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Diastematomyelia

– Clinical Manifestation and Treatment Outcome –

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Diastematomyelia is a rare congenital anomaly characterized by a division of the spinal cord or the filum terminale into two parts. In Korea, only one case has been reported. The authors have operated on 5 cases of diastematomyelia with septum since July, 1978. The ages ranged from 1 to 44 years (median ; 11 years). There were 2 boys, 2 girls and an adult man. The disease manifested by cutaneous abnormalities and neurological or orthopedic deficits. Pain was a chief complaint in the adult patient. The symptoms had progressed in 3 cases. The diagnosis was made correctly by CT myelography or MRI in 4 cases. The median septum was located at the lumbar area in 4 cases and at the lumbosacral region in 1 case. Associated abnormalities included low lying conus (5 cases), lipoma (2 cases), thickened filum terminale (1 case), hemilipomyelomeningocele (1 case) and syrinx (1 case). The median septum was removed. The dural sleeve adjoining the septum was resected and the dural sac was reconstructed.

The role of MRI in the diagnosis and planning of surgery and the high frequency of associated low lying conus were emphasized. Though the surgical treatment relieved pain, it did not reverse the neurological deficits or orthopedic deformities significantly, which suggests the beneficial effects of early surgical intervention in the cases with progressive symptoms.

Key Words : *Diastematomyelia, Median septum, Low lying conus.*

INTRODUCTION

Diastematomyelia is a congenital anomaly in which the spinal cord or the filum terminale is divided into lateral halves. In some cases, the hemicord is in each of two dural sacs which are separated by a septum of bone, cartilage, or fibrous tissue. The septum runs anteroposteriorly from the posterior surface of one or more vertebral bodies through the center of the neural canal to be attached dorsally to the dura and even to the neural arch (Moes and Hendrick, 1963).

Although the embryogenesis of diastematomyelia has not been clearly understood, the theory of persistent accessory neurenteric canal (Bremer, 1952) is widely accepted.

Diastematomyelia may present with cutaneous,

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skeletal and neurological abnormalities. Among these, it is believed that the neural damage results from traction and repetitive trauma (Russell et al., 1990-1991). It is emphasized that the tethered spinal cord is adequately released only if the dural sleeve surrounding the septum is also resected flush with the anterior wall of the spinal canal, and the two halves of the cord are allowed to reside in a single-chambered thecal sac (Pang and Parrish, 1983).

Diastematomyelia can be associated with other forms of occult spinal dysraphism. Although it is a rare disease, of which only one case has been reported in Korea (Shin et al., 1992), the incidence of diastematomyelia will increase due to recent advances in radiological diagnostic methods.

In an attempt to understand the pathophysiology and the principle of treatment, the authors analyzed the clinical manifestation and treatment outcome in 5 cases of diastematomyelia with septum which had been operated on over a period of 15 years.

CLINICAL MATERIAL AND METHODS

From July, 1977 to February, 1993, 5 cases of diastematomyelia with septum were operated on at the Department of Neurosurgery, Seoul National University College of Medicine. Cases of diastematomyelia without septum are excluded from this study. Retrospectively, clinical manifestation, radiological findings, operative findings, and treatment results were reviewed.

For clinical manifestation, age, gender, presenting symptoms, progression of symptoms, physical examination and neurological findings on admission were investigated. Visualization of septum, split cord and associated anomalies were evaluated by the plain radiographs, CT myelography and MRI. Also the operative management for the lesion and the postoperative outcome were investigated.

Table 1. Clinical features in 5 cases of diastematomyelia

Case No.	Sex/Age (yrs)	Septum Location	Presenting Symptoms	Symptom Duration (mos) and Progression	Cutaneous Anomalies	Skeletal Anomalies	Neurologic Abnormalities
1	M/44	L_4-S_1	paresthesia of left leg (after trauma)	2 progressed			diminished muscle power, left leg diminished pain sense below left L_5 absent ankle tendon reflex, left sphincter dysfunction(+): hydronephrosis
2	M/1	L_4	delayed development of walking	9 stable	subcutaneous lipoma	Sprengel's deformity scoliosis bilateral foot deformity	spastic paraparesis ankle clonus(+/+) sphincter dysfunction(+)
3	M/11	L_{3-4}	scoliosis	132 stable	hypertrichosis	scoliosis right foot deformity	wasting of muscles of right leg diminished pain sense below right S_1 diminished knee and ankle tendon reflexes, right sphincter reflexes normal
4	F/3	L_4	weakness of muscles, right leg	1 progressed	dermal sinus tract	right foot deformity	wasting of muscles and diminished muscle power, right leg diminished ankle tendon reflex, right sphincter reflexes normal
5	F/15	L_3	back pain	1 progressed	subcutaneous lipoma		no motor or sensory disturbances reflexes normal

RESULTS

Age and gender

The ages ranged from 1 to 44 years (median ; 11 years). There were 2 boys, 2 girls and an adult man (Table 1).

Clinical features

The clinical features of the 5 cases are summarized in Table 1. The presenting symptoms were paresthesia of lower extremity, delayed development of walking, scoliosis, paraparesis, and back pain. The duration of symptoms ranged from 1 month to 11 years (median ; 2 months). Four cases had cutaneous anomalies. There was a subcutaneous lipoma in 2 cases, hypertrichosis and dermal sinus tract in 1 case, each. In 3 cases, skeletal anomalies (foot deformity in 3 cases, scoliosis in 2 cases and Sprengel's deformity in 1 case) were noted. Neurological examination revealed paraparesis in 4 cases, sensory changes in 2 cases, hyporeflexia in 3 cases, sphincter dysfunction in 2 cases and hyperreflexia in 1 case.

Radiological findings

The radiological findings are summarized in Table 2. Plain radiographs showed wide interpedicular distance, spina bifida, scoliosis, bony spur, narrow intervertebral disc space and laminar fusion (Fig. 1). The septum was detected in 2 cases which was at the same level with the laminar fusion.

CT myelography was performed in 2 cases. It revealed splitting of the spinal cord in 2 cases, one of which had a bony septum (case 3, Fig. 2 and 3) while the other did not (case 1). In the latter case, a fibrocartilaginous septum was identified during the operation. An associated hemilipomyelomeningocele was seen in 1 case.

MRI was taken in 3 cases. In two of these, a bony spur and the split spinal cord with tethering of the spinal cord were identified (Fig. 4). MRI in case 2 which was taken by a primitive machine, demonstrated a lipoma at the lumbar level and a syrinx at the thoracic level only. Possible reasons for the invisible septum by MRI in this case are ; 1) low resolution of the primitive MRI machine used, 2) obtaining sagittal images only, and 3) the same location of the septum and the associated lipoma.

Operative findings

In 4 cases, a correct diagnosis could be made preoperatively. One was considered to be a case of lipomyelomeningocele combined with syringomyelia

Table 2. Radiologic findings in 5 cases of diastematomyelia

Case No.	Plain Radiographs	CT Myelography	MRI Scan
1	no bony abnormalities	fibrocartilaginous septum L_{4-5}, L_5-S_1 filling defect L_{4-5}, L_5-S_1 duplicated cord L_{4-5}, L_5-S_1	
2	interpedicular distance widened L_{3-4} spina bifida L_{3-5} deformed abnormal expansion of lamina with fusion L_{3-4} complete or incomplete fusion defect T_{6-8} scoliosis		syrinx T_{1-5} lipoma L_{3-4}
3	posterior arch fusion L_{3-4} scoliosis narrowing of T_{7-8}, T_{8-9}, L_{3-4} interspace	bony septum L_{3-4} filling defect L_{3-4} duplicated cord T_8-L_3 right hemilipomyelomeningocele	
4	interpedicular distance widened L_{3-5} spina bifida L_{3-5} bony spur L_{4-5} narrowing of L_{4-5} interspace		bony septum at L_4 cord splitting L_{3-5} low lying conus S_1
5	interpedicular distance widened L_3-S_1 spina bifida L_2-S_1 bony spur L_3		bony septum at L_3 cord splitting L_{2-4} low lying conus lipoma L_{2-4}

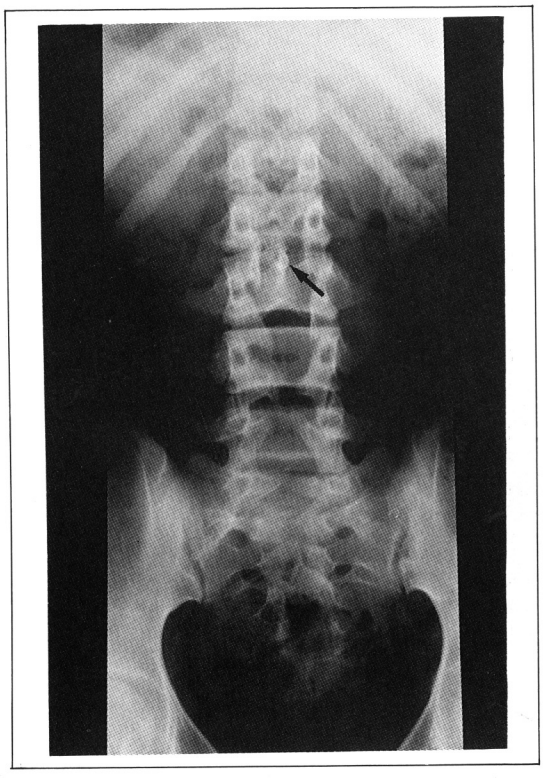

Fig. 1. Case 5. The plain radiograph of the lumbar and sacral spine anteroposterior view shows spina bifida from L_2 to S_1 with widening of the interpedicular distance from L_3 to S_1. The arrow points to the bony spur.

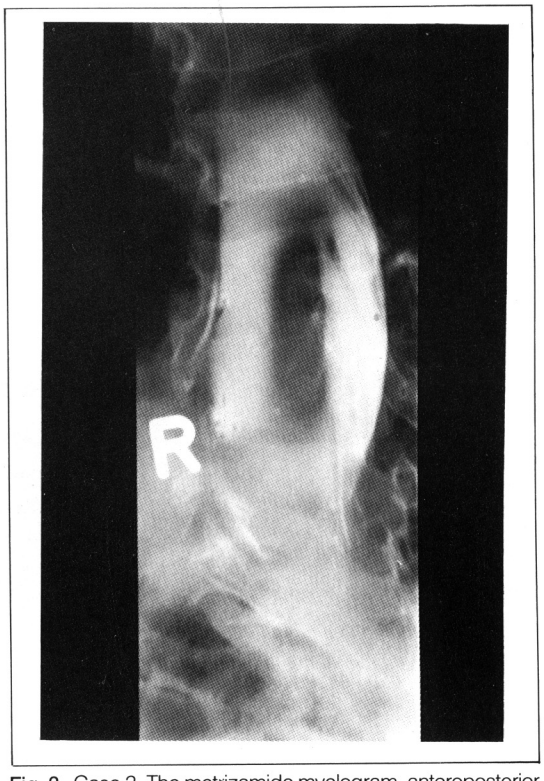

Fig. 2. Case 3. The metrizamide myelogram, anteroposterior projection reveals a splitting of the contrast medium into lateral halves by a midline oval filling defect at the L_{3-4} level.

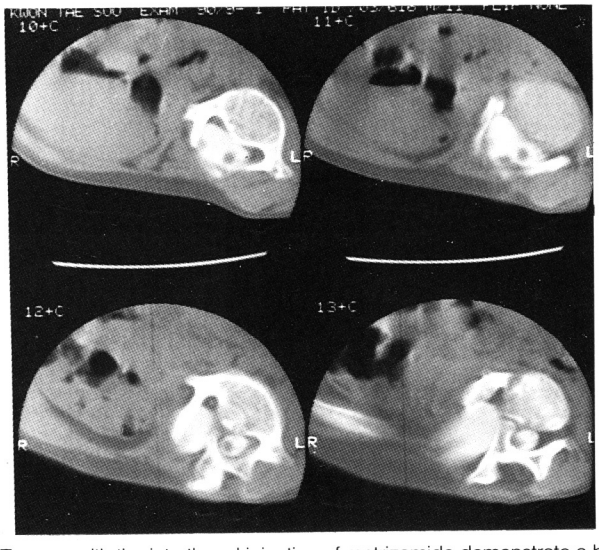

Fig. 3. Case 3. CT scans with the intrathecal injection of metrizamide demonstrate a bony septum splitting the lumbar spinal cord.

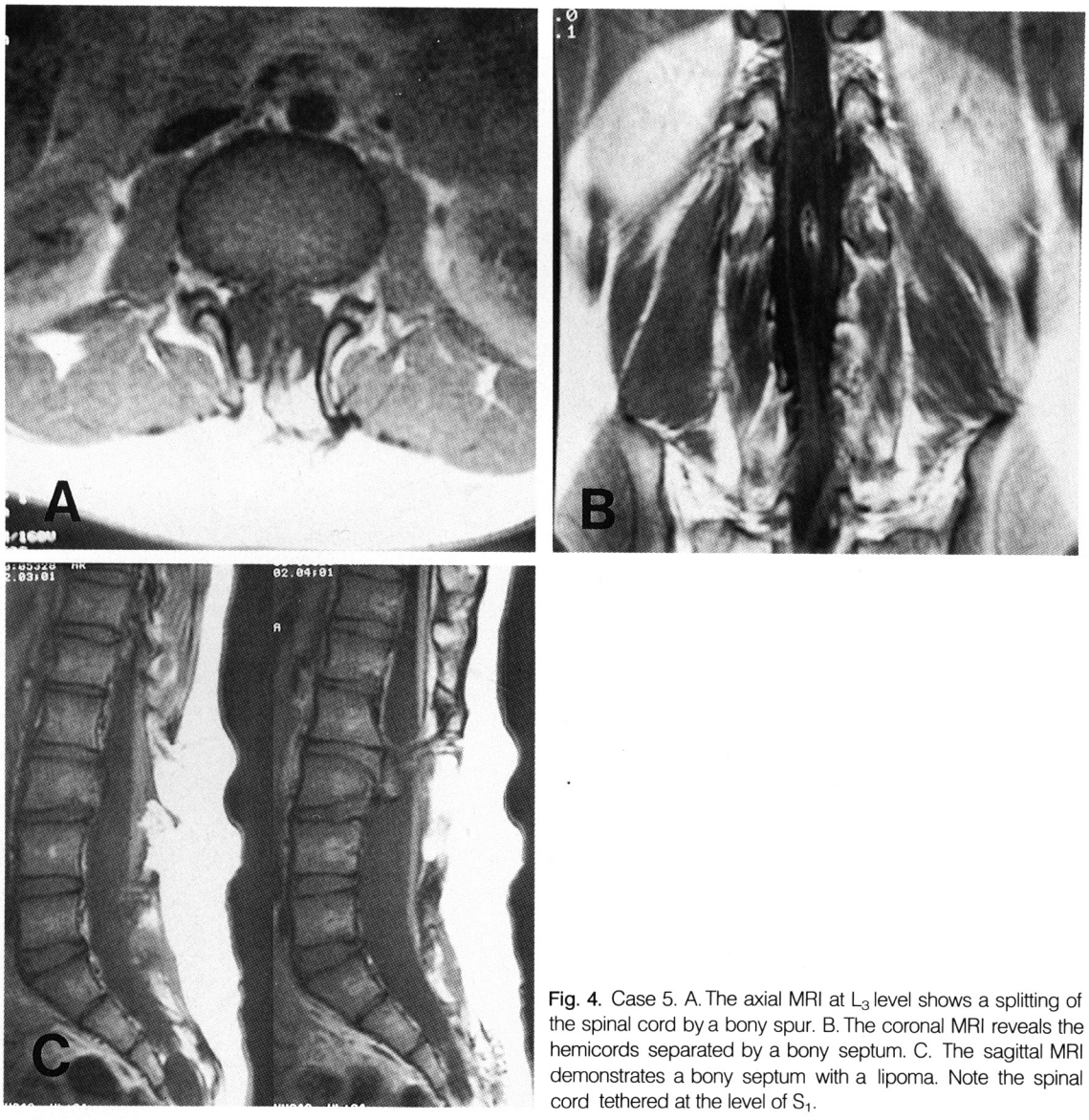

Fig. 4. Case 5. A. The axial MRI at L_3 level shows a splitting of the spinal cord by a bony spur. B. The coronal MRI reveals the hemicords separated by a bony septum. C. The sagittal MRI demonstrates a bony septum with a lipoma. Note the spinal cord tethered at the level of S_1.

preoperatively because the septum or split spinal cord was not detected on MRI while a lipoma, tethered spinal cord and a syringomyelic cavity were found.

All the patients were operated on in the same manner ; the standard laminectomy, decompression of the neural element, removal of bony spurs, division of fibrous bands and lysis of adhesions. In addition, duraplasty was performed in order to allow the two halves of the spinal cord to reside within a single-chambered thecal sac (Table 3, Fig. 5). Three cases had paramedian nerve rootlets that seemed nonfunctional and atrophic. Paramedian nerve rootlets were removed in an attempt to widen the operative exposure and to remove the septum. No postoperative permanent deficits occurred. Among the 5 patients, 4 had bony septa and 1 had a fibrocartilaginous septum. The two hemicords appeared symmetrical in 2 cases while they were asymmetrical in 3 cases. The hemicords of the side

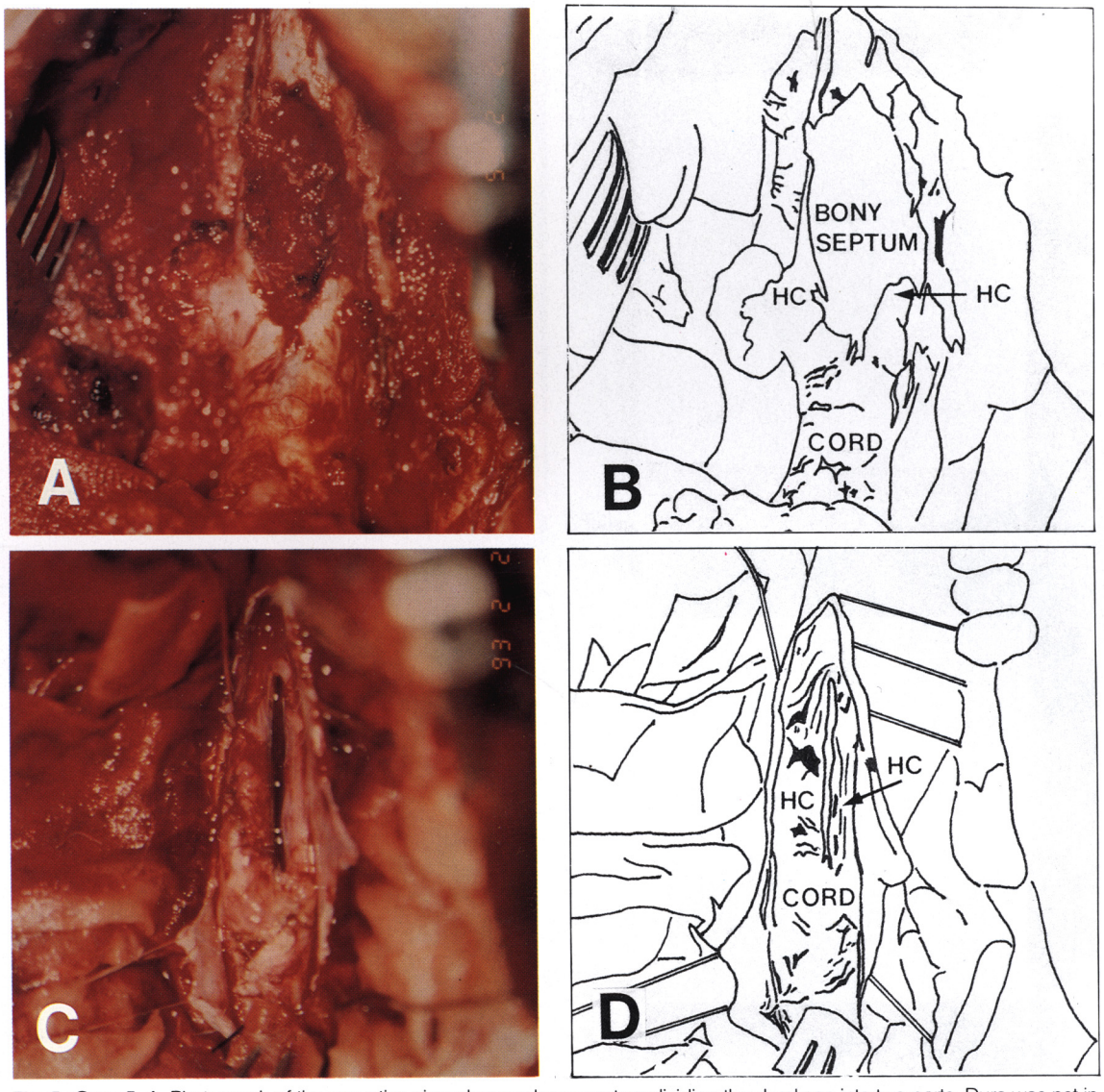

Fig. 5. Case 5. A. Photograph of the operative view shows a bony septum dividing the dural sac into two parts. Dura was not incised yet. B. Schematic drawing of A. HC=dural sac of each hemicord, cord=dural sac of the reunited cord C. Photograph of the split cord seen at operation, after removal of the bony septum. D. Schematic drawing of C. HC=hemicord

in which more neurological deficits of lower extremity were seen, were thinner than their fellows.

Low lying conus was suspected preoperatively in all the cases. However, in 3 cases (case 1, 2 and 5), untethering of conus was not performed due to wide separation of the septum and conus. Untethering of conus was also impossible in 1 case (case 4) because symptoms of malignant hyperthermia such as high fever, tachycardia and hypertension occurred during the surgery. In these cases, untethering of low lying conus will be done at the second stage if necessary. Untethering of conus was done in only

Table 3. Operative findings and outcome in 5 cases of diastematomyelia

Case No.	Operative Findings	Associated Spinal Abnormalities	Complication	Follow-up Period(mos)	Clinical Outcome
1	laminectomy midline dural incision left hemicord thinner than its fellow fibrocartilaginous spur extending from arch of S_1 to dura excised dorsally		CSF leak, transient right leg pain	2	improved improvement of motor and sensory function paresthesia was relieved
2	laminectomy paramedian nerve roots(+) fibrosseous spur removal (L_4) dural sleeve resection	low lying conus lipoma	wound dehiscence	67	improved seven months postoperatively, able to walk scoliosis had progressed
3	laminectomy bony spur removal (L_3-L_4) dural sleeve resection right hemicord thinner than its fellow repair of right hemilipomyelomeningocele untethering	hemilipomyelomeningocele thickened filum terminale	transient left leg pain	4	stable three months postoperatively, able to walk
4	laminectomy bony spur removal(L_4) dural sleeve resection right hemicord thinner than its fellow	low lying conus	malignant hyperthermia (intraoperatively)	1	stable
5	laminectomy paramedian nerve roots(+) bony spur removal (L_3) dural sleeve resection lipoma removal	lipoma	CSF leak, transient	2	stable low back pain was relieved

1 case. Other associated intraspinal anomalies included lipoma (2 cases), lipomyelomeningocele (1 case), and spondylotic bar (1 case). Corrective operations for these conditions were done. In 1 syringomyelia case, syringotomy was performed later.

Outcome

The treatment outcomes are summarized in Table 3. The duration of follow-up ranged from 1 to 67 months (median ; 2 months). There was an accumulation of cerebrospinal fluid at the wound site in two cases. These were managed successfully by conservative methods. Two cases with postoperative leg pain improved. All the patients did well after surgery, although there was no remarkable improvement or deterioration in the neurological status. Back pain was relieved by operation. A child with delayed development of walking could start walking by himself at 7 months after surgery.

DISCUSSION

In 1837, Ollivier first used the term diastematomyelia (Greek ; diastema : cleft ; and myelos : medulla or spinal cord) in order to describe a form of spinal dysraphism. Diastematomyelia is defined as a congenital anomaly in which the spinal cord or the filum terminale or both are split dorsoventrally into two parts usually separated by a septum. Diastematomyelia takes various forms ranging from a simple division of the spinal cord to the hemicords separated by a cleft containing bone and soft tissue. It may be also associated with tethering of conus medullaris, lipomas, teratomas, and myelomeningocele (Ugarte et al., 1980 ; Walsh and Markesbery, 1980 ; Gower et al., 1988 ; Shin et al., 1992).

The embryogenesis of diastematomyelia has not yet been clearly elucidated. During the early stages of the embryogenesis, a primitive neurenteric canal

joins the yolk sac to the amniotic cavity through the primitive knot (Hensen's node) transiently. Then the knot comes to lie at the tip of the coccyx in the fully developed embryo. In 1952, Bremer suggested that the neural plate might become divided by an accessory neurenteric canal which is located cranial to the Hensen's node. With the lateral growth of the embryo, the yolk sac is compressed bilaterally, and a dorsal herniation of the entodermal lining travels up the accessory neurenteric canal and divides the notochord, neural groove and the medial part of the mesenchymal vertebral body precursors into halves. Even a temporary persistence of the part of the fistula will result in the formation of two hemivertebral bodies which come together in the midline at a later date. The cuff of mesenchymal cells surrounding the fistula may then converge to form a midline spur that is made of bone, cartilage or fibrous tissue and project into the spinal canal between the two hemicords, depending on the degree of dorsal extension of the entodermal fistula. In the spinal cord, there is little attempt to close the clefts, and the mesenchymal tissue spreads around each half, investing it in a separate meningeal covering. In 1960, Bentley and Smith suggested another theory of pathogenesis, the split notochord syndrome. According to the theory, an adhesion between the ectoderm and entoderm in the early stage of embryogenesis forces the notochordal process to be divided into two parts, from which formation of neural plate and surrounding tissue occurs. At the later stage, depending on the range of healing, anterior and posterior spina bifida, diastematomyelia, neurenteric cyst, and congenital dermal sinus are created.

Approximately 310 cases of diastematomyelia have been reported (French, 1990). One Korean patient of diastematomyelia associated with cystic teratoma has also been reported (Shin et al., 1992). In diastematomyelia, the spinal cord reunites just below the level of the split. The spinal cord above and below the split is histologically normal (French, 1990). One hemicord may be smaller than its fellow, and the roots can be hypoplastic at the side of the smaller hemicord. The smaller hemicord has a correlation with the symptomatic lower extremity (Guthkelch, 1974), which was also supported by the present study. In 1990, Hilal reported that the lumbar region is the most common site for diastematomyelia (47%) while 27% are located at the thoracolumbar area, and 23% at the thoracic region. Only 1.5% of the lesions are at the cervical and sacral regions, each. In the present study, 4 out of 5 septa were located at the lumbar area and only 1 lesion was at the lumbosacral area. Septum may be a fibrous and/or cartilaginous tissue (25%) or a bony tissue (75%) (French, 1990). In the present study, the septum was a bony spur in 4 of 5 patients.

Diastematomyelia occurs more often in females than in males in a ratio of 3.5 : 1 (French, 1990). In contrast, male to female ratio was 3 : 2 in the present small series. Although adult cases have been reported (Russell et al., 1990-1991), most of the patients were younger than 20 years. In the present study, 1 case was older than 15 years. Earlier theories attempted to explain the development of symptoms in diastematomyelia on the basis of differential growth between the transfixed spinal cord and the vertebral column. However, this explanation is rather controversial. Most of the cord ascent occurs before the age of 2 months. However, the symptoms usually manifest after the age of 2 months and before adolescence during which the linear growth is accelerated (Guthkelch, 1974 ; Gower et al., 1988). Recently it is believed that the neural damage results from the sustained traction by the septum, fibrous bands and adhesions, or by the associated anomalies such as low lying conus. This may leave the neural tissue vulnerable to ischemic injury by the repeated stretching. Therefore, the tethering and the cumulative effect of repetitive trauma may be combined and result in the neurological deficits (Guthkelch, 1974 ; Yamada et al., 1981 ; Maroun et al., 1982 ; Russell et al., 1990-1991).

Diastematomyelia is usually combined with abnormalities of the cutaneous, skeletal, and nervous system (Moes and Hendrick, 1963). In children, the common modes of presentation are : 1) a newborn with a myelomeningocele, 2) cutaneous abnormalities such as hypertrichosis, hemangioma, skin dimple, or lipoma, 3) what we call "orthopedic syndrome" (James and Lassman, 1960) which consists of scoliosis and foot abnormalities, and 4) neurological deficits such as sphincteric dysfunction, reflex abnormalities, and weakness associated with muscle atrophy (Gower et al., 1988). For adults, two modes of presentation are : 1) back pain, and 2) progressive myelopathy or sudden onset of neurological deterioration following minor trauma (Gower et al., 1988). In the present study, progression of the symptoms was noted in 3 out of 5 cases. Of those, an adult patient showed a neurological deterioration

after trauma.

In the plain radiographs of the spine, the spinal canal is almost always widened, as shown by an increase in the interpedicular distance at the level of the lesion. The septum is seen as a spindle or oval-shaped density located at the approximate center of the wide spinal canal (Moes and Hendrick, 1963). In the present series, the interpedicular distance was increased in 3 of 5 patients and the bony spur was visible by plain radiographs in 2 of 5 cases. The neural arches themselves may be malformed, showing lack of fusion, abnormal fusion, or bizzare bony overgrowths. Of these, spina bifida occulta is the most common associated findings (75%) (Herren and Edwards, 1940). In the present study, it was noted in 3 of 5 cases. The combination of intersegmental laminar fusion and spina bifida is a more accurate indicator of the level of the septum than are the cutaneous anomalies or deformities involving the vertebral bodies (Hilal et al., 1974). In the present study, the septum was identified at the level of intersegmental laminar fusion intraoperatively. There may be associated anomalies of the spine such as hemivertebrae, unsegmented or hypoplastic vertebral bodies, narrow intervertebral disc space, kyphosis and scoliosis (Moes and Hendrick, 1963). Myelography reveals the split contrast medium into lateral halves seperated by a round or oval midline filling defect. Also CT myelography provides a better outline of the bony spur and its abnormal anatomy (Harwood-Nash and McHugh, 1990-1991). MRI shows the sagittal and coronal images of diastematomyelia and can detect associated anomalies accurately (Harwood-Nash and McHugh, 1990-1991). However, it may not show a bony spur that does not contain marrow, and its effectiveness may be reduced significantly by scoliosis (Thron and Schroth, 1986 ; Gower et al., 1988).

It is controversial whether the surgical management of diastematomyelia should be performed prophylactically within the first two years of life (Guthkelch, 1974), and whether the surgical correction of the lesion prevents progression of the neurological deficits and may even result in some improvement (Moes and Hendrick, 1963 ; Russell et al., 1990-1991). The surgical treatment of diastematomyelia includes decompression of the neural structures, removal of the bony spur, and lysis of adhesions. Resection of the double dural sleeves should be started from the upper margin of the cleft where the hemicords are less adherent and set widely apart, and then continued caudally (Pang, 1991). The double dural tubes are then converted into a single-chambered thecal sac with the midline closure. In the operative approach to diastematomyelia, the resection of the dural sleeves along with the bony spur is important and its advantages are : 1) an adequate release of the conus could be achieved by the complete removal of the stiff inferior edge of the dural sleeve which exerts potentially deleterious effects on the tightly apposed neural tissue, and 2) the intradural exposure affords the advantage of incising the fibrous adhesions which seem to anchor the lateral surfaces of the hemicords to the dural wall and of seeking and eliminating other assocated anomalies such as a thickened filum (Pang and Parrish, 1983). After a simple extradural removal of the septum, residual mesenchymal cells may persist and be reactivated to cause complete regeneration of a new septum (Pang and Parrish, 1991). In the present study, all the patients were treated with the removal of septa and dural sleeve resection. Also the operative findings showed that all the septa were located at the distal part of the cleft between the hemicords and the spinal cords above the splits were not compressed or tethered while the spinal cords which reunited just below the level of the split were compressed and tethered by the septa and surrounding dura. Because the symptoms of diastematomyelia are produced not only by the septum but also by the associated anomalies, untethering of other sources should be done if possible. In the present study, all the patients were suspected of having a low lying conus. In 4 of these, untethering of the conus was not done due to the limited operative exposure and the malignant hyperthermia. The long term follow-up data in a larger number of cases are necessary for the adequate evaluation of the role of untethering of associated lesions.

The majority of patients remain neurologically stable or improved after surgery. Scoliosis, however, may continue to progress postoperatively (Gower et al., 1988). In the present study, 2 cases had some degree of improvement. But remarkable neurological changes were not shown which supports early surgical intervention in the cases with progressive symptoms.

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