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Occult dysraphism in adulthood: clinical course and management

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Abstract We present a series of 23 patients with dysraphic malformations and adult onset of symptoms (4 meningoceles, 19 spinal hamartomas). Mean age at presentation was 39±21 years (range 23–67 years). Patients were followed up for a mean period of 19 months (range 0.5–68 months). Only patients with progressive neurological disease were operated on (3 meningoceles and 16 spinal hamartomas). The remaining patients were treated conservatively and continue to be observed clinically. Two of three patients operated for meningoceles improved without recurrence of symptoms. Patients with spinal hamartomas could be divided into two groups according to their main symptom: paraparesis (group A, $n=8$) or pain (group B, $n=11$). Malformations in group B were typically associated with a tethered cord and tended to be more complex than in group A. The majority of patients in group A showed better long-term results than patients in group B, due to their considerably lower rate of recurrence.

Key words Tethered cord
Diastematomyelia · Lipoma
Dermoid cyst · Meningocele
Dysraphic malformation
Syringomyelia

Introduction

The natural history of spinal dysraphism has not been determined precisely. According to many authors about one-half of patients will eventually become symptomatic due to interference with spinal cord blood flow and metabolism [16, 46, 48, 52]. This leaves the other half of them without neurological problems, and for these surgery would not offer any benefit. It follows that the indication for and timing of surgical procedures in children with dysraphic malformations is a very controversial issue. While several authors recommend prophylactic surgery [31, 43, 53] others perform surgery as soon as neurological progression occurs [21, 32, 33, 48].

The majority of studies on dysraphic malformations in adults have been case reports or reports on small series [3, 7, 9–11, 14, 23, 26–28, 34, 37, 40–42, 49–52]. Only six papers describe between 9 and 23 adult patients with spinal dysraphism [19, 22, 25, 30, 35, 53].

We have undertaken a retrospectice analysis of dysraphic malformations seen in our department during the period from 1977 to 1992. We wanted to examine at what age these patients with congenital anomalies became symptomatic, to study their clinical course before and after operation in order to optimize surgical procedures for this patient group, and to extrapolate from this whether guidelines for the treatment of children can be deduced.

Patients and methods

A total of 23 patients with onset in adult life of symptoms of occult dysraphic malformations were identified. Case records, operation

Table 1 Clinical grading system

Score	Sensory deficits, pain, dysesthesias	Paresis	Gait	Bladder function	Bowel function
5	No symptom	Full power	Normal	Normal	Normal
4	Present, not significant	Movement against resistance	Unsteady, no aid	Slight impairment, no catheter	Slight impairment, full control
3	Significant, function not restricted	Movement against gravity	Mobile with aid	Residual function, no catheter	Laxatives, full control
2	Some restriction of function	Movement without gravity	Few steps with aid	Catheter sometimes	Loss of control sometimes
1	Severe restriction of function	Contraction without movement	Standing with aid	Catheter often	Loss of control often
0	Incapacitation of function	Paraplegia	Wheel-chair	Permanent catheter	No control

Table 2 Summary of patients with adult dysraphism

Type	*n*	Sex ratio M/F	Age (years)	Length of history (months)	Follow-up (months)
Meningocele	4	2/2	40±10	9±13	49±25
Dysraphic hamartoma	19	8/11	39±14	101±112	14±17
Pain (group B)	11	4/7	41±14	116±113	14±19
Paresis (group A)	8	4/4	37±15	81±111	15±15
Total	23	10/13	39±13	90±108	19±21

Table 3 Patients with meningoceles

Sex	Age (years)	Spinal level	Meningocele type	Comments
F	30	T2–3	Lateral	Neurofibromatosis
M	50	S1–5	Dorsal	Tethered cord
M	45	L1–2	Lateral	
F	33	S1–4	Ventral	No operation, neurofibromatosis

notes, and radiographs were evaluated and follow-up examinations were performed. We encountered two forms of dysraphic malformation: meningoceles and dysraphic hamartomas. The latter consisted of either lipomatous or dermal hamartomas [33] and one neurenteric cyst.

Only patients with progressive neurological disease were operated on. Surgery consisted of a laminectomy, tumor removal or closure of the communication between spinal canal and meningocele, and dissection of arachnoid adhesions if present. The filum terminale was not cut in every instance, due either to inexperience of the surgeon or severe arachnoid adhesions.

For monitoring of the postoperative course a grading system was used for each individual symptom: gait ataxia, motor weakness, sensory deficits, dysesthesias, pain, and bladder and bowel function (Table 1) (J. Klekamp, M. Samii, in preparation). Scores between 3 and 5 indicate satisfactory to normal function, whereas scores of 2 to 0 describe unsatisfactory to incapacitated functional levels. Only the worst affected limb was analyzed in this manner. Patients were also graded according to the Karnofsky scale [18].

For testing of statistical significance, Student's *t*-test was employed. The rates of recurrence were determined by Kaplan-Meier analysis [17]. A recurrence was defined as re-onset of neurological progression. Surgical morbidity was defined as a new, permanent postoperative deficit or a permanent aggravation of a pre-existing deficit. For Kaplan-Meier analysis cases with surgical morbidity were classified as a recurrence on the day of surgery.

Results

Of 23 patients presenting at our hospital, 19 showed signs of neurological progression and were operated on. The remaining 4 patients did not show neurological progression and continue to be observed clinically. Table 2 gives an overview of patient sex distribution, mean age, length of history, and follow-up.

Three of four patients with meningoceles had neurological progression and were operated on. Two of them presented with radicular pain and improved postoperatively without subsequent recurrence. One patient with a sacral meningocele presented with bladder dysfunction which progressed despite surgery. Apart from bladder function every other symptom either improved or remained unaffected. The Karnofsky score increased postoperatively (Tables 3, 5).

Of 19 patients with spinal hamartomas, 16 were operated on. According to their main symptom they could be divided into two groups: group A, with paraparesis as the main symptom ($n=8$), and group B, in whom the main symptom was pain ($n=11$). In both groups the ratio between intra- and extramedullary tumors was 2:1. Table 4 gives an overview on type of operation performed, presence of a tethered cord, arachnoid adhesions, sy-

Table 4 Patients with dysraphic tumors. (*PR* Partial removal, *TR* total removal, *c* filum cut, *nc* filum not cut)

Hamartoma type	Spinal level	Sex	Age (years)	Surgery		Tethered cord	Arachnoid adhesion	Syrinx	Diastemato-myelia	Dermal sinus	Spina bifida
				Type	no.						
Pain group (B)											
Dermoid	T12–L2	F	39	PR	2	+ nc	+	–	+	+	+
Dermoid	L3–5	F	61	PR	2	c	+	–	–	–	+
Dermoid	T10–L3	F	42	PR	2	–	+	–	–	–	–
Dermoid	L5–S1	F	25	PR	1	+ nc	–	–	+	–	+
Dermoid	L3–5	M	44	–		–	+	–	–	–	+
Dermoid	L1–2	F	42	PR	1	+ c	–	–	–	–	–
Lipoma	S3	F	37	PR	1	+ nc	–	+	–	–	–
Lipoma	S2–5	M	23	PR	1	+ nc	–	–	–	+	+
Lipoma	L1–3	M	63	–		+	+	–	–	–	+
Lipoma	L2–3	F	31	TR	1	+ c	+	–	–	–	–
Lipoma	L1–4	M	24	–		+	–	+	+	–	+
Paresis group (A)											
Dermoid	T12–L1	M	30	PR	3	–	+	+	–	–	–
Dermoid	T11–12	M	29	PR	1	–	–	–	–	–	–
Dermoid	T12–L1	M	19	TR	1	–	–	–	–	–	–
Dermoid	T3–5	F	32	PR	2	–	+	–	–	–	+
Lipoma	L3–5	F	67	PR	1	+ c	–	+	+	–	+
Lipoma	L3–5	M	44	PR	1	–	+	–	–	–	–
Lipoma	L2–3	F	44	PR	3	–	+	+	–	–	–
Neuroenteric cyst	C1–2	F	29	TR	1	–	–	+	–	+	–

ringomyelia, diastematomyelia, dermal sinus, or spina bifida occulta. In general, dysraphic malformations were more complex in group B. These patients were more likely to have an additional dermal sinus or diastematomyelia. The most prominent difference was the higher proportion of patients with a tethered cord and spina bifida occulta compared to group A. Furthermore, the spinal level of the malformation tended to be lower. Syringomyelia and arachnoid adhesions were seen in equal proportions (Figs. 1–4).

We observed a postoperative cerebrospinal fluid leak in two cases, which in one case responded to a lumbar drain and in one necessitated reoperation. No other complications were encountered.

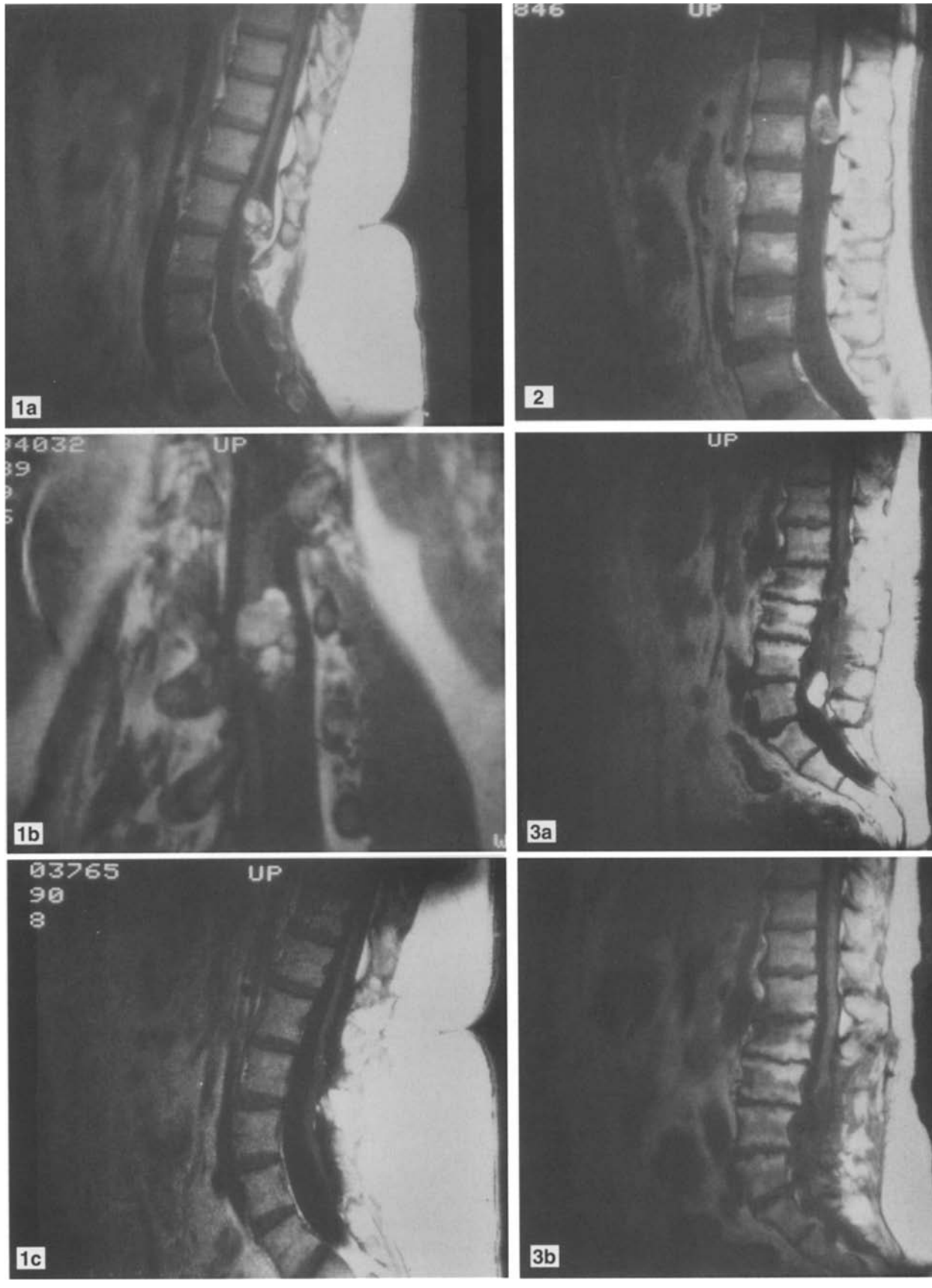
Analysis of the postoperative course revealed that the majority of patients in group A benefited from the operation. Dysesthesias, motor weakness, and gait ataxia improved, while sensory deficits, pain, and impairment of bladder and bowel function slightly increased. Overall, the Karnofsky score in group A remained unchanged (Table 5). In group B pain improved postoperatively but tended to increase again after 6 months, so that most of the benefit had been lost again at the end of 1 year. Similarly, we observed a tendency to deterioration of sensory deficits, dysesthesias, and motor weakness starting 6–12 months after surgery, so that the initial benefit from operation was lost after 1 year. Gait ataxia and bladder and bowel function continued to deteriorate despite surgery. In this group the Karnofsky score decreased postoperatively (Table 5).

Kaplan-Meier analysis revealed a higher rate of recurrence in group B patients than in group A. We observed no surgical morbidity in group A, as against 9% in group B. The subsequent rates of recurrence were 25% and 47%, respectively. No recurrences were observed more than 12 months postoperatively (Fig. 5).

Fig. 1a–c A 39-year-old woman with a 4-year history of sacral pain. Magnetic resonance imaging (MRI) identified a dermoid cyst and a lipoma from T12 to L2, diastematomyelia, a tethered cord, a dermal sinus, arachnoid adhesions, and spina bifida occulta (**a, b**). At operation, dissection of nerve roots and tumor were extremely difficult due to arachnoid adhesions. Part of the lipoma could be resected. The filum terminale was not cut. Postoperatively, pain improved for about 6 months and then became progressively worse again. The postoperative scan (**c**) demonstrates arachnoid scarring at the site of surgery. At present, the patient is in a stable neurological condition

Fig. 2 A 19-year-old male patient presenting with a 1-year history of progressive paraparesis. MRI disclosed a dermoid cyst from T12 to L1. After complete removal of the cyst the paraparesis improved

Fig. 3a, b A 61-year-old woman presenting with a 5-year history of low back pain and a moderate degree of motor weakness. MRI demonstrated a dermoid cyst from L3 to L5, a tethered cord, arachnoid adhesions, and a spina bifida occulta (**a**). Despite partial removal of the hamartoma and section of the filum terminale, pain reappeared after 6 months. The postoperative MRI scan showed retethering of the cord (**b**). Please note the severe degree of degenerative changes of the lumbar spine


1a
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1b
3a
1c
3b

Discussion

We have undertaken a retrospective analysis of patients with occult dysraphism presenting in adulthood at our institution from 1977 to 1992. A total of 23 patients were reviewed, of whom 19 were operated on for progressive neurological symptoms. For a detailed description of neuroradiological findings in spinal dysraphism we refer to reviews by Scatliff et al. [38], Szalay et al. [44], and Tortori-Donati et al. [47]. Not every patient experienced a long-term benefit from surgery.

Meningoceles

Two of four patients with meningoceles presented because of pain which followed a root distribution. They were operated on with a good response and no recurrence. One patient presented with bladder dysfunction which continued to deteriorate postoperatively (Table 5). Similar results have been reported in the literature [1, 6, 24, 36]. In the same way as noted by previous authors [4, 5, 12, 20], two of our patients had associated neurofibromatosis.

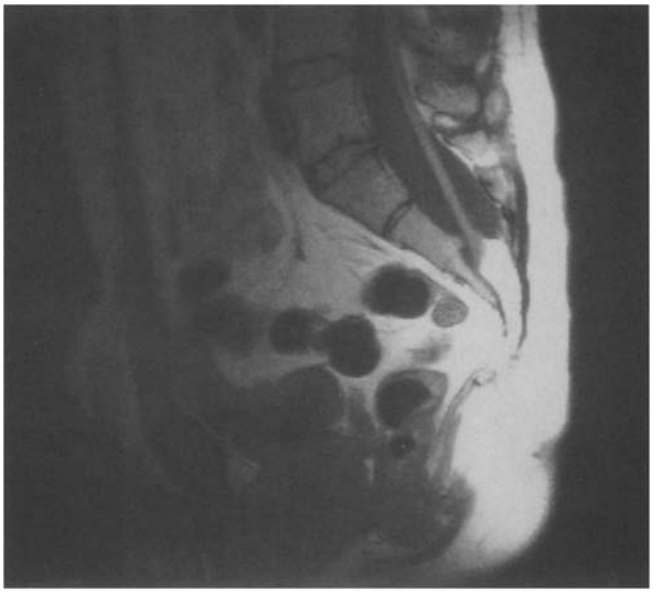

Fig. 4 A 23-year-old man presenting with a 2-year history of sacral pain. MRI disclosed a lipoma from S2 to S5, a tethered cord, a dermal sinus, and a bony defect of the sacrum. After partial removal of the lipoma without section of the filum terminale, pain improved. The patient is free of symptoms 5 years postoperatively and the father of two children

Hamartomas

Among the patients with occult dysraphism associated with a spinal hamartoma two groups could be distinguished according to their main complaint: paraparesis (group A) or pain (group B). A number of differences between these two groups were noted. Patients in group A tended to be slightly younger, and the malformation was at a higher spinal level and less complex (Fig. 2). The difference was most striking in regard to the presence of spina bifida occulta and tethered cord (Figs. 1, 3). The incidences of associated syringomyelia or arachnoid adhesions were similar (Table 4).

Results of treatment were better in group A than in group B: while stabilization of disease was achieved in the former, with even a slight increase in Karnofsky score during the 1st postoperative year, this was found in the latter group only in regard to pain. However, all symptoms – pain in particular – demonstrated a tendency to deteriorate after about 6 months (Table 5). Kaplan-Meier analysis revealed a recurrence rate of 25% in group A and 47% in group B, with surgical morbidity rates of 0% and 9%, respectively (Fig. 5).

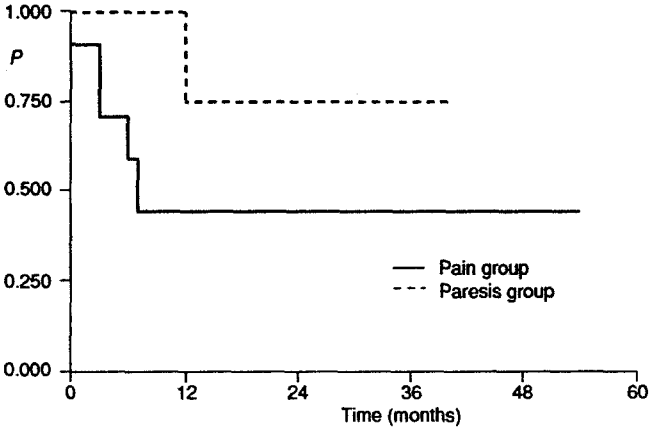

Fig. 5 Recurrence of neurological deterioration for adult spinal hamartomas: Kaplan-Meier analysis of patients with dysraphic hamartomas

These results are somewhat poorer than those reported in the literature. Lesoin et al. [22] and Kondo et al. [19] concentrated on urological problems and reported improvement in 7 of 8 and 7 of 15 operated patients, respectively. Kondo et al. assessed sphincter function with urodynamic testing and followed their patients for 3–54 months (mean 20 months), whereas Lesoin et al. do not state how sphincter function was evaluated or the length of follow-up. Similar to us, Kondo et al. commented that a satisfactory result was only achieved in adults without complex malformations.

Table 5 Postoperative course of patients[a] with dysraphic malformations: mean clinical scores for function

Patient group	*n*	Before operation	At discharge	After 3 months	After 6 months	After 1 year
Sensory deficit						
Meningocele	4	4.3±1.2	4.0±1.0	4.3±0.6	4.3±0.6	5.0±0.0
Hamartoma						
Pain	6	4.3±1.0	3.8±1.6	4.0±1.3	4.5±0.8	4.3±0.8
Paresis	5	3.6±1.3	3.8±1.3	3.8±1.3	3.8±1.3	3.4±1.1
Dysesthesias						
Meningocele	4	4.7±0.7	5.0±0.0	5.0±0.0	5.0±0.0	5.0±0.0
Hamartoma						
Pain	6	3.8±1.3	4.5±0.5	4.2±1.2	4.3±1.2	3.8±1.2
Paresis	5	4.1±1.1	4.1±1.1	4.4±0.9	4.4±0.9	4.4±0.9
Pain						
Meningocele	4	3.3±0.6	3.3±0.6	3.7±0.6	4.3±0.6	4.7±0.6
Hamartoma						
Pain	6	2.5±0.5	3.0±0.9	3.3±1.2	3.2±1.2	2.7±1.5
Paresis	5	5.0±0.0	4.4±1.3	4.6±0.9	4.6±0.9	4.6±0.9
Motor weakness						
Meningocele	4	5.0±0.0	4.7±0.7	5.0±0.0	5.0±0.0	5.0±0.0
Hamartoma						
Pain	6	3.8±0.8	3.8±0.8	4.3±0.5	4.2±0.8	3.8±0.4
Paresis	5	1.8±1.1	2.2±1.3	2.0±1.0	2.0±1.0	2.2±1.1
Gait ataxia						
Meningocele	4	5.0±0.0	5.0±0.0	5.0±0.0	5.0±0.0	5.0±0.0
Hamartoma						
Pain	6	4.5±0.5	4.3±0.8	4.3±0.8	4.3±0.8	4.0±1.1
Paresis	5	2.4±1.8	2.4±1.8	2.6±1.9	2.6±1.9	2.6±1.9
Bladder function						
Meningocele	4	4.3±1.2	4.0±1.7	4.0±1.7	4.0±1.7	4.0±1.7
Hamartoma						
Pain	6	3.5±0.8	2.2±1.3	3.7±1.0	3.5±0.5	3.3±0.5
Paresis	5	4.4±1.3	4.2±1.1	4.2±1.1	4.2±1.1	4.2±1.1
Bowel function						
Meningocele	4	5.0±0.0	5.0±0.0	5.0±0.0	5.0±0.0	5.0±0.0
Hamartoma						
Pain	6	4.0±0.9	2.7±1.5	3.7±1.0	3.8±0.8	3.7±0.8
Paresis	5	4.6±0.9	4.2±1.1	4.4±0.9	4.4±0.9	4.4±0.9
Karnofsky score						
Meningocele	4	83± 6	83± 6	87± 6	87± 6	90±10
Hamartoma						
Pain	6	76± 6	72±12	77±10	75±11	72±17
Paresis	5	64±22	58±24	64±27	64±27	64±27

[a] Only cases with a complete 1-year follow-up are included

Pang and Wilberger [30] and Zumkeller et al. [53] reported improvements in regard to pain, motor weakness and stabilization of bladder function in the great majority of cases. However, exact data on the length of follow-up were not given. Also, in a number of case reports, improvements of preoperative symptoms were described – for pain in particular – but follow-up information covering at least 12 months postoperatively was not available [3, 11, 37]. Several authors [2, 7, 9, 40] have emphasized the importance of arachnoid changes as a prognostic factor in regard to stabilization of the neurological status of their patients. Similarly, reports about children with spinal dysraphism and follow-up information over several years state that stabilization of the disease process was achieved after variable degrees of early but transient postoperative improvement [15, 21, 31, 39, 43, 53]. Hogen Esch et al. [15] described 22 patients with a tethered cord syndrome after surgery for spina bifida aperta with a

follow-up of 6 months to 8 years. The great majority of patients presented with motor weakness, whereas less than half complained about pain and only 14% demonstrated bladder dysfunction. Bladder function and pain remained unchanged in the majority of patients, whereas motor function tended to improve. Schmidt et al. [39] published a series of 46 patients between 7 months and 18 years of age with a mean follow-up of 7.3 years. Twelve patients had a tethered cord. Surgery led to stabilization of neurological symptoms but failed to achieve long-lasting improvements. Stolke et al. [43] described 26 children between 1 month and 12 years of age. The majority presented with skin lesions, foot deformities, and sphincter disturbances. They reported improvement in 7 and stabilization in the remaining 19 children but did not give the length of follow-up. They recommended prophylactic surgery. Similarly, Zumkeller et al. [53] reported on 12 cases with improvement of pain in all 6 children affected, improvement of bladder dysfunction in 6 of 8 patients affected, improvement of motor weakness in 5 of 7 affected, and improvement of sensory deficits in 5 of 10, but did not state the length of follow-up.

Peter [31] described 88 children less than 12 years of age with occult dysraphism. He observed an increasing frequency of neurological symptoms attributable to dysraphism with increasing age. Only three children showed postoperative improvement, the rest of the children remaining unchanged. Peter recommended prophylactic surgery. On the other hand, Lagae et al. [21] compared 20 children with release of the tethered cord to 21 children with known tethered cord without surgery. The children were followed for between 8 months and 13 years. Surgery achieved stabilization of neurological signs and symptoms but did not improve deficits. Three children who underwent operation showed radiological evidence of retethering. In the nonsurgical group only two children showed neurological progression.

Pain and arachnoid scarring

In our study, every recurrence was marked by reappearance of back pain with radiation into sacral dermatomes. The pain was reported to be very severe, even worse than preoperatively. This is indicated by an average pain score of 2.7 after 1 year, with a tendency to further progression. Magnetic resonance scans of these patients did not disclose regrowth of the partially removed hamartoma, but showed extensive arachnoid scars at the level of operation. In general, our observation was that spinal hamartomas are associated with more severe postoperative arachnoid scarring than other spinal processes (Figs. 1, 3). The aggravation of arachnoiditis has to be regarded as the major cause of the reappearance of pain.

Pain and tethered cord

All four patients with postoperative deterioration had a tethered cord. The filum terminale had been cut only in two patients, with a retethering of the cord due to arachnoid scars in both (Fig. 4). Among different authors there is general agreement that the filum terminale should be cut if a tethered cord is present. However, the rate of retethering due to postoperative arachnoid scars is generally believed to be of the order of at least two-thirds of the patients [8, 13, 45]. Furthermore, several reports mention that arachnoid adhesions may render surgery extremely difficult in adults [11, 45], raising the probability of retethering even further. In our study, every patient with a recurrence also had a spina bifida occulta. Orthopedic problems due to the bony anomaly might have contributed to the recurrence of pain [41] (Fig. 3). Nevertheless, we would recommend untethering the spinal cord and cutting the filum terminale whenever a tethered cord is present and this can be done safely.

Differences between children and adults

In comparison to children, the adult patient population shows a number of differences. In general, adults present with pain more often and demonstrate bladder dysfunction less often than children [30, 35, 52]. For children, bladder function, motor weakness, and scoliosis are the most prominent symptoms of dysraphic malformations [32, 53]. In adults the spinal level tends to be higher – thoracic and cervical – and degenerative changes of the lumbar spine are encountered as well. These factors may explain the higher proportion of adult patients who complain of pain as compared to children.

Clinical observation vs surgery

There was no surgical mortality in this series. Surgical morbidity was low. In terms of complications, cerebrospinal fluid leak occurred in two cases one of which required operation while the other responded to a lumbar drain. One patient in group B suffered postoperative deterioration without regaining the preoperative level of function. This leaves exacerbation of spinal arachnoiditis as the major risk of surgery for spinal hamartomas in adults.

Our retrospective analysis has shown that a considerable number of patients with occult dysraphism reach adulthood or even old age before becoming symptomatic. This implies that not every dysraphic process inevitably leads to progressive neurological problems. Oi et al. [29] have shown that patients without a tethered cord are not at significant risk of neurological deterioration. In a com-

parative study, Lagae et al. [21] observed clinical progression in only 2 out of 21 children with a tethered cord who did not undergo surgery. In the operated group of 20 children, disease progression was stopped, with 3 children subsequently developing signs of retethering.

We consider delay of surgery in children to be justified if no progression of symptoms occurs – particularly in patients without a tethered cord. The risk of causing a neurological deficit or exacerbation of arachnoiditis should be weighed carefully when prophylactic surgery is considered.

Adults and children should be operated on as soon as neurological symptoms appear or progress (for those presenting to the surgeon the first time with established deficits), because in the majority of cases only stabilization of the disease is achieved. To prevent postoperative neurological deterioration in adults we suggest removal of the associated mass, dissection of arachnoid adhesions as far as can be done safely, section of the filum terminale in patients with a tethered cord, and decompression of the spinal canal by laminectomy and duraplasty if appropriate.

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