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Split cord malformation

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Split cord malformation is one of the rare occult dysraphisms. The exact incidence is difficult to quote but in our experience, we find the incidence of 33%, slightly more than reported in other series. With advent of different MRI protocols and increasing diagnostic armamentarium, the incidence continues to rise.

In 1992 Pang revolutionalized the concept and nomenclature of split cord malformation. His unified theory of embryogenesis unravelled the long-lasting conundrums in understanding why some septum is bony and other fibrous. After this concept, words like "diplomyelia" and "diastematomyelia" became history. According to Pang's unified theory, persistent accessory neuroenteric canal leads to formation of septum. Actually, it was Bremer et al. [1] who first proposed the term accessory neuroenteric canal. He said that the persistent adhesion of the ectoderm-endoderm and cleft notochord favours the formation of neuroenteric canal. Later on, Pang added concept that the failure of "anlagen" to integrate in midline and formations of endomesenchymal tract contribute to septum in split cord malformation. Further, incorporation of "menix primitiva" into the tract leads to bony septum and non-incorporation leads to fibrous septum.

Besides type of septum, the surrounding thecal sac and possibility of whether lamina is fused or hypertrophied is to be kept in mind. In SCM type 1 there are two dural sleeves with or without fused lamina. In SCM type 2 there is only one dural sleeve. In our experience of 16 cases, we described that the patients with SCM are rarely associated with meningomyelocoele and their clinical, embryological and prognosis profiles differ from those who are not associated with same. We coined the term "Complex spina bifida" for SCM associated with meningomyelocoele highlighting the fact that they deserve special attention in order to fetch an optimum outcome. Mahapatra et al. [2] has modified the Pang's classification further into type 1a-d to avail safe excision of septum. McLone unified theory further highlights the existence of rare exceptions to pangs embryological hypothesis like dorsal spur. To add on Pangs classification, we proposed a sub classification into type A and B on basis of whether Split cord malformation (SCM) is associated with meningomyelocoele or not. This classification highlights the importance of pure or combined occurrence. Pang and Ersahim have also pointed on high incidence of type 1 SCM as association with MMC. The series from Mahapatra et al. [2] accentuates the importance of dorsal spurs. In our retrospective experience of 5 years' study in 60 patients of Split cord malformation, we found 6 cases of dorsal spur and all patients were operated and de-tethering and drilling of bony spur was done. We found that patients with dorsal spur have higher incidence of associated anomalies (50%) when compared with their "ventral" counterpart (33%). Based on our experience, we highlighted the importance of single ontogenic error in embryogenesis and association with other spinal dysraphisms.

The patients with SCM are usually asymptomatic and may presents with back pain, difficulty in walking, urinary difficulties and stigmata of dysraphism. In our series, we found that the presence of hypertrichosis and orthopaedic syndromes should raise index of suspicion. Mechanical tethering of cord may be the central cause so dealing with tethering is also equally important. These patients may have dorsal bands which are extension of neural elements from "menix primitive" to extradural space. The main problem of Split cord malformation arises due to tethering of cord either at level of spur or below it. Tethering can be at a single level or multilple level. We defined the entity "spina bifida multiplex" for patients with more than two level tethering. In our series of 41 patients, we showed that these patients of spina bifida multiplex behave in different way and need separate place in classification system because the prognostication and outcome also depends on such associations.

We investigate all patients of spinal dysraphism with cranial and spinal MRI, chest Xray, Ultrasonography, ECHO screening, uroflowmetry and post voidal residual volume as our departmental standard policy wherever possible. CT Spine is done for better bony delineatio for cases of bony split cord malformation. Magnetic resonance imaging is the most common and the investigation of choice for split cord malformations. The spur may be isointense or slightly hyperintense compared to CSF on T1-weighted images. Bony, cartilaginous and fibrous spurs all appear hypointense on T2 weighted images. The osseous spurs can be missed on T1-weighted images.

Surgery is the treatment of choice wherein the bony spur should be excised microsurgically. All the associated tethering lesions like arachnoid bands, adhesions, aberrant nerve roots, thick filum terminale, etc must be thoroughly studied and dealt with. Repairing the dura in most meticulous manner in order to re-construct the normal anatomy is a very important step. Cutting of fibrous septum should be started with distal most point and complete excision is thumb rule. Bony septum is difficult to deal with especially when spur is dividing at certain levels and going high up. We usually extend our laminectomy to have a proper exposure. Mahapatra et al. [2] has also emphasized on use of drills for dorsal spur.

In conclusion, the understanding of embryology, thorough investigations, study of associated lesions, and use of recent concepts

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and classification in dealing split cord malformation are key modulators for better surgical results and may vicissitude the outcome. Early surgical intervention even in asymptomatic presentation has shown to improve long term outcome.

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