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RESEARCH ARTICLE

CONGENITAL SCOLIOSIS DUE TO HEMIVERTEBRA: A REVIEW OF LITERATURE AND MANAGEMENT STRATEGIES

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ABSTRACT

Background: Hemivertebra is a common congenital problem resulting in varied deformities of the spine. Etiopathogenesis of hemivertebra is still not clear. But the spectrum of cases resulting from hemivertebra pose a clinical challenge especially in growing skeleton. The progression of deformity also depends upon part of spine involved. The treatment can be conservative or surgical. Multiple factors are taken into consideration before embarking on any treatment modality.
Aims and objective: To evaluate the the role of conservative management and surgical treatment for the management of congenital scoliosis resulting from hemivertebra and draw an inference based on our observations.
Material and Methods: We identified 3 cases from our database. One case was a 12 year old child with upper dorsal scoliosis who was managed conservatively. 2nd case was a 23 year patient with l2 hemivartebra who was also managed conservatively. And 3rd case was a 11 yr patient had progressive dorsolumbar kyphoscoliosis who was managed by surgery.
Results: In in first 2 cases, bracing and serial follow ups showed progression of not more than 2 degrees per year and resulted in stable spine with no deficits. In the 3rd case, timely surgical intervention with a stable construct resulted in halting of progression of deformity.
Conclusion: Regular follow up with imaging plays a vital role in management of these cases. Assessment of progression and choosing appropriate line of management will benefit these cases effectively. A systematic review of literature and understanding the biomechanics and progression of deformity is very essential in deciding the treatment options.

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INTRODUCTION

Congenital scoliosis is a spectrum of segmental anomaly resulting from lack of formation of one half of vertebral body. It is a rare congenital malformation of spine where only one side of the vertebral body develops resulting in laterally based wedge shaped vertebra with half vertebral body, single pedicle, and hemilamina[1]. Scoliosis in hemivertebra is considered as congenital scoliosis,as the deformity is present since birth. However, the diagnosis is made at a later age when the patient is brought to the doctor by the parent or educational worker who observes the defective spinal curve after it becomes prominent with growth.

Deformities due to hemivertebra[7]

- Scoliosis- lateral hemivertebra
- Lordosis- dorsal hemivertebra
- Kyphosis-ventral hemivertebra

Incidence[7]: Congenital scoliosis due to hemivertebraoccurs in 1 in 10,000 newborns.

Causes of congenital scoliosis are[7]

- Incomplete formation of vertebra : hemivertebra
- Failure of separation of vertebra: block vertebrae
- Combination of bars and hemivertebra.

Types of scoliosis in hemivertebra[1]

- Failure of formation
- Failure of segmentation
- Mixed

Cause[2]

Congenital scoliosis is believed to be a result of insult to the fetus during spine embryology development between 5th and

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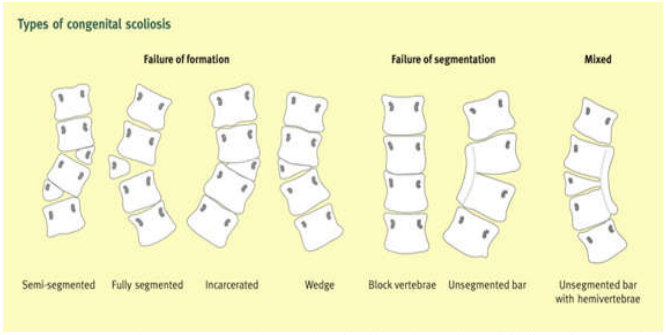
8th week of gestation & other malformations such as congenital heart diseases, spinal column dysraphism, or kidney malformations are frequently associated.

Figure 1:

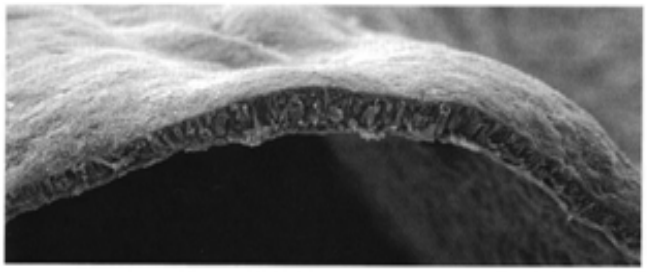


Courtesy: Fender, D. *et al*. "Spinal disorders in childhood II: spinal deformity", Surgery (Oxford), Vol. 32, Nr1, pp 39–45, January 2014.[1]

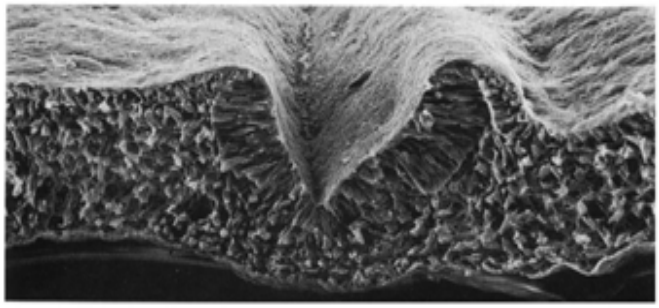
Embryology[6]

The spinal vertebrae develop during 6th week of gestation with 2 lateral chondrification centers arising in the developing vertebral bodies. These centers fuse to form primary ossification center by 8thweek of gestation. These centers are transiently separated by notochordal elements into anterior and posterior aspects. Lateral hemivertebra results from failure of development of one of paired chondral centers. Less commonly posterior hemivertebra results from failed anterior ossification.

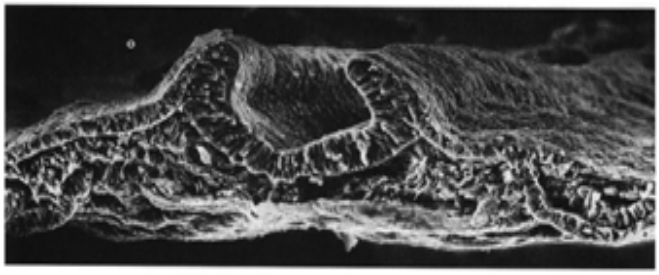
Figure 2:



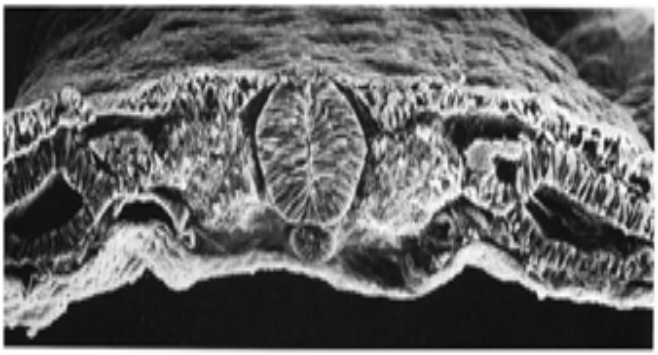
Formation of neural palte from blaminar embryonic disc



Formation of neural groove from ventral aspect of the neural plate



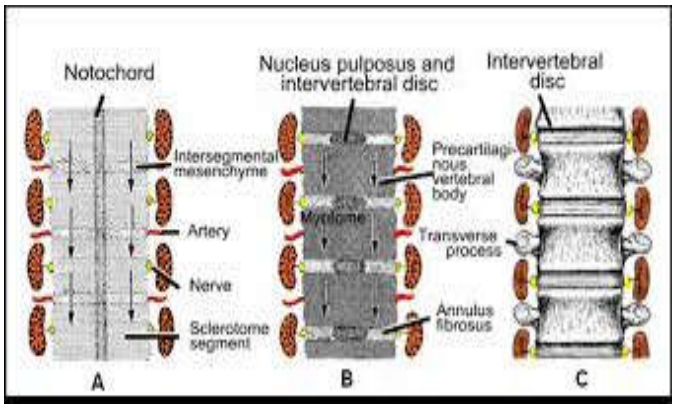
Formation of neural true



Migration of sclerotomes around the notochord

Migration of cells of sclerotomes around the notochord-ventromedially to form body of vertebra and disc

- Ventrolaterally to form costal processes
- Dorsally to form vertebral arch.



COURTESY: LANGMANS TEXT BOOK OF EMBRYOLOGY[6]

Fig. 3.

Lateral malfromations leads to scoliosis; Posterio-lateral defects causing lordo-scoliosis; dorsal causing lordosis; antero-lateral causeskyphoscoliosis; ventral causing kyphosis.

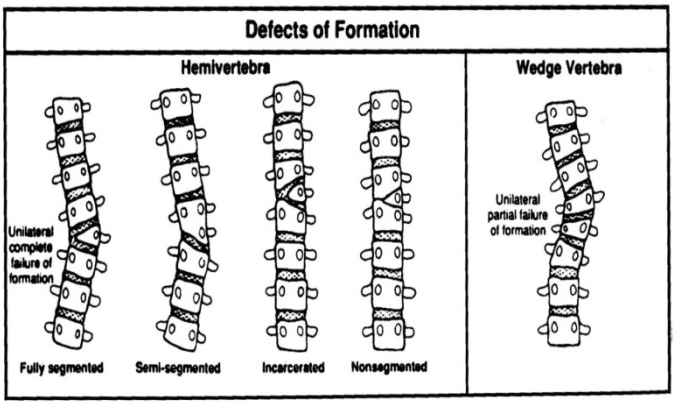
Classification of hemivertebra[1,5]

Fully segmental: Not attached with either vertebra. Usually triangle shaped with disc spaces above and below appearing normal. A single unilateral hemivertebra always lay at the apex of a definite scoliosis and usually progresses as the child grows.

Partially segmental: Segment fused with vertebra above or below. They are commonest in lumbar region and tended to be single.

Non segmental: Not separated from above or below vertebral bodies.

Incarcerated: Joined by pedicles to levels above and below. Least common type. Most frequent in thoracic region and tend to be single. They are ovoid in shape and smaller than a fully segmented one. Disc spaces above and below tend to be narrow and poorly formed. They usually have single pedicle and in thoracic region they are associated with a rib.



Courtesy: Goldstein I. (2005). Hemivertebra: prenatal diagnosis, incidence and characteristics, Fetal DiagnTher,.20 (2), pp 121-6. [11]

Fig 4.

Courtesy: Journal of Medicine and life.Vol IX, Isse 2, April-June 2016; Baneasa Hospital – "Regina Maria" Private Health Care Network, Bucharest, Romania Department of Pediatric Surgery,Brasov, Romania.[12.]

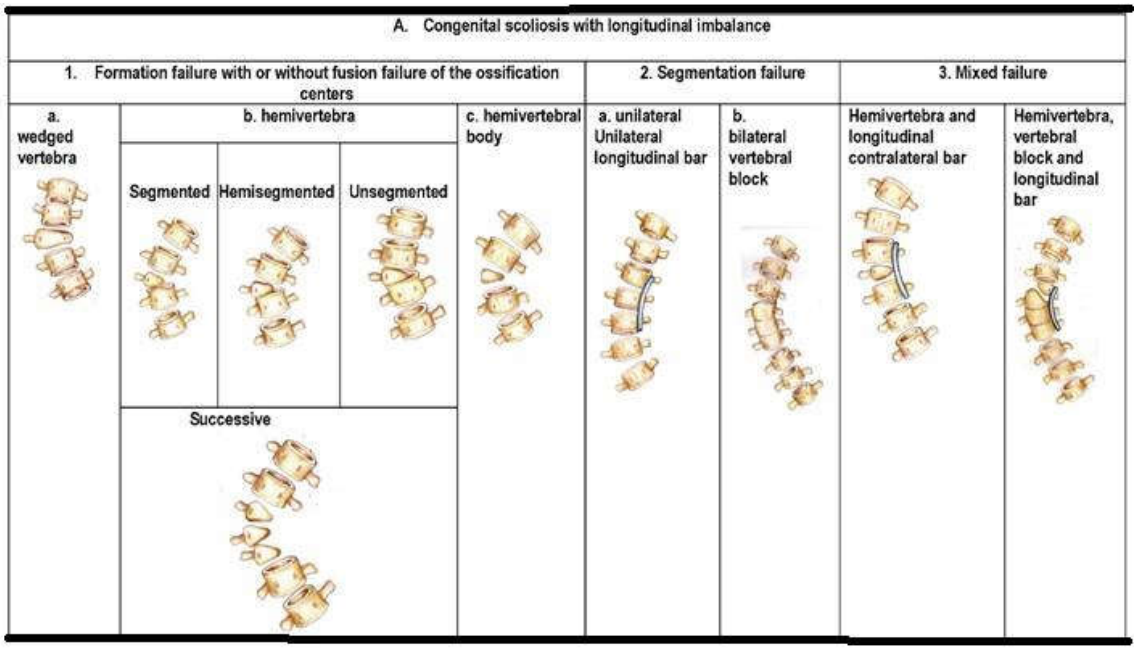
Associated anomalies with hemivertebra[4]

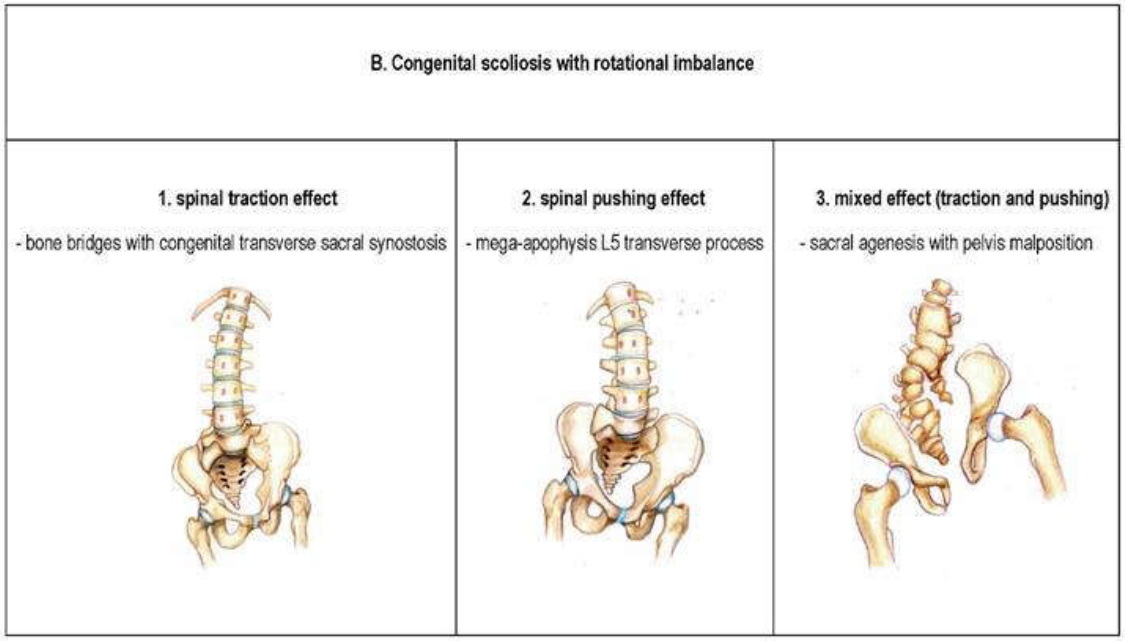
Some of the anomalies associated with hemivertebraare Klippelfiel syndrome, Tethered cord, Diastometomyelia, VACTREL anomaly, congenitalhydronephrosis, musculoskeletal problems like club feet and DDH. Hemivertebra can be seen in patients with mucopolysacharidosis.

Factors of progression of scoiliosis:[3]

25% of congenital curves do not progress. 25% undergoes mild progression and 50% evolve rapidly and require treatment.

Burnei-gavriliu classification of hemivertebra





The natural history of progression is highly unpredictable and depends on various factors. Mc Master and Ohtsukawere first to focus on the natural progression congenital scoliosis and define the risk of progression in relation to 4 key factors:

- Type of hemivertebra
- Site
- Number of hemivertebrae
- Age of patient

Type of Hemivertebrae

Commonest pathological variant is fully segmented and non-incarcerated (65%). There is absence of growth plates on the unformed side of hemivertebra in contrast to two relatively normal growth plates, one on each surface of the developed part of the vertebra. This causes the hemivertebra to grow as an enlarging wedge. Incarcerated hemivertebra is least common (13%) and it has poor growth potential and usually does not require treatment. Partially segmental vertebra (22%): Two growth plates are absent on the on the convexity of the curve. This tends to balance the growth plates on 2 sides of hemivertebra. The hemivertebra itself causes tilting of spine and can induce progressive scoliosis. These curves usually do not exceed 40 degrees at skeletal maturity and treatment is usually not required except when the hemivertebra is at lumbo-sacral junction.

Site:

Upper thoracic curves: (T1TO T4): deteriorate slowly, not severe, cause significant cosmetic deformity.progresses at rate of 1-2 degrees before 10 years and 2.5 degrees after 10 years. Lower thoracic spine: (T5 to L10): deteriorate more rapidly and exceed 45 by skeletal maturity. These usually do not produce significant cosmetic deformity. Rate of progression is 1 to 2 degrees per year before puberty and 3 degrees after puberty.

Thoraco-lumbar: Rate of progression is 2-3 degrees before puberty and 3-4 degrees after puberty resulting in substantial trunk imbalance.

Lumbar curves: (L2-L4): Deteriorate relatively slowly and cause mild cosmetic deformity.

Lumbo-sacral hemivertebrae: Results in large secondary structural thoraco-lumbar curve. Causes moderate to severe cosmetic deformity. These patients require early prophylactic treatment before they begin to list and before secondarycurve becomes structural.

Number

2 unilateral fully segmental hemivertebrae cause greater imbalance and progress more rapidly than single fully segmental hemivertebra and hence require prophylactic fusion as soon as they are diagnosed. If hemivertebra are in different locations, the resulting curves are often unbalanced and cause the trunk to list on one side. Hence this requires prophylactic treatment. Multiple hemivertebrae are often of mixture types but the spine usually remains balanced and hence there is no deformity apart from stunting of trunk. No treatment is required. In cases of unilateral unsegmented bar and a contralateral hemivertebra the rate of progression is very rapid with 14 degrees per year. They occur most commonly in thoracic spine and are most severe of all scoliosis. If treatment is necessary, this should be anticipated and carried out at an early age and it is much easier to prevent than to correct the severe deformity resulting from hemivertebra. In planning of prophylactic treatment, treatment should be directed to prevent the deforming growth of hemivertebra.

Clinical examination[4]

- Look for neurocutaneousmarkers.E.g: tuft of hair, café au lait spots
- Sagittal and coronal plane balance
- Shoulder mal-alignment
- Assessment of structural and compensatory curve
- Neurological examination- muscle atrophy, muscle strength, reflexes.
- Flexibility of curve
- Gait assessment
- Associated anomalies.

Investigations:[3,5]

X ray: gold standard investigation of choice for the initial assessment. Best time to do in a scoliosis with hemivertebrae is before 4yrs of age.Things to be seen in hemivertebrae with scoliosis are presence and spacing of pedicles, fused or absent ribs, type and location of hemivertebra, assess pedicle width and Cobbs angle.

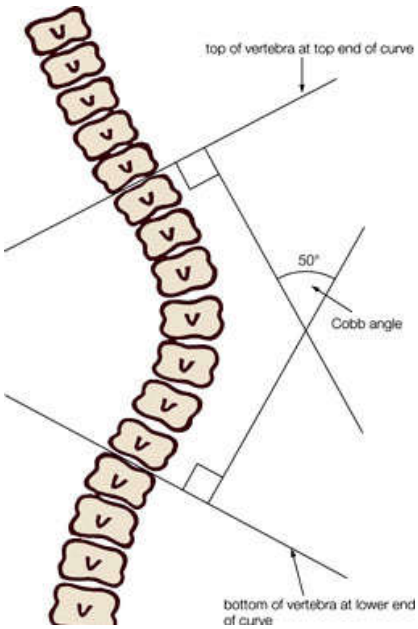


Fig. 5.

It remains standard for measuring curve magnitude, progression and assessing growth potential of vertebral anamoly. Cobb's method is used to evaluate for degree of scoliosis and for serial follow-ups.X ray is the early and basic investigation for monitoring the curve progression in a conservatively treated scoliosis in hemivertebra. Surgery is indicated if the curve is progressive and cobbs angle is > 40^{o}

CT scan: useful in complex scoliosis inhemivertebra

- Where visualization is difficult with x rays due to overlying structures. Eg: cervico thoracic junction.

- To know the type of spinal abnormalities
- To know the pedicle width for operative planning.
- To know the bone and soft tissue plane for dissection

MRI: Useful in identifyingintra-spinal anomalies eg: tethered cord, syringomyelia, spinal dysraphism, lipoma of cord. It is also useful in detecting associated anomalies eg: renal anomalies. Usually not needed unless the curve is progressive or a high grade curve associated with neurological deficits in patients whom surgical procedure is considered.

Treatment options:

- Serial observations
- Bracing until skeletal maturity
- Surgery for correction and fusion or stabilization of severe or progressive deformity.

Non-operative method[2,3]

Observation should be applied only for non-progressivecurves with balanced spine.

Indications for conservative management[2]

- Non-incarcerated or semi-incarcerated defects
- Non-progressive curves with rate of progression less than 2 degrees before puberty.
- Stable balanced spine with no neurological deficits.

Contra-indications for conservative treatment[2]

- Unilateral unsegmented bars
- Curves greater than 20 degrees in infancy
- Progressive curves
- Loss of sagittal balance with development of secondary curves
- Evolving neurological deficits or worsening of existing deficits.
- Significant pain and gross cosmetic deformity
- Management of hemivertebra is controversial as the progression of deformity is unpredictable and requires continuous evaluation.

Bracing: Most congenital scoliosis are not flexible and therefore are resistant to repair with bracing. For this reason bracing is effective in long flexible curves or for rebalancing the spine after surgery until fusion is solid.

Braces for treatment[3]

- The goal of brace treatment is to prevent or minimize progression of the deformity during longitudinal spine growth, but despite the improvement of curve magnitude in the brace, it is not anticipated to reduce scoliosis magnitude once the brace treatment is discontinued.
- As bracing is an attempt at deformity prevention by modulation of growth, it should not be offered to patients who are skeletally mature.
- In general, braces can be classified based on the area of the spine that they contact. A TLSO (thoraco-lumbar-sacral orthoses) spares the cervical spine while a CTLSO (cervical-thoracic-lumbar-sacral orthosis) includes the cervical spine.
- Currently, the most commonly prescribed braces are thermoplastic rigid TLSOs that are either “off the shelf” and can be modified with foam pads to optimize correction of the scoliosis in the brace (eg: the Boston brace), or the TLSO can be custom moulded to the patient as the patient is held in longitudinal traction to correct their scoliosis.
- An alternate to full time brace wear is the night time bending rigid TLSO brace, where the goal is for scoliosis overcorrection in the thermoplastic orthosis, and with resultant brace wear in the evening and during sleep where the dramatic trunk bend and curve correction would be tolerated by the patient.
- In order for a scoliosis brace to be effective, it must be worn. Compliance has been an important problem with every brace design. Patients may be advised to wear their rigid TLSO braces for more than 12 h per day.

Success of non-operative treatment depends on[3]

- Etiology for the deformity
- Close follow-up
- Monitoring for evolution of deformity
- Patient compliance with their treatment regimen

Operative treatment: In patients whose spinal deformities have not been controlled with non-surgical management, and where the scoliosis in hemivertebra is either relentlessly progressing or is over 40°, surgical intervention may be required.

Goals of surgery:

- To achieve straight spine
- Physiological sagittal profile while maintaining flexibility
- To arrest the progression of curve
- As short a segment fusion as possible preserving much of normal spine growth as possible.

Surgery for congenital scoliosis carries greater neurological injury risk than in idiopathic spinal deformities.

Early treatment helps in

- Minimizing the risks of surgery
- Allows better correction
- Prevent development of secondary structural and compensatory curves.

Pre-op evaluation

- Comprehensive history particularly pre-natal history
- Physical examination
- Neurological examination
- Signs of spinal dysraphism
- Spinal balance
- Rib cage deformities and pulmonary function tests.
- Cardiac and renal anomalies evaluation.
- Plain radiographs measuring curve magnitude
- CT (2D and 3D) to define anatomy.

- MRI: to evaluate for spinal dysraphism, tethered cord, lipoma of cord, syrinx and Chiari malformations

Surgical procedures

Posterior fusion[8]: Posterior fusion isdone for middle and lower thoracic, dorsolumbar junction and lumbar spine. Pediatric size implants should be used for age below 2 yrs.

Types-1) Only posterior fusion without hemivertebra excision-doesnotcorrect deformity but prevents further progression.
2) Posterior fusion with hemivertebra excision corrects deformity and outcome is better. Disadvantage- chances of nerve root injury especially in lumbar spine.

Procedure: Transpedicular eggshell hemivertebrectomy and removing periosteum is mandatory to prevent recurrence.

- Costotransversectomy in thoracic hemivertebra, subperiosteally making a plane by blunt dissection with finger prevents pleural and adjacent vessels injury.
- Compressionon convex side and distraction on concave side will give a better reduction of curve.Can be used in multilevel hemivertebra (upto 3 levels).

Advised in patients with less growth potential.

Disadvantages: If done in younger age <4 yrs, at thoracic level, will compromise thoracic cavity growth and hence pulmonary compromise : Crank shaft phenomenon

Combined anterior and posterior fusion[8]

Anterior approach can be done in cervical spine, cervico thoracic junction, upper thoracic and lumbar spine (L1-L4)

Advantages:

- More substantial correction
- Prevent crankshaft phenomenon and pseudoarthrodesis

Disadvantages:

- More extensive procedure
- Increase iatrogenic neurovascular injury

Single and dual growing rods:

In young patients, primary congenital curve can be treated with in situ fusion and correction can be maintained with growing rods until child attains skeletal maturity by increasing the rod length by monitoring appropriately at every 6 months.

Advantages:

- Can be offered at young age.
- Prevent thoracic insufficiency and crankshaft phenomenon

Disadvantages:

- Cannot be used in hemivertebra at cervicothoracic and dorsolumbarjunctions.
- Due to growth rod disproportionate expansion, the posterior construct may be damaged and pedicle screw cut through which might lead to nerve root injuries.

Expansion thoracoplasty and VEPTR[13]:

This procedure is used for scoliosis in hemivertebrawith fused ribs to prevent thoracic insufficiency syndrome in young age <8 yrs.

Mechanism: expanding the thorax by rib distractionon concave side achieves indirect correction of curve.

Convex epiphysiodesis:

Allows growth on concave side and arresting the growth on convex side.

Advantages: This procedure can be used in children <5 years of age with good growth potential

Disadvantage: Unpredictable concave side growth.

Intra op Neuro monitoring[8]:
Intraopneuro-monitoring is done to identify and prevent the development of neurologic deficit or worsening of pre-existing neurologic injury in patients who is undergoing surgery by monitoring SEP and MEP.

Complications of surgery:

- Amyotrophy
- Lower limb radiculopathy
- Sphincter disturbances
- Paraparesis of lower limbs
- Rod breakage and screws backout
- Restrictive lung diseases and pulmonary insufficiency

Drawbacks of early fusions:

- Patients with early thoracic fusions had significantly lower physical functioning and physical activity compared with healthy children.
- Lung growth was reduced in cases of early posterior spinal fusions leading to pulmonary compromise.
- Crank shafting was observed in 15 % of cases with large curves and early fusions.
- Continued low back pain with multilevel degeneration.
- Pseudoarthrosis requiring revision surgeries and bone grafting.
- Growth deficits at the end of growth after fusion of 5 vertebrae when surgery is performed at early age.
- Compressive forces on ring apophysis of growing vertebra leads to iatrogenic vertebral stenosis (**Heuter-Volkman Principle**-The rate of epiphyseal growth is affected by pressures applied to its axes: increased pressure inhibits growth; decreased pressure accelerates growth) leading to development of kyphosis.

Outcome of surgery[5]:

While hemivertebra resection has higher complication rate compared to hemiepiphysiodesis or instrumented fusion without resection, posterior hemivertebra resection in younger patients resulted in better correction than the other 2 techniques. Posterior hemivertebra resection with transpedicular instrumentation is a safe and effective procedure in young patients. Surgeries can achieve short fusions and more mobile segments. However complications associated

with implants and spinal growth still remain concern.Vertebral fusion will affect future growth but surgery at right age will result in longer spine. Addressing the hemivertebra with scoliosis at an early age will prevent deformity and pain in adulthood.

Prognosis[4,5]

The prognosis is directly related to presence or absence of associated anomalies. The prognosis of isolated hemivertebra is relatively good. If left untreated 25% will have no progression, 50% progress slowly and 25% progress rapidly during growth.The occurrence of hemivertebra in siblings is very uncommon. Occasional associations have been reported with neural tube defects in the range of 0-4%. There is increased incidence of neural tube defects in siblings with single hemivertebra as well as those with multiple vertebral defects. Therefore it would be reasonable to offer genetic counseling and prenatal diagnosis for neural tube defects to those patients with a previous child with vertebral anomalies.

Case 1

12 year old child with upper dorsal hemivertebrae with scoliosis of 25 degrees with convexity on right with segmented vertebra at the apex of scoliosis is managed by conservative method by serial observations with regular follow-up. The child does not have any neurological deficits with only mild cosmetic deformity. Sagittal balance well maintained and spine is well balanced with good flexibility. Her pulmonary function is well preserved. This will also help her to accommodate for skeletal growth and improve lung compliance and prevent restrictive lung pattern. Patient is being evaluated by serial radiographs till puberty.

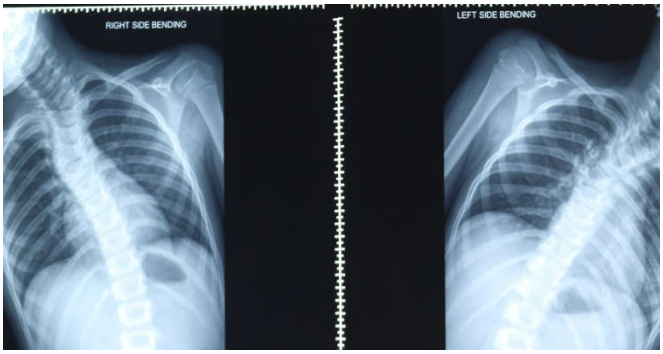
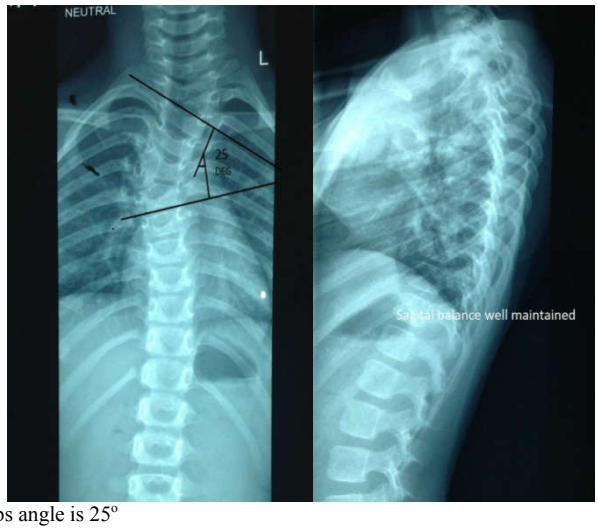


Fig 6.



Cobbs angle is 25°

Fig. 7.

Cobb's method was used to asses the deformity and degree of angulation. Serial radiographs showed increase in curvature not more than 1 degree per year.

Ct Scan Showing Wedge Shaped Segmented Lateral Hemivertebra

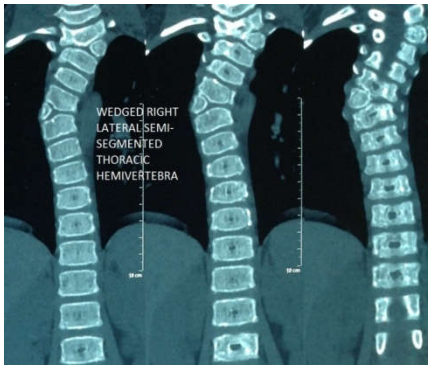


Fig. 8.

MRI images showing wedged hemivertebra with no cord compression, no syrinx formation and no tethering of cord. Hence the patient was managed conservatively.

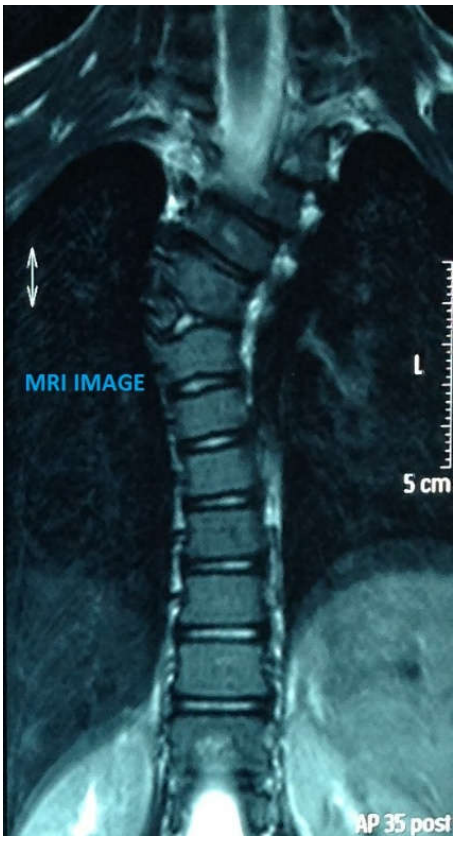


Fig. 9.

Case 2

23 year old male with lumbar scoliosis with L2 hemivertebra with no deficits and no pain. X-ray showed left lateral wedged incarcerated type of hemivertebra with Cobb's angle measuring 30 degrees. Patient was successively managed conservatively by serial imaging which showed progression of curve not more than 2 degrees per year .Sagittal balance was well maintained. Cord was ending at normal level with no tethering on MRI imaging. No significant cosmetic deformity. No significant change in gait pattern.

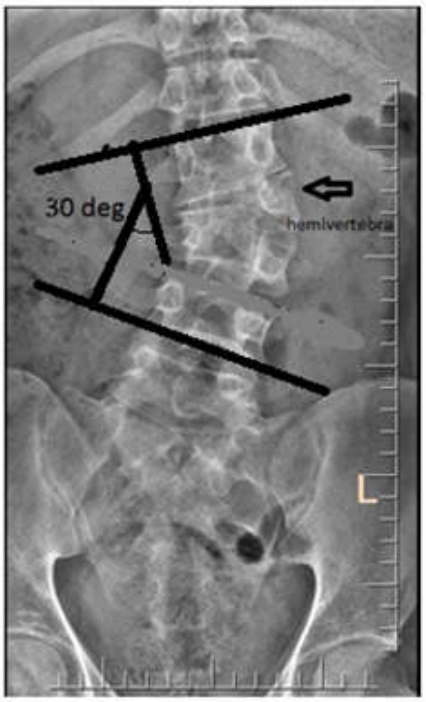


Fig. 10.

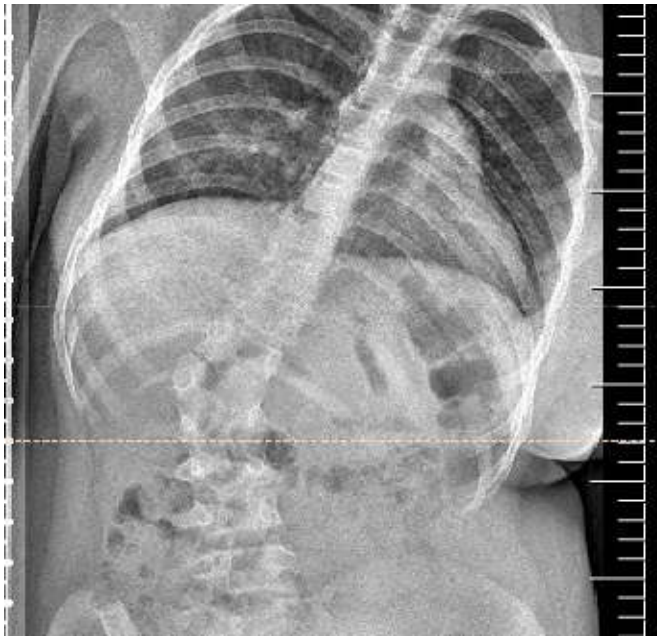
Fig. 11.

CASE 3

11 year old female patient with dorsolumbarkyposcoliosis with back pain since 1 year and increased since 4 months. Patient was serially followed up every 3 months with serial radiographs which showed progression of curvature with increasing deformity.

Fig 12.

There was progression of curvature at D12-L1 levels. Patient had right side radiculopathy with pelvic tilt and significant cosmetic deformity. There was L1 hemivertebra which was partially segmented with apex at the convexity curve. On lateral bending X-ray there was no significant correction indicating rigid spine. Since the deformity was at the junction where spine curvature changes from normal dorsal curve to ventral curve there was progressive rotational abnormality leading to significant kyphoscoliotic deformity.



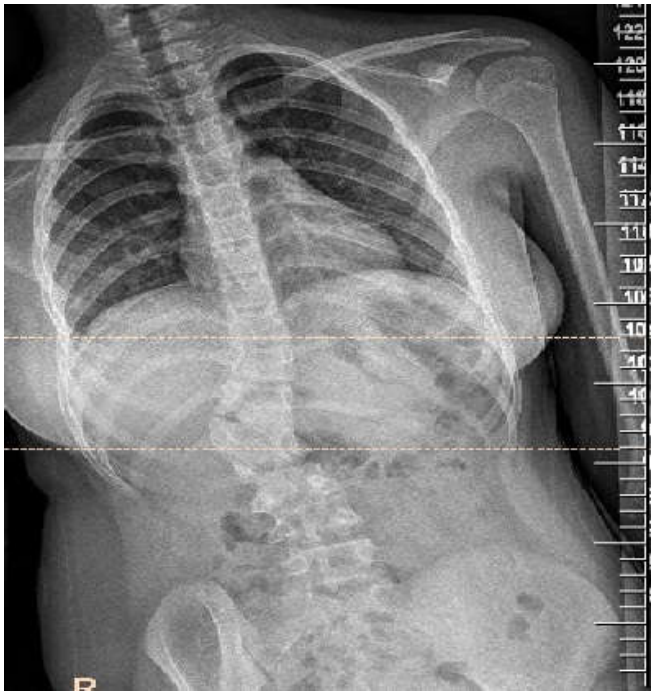


Fig. 13.

On 3D reconstruction CT images, there waspostero-lateral angulation on right side due to semi-segmented incarceratedL1 hemivertebra with significant rotational abnormality with convexity on right postero-lateral side.

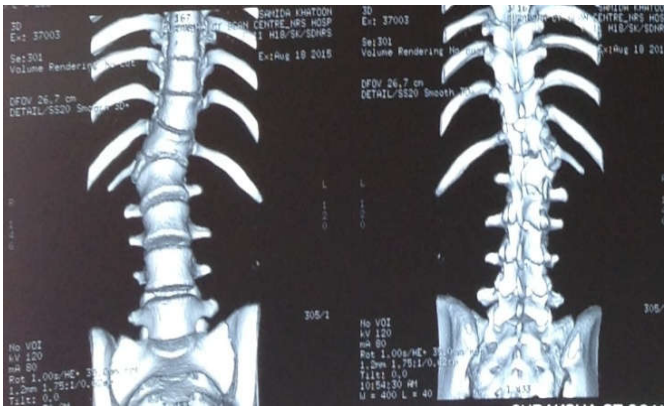


Fig.14.

On axial CT scan there was semi-semented right postero-lateral L1hemivertebra with absence of left sidebody,lamina and pedicle.

Fig. 15.

MRI images showing significant compression of exiting root on left side at the level of D12-L1 level.

On whole spine screening there was evolution of secondary curves in proximal segments with loss of sagittal balance.

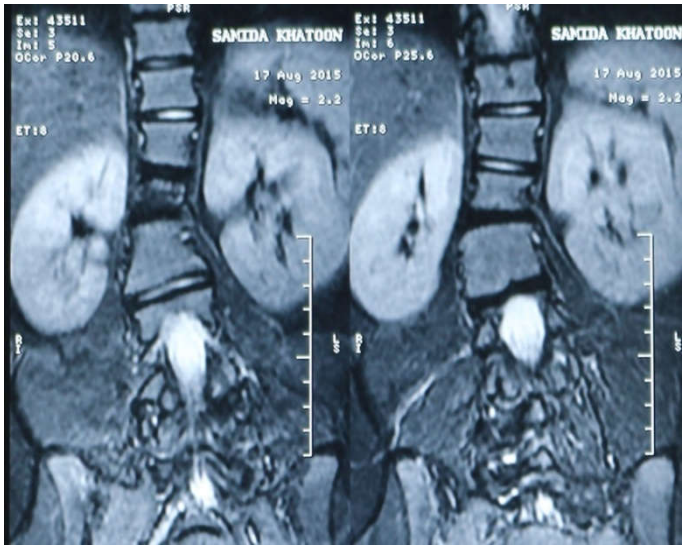


Fig. 16.

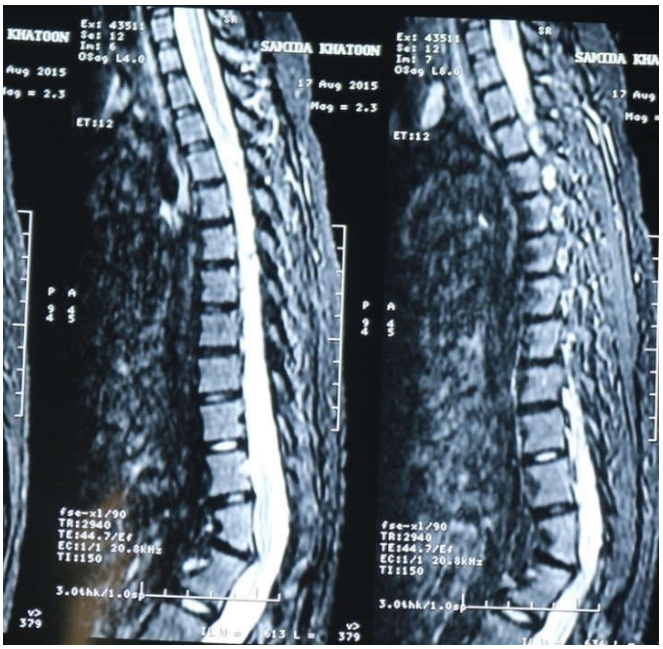


Fig. 17.

At the level of D12-L1 there was compression of cord with low lying cord with tethering of cord with focal syrinx formation indicating compression effect necessating surgery.With this rationale, patient was undertaken for surgery. Pre-op evaluation done and correction of kyphoscoliosis by excision of L1hemivertebra with pedicle screw rod fixation was planned. Intra-op neuro-monitoring done especially during correction of curvature to avoid neuro-vascular compromise.

Fig. 18.

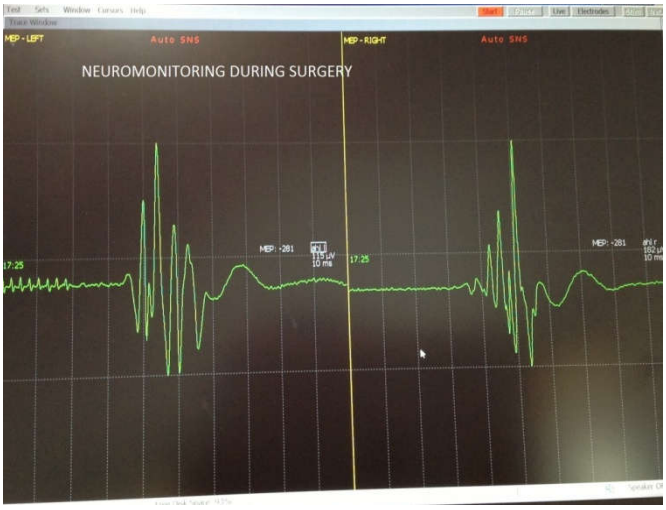


Fig. 19.

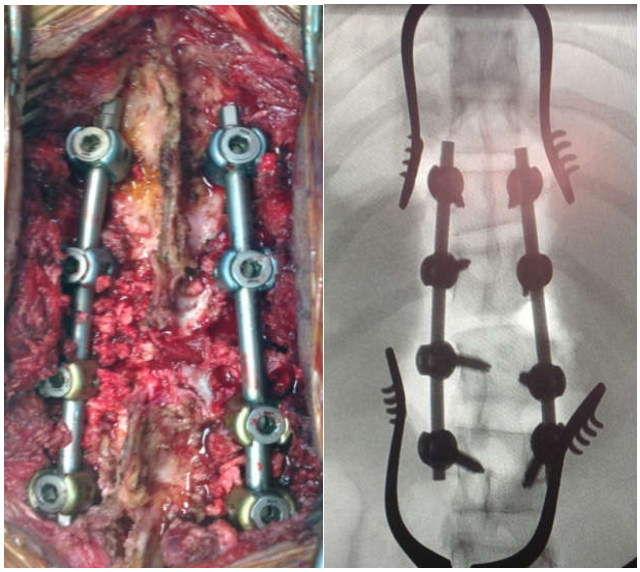


Fig. 20.

Immediate post op CT showed significant correction of kyphoscoliosis with perfect pedicle screw placement. There was significant reduction of radicular pain.

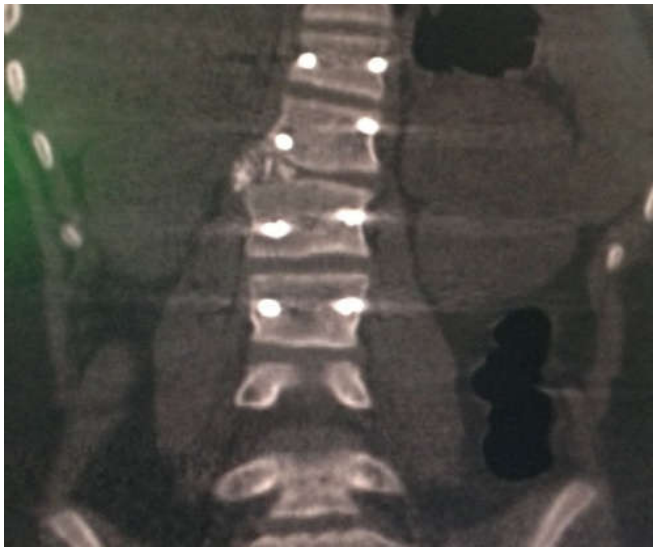


Fig. 20.

Post op Axial images showing L1 scolio-hemivertebrectomy with pedicle screws in situ. Patient was followed up every month which showed maintainance of alignment with nochange in sagittal and coronal balance. Patient was neurologically stable with no pain.

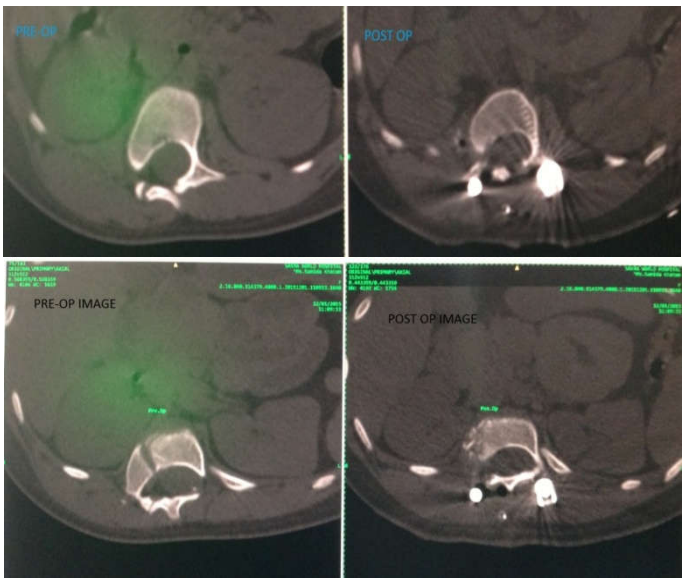


Fig. 21.

Follow-up X-ray at 3 months:

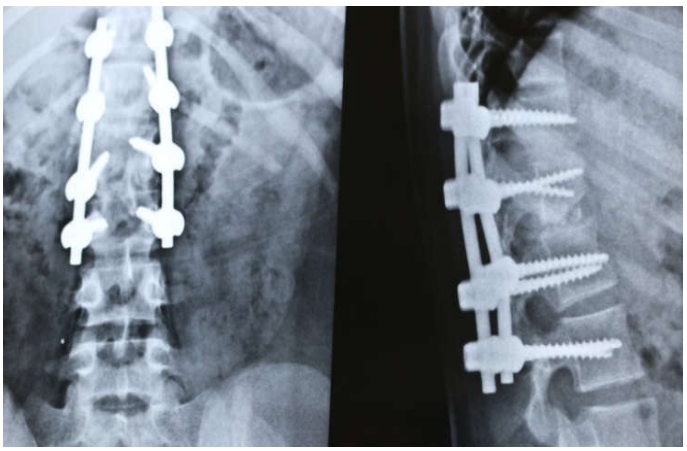
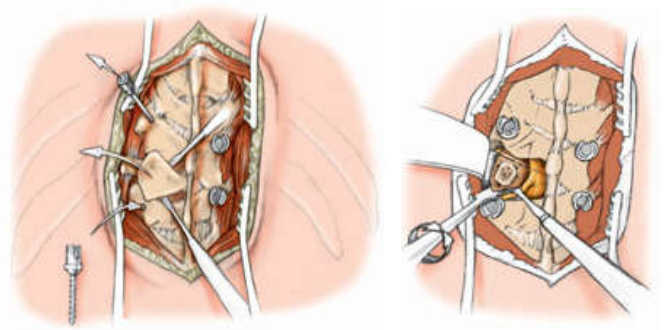
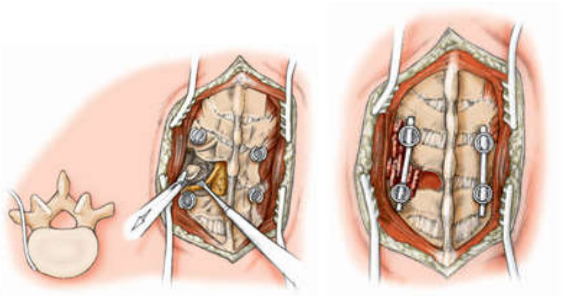


Fig. 22.

Steps of operative procedure of pedicle screw placement with excision of hemivertebra and correction of scoliosis:[13,8]





Courtesy: Prof. Dr. med. Jürgen Harms KlinikumKarlsbad-Langensteinbach ·Guttmannstrabe 1 · 76307 Karlsbad HARMSSPINE SURGERY.[13,8]

Fig. 23.

Conclusion

A pragmatic review of literature is very essential in deciding the line of management of these cases. Not all cases need surgery. Regular follow up and assessment of progression based on imaging is vital. Patience and perseverance on the part of both treating surgeon and the patients is of utmost importance to draw an optimum line of management. Undue surgery may hamper growing skeleton and alter the physiology. At the same time in cases with rapid progression, proper surgical measures to halt the progression is mandatory to avoid rigid severely deformed and compromised spine.

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