone early prophylactic surgical intervention. The authors suggest that nonsurgical management is an alternative to early surgical intervention for asymptomatic patients.

The authors refer to these lesions as lipomas. A lipoma is a benign neoplasm in which a progressive increase in the number of adipose cells occurs. These lesions are really hamartomatous lipomatous malformations. A number of other terms, such as lipomyelomeningocele, are also used and are not completely accurate either.

The authors used as a historical control cohort 100 patients who were surgically treated between 1972 and 1994. Obviously, the usual problems with historical control subjects can be noted. In addition, there was the problem of limited numbers of patients with multiple variables (such as the presence or absence of sacral agenesis, the size of the lipomatous malformation, the way in which the lesion involved the spinal cord, whether the spinal cord was beyond the spinal canal, and the presence or absence of a syrinx), yielding smaller subsets of patients and making it harder to note significance. Interestingly, there was a 35:18 female-to-male ratio in this series, whereas the historical control group exhibited a ratio of 56:54. We might wonder why there was a 2:1 predominance of female patients in this study. The authors noted that, in sequential magnetic resonance imaging studies, the syrinx became larger in two cases, disappeared in one case, and developed anew in four cases. This was not at all surprising. What was surprising, however, was a decrease in the size of the lipoma in four cases, with two having "almost disappeared." That is an unusual finding and is difficult to explain.

The longest follow-up period was 9 years. A better assessment would involve follow-up periods encompassing many decades, because there may be changes that develop at longer intervals. Another question that could be asked regarding the patients who underwent prophylactic surgery, despite being asymptomatic, is whether their loss of function would have been greater if they had not undergone early intervention.

This study definitely adds more information but does not fully answer the question. To do so would require a multiinstitutional effort encompassing many decades.

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