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Original Article

Adult Versus Pediatric Tethered Cord Syndrome: Clinicoradiological Differences and its Management

Abstract

Background: Dysraphic lesions in adults, presenting clinically as tethered cord syndrome (TCS), are relatively rare, and their optimal management remains controversial. **Patients and Methods:** We performed a retrospective analysis of our pediatric database over a period of last 7 years to focus on the adult TCS. Our aim was to determine the clinicoradiological and etiopathological differences between adult and pediatric patients as well as to determine the results of surgery in adult TCS. **Results:** Adult spinal dysraphisms constituted 15.4% of our patients (20 out of 130). Motor weakness, sphincteric dysfunction, and backache (n = 13, 65.0% each) predominated in adults unlike children who presented with subcutaneous swellings (n = 74, 67.6%) followed by motor weakness (n = 40, 46.4%), backache being reported by only three patients. The different pathologic substrates underlying adult dysraphisms were lipomeningocele (n = 8), split cord malformation (total = 7; Type 1: n = 5; Type 2: n = 2), dermal sinus (n = 2), and fatty filum (n = 3). On the other hand, meningomyelocele/meningocele (61, 54.9%) followed by split cord malformation Type 1 and 2 (n = 29, 26.1%) predominated in children. The radiological differences between the two groups were a higher incidence of vertebral body defects (hemivertebrae and butterfly vertebrae) and lack of intracranial anomalies in adults. At a mean follow-up of 20.5 months, the most common symptoms to improve following detethering were pain (11 out of 13, 84.6%) followed by motor weakness (six out of 13, 56.2%) and sphincteric control (7 out of 13, 53.8%). **Conclusion:** Most common symptoms to improve following detethering in adult TCS were pain followed by motor weakness. The major radiological differences between these two groups were a higher incidence of vertebral body defects (hemivertebrae and butterfly vertebrae) and lack of intracranial anomalies in adults.

Keywords: *Adult spinal dysraphism, lipomyelomeningocele, split cord malformation, tethered cord syndrome*

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Introduction

Tethered cord syndrome (TCS) is basically a condition where anchorage of a part of the spinal cord, most commonly the area of the conus, by some inelastic structure, which results in functional dysfunction involving the adhered segment of the spinal cord or at times segments beyond that.[1] This entity was first described by Garceau (1953) and subsequently named by Hoffman *et al*. (1976) as "tethered spinal cord."[2] Yamada *et al*. broadened the condition further in 1981.[3] Myelomeningocele, lipoma, lipomyelomeningocele, diastematomyelia, meningocele manque', and dermoid sinus are the "usual suspects" when such a clinical condition surfaces. TCS remains a well-defined entity in children and there is hardly any controversy in their surgical treatment, which involves elective excision of the underlying cause and detethering of the cord. However, no such clear-cut policy exists for the adult patients with TCS. A relative rarity of this condition and a lack of large number of studies have resulted in a lack of clarity as far as the different clinical and management aspects of adults TCS is concerned. In this study, we tried to analyze the clinicoradiological and etiopathological features of adult TCS comparing them with our pediatric database and also analyzed their outcome to surgical intervention. We also tried to inquire into the reasons for the delay in seeking treatment which results in them seeking treatment at adulthood.

Patients and Methods

We retrospectively reviewed medical records as well as radiographic and operative details of 130 consecutive patients operated primarily for spinal dysraphism

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at our center between 2007 and 2013. It yielded twenty adult patients (≥16 years and) and 110 pediatric patients (<16 years). Patients operated in childhood and presenting with retethering in adulthood were excluded. Patients without proper records and follow-up details were also not considered for analysis. A set of questionnaire was given to all patients [Tables 1 and 2] to determine the reasons for the delay in seeking surgical treatment and also to find out if there were any precipitating factors. These patients were evaluated using magnetic resonance imaging (MRI) of the spinal cord with cranial screening, as well as computed tomography scans, if any bony anomaly was suspected on MRI. Follow-up data were obtained from outpatient department visits/telephonic conversations/postal communication. Data were analyzed using the SPSS Statistics for Windows, Version 17.0. Chicago.

Table 1: The questionnaire given to patients to find out reasons for delay

Q. no	Question
1	Are the symptoms/signs present since birth?
2	Did parents consult a clinician before presenting to us?
3	If yes, what was the reason for not undergoing surgery?

Table 2: The precipitating factors leading to production or exacerbation of symptoms in adult tethered cord syndrome in our series

Q. no	Precipitating cause
1	Very active lifestyle/yoga/exercise
2	Prolonged sitting/forward bending
3	Pregnancy/childbirth/Sexual intercourse
4	Trauma

Results

Of the total 130 patients who met the inclusion criteria, we could find 20 (15.4%) adult patients with TCS in our series. There was a male predominance in adults (M:F = 3:2). The patient's age ranged from 16 years to 56 years (mean 24 years). The mean age of the pediatric patients was 3.8 years (1 month to 16 years). Children also displayed a male predominance similar to the adults (M:F = 1.7:1). The clinical features in adults and pediatric population are depicted in Table 3.

The presenting complaints in adults included motor symptoms in 13 (65.0%), sphincteric dysfunction in 13 (65.0%), backache in 13 (65.0%) patients, 7 (35%) of whom had radiating nondermatome pain, sensory loss, or paresthesia was present in three (15.0%) patients, and swelling at back was present in eight (40.0%) patients. The orthopedic deformities (n = 6, 30%) included scoliosis and foot deformity. On the other hand, pediatric spinal dysraphism mainly presented with swelling at back (n = 74, 67.3%), followed by motor deficits (n = 40, 46.4%), sphincteric disturbances were present in 29 (18.2%) patients, orthopedic deformities were present in 18 patients (16.3%), and only three patients complained of backache (2.7%). Hence, in adults, pain, sphincteric dysfunction, motor weakness, and orthopedic deformities tend to be more common than children. The incidence of cutaneous stigmata is, however, nearly similar to the children.

From the point of etiopathological substrates underlying the tethering, in adults, there was a predominance of lipomyelomeningocele (n = 8, 40%) and split cord malformation (n = 7, 35%: Type 1: 25% and Type 2: 10%). Dermal sinus was seen in two (10.0%)

Table 3: Clinical presentation and etio-pathological distribution among adult and paediatric patients of tethered cord syndrome

	Clinical presentation	Adults patients (total, n=20)	Pediatric patients (total, n=110)
Clinical characteristics	Pain	13 (65.0%)	3 (2.7%)
	Swelling	8 (40.0%)	74 (67.3%)
	Leg weakness	13 (65.0%)	40 (46.4%)
	Sensory impairment	3 (15.0%)	20 (18.2%)
	Sphincteric dysfunction	13 (65.0%)	29 (18.2%)
	Orthopedic deformity	6 (30.0%)	18 (16.3%)
	Cutaneous stigmata	18 (90.0%)	100 (91.0%)
Etiopathogical characteristics	Lipomeningocele	8 (40%))	15 (13.5%)
	Meningocele	0	10 (9.0%)
	Split cord malformation type-1	5 (25 0.0%)	23 (20.7%)
	Split cord malformation type-2	2 (10%)	6 (5.4%)
	Dermal sinus	2 (10.0%)	1 (0.9%)
	Thickened fatty filum	3 (15%)	4 (3.6%)
	Meningomyelocele	0	51 (45.9%)

patients. Three patients had fatty filum (15%). These patients had associated lesion in the form of a lumbar arachnoid cyst and neurenteric cyst and teratoma, respectively. Whereas in pediatric patients, there was predominance of meningomyelocele (*n* = 10, 9.0%)/ myelomeningocele (*n* = 51, 45.9%) followed by split cord malformation (*n* = 29, 26.1%, SCM Type 1 and 23, 20.7% SCM Type 2). Hence, as opposed to meningomyelocele and meningocele in children, adult TCS is most commonly attributable to lipomeningocele, split cord malformation, and dermal sinus. The occult nature of the dysraphic state is one reason for not getting the desired clinical attention in these patients during their childhood.

Majority of the adult patients had the cutaneous stigmata of dysraphism (*n* = 18, 90.0%). These included tuft of hair with or without depression/dimple (*n* = 7, 35.0%), subcutaneous lipoma (*n* = 6, 30.0%), thick scar tissue like skin (*n* = 4, 20.0%), and hard bony swelling in one (5.0%) patient. These cutaneous stigmata were disregarded and ignored in childhood. Figure 1 shows the lesion shown in figure is a dermal sinus with skin puckering. The cutaneous stigmata are more common in occult dysraphisms.

On comparing radiological features, there was a predominance of occult spinal dysraphism like lipomyelomeningocele with or without subcutaneous lipoma, (*n* = 8, 40%) and split cord malformation Type 1 and 2 (*n* = 5 and 2, respectively), whereas pediatric patients had a predominance of meningocele or myelomeningocele, with or without skin defect, with protrusion of nerve roots, and cord tissue into protruding thecal sac. Major vertebral body anomalies excluding posterior arch defect were present in 9 (45.0%) adult patients, whereas only 25 (20%) pediatric patients had similar defects. Figure 2 depicts a case of lipomeningocele causing tethered cord.

The results of the cranial screening MRI revealed a striking difference between the two groups. Whereas, none of the adult patients had any abnormality, as high as 25.4% of the children (*n* = 28) had some or the other congenital anomalies of the brain. These cranial anomalies included Chiari malformation (Type 1) with hydrocephalus in 19 (17.23%) patients; aqueductal stenosis in 3 (2.72%) patients; and posterior fossa cyst, split medulla, craniosynostosis, split 4th ventricle, small posterior fossa, and porencephalic cyst was present in one patient (0.9%) each. Table 4 shows the radiological differences between adult and pediatric spinal dysraphism observed in our study.

Results of electrophysiological tests carried out in adults

Ten (50.0%) adult cooperative patients underwent electrophysiological studies (nerve conduction velocity). The findings were normal study present in six (30.0%) patients, three patients (15.0%) had common peroneal nerve involvement, and one patient (5.0%) having deep peroneal nerve involvement. The abnormal electrophysiological studies (increased latency and decreased nerve conduction velocities) correlated with motor weakness in the preoperative period and chances of improvement in postoperative were also poor in these patients.

Surgical complications

Complications of surgery were cerebrospinal fluid (CSF) leak (*n* = 3, 15%) and wound infection in 1 (5%). Two patients (10.0%) had pseudomeningoceles, while one patient (5.0%) had active CSF leakage from the wound. Barring the last patient who required lumboperitoneal shunt, all were managed conservatively.

Results of surgery in adult spinal dysraphism

Surgery was performed in all twenty patients. The goal of surgery was to excise the etiological cause (lipoma,

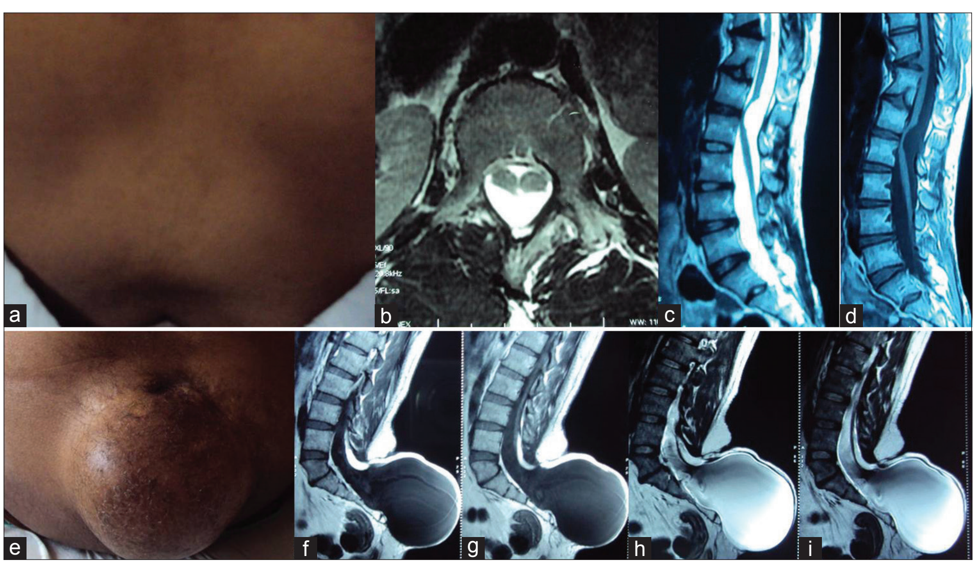

Figure 1: (a) Cutaneous stigmata of swelling and dimple in lumbar region with (b) split cord malformation Type 1 with (c and d) arachnoid cyst at L1 level. (e) Another patient with swelling at lumbosacral region with (f-i) sacral meningomyelocele with intraspinal lipoma

dermal sinus, lipomyelomeningocele, and bony spur) that was causing tethering and also associated lesions such as arachnoid and neurenteric cyst, and the filum was sectioned in all patients. The mean follow-up duration was 20.5 months (range 2 months to 7 years). Of the 13 patients with pain, 11 (84.6%) improved. Of the 13 patients with sphincteric dysfunction, 7 patients (53.8%) improved, 1 patient worsened (7.8%), and 5 of them (38.4%) had their sphincteric function unchanged. Of the 13 patients with motor complaints, 6 (56.2%) patients showed improvement of at least 1 MRC grade and in remaining 7 patients (53.8%) power remained stable after surgery. Sensory hypoesthesia improved after surgery in two (66.7%) patients and remained same in one (33.3%) patient. Among all symptoms, urinary urgency was the earliest to improve (average 2 days after surgery). Figure 3 shows the graphical presentation of the outcome to treatment.

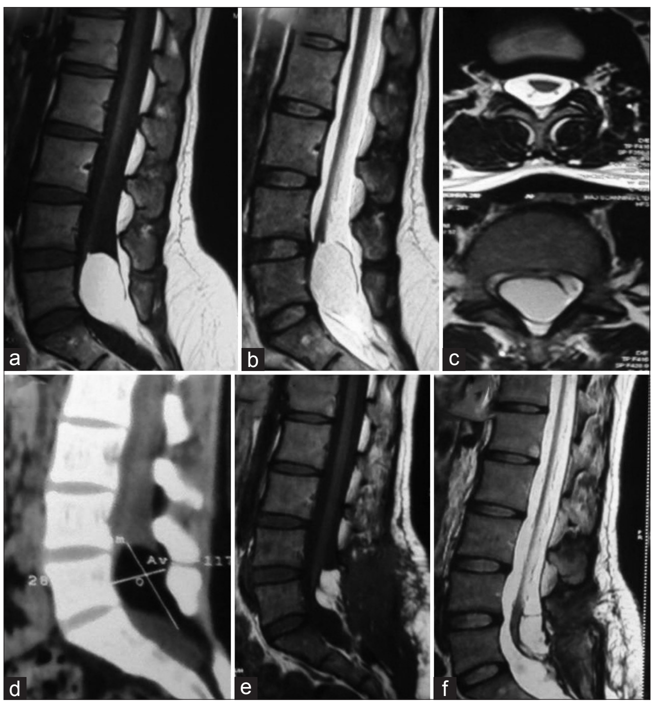

Figure 2: (a) T1 hyperintense (b) T2 hyperintense lobular mass in the conus region with extension along the filum (c) with tethering of the cord suggestive of transitional lipoma. A subcutaneous lipoma is also seen. On computed tomography, the mass is hypodense, no posterior element deficit is seen (d). Postoperative scan shows small residual fat along filum with detethering of the cord (e and f)

We also interviewed the adults in our effort to enquire into the possible causes that could have led to nonaddressal of this condition in their childhood itself. We also tried to find out if there were any precipitating factors leading to the onset/worsening of the symptoms that could have potentially forced these patients to seek treatment at this unusual age. The responses are shown in Tables 1 and 2. Interestingly, 60% of these patients were symptomatic since childhood (*n* = 12) and a large majority of them (*n* = 11, 55%), in fact, consulted a physician for the same. The major reason for not seeking surgical treatment was a lack of awareness of the condition and the necessity to get it treated (*n* = 6, 30%). Pessimistic counseling by the primary physician regarding postoperative outcome led to the refusal of surgery in five patients (25%). Some of them chose indigenous medications for their treatment (*n* = 2, 10%) instead. Three (15.0%) females sought surgery at adulthood as their parents thought these girls were nearing their marriageable age and the disease needed treatment so that they could marry like other girls.

The precipitating factors noted in our study were an active lifestyle, yoga and exercise (*n* = 7, 35%), prolonged sitting due to the inherent nature of their jobs (*n* = 6, 30%), pregnancy and childbirth (*n* = 2, 10%), and trauma (*n* = 1, 5%).

Discussion

Spinal dysraphic lesions in adults are rare. The incidence of adult spinal dysraphism was 15.4% in our series. Klekamp

Figure 3: (a) T1 and T2 hyperintense subcutaneous mass with extension intradurally s/o lipomeningocele (a and b). The mass is continuing on the dorsal aspect of a low-lying cord. Bony defects are visible. Postoperative scan shows excision of majority of the mass with detethering of cord (c)

Table 4: Distribution of radiological findings among patients of tethered cord syndrome

Radiological findings	Adults patients (total, *n*=20)	Pediatric patients (total, *n*=110)
Lipomyelomeningocele	8 (40%)	15 (13.5%)
Dermal sinus	2 (10%)	1 (0.9%)
Split cord malformation type-1 (Bony spur)	5 (25%)	23 (20.7%)
Split cord malformation type-2 (fibrous septum)	2 (10%)	6 (5.4%)
Fatty filum	3	-
Vertebral body defect (hemivertebrea, butterfly vertebrea)	9 (45.0%)	25 (20.0%)
Associated Intracranial anomalies	None	28 (25.4%)

reported 85 patients over a period of 18 years.[4] Hüttmann *et al*. reported 54 patients over a span of 16 years.[5] These lesions clinically present with TCS. Adult TCS can present in either of the three possible scenarios: (1) Those who were normal in childhood but developed symptoms in adulthood, (2) those with static neurological deficits or skeletal deformities diagnosed in childhood but who remained well until the onset of new and progressive neurological deficits in adulthood, and (3) those who had gradually progressive neurological deficit since childhood.[6] It is indeed strange that congenital anomalies like this often reach adulthood without getting medical attention. The reasons for the delay in seeking medical attention in our series are shown in Table 5. Lack of awareness and being explained the poor prognosis were the major reasons for not seeking treatment in our series (n = 11, 55%).

The pathogenesis of TCS symptoms in adults is unclear; evidence suggests that accumulative injury and precipitating factors play an important role. Breig reported that flexion of the head and neck results in sudden upward movement of the cord.[7] Gupta *et al*. suggested that the cumulative effect of repeated cord traction from years of natural head and neck flexion could ultimately lead to "punch-drunk" injury to the conus.[8] Strenuous exercise, childbirth in lithotomy position, sexual intercourse, forced flexion of legs at hip joints, straight leg raising exercise, forward bending, prolonged sitting, heavy lifting, lumbar spondylosis, disc herniation, trauma, and colonoscopy, etc., have been reported as precipitating factors in adult patients with TCS.[5,9-13] The precipitating factors noted in our study were very active lifestyle, yoga and exercise (n = 7, 35%), prolonged sitting due to the nature of their jobs (n = 6, 30%), pregnancy and childbirth (n = 2, 10%), and trauma (n = 1, 5%) [Table 6].

There was a male predominance in our series in both children and adults. This was in contrast to the observations by other authors.[11,14] The presenting symptoms in adult TCS include pain in the back with or without leg pain, sphincteric disturbance, and sensory-motor deficits.[11] Although pain is an uncommon symptom in children, it is almost the most common symptom in adults. Moreover, sensorimotor deficits are more frequently found in adults, occasionally with a combination of lower and upper motor neuron signs. In our series, the most common symptoms were motor weakness and bladder dysfunction followed by backache. The backache was localized in 12 (60.0%), while 8 (40.0%) patients had nondermatomal leg pain. Cutaneous stigmata of underlying dysraphic lesions were seen in 18 patients (90.0%). Cutaneous stigmata were present in 21 of 34 patients reported by Iskandar *et al*.[11] The most common cutaneous stigmata reported by them were hypertrichosis, followed by subcutaneous lipoma, scar of previous surgery, and hemangioma. It is indeed interesting that telltale signs of underlying dysraphism are as common in adults as in children. In spite of these stigmata, patients remain undiagnosed and untreated till adulthood.

Table 5: The reasons for delay in seeking treatment in adult spinal dysraphism

Q. no	Question	Patient response
1	Are the symptoms/signs present since birth	12 (60%) patients say yes and 8 patients 40% say no
2	Did parents consult a clinician before presenting to us?	11 patients 55% say yes and 9 (45%) patients say no
3	What was the reason for not undergoing surgery?	
	Poor prognosis explained	5 (25.0%)
	Lack of information	6 (30.0%)
	Social factors	4 (20.0%)
	Taking herbal or ayurvedic treatment	2 (10.0%)

Table 6: Precipitating factors in adult spinal dysraphism

Q. no	Precipitating cause	Number of patients
1	Very active lifestyle/yoga/exercise	7 (35.0%)
2	Prolonged sitting/forward bending	6 (30.0%)
3	Pregnancy/childbirth/Sexual intercourse	2 (10.0%)
4	Trauma	1 (5.0%)

The etiopathologic substrates underlying adult TCS in our study were lipomeningocele (n = 8), split cord malformation (Type 1 n = 5, Type 2, n = 2), dermal sinus (n = 2), and thickened fatty filum (n = 3). On the other hand, the lesions in pediatric patients included meningomyelocele/meningocele (61, 54.9%) followed by split cord malformation Type 1 and 2 (29, 26.1%). Iskandar *et al*. reported fatty filum (19/34), lipomeningocele (15/34), meningocele manqué (8/34), split cord malformation (13/34), and dermoid tumor in two cases.[11] Klekamp found split cord malformations, tight filum, conus lipoma, and a combination of these as etiopathological lesions leading to cord tethering.[4]

Two major radiological differences between children and adults that emanated from our study were the vertebral body defects and intracranial anomalies. Vertebral body anomalies were also reported in a very high number of patients by Iskandar *et al*. (30/34).[11] It is probably also the lack of intracranial anomalies associated with dysraphic states that cause a delay in clinical suspicion and hence the clinical diagnosis. It also indicates that cranial screening MRI may be avoided altogether in adults.

Probably, the most controversial issue with adult spinal dysraphism is their surgical treatment. With children, it is commonly believed that surgical detethering of the cord prevents neurological deterioration with advancing age.[15-18] In adults, however, the issue of growth with subsequent stretching of cord does not hold ground. However, the issue of mechanical stretching of the spine due to trauma or specific postures plays an important role in the development of new symptoms in these patients. In fact, a significant number of our patients, as well as patients reported in other studies, did have such a history.[6,19] Hence, controversy has surrounded the issue of surgery in newly diagnosed adult TCS. However, one more aspect that needs to be considered is that in a situation like ours, certain socioeconomic factors come into play. In female patients, the initial part of the disease in the childhood is often overlooked, unlike their male counterparts, until deficits increase and stabilize or the patient attains marriageable age when the family usually seeks medical attention.

The surgical complications in our experience were low (n = 3, 15%) and were mostly minor. CSF leakage occurred in two patients (10.0%) postoperatively. There were wound gape and local wound infection in one patient (5.0%). Two patients (10.0%) experienced transient postoperative urinary retention that resolved within 2 days. This may have been related to either the operation or the anesthesia. However, many authors have noted a higher incidence of complications following surgery in adults compared to children.[15-17,20] Lad *et al*. analyzed the surgical outcomes and complication rates after surgery for TCS in the United States between 1993 and 2002.[21] According to their observation, patients who were 65 years of age or older experienced a nearly 3-fold increase in complication rate (20.3%) compared to patients who were younger in age. Other authors have reported a similar adverse association between age and lumbar spine surgery.[22] In adult patients, intradural structures are frequently scarred and surrounded by significant arachnoidal adhesions that add to operative difficulty with subsequently higher postoperative complications. Some of the surgical difficulties include difficulty in dissecting the edge of the spinal cord from the surrounding dura and unusual, often confusing, anatomy. Iskaander *et al*. also noted a very high incidence of CSF leak related complication in their series.[11] Hüttmann *et al*. reported 19% subcutaneous CSF collection, 4% wound infection, 2% extradural hematoma, 5% revision CSF leakage and wound infection, and 2% permanent neurological deficit.[5]

The long-term results of surgery in adult TCS in our series are shown in Figure 4. All symptoms showed improvement or stabilization after surgery. Improvements in symptoms in descending order were pain (n = 11, 84.6%), sensory hypoesthesia (n = 2, 66.7%), motor weakness (n = 6, 56.2%), and sphincteric control (n = 7, 53.8%). Lee *et al*. also reported a low risk of neurologic complications in their series of sixty patients with adult TCS. In this series, back pain and leg pain improved significantly (78–83%) regardless of the origin of the tethering.[23] Rajpal *et al*. also reported that the symptoms that improved the most after surgical correction were back pain (65%) and bowel/bladder dysfunction (62%).[24] Garg *et al*. reported that 15 (83.3%) patients had shown improvement in a backache. Weakness improved by at least one grade in seven (77.8%) patients. Bladder symptoms improved in six (50%) patients.[14] Hüttmann *et al*. observed improvement in pain (86%), spasticity (71%), sphincteric dysfunction (44%), and sensorimotor deficits (35%) in descending order. Analysis of the factors affecting outcome reveals that delay in diagnosis (>5 years) and incomplete tethering are often associated with poor outcome.[5] Klekamp also stressed the importance of complete detethering for good long-term outcome. He additionally observed that the presence of lipoma and cystic lesions such as epidermoid/dermoid or neurenteric cysts was associated with poorer outcomes, especially in surgeries for retethering.[4]

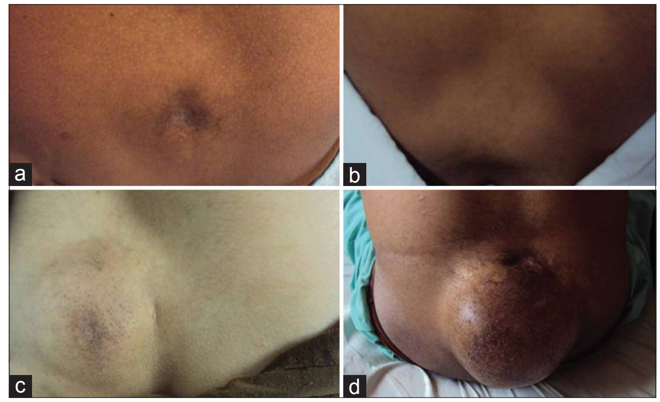

Figure 4: It depicted various types of skin stigmata associated with occult spina bifida: (a) Swelling with midline pits (depression); (b) overlying soft swelling; (c) cafe lau-spots with midline dermal sinus; (d) overlying hypopigmented skin

There were certain limitations of the study. The analysis was retrospective and follow-up was only 20 months, unlike other studies. However, patients in developing milieu do not turn up for follow-ups because of a variety of reasons; hence, a number of patients remain an inherent problem in studies involving follow-ups. However, this study has given some insight into the causes, clinical profile, and outcome profile of these groups of patients in a developing milieu.

Conclusion

Adult spinal dysraphic lesions constituted 15.4% of all cases in our series. These patients differed from their pediatric counterparts in having pain as a predominant symptom, having predominance of lipomyelomeningocele and split cord malformation as the cause of tethering, and having a higher incidence of vertebral body defects. Lack of information, pessimistic counseling by physicians, and certain social factors contributed to the delayed clinical presentation and treatment. Surgical detethering drastically relieves pain, stabilizes or improves sphincteric, and sensorimotor deficits. Considering the low complications following surgery, we recommend surgery in all symptomatic patients.

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Conflicts of interest

There are no conflicts of interest.

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