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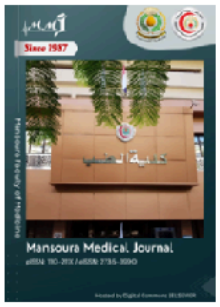

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Spinal cord lipomas in children: surgical resection and outcome

Ramy Omar Omar El Sakhry

Neurosurgery department of Mansoura specialized Hospital., ramineuro1982@gmail.com

Hatem Ibrahim Badr

Neurosurgery department, Faculty of Medicine, Mansoura University

Ibrahim El Sayd Ahmed Abdel-Aal

Neurosurgery department, Faculty of Medicine, Mansoura University

Amr Farid Khalil

Neurosurgery department, Faculty of Medicine, Mansoura University

ORIGINAL STUDY

Spinal Cord Lipomas in Children: Surgical Resection and Outcome

Ramy Omar Omar El Sakhry, Consultant of Neurosurgery[a,*],
Hatem Ibrahim Badr, Professor of Neurosurgery[b],
Ibrahim El Sayd Ahmed Abdel-Aal, Lecturer of Neurosurgery[b],
Amr Farid Khalil, Associate Professor of Neurosurgery[b]

[a] Neurosurgery Department, Mansoura Specialized Hospital, 35516, Egypt
[b] Neurosurgery Department, Mansoura University Hospitals, Mansoura University Faculty of Medicine, 35516, Egypt

Abstract

Background: **Management of spinal lipomas is considered a challenging task for pediatric neurosurgeons.**
Study design: **A prospective, clinical study was performed.**
Objectives: **To assess the neurological outcomes after surgical resection of spinal lipomas.**
Patients and methods: **The study was performed on 30 patients who underwent surgical resection of spinal cord lipomas. The cases were followed up postoperatively in the outpatient clinic for at least 24 months.**
Results: **This study included 30 patients with spinal lipomas (12 boys and 18 girls) with age of less than 2 years at surgery. At presentation, all the 30 cases had low back swelling. A total of 19 (63.3%) cases had bladder dysfunction, of whom 14 (73.7%) had incomplete dysfunction, whereas five (26.3%) had complete dysfunction. Moreover, 16 (53.3%) cases had motor weakness. Two (6.6%) cases presented with orthopedic deformities. All 30 cases had no previous surgery for spinal lipoma. Three types of spinal lipomas were detected in preoperative MRI: transitional ($n = 24$), dorsal ($n = 5$), and caudal ($n = 1$). Excision of lipoma was near total in 21 (70%) cases and subtotal in nine (30%) cases. Postoperative complications were cerebrospinal fluid leakage in five (16.6%) cases and wound infection in four (13.3%). Early outcome showed that two (6.6%) cases acquired a new but transient motor deficit. Out of the 16 patients who experienced preoperative motor weakness, 13 (81.3%) improved, whereas three (18.7%) cases remained the same. Incomplete bladder dysfunction was regained in nine (64.3%) patients, whereas only one (7.1%) patient progressed to have complete dysfunction (out of 14 patients). None of the patients with complete bladder dysfunction showed improvement. Long-term outcomes showed that of the 16 patients who presented with preoperative motor weakness, 11 (68.7%) patients had improved, whereas five (31.3%) patients developed worsening ($P < 0.001$). Concerning the bladder function, nine (81.9%) of the 11 patients with normal preoperative bladder function maintained normal function, whereas worsening of bladder function was noted in two (18.1%) patients. All the patients with preoperative complete bladder dysfunction remained on clean intermittent catheterization (CIC), whereas in 13 (92.9%) of 14 patients with incomplete bladder dysfunction, normal bladder function was regained, and only one case developed worsening ($P < 0.001$), that is, the untethering surgery resulted in significant improvement in the preoperative neurological deficits (motor and urological), as a P values less than 0.001. On long-term follow-up of our patients, five (16.7%) patients developed symptomatic retethering (neurological deterioration) and underwent partial excision of spinal lipomas. The mean time interval from appearance of first symptom and surgery in those who improved was 10 months (range, 4−18 months), whereas in those who did not improve was 16 months (range, 10−24 months), with P value less than 0.001, indicating the beneficial role of early surgery.**
Conclusion: **Better long-term neurological outcomes could be achieved after near-total than partial excision of spinal lipomas.**

Keywords: Occult spinal dysraphism, Spinal lipoma, Surgical outcome

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* Corresponding author. Faculty of Medicine, Mansoura University, Mansoura, Egypt.
E-mail addresses: ramineuro1982@gmail (R.O.O. El Sakhry), hatembadr1@gmail.com (H.I. Badr), iabdelaal@mans.edu.eg (I.E.S.A. Abdel-Aal), dr_amr77@yahoo.com (A.F. Khalil).

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1. Introduction

Spinal lipoma is defined as a soft subcutaneous swelling in the lumbosacral region, which presents at birth and results in spinal cord tethering that could lead to variable and progressive neurologic deficits in the form of motor impairment of the lower limbs and sphincter dysfunction. It proportionally increases in size with the body growth in general (Pang, 2019).

Moreover, conus lipomas were subcategorized by Chapman into three categories: caudal, dorsal, and transitional (Muthukumar, 2009).

As the other types of occult spinal dysraphism, early surgical intervention is required for untethering of the spinal cord. The surgery aims to relieve the mechanical traction of the lipoma on the conus. However, near-total resection of spinal lipomas in young age is a significant predictor for good long-term outcomes; the presence of preoperatively motor or sphincter dysfunctions has only a small chance to improve with surgery (Tarang et al., 2021).

The assessment of the extent of resection during surgery is guided by intraoperative neuromonitoring (IONM) of the residual lipoma and the ability to neurulate the placode (Charalampidis et al., 2020).

The performing of routine postoperative MRI to evaluate the residual lipomatous tissue is controversial. Although it is recommended by some authors (Pang et al., 2013; Talamonti et al., 2014), others suggest the usefulness of routine postoperative MRI because the decision to interfere is not only based on the presence of residual lipoma on an MR but also on clinical presentations (Collmann, 2011; Halevi et al., 2011).

Furthermore, other authors recommend routine postoperative MRI in particular situations such as an immediate postoperative complication related to the surgical technique (Steinbok, 2011; Thompson, 2011).

2. Aim

The aim was to assess the neurological outcomes after surgical resection of spinal lipomas.

3. Patients and methods

This was a prospective, clinical study that was approved by the ethics committee of Faculty of Medicine, Mansoura University. It was conducted at Department of Neurosurgery of Mansoura University Hospital in the period between July 2019 and October 2022. The study was performed on 30 patients who underwent surgical resection of spinal cord lipomas for the first time. The cases were followed up postoperatively in the outpatient clinic for at least 24 months. Patients were managed according to the study procedure mentioned later.

Inclusion criteria were as follows: the age during the time of study not exceeding 18 years, patients with lumbosacral lipomas who did not undergo untethering surgery for spinal lipoma before, and any patient with spinal dysraphism containing lipomas. However, any patient having other neuromuscular disorders was excluded from the study.

Study procedure: after receiving the approval, full detailed history, clinical examination, preoperative MRI, and urodynamic evaluation (history taking, clinical examination, and ultrasonography) were done (Tarang et al., 2021).

Surgical resection of the spinal lipomas was done with untethering and decompressing the spinal cord, while preserving the functional nerve roots. Under general anesthesia, the patient was operated in the following steps: elliptical skin incision, dissection of lumbosacral and thoracolumbar fascia, dural opening with near-total or subtotal resection of the intradural lipoma (according to the operative field), detethering of the cord, laminotomy was done if needed, identification of the filum, closure of the dura without graft in all patients, and finally, closure of the fascia and skin (Pang, 2020).

IONM was used in all patients for proper monitoring of the cord and minimizing postoperative morbidities (Charalampidis et al., 2020; Bello et al., 2015).

Postoperative evaluation of the neurological outcomes was performed in the outpatient for at least 24 months. Early outcome was observed in the first 12 months after surgery, whereas long-term outcomes were evaluated within the next 12 months after surgery (recognition of cord retethering).

3.1. Ethics of research

After approval of the ethical committee, consent was taken from the parents while respecting their personal privacy.

3.2. Statistical analysis

We used SPSS (Statistical Package for the Social Sciences) version 29 for Windows (SPSS Inc., Chicago, IL, US) program for performing statistical analysis. The appropriate statistical tests were used when needed. The end point of our study was

neurological deterioration. We considered P value less than 0.05 (5%) as statistically significant.

4. Results

This study included 30 patients with spinal lipomas (12 boys and 18 girls), with age of less than 2 years at surgery. At presentation, all the 30 cases presented with low back swelling. A total of 19 (63.3%) patients presented with bladder dysfunction, of whom 14 (73.7%) had incomplete dysfunction, whereas five (26.3%) had complete dysfunction. Sixteen (53.3%) cases had motor weakness. Spinal lipomas were transitional ($n = 24$), dorsal ($n = 5$), and caudal ($n = 1$) in preoperative MRI. Lipoma excision was near total in 21 (70%) cases and subtotal in nine (30%) cases. Postoperative complications were as follows: cerebrospinal fluid (CSF) leakage occurred in five (16.6%) cases and managed by resuturing of the wound, whereas wound infection was noted in four (13.3%) cases and managed by debridement and proper antibiotics (Table 1).

Early outcome were as follows: two (6.6%) cases acquired a new but transient onset motor weakness. Of the 16 patients who experienced preoperative motor weakness, 13 (81.3%) patients improved, whereas the preoperative motor weakness got worse in the other three (18.7%) cases. Incomplete bladder dysfunction was regained in nine (64.3%) patients, whereas only one (7.1%) patient developed worsening (of 14 patients) (Table 2).

Table 1. Patients, clinical presentations, preoperative MRI, surgical procedure, and postoperative complications.

Patients' characteristics	n (%)
Sex	
Male	12 (40.0)
Female	18 (60)
Age group (years)	
0–1	14 (46.7)
1–2	16 (53.3)
Clinical presentation	
Low back swelling	30 (100)
Motor dysfunction	16 (53.3)
Bladder dysfunction	
Complete	5 (26.3)
Incomplete	14 (73.7)
Preoperative MRI	
Type of lipoma	
Transitional	24 (80)
Dorsal	5 (16.7)
Caudal	1 (3.3)
Surgical procedure	
Near total excision	20 (66.7)
Partial excision	10 (33.3)
Postoperative complications	
CSF leak	5 (16.6)
Wound infection	4 (13.3)

CSF, cerebrospinal fluid.

Long-term outcomes were as follows: of the 16 patients who presented with preoperative motor weakness, 11 (68.7%) patients had improved, whereas five (31.3%) patients developed worsening ($P < 0.001$). Concerning the bladder function, nine (81.9%) of the 11 patients with normal preoperative bladder function maintained normal, whereas worsening of bladder function was noted in two (18.1%) patients. All the patients with preoperative complete bladder dysfunction remained on clean intermittent catheterization (CIC), whereas in 13 (92.9%) of 14 patients with incomplete bladder dysfunction, normal bladder function was regained and only one case developed worsening ($P < 0.001$), that is, the untethering surgery resulted in significant improvement in the preoperative neurological deficits (motor and urological), as P values less than 0.001 (Table 2).

On long-term follow-up of our patients, five (16.7%) patients developed postoperative neurological deteriorations, considered to develop retethering. All the deteriorated cases underwent subtotal excision of spinal lipomas. The mean time interval from appearance of first symptom and surgery in those who improved was 10 months (range, 4–18 months), whereas in those who did not improve was 16 months (range, 10–24 months), with P value less than 0.001, indicating the beneficial role of early surgery.

5. Discussion

Spinal lipoma is defined as a soft subcutaneous swelling in the lumbosacral region, which presents at birth and results in spinal cord tethering that could lead to variable and progressive neurologic deficits (motor and sphincter dysfunction). It proportionally increases in size with the body growth in general (Pang, 2019).

As with other types of occult spinal dysraphism, early surgical intervention is required for untethering of the spinal cord and guided by IONM. The surgery aims to relieve the mechanical traction of the lipoma on the conus. However, near-total resection of spinal lipomas in young age is a significant predictor for good long-term outcomes, and the presence of preoperatively motor or sphincter dysfunctions has only a small chance to improve with surgery (Charalampidis et al., 2020).

The performing of routine postoperative MRI to evaluate the residual lipomatous tissue is controversial. Although it is recommended by some authors (Pang et al., 2013; Talamonti et al., 2014), others

Table 2. Early and long-term postoperative neurological outcomes.

	Outcomes		Same [*n* (%)]	Worse [*n* (%)]	Improved [*n* (%)]
Preoperative motor weakness	Early ($n = 16$)		3 (18.7)	–	13 (81.3)
	Long-term ($n = 16$)		–	5 (31.3)	11 (68.7)[a]
Preoperative bladder function	Early ($n = 30$)	Normal	11 (100)	–	–
		Incomplete	4 (28.6)	1 (7.1)	9 (64.3)
		Complete	5 (100)	–	–
	Long-term ($n = 30$)	Normal	9 (81.9)	2 (18.1)	–
		Incomplete	–	1 (7.1)	13 (92.9)[a]
		Complete	5 (100)	–	–

[a] *P* value less than 0.001.

suggest that decision to interfere is not only based on the presence of residual lipoma on an MR but also on clinical presentations (Collmann, 2011; Halevi et al., 2011).

Consequently, the present study aimed to prospectively evaluate the neurological outcomes after near-total and subtotal surgical resection of spinal lipomas.

Regarding patients' criteria and initial presentation, our study included 30 patients with lumbosacral spinal lipomas (12 boys and 18 girls) with age less than 2 years at the time of surgery. All the 30 cases presented with low back swelling. A total of 19 (63.3%) patients presented with bladder dysfunction, of whom 14 (73.7%) had incomplete dysfunction, whereas five (26.3%) had complete dysfunction. In addition, 16 (53.3%) cases had motor weakness. All 30 cases had no previous surgery for spinal lipoma. Our patients' criteria coped with the patients' criteria of the trial by Manish et al. (2007), in which cutaneous manifestations were found in 50%, bladder dysfunction in 38%, and motor deficits in 54%.

In preoperative MRI, spinal lipomas were transitional ($n = 24$), dorsal ($n = 5$), and caudal ($n = 1$). Wykes et al. (2012) conducted a retrospective study on a cohort of children with asymptomatic lipomas of the conus and reported that preoperative MRI revealed a transitional, caudal, and dorsal lipoma in 69, 19, and 12% of patients respectively. This was comparable to our results.

In the present study, excision of lipoma was near total in 21 (70%) cases and subtotal in nine (30%) cases. However, other studies reported higher rates of total (90%) and near-total excision (100%) (Pang, 2015; Pang et al., 2009, 2010). In contrary, the subtotal excision was the traditional surgical technique of the spinal lipomas in the historic views (Arai et al., 1992; Byrne et al., 1995; La Marca et al., 1997).

Regarding the postoperative complications, CSF leak occurred in five (16.6%) cases, and wound infection was noted in four (13.3%) cases. This is in agreement with the reported rates of other multiple series (Wykes et al., 2012; Cochrane et al., 2000; Xenos et al., 2000). On the contrary, lower rates of CSF leakage and wound complications were reported in other published series (Pang, 2015, 2019; Manish et al., 2007; Pang et al., 2010).

Regarding the early outcome of the present study, two (6.6%) cases acquired a new, although, transient motor deficit. Of the 16 patients who experienced preoperative motor weakness, 13 (81.3%) patients improved, whereas the preoperative motor weakness got worse in the other three (18.7%) cases. Incomplete bladder dysfunction was regained in nine (64.3%) patients, whereas only one (7.1%) patient progressed to have complete dysfunction (out of 14 patients). None of the patients with complete bladder dysfunction showed improvement. Likewise, other studies reported the incidence of a new neurological deficit of 4.2 and 5.2% after total and subtotal excision, respectively (Pang, 2019; Pang et al., 2010). This also coped with the reported rates of symptomatic improvement or stabilization in several published series (94, 83, 80, and 70%) (Pang et al., 2010; Cochrane et al., 2000; Xenos et al., 2000; Kulkarni et al., 2004). On the contrary, others reported rare or even partial early neurological improvement in about 20–30% of the patients only (Pang, 2020; Manish et al., 2007; Pang et al., 2009, 2010).

Regarding the long-term outcomes, of the 16 patients who presented with preoperative motor weakness, 11 (68.7%) patients had improved, whereas five (31.3%) patients developed worsening ($P < 0.001$). Concerning the bladder function, nine (81.9%) of the 11 patients with normal preoperative bladder function maintained normal, whereas worsening of bladder function was noted in two (18.1%) patients. All the patients with pre-operative complete bladder dysfunction remained on CIC, whereas in 13 (92.9%) of 14 patients with incomplete bladder dysfunction, normal bladder function was regained and only one case developed worsening

($P < 0.001$), that is, the untethering surgery resulted in significant improvement in the preoperative neurological deficits (motor and urological), as P values less than 0.001. Other published series reported ranges between 3 and 30% of delayed neurological (motor and bladder) worsening on long-term follow up, which was comparable with us (Pang et al., 2013; Andrew et al., 2009; Lee et al., 2020). Moreover, other authors stated comparable rates of improvement in urological function after surgical cord untethering (Kim et al., 2022; Morizawa et al., 2022).

On the contrary, others reported no neurological worsening throughout the follow up period (Manish et al., 2007; Seki et al., 2018).

On long-term follow-up of our patients, five (16.7%) patients developed postoperative neurological deteriorations, considered to develop retethering. All the deteriorated cases underwent subtotal excision of spinal lipomas. The mean time interval from appearance of first symptom and surgery in those who improved was 10 months (range, 4–18 months), whereas in those who did not improve was 16 months (range, 10–24 mo;nths), with P value less than 0.001, indicating the beneficial role of early surgery.

Regarding the retethering rate, our study agreed with rates published in several series from 3 to 30% or more, especially with subtotal compared with near-total excision (Pang, 2015, 2019; Tarang et al., 2021; Pang et al., 2009, 2010, 2013; Wykes et al., 2012; Cochrane et al., 2000; Andrew et al., 2009; Lee et al., 2020; Dorward et al., 2002).

Similarly, several authors encouraged early and complete surgical interference even if the patient was asymptomatic before the development of established neurological damage (Tarang et al., 2021; Pang, 2020; Manish et al., 2007; Xenos et al., 2000; Kim et al., 2022; Hayashi et al., 2020).

5.1. Recommendations

We recommend other larger studies to confirm the results of the present study.

5.2. Conclusion

Better long-term neurological outcomes could be achieved after near-total than subtotal resection of spinal lipomas. Early surgical resection of spinal lipomas has a beneficial role in the outcome.

Conflict of interest

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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