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Pan Sacral Agenesis of the Dorsal Wall of Sacral Canal – A Case Report.

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Introduction

Sacrum is the last part of vertebral column and is triangular in shape, it is formed by the fusion of five sacral vertebrae. Its broad upper base articulate with fifth lumbar vertebra to form prominent scaro vertebral angle while lower narrow apex articulate with coccygeal vertebrae.

Ventral surface of sacrum is smooth and form posterio superior wall of pelvic cavity and dorsal surface shows sacral canal which is continuation of vertebral canal is formed by fusion of posterior elements namely lamina, ossified ligament flava and spinous process^[1].

Sacral canal contains cauda equina, filum terminale and spinal meninges. Dura and arachnoid matter end at the level of S2 vertebrae while pia matter continues up to coccyx as filum terminale. There is opening at the caudal end of sacral canal called as sacral hit us which is formed by failure of fusion of lamina of S5 vertebrae, this hiatus opening is covered by scarococygeal membrane, fatty tissue and skin^[2]. There are various variations observed in the formation of dorsal sacral canal which may show open sacral canal throughout or may be low lying lamina of 1st sacral vertebra. Clinically sacral canal has been used for administering the caudal epidural block (CEB), which is important tool for spinal surgeons and domain of pain reduction management associated with lumbar and sacral nerve roots in lumbar spinal disorders.

Hence a thorough knowledge of different anatomical features of dorsal wall of sacrum in male and female lead of reduction in failure of administration of caudal analgesia^[3].

Case report

During regular osteology illustration classes for undergraduate students of Ist year MBBS, a dry human sacrum showed complete agenesis of the dorsal wall of sacral canal. Sacral canal was open throughout its length extending from S1 to S5 sacral vertebra.

The lamina was fused laterally but were open in midline. Other features such as intermediate sacral crest which represent the fused superior articular facets and lateral

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sacral crest which represent the fused transverse process were nor mal. Dorsal sacral foramina were very well observed. (Fig 1 and Fig 2)

Figure 1: Posterior View of Sacrum

Dotted double sided vertical arrow represents complete agenesis of dorsal sacral canal wall

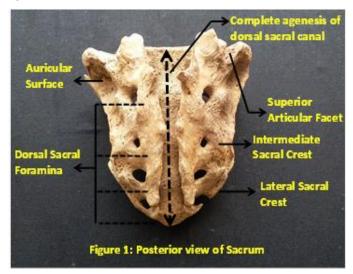
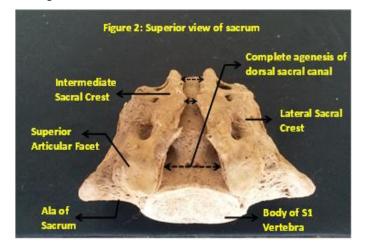


Figure 2: Superior View of Sacrum

Dotted double sided horizontal arrow represents comp lete agenesis of dorsal sacral canal wall.



Discussion

Variations in sacral hiatus like its shape, size etc has been reported several times in previous literature, but complete agenesis of dorsal wall of sacral canal is a scare finding. Seema et al., from her study on 159 scara reported complete agenesis of dorsal scaral canal in 3.14% ^[4]. Jhenneberg et al., observed 3% of scara with complete Kiran V P et al also reported 2% incidence of complete agenesis of dorsal wall^[6].

Doral wall agenesis out of 124 adult sacral vertebrae^[5].

Embryological explanation

Vertebra develops from ventromedial sclerotome part of somite. Spina bifida oculta occur as a fallacy in tissue separation during secondary neuralization resulting in dorsal nonunion of lamina. In this case laminas of all the 5 sacral vertebrae were present but they failed to fuse in the midline. Primary centers for the centrum and each half of neural arch appear between 10 and 20 weeks of intrauterine life.

Additional primary centers for costal elements are present. Each costal element unites with its half of neural arch at 2^{nd} & 5^{th} year and the conjoined element so formed unites anteriorly with centrum and posteriorly with its opposite fellow at about 8 years.

After puberty the fused neural arches and costal elements of adjacent vertebrae begins to coalesce from below upwards.

At the same time individual epiphyseal centers develop for bodies, spinous tubercles and costal elements. The primary centre for neural arch appears near the root of trans verse tubercles and then spreads anteriorly towards pedicles and posteriorly towards lamina and spine. Complete fusion of five vertebrae as single piece of bone was observed after puberty.

Any defect in fusion leads to in complete formation of sacral canal and incomplete ossific ation of laminae. Sacral Spina bifida oculta (SSBO) represents an open neural arch due to insufficient fusion of posterior elements of spine ^[7]. Kettler and Wilke categorised the SSBO defects on scale of 0 to III depending upon percentage of opening in the posterior neural arch.

Grade 0 being unaffected complete posterior arch to grade III "pan- sacral" was defined as failure of closure

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from S1 to S5^[8]. The overall incidence of pan sacral agenesis of scaral canal ranges from 0.98% to 4.8% ^[10].

Molecular Reasoning

Failure of fusion of lamina of sacral vertebra in midline to form median scaral crest result is this condition although embryo logical basis is still not clear and may be multifactorial, but the most acceptable cause is faulty induction of vertebrae formation at this level due to altering sonic hedgehog signaling. Genetic causes of these defects remain elusive although according to Lang man factors responsible for this condition are a mutation in VANGL gene and HOX gene.

VANGL genes are part of the planar cell polarity pathway that regulates convergent extension, the process that lengthens the neural tube and is necessary for normal closure to occur. Homeobox gene code for transcription factors activates the cascades of genes regulating pheno mena such as segmentation and axis formation^[10].

Clinically caudal epidural block (CEB) has been extensively used both for diagnostic and therapeutic management of lumbo sacral disorders.

The CEB in volves passing a spinal needle through the sacral hiatus to distribute medications into the sacral epidural space. In case of pan sacral agenesis of the dorsal wall of sacral canal, the CEB is possible, but this procedure becomes risky to complications like an accidental puncture of dura mater. Similarly, the spine surgeons may face difficulty in the exposure of the sacrum for the surgical procedure including instru mentation using the transpedicular screw for spinal fusion. Dorsal agenesis of sacral canal also places the sacral spinal nerves to risk to injury during such internal screw fixation^[9].

Conclusion

Awareness of morphological variations of sacrum like complete or partial agenesis of the dorsal wall of the sacral canal is important to diagnose low back pain, sciatica, caudal regression syndrome and to avoid comp lications related to caudal epidural block and other spinal surgeries like in placement of screw for spinal fusion. Know ledge about this abnormality is also im port ant in the field of pediatrics to deal with the associated congenital abnormalities.

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