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Preventive Surgery for Asymptomatic Spinal Lipomas in Children

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ABSTRACT

AIM: To explore the clinical effect and significance of preventive surgery for asymptomatic spinal lipomas in children.

MATERIAL and METHODS: We retrospectively analysed the clinical data of 168 patients with asymptomatic spinal lipoma from April 2001 to June 2019, Shanghai Department of Neurosurgery, Shanghai Children's Medical Center, Shanghai Jiao Tong University School of Medicine. The patients were aged from 1.5 months to 15 years (the average age was 7 months), and there were no neurological symptoms, such as pain, incontinence, and/or bilateral lower limb dysfunction, before surgery. The surgical procedure included completely removing the lipomas in subcutaneous and extramedullary tissues of the spinal cord, subtotal resection of intraspinal fat, and separating the spinal cord, including the medullary conus from the dura sac, to release the tethered cord.

RESULTS: For the 168 children with spinal lipomas included in the study, complete resection was undertaken for the dorsal spinal lipomas, and subtotal resection was performed for the transitional lipomas. Subcutaneous effusion caused by cerebrospinal fluid leakage occurred in 5 cases after surgery and was cured after multiple punctures and aspiration. Six patients developed mild incontinence immediately after the operation, 5 of whom completely returned to normal within 1 month, and 1 had no relief of symptoms. A total of 159 of the 168 patients were followed up for 3 to 19 years (the median follow-up time was 76 months). Long-term postoperative symptoms were observed in 13 patients (7.7%), including 12 cases of spinal cord retethering and 1 case of lipoma enlargement.

CONCLUSION: Preventive surgery can reduce the future incidence of neurological dysfunction in children with asymptomatic spinal lipomas.

KEYWORDS: Spinal cord, Lipoma, Asymptomatic, Neurosurgery procedures, Prognosis

INTRODUCTION

Lipoma of the conus medullaris (LCM) is the most common congenital disease of spina bifida with slow onset. Many children can be asymptomatic, or they can gradually suffer from urinary incontinence, lower limb dysfunction and foot deformity due to spinal cord tethering or lipoma compression (10). There is a consensus regarding timely surgical treatment for LCM with pre-existing neurological dysfunction (5). However, it remains controversial whether preventive surgery is needed for children with LCM without neurological impairment (16). One view is that some children with asymptomatic LCM may remain without neurological impairment for their entire life, even if they are not treated. On the other hand, surgery may increase the risk of aggravating neural damage by itself (8). At the same time, some children may still have neurological dysfunction within several years after surgery (15). Another view is that the conservative treatment of asymptomatic children with LCM is more likely to cause neurological impairment, and once the clinical symptoms appear, up to 70% of children with LCM cannot recover even with surgical treatment (8). Based on the different viewpoints mentioned above, this paper retrospectively

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analysed the prognosis and recurrence of 168 asymptomatic LCM patients who underwent surgical treatment in the Shanghai Department of Neurosurgery, Shanghai Children's Medical Center, Shanghai Jiao Tong University School of Medicine from April 2001 to June 2019 and investigated the value of preventive surgery in the treatment of asymptomatic LCM.

MATERIAL and METHODS

Clinical Data

A total of 168 children with asymptomatic LCM were included in the study. There were 78 males and 90 females. The operative age ranged from 1.5 months to 15.0 years with an average age of 7 months. The clinical manifestations were subcutaneous lipoma of the central lumbosacral region of the back and/or superficial skin with nevus, local pits or neoplasm. None of the patients had obvious neurological symptoms, such as pain, incontinence, lower limb dysfunction or clubfoot deformity, before the operation, and the muscle strength of the lower limbs was normal. The family members of the children all signed informed consent forms for surgery (Approval number: SCMCIRB-K2023186-1; Date:11.06.2023).

Imaging Data

All children underwent MRI examination before the operation. We found dorsal spinal lipoma (lipoma attached only to the dorsal side of the spinal cord) in 132 cases (Figure 1A) and transitional spinal lipoma (lipoma extending from the back to one or both sides of the spinal cord) in 36 cases (Figure 1B).

Surgical Method

The surgical procedure includes completely removing the lipomas in subcutaneous and extramedullary tissues of the spinal cord, subtotal resection of intraspinal fat, and separating the spinal cord, including the medullary conus from the dura sac, to release the tethered cord. First, we separate the subcutaneous lipoma from the spinal canal defect, cut the lamina from the head of the defect to reveal the normal anatomical structure, and then carefully and gradually remove the subcutaneous lipoma from the head to the tail and enter the spinal canal cavity. We cannot suture the paraspinal muscles and the sacral fascia because the sacral canal defect is usually large. Therefore, too much of the lipoma outside the sacral fascia should not be removed. The lipoma can be used to cover and suture the sacral canal defect to prevent subcutaneous fluid accumulation. Then, the dura mater is cut from the top of the normal tissue until the lipoma is fully exposed, cutting the lipoma capsule under neuroelectrophysiological monitoring and using microscissors to remove the lipoma piece by piece from the spinal cord surface. When most of the spinal lipoma is removed and the spinal cord is decompressed, the pressure-relieved spinal cord is gradually lifted from the ventral side of the spinal lumen. At this time, since the spinal cord boundary has not been separated, we should not remove too much lipoma on the surface of the spinal cord to avoid injury of the spinal cord below. The spinal cord should be slightly retracted to one side and separated from the attached bilateral dural membranes from the head to the tail with microscissors to reveal the spinal cord boundary. The spinal nerves below the spinal cord can be seen later. Subsequently, the lipoma is safely removed from the surface of the spinal cord. Finally, the tether between the end of the dural sac and the conus medullaris is separated.

Follow-Up Methods and Evaluation Criteria

All patients were followed up in the outpatient department. Evaluation methods included preoperative and postoperative clinical examination and urinary system b-ultrasound examination. Clinical examinations include inquiries on the frequency and nature of stools, whether there are leaking stools, whether the urine is strong, whether there is urinary incontinence and enuresis, feeling, and muscle strength and muscle tone of both lower limbs. B-ultrasound examination includes determination of hydronephrosis in the kidneys, bladder capacity, and residual urine. Urodynamic examination

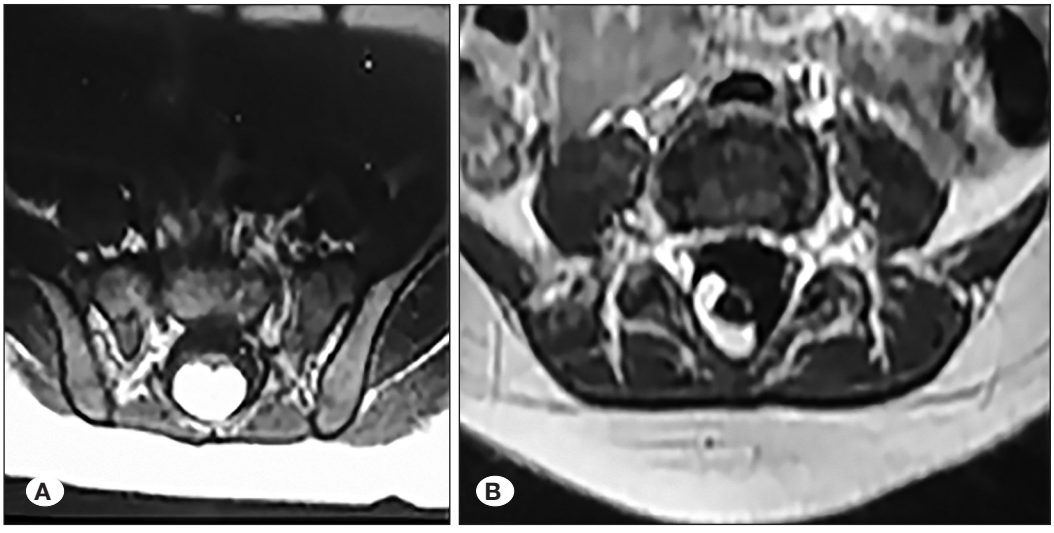

Figure 1: Magnetic resonance imaging (MRI) showed the relationship between lipoma and the spinal cord.
A) Dorsal type: lipoma attached only to the dorsal side of the spinal cord;
B) Transition type: lipoma not only located on the dorsal side of the spinal cord but also growing laterally.

or lower extremity electromyography examination should be performed in suspicious cases according to the pathological condition.

RESULTS

Surgical Results

All children with dorsal spinal lipoma were completely resected, and the transitional lipoma was sub-totally resected. Subcutaneous effusion caused by cerebrospinal fluid leakage occurred in 5 cases after surgery and was cured after multiple punctures and aspiration. Six patients (3.7%) developed mild incontinence immediately after the operation, 5 of whom completely returned to normal within 1 month, and 1 had no relief of symptoms.

Follow-Up Results

A total of 159 of the 168 children with asymptomatic LCM were followed up (94.6%), and 9 were lost to follow-up. The follow-up period ranged from 3 to 19 years, with a median follow-up time of 76 months. MRI re-examination 3 months after the surgery showed that the lipoma disappeared, and only a small amount of fat remained on the surface and inside the spinal cord. Among the 22 patients with syringomyelia before surgery, 11 cases improved, 5 cases disappeared, 4 cases were unchanged, and 2 cases were enlarged. Six of the 137 children without syringomyelia before surgery developed syringomyelia after the operation. There were 14 cases (8.8%) with long-term recurrence and 13 cases (8.2%) with symptoms. Of the 2 children with lipoma recurrence, 1 case was asymptomatic all the time, and the other case had right foot deformity with decreased muscle strength. Neurological symptoms were found in 12 patients with spinal cord retethering, including faecal incontinence and/or urinary incontinence and/or foot deformity with sensory muscle loss. Among them, 9 cases received spinal cord adhesion lysis again, 6 cases were improved, 2 cases remained stable, and 1 case was aggravated. The other 3 patients underwent a second operation.

DISCUSSION

Children with LCM had neurological dysfunction of different severities, primarily manifested as defecation and urination dysfunction, lower limb muscle strength change or pain, foot deformity and scoliosis, occult onset, slow progression, or were completely asymptomatic. According to reports, the incidence of asymptomatic LCM is 4 ~8/100,000 (12). There is currently controversy regarding whether asymptomatic LCM requires prophylactic surgical treatment. The focus of controversy is whether preventive surgery is superior to conservative treatment in avoiding the occurrence of neurological disorders. To solve this problem, it is necessary to understand the natural history of LCM and observe the incidence of neurological dysfunction.

The author has retrieved few literature reports regarding the natural history of LCM. Kulkarni et al. reported 53 untreated asymptomatic LCM children in 2004, of whom 33% developed neurological symptoms after 9 years of follow-up (8). In 2012, Wykes et al. reported 56 untreated asymptomatic children with LCM. After follow-up for 1 to 19 years (mean time was 5.9 years), 16 of them developed symptoms, with an incidence rate of 28.57% (15). Talamonti et al. reported 24 untreated asymptomatic LCM patients in 2014. Seven patients developed symptoms after 4-16 years of follow-up (10.4 years on average), with an incidence of 29.1% (13). At present, we have retrieved 9 articles on the surgical treatment of asymptomatic LCM in a total of 269 patients, of which 23 patients had postoperative symptoms, with an incidence of 8.55% (1,3-7,9,11,16). Pang et al. reported a set of data with the largest number of cases and the longest follow-up time retrieved from the English literature. During the follow-up period of more than 16 years, progression-free survival of surgical treatment of asymptomatic LCM was as high as 98.4% (1.6% with symptoms) (11). A total of 410 LCM cases with both surgical and conservative treatment were included in the 5 studies included in the meta-analysis. The results showed that the incidence of neurological dysfunction in untreated children with LCM was 28%, and the incidence of neurological dysfunction in preventive surgery was 11.22% (2,8,10,13,14). In this study, 168 children with asymptomatic LCM were all treated with preventive surgery. Except for 9 (5.4%) children who were not followed up, 1 of the other 159 children had irreversible nerve injury caused by surgery, and 13 patients presented long-term neurological injury symptoms after surgery. The incidence rate of nerve injury was 8.2%, which was similar to the foreign literature. Thus, the incidence of neurological dysfunction in conservative observation is generally higher than that in prophylactic surgery.

The pathological anatomy of the spinal lipoma is that the subcutaneous lipoma grows into the spinal lumen through the spinal canal defect and connects to the spinal cord lipoma, causing compression and tension to the spinal cord. However, with the growth of children, some of them develop symptoms of nerve lesions due to the tethered spinal cord caused by pulling and the compression of the lipoma. Therefore, we recommend early surgical treatment for asymptomatic patients with LCM. Due to incomplete differentiation of LCM in the embryonic stage, lipoma and the spinal cord grow mixed with each other without boundaries. Even in some cases, the lipoma grows around the spinal cord, which leads to a greater risk of surgery and is more likely to injure the nerve accidentally. Among the 168 children in this group, 6 had nerve injury immediately after surgery, with an incidence rate of 3.7%. Therefore, how to reduce surgical complications has become particularly important. The author believes that the following points need to be noted: First, the key of the surgery is to find the boundary of the conus and separate it from the dural membrane to release the tethered spinal cord to help the surgeon fully observe the boundary between the spinal cord and the lipoma to maximize the removal of the lipoma in the conus. For lumbosacral lipoma (Figure 2A, B), since the conus grows on the middle segment of the lumbosacral dural membrane and its boundary is easy to find, we only need to dissect the normal sacral dural membrane under the lipoma and cut it upward to see the conus and separate it from the

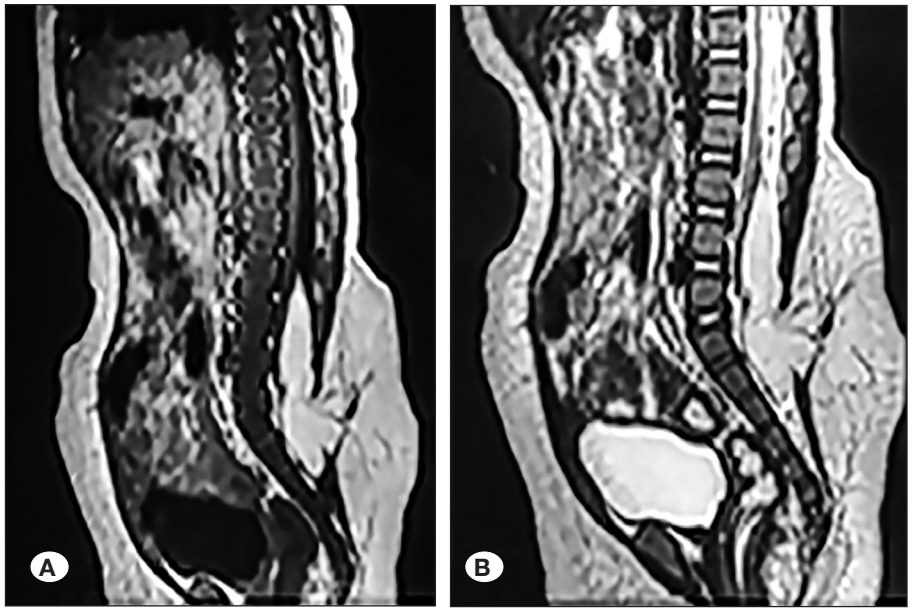

Figure 2: Sagittal MRI data of children with spinal lipoma. **A)** T1 image shows that subcutaneous lipoma extends internally, enters the spinal canal lumen through the spinal canal defect, and grows mixed with the lower spinal cord; **B)** The T2 image shows that the lower conus was located in the lumbosacral region.

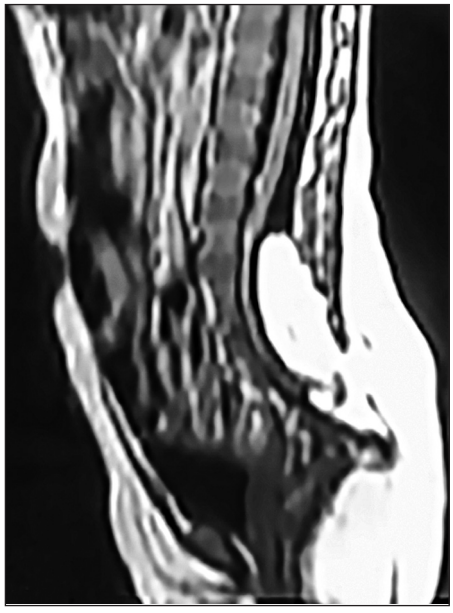

Figure 3: T1 sagittal MRI shows that the subcutaneous lipoma extends inwardly through the sacral canal into the spinal canal lumens and grows mixed with the lower spinal cord. The lower conus was located at the end of the dural sac.

dural membrane. For the sacrococcygeal lipoma (Figure 3), the lipoma on the surface of the conus not only grows with the conus but also grows outside the sacral canal. It is connected to the normal adipose tissue of the sacral tail and has no boundary, so it is difficult to find the conus. In this study, we first reduced the fat on the conus surface, separated the connection between the conus and the two sides of the dura mater, and then continued to thin the fat on the conus surface when part of the conus boundary was exposed. When tapering to the fibrous fat layer, gently retract the conus to one side to further confirm the end of the dural sac from the sacral lumen, as the end of the dural sac in the sacral lumen is inclined upward. After reaching the end of the dural sac, the conus is separated from the end of the dural sac from both sides to the middle to completely release the tethered spinal cord. Second, the technique for removing the lipoma is to cut the capsule of lipoma first to make lipoma expand gradually, separate the lipoma from the capsule and then cut it off layer by layer with fine scissors until the lipoma outside the spinal cord is excised. The fat in the spinal cord has different excision methods due to the pathological types. The dorsal lipoma, which is separated from the spinal cord by a layer of fibrous and glial tissue, can be completely removed along the surface of the spinal cord. It is worth noting that the lipoma should be excised layer by layer, with not too much deep mass resection, to avoid damage to the conus below. In contrast, it was found that the transition was not located on the outer side of the spinal cord but grew into the spinal cord and grew around the central tube of the spinal cord. Therefore, this part of the lipoma could only be largely removed to reduce the tension in the spinal cord and protect the function of the spinal cord (1,3,5,10). In 6 patients in this group, the reason for the aggravation of postoperative symptoms was that excessive removal of spinal cord fat and incision of the nerve lamina resulted in mild spinal cord injury.

Delicate surgical operation combined with neuroelectrophysiological monitoring is also the guarantee of successful LCM surgery. In addition, the type of lesions is also related to the risk of surgery; dorsal-type LCM operations are relatively easy, and the risk is relatively low. In this group, 132 cases of dorsal type accounted for most cases. However, transitional-type LCM with greater surgical difficulty and risk was relatively few in this group, with 36 cases. Objective results showed that the pathological types of asymptomatic LCM were relatively simple and that the surgical difficulty and risk were relatively low.

The purpose of LCM surgery is to remove the lipoma and release the tethered spinal cord. The data for this group showed that most patients with long-term neurological symptoms after surgery were caused by nerve adhesion. To

avoid retethering after surgery, the author believes that the following points need to be noted: 1) The tethered spinal cord must be completely released during the first operation. 2) The lipoma should be resected to the maximum extent. Pang et al. believed that partial resecting of lipoma in children would lead to more extensive fat scarring and adhesion after surgery and is more likely to produce secondary spinal cord tethers (11). Therefore, this study believed that LCM should be resected to the maximum extent. The dorsal lipoma can be almost completely removed, while the lipoma in the spinal cord can be sub-totally removed as much as possible. 3) When suturing the dura mater, the subdural space should be large enough, and if necessary, a patch should be used to expand the suture. In addition, the dural sac needs to be filled with water before the end of the dura suture so that the spinal cord is completely immersed in the cerebrospinal fluid, which can reduce the adhesion of the spinal cord by 85% (13).

Foreign scholars have proposed that prophylactic surgery is not recommended for asymptomatic LCM in older children because of the greater risk of surgery and the higher risk of postoperative neurological injury complications (8). The author agrees with this view. In this study, it was found that the fibre composition of asymptomatic LCM in older children increased, the toughness was higher than that of infant lipoma, and the boundary between lipoma and spinal cord was not as clear as in infants; therefore, it was difficult to remove the fat in the spinal cord. In addition, the long-term compression and tethering of LCM in older children may make spinal cords less tolerant to surgery than in infants and young children. Therefore, the author believes that the prophylactic operation of asymptomatic LCM in older children requires caution.

CONCLUSION

Asymptomatic LCM can be performed as early as possible at a qualified and experienced hospital to minimize intraoperative nerve damage. LCM should be removed as much as possible, and the tethered spinal cord should be completely released.

AUTHORSHIP CONTRIBUTION

Study conception and design: DG, NB
Data collection: DG, SQS
Analysis and interpretation of results: DG, BY, YHS
Draft manuscript preparation: DG
Critical revision of the article: NB
Other (study supervision, fundings, materials, etc...):
All authors (DG, NB, BY, YHS, SQS) reviewed the results and approved the final version of the manuscript.

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