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RESEARCH ARTICLE

AN UNUSUAL CAUSE OF INFANTILE TORTICOLLIS

Suchitra Sivadas[1], Sajitha, S.,[2] Suhas Udayakumaran[3], Aravind, S.,[4] Gayatri Sajeevan[5] and Jayakumar, C.[6]

[1]Senior Resident Department of Paediatrics, Amrita Institute of Medical Sciences, Kochi, Kerala
[2]Professor, Department of Paediatrics, Amrita Institute of Medical Sciences, Kochi, Kerala
[3]Professor, Department of Neurosurgery, Amrita Institute of Medical Sciences, Kochi, Kerala
[4]Junior Resident, Department of Paediatrics, Amrita Institute of Medical Sciences, Kochi, Kerala
[5]Senior Resident, Department of Paediatrics, Amrita Institute of Medical Sciences, Kochi, Kerala
[6]Professor, Department of Paediatrics, Amrita Institute of Medical Sciences, Kochi, Kerala

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ABSTRACT

Torticollis is a clinical symptom and sign characterized by a lateral head tilt and chin rotation toward the side opposite to the tilt. Torticollis in infants can be due to congenital and acquired causes. Congenital muscular torticollis is the commonest cause detected in infants. Acquired causes range from ocular causes, retropharyngeal infections and cervical spinal cord tumors.Benign and malignant neoplasms of the upper cervical spine are rare causes of torticollis in children.We present here a 10 month old infant presenting with loss of acquired gross motor developmental milestones, neck flop and irritability since past 4 months .Clinical examination revealed torticollis to the right along with weakness of right upper limb. Imaging -MRI Spine revealed expansile intramedullary tumour extending along the cervicomedullary junction till C7 vertebral level .Surgical excision was done and histopathology revealed features of a cervical cord astrocytoma. Child is currently recovering power with physiotherapy and is on follow up.

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INTRODUCTION

Torticollis is a clinical symptom and sign characterized by a lateral head tilt and chin rotation toward the side opposite to the tilt. Many conditions cause torticollis. The differential diagnosis is different for infants than for children and adolescents (Herman, 2006). Congenital muscular torticollis associated with a contracture of the sternocleidomastoid muscle is the most common etiology of torticollis in infants.Other congenital causes include birth trauma, cervical spine abnormalities: hemivertebra, C1 C2 subluxation, spina bifida, uterine tumours. Unusual nonmuscular causes of torticollis in the infant also must be considered and include ocular torticollis caused by eye muscle weakness, Sandifer's syndrome resulting from gastroesophageal reflux, neural axis abnormalities, and benign paroxysmal torticollis. Retropharyngeal abscesses and pyogenic cervical spondylitis are unusual infectious causes of torticollis (Visudhiphan, 1982). Intermittent torticollis associated with headaches, vomiting, or neurologic symptoms may be caused by tumors of the posterior fossa. Benign and malignant neoplasms of the upper cervical spine are rare causes of torticollis in children. We present here a 10 month old infant presenting with torticollis and developmental regression who was found to have a rare cause for the same.

Corresponding author: **Suchitra Sivadas,**
Senior Resident Department of Paediatrics, Amrita Institute of Medical Sciences, Kochi, Kerala.

Case Report

A 10 month old female infant born of non consanguineous parentage, apparently asymptomatic till 6 months of age, was brought with loss of acquired gross motor developmental milestones, neck flop and irritability since past 4 months. This baby who had an

uneventful antenatal and birth history was born term by normal vaginal delivery with a birth weight of 2.9 kg. She was apparently asymptomatic till 6 months of age and had acquired the age appropriate developmental milestones till then. Subsequently she was noted to have a slowly progressive loss of head control and inability to turn over. She had a weak grasp using her right hand. Social and linguistic domains of development were preserved.

There was no history of birth injury, trauma, seizures, projectile vomiting, vision or hearing abnormalities, abnormal odour, exaggerated startle response, administration of medications, fever or recent vaccination. She had been to the paediatric outpatient clinic of multiple hospitals, was evaluated and advised physiotherapy, with no relief of symptoms. Clinical examination revealed an alert, irritable baby, with torticollis to the right. Weight ,length and US L S ratios were normal. Neurological examination revealed torticollis to the right, hypotonia and weakness of right upperlimb, both proximal and distal, with normal power of left upper limb and both lower limbs. Reflexes were not elicitable over the right upper limb,but were brisk in left upper limb and both lower limbs.

There were no cerebellar signs or signs of meningeal irritation. Skull, spine, vision and hearing were normal. Rest of the systems examination was normal. There was no organomegaly. Fundus examination was normal. Baseline blood investigations were normal. In view of the torticollis to the right side, with loss of acquired gross motor milestones and right upperlimb monoplegia, an MRI brain with cervical spine screening was done. Sagittal T2 weighed sequences on MRI showed an expansile intramedullary tumour extending along the cervicomedullaryjunction till C7 vertebral level (Figure 1,2) with a cystic component superiorly showing post contrast enhancement in the periphery, with no bleed or calcification and causing early obstructive hydrocephalus.

The possibility of a cervical cord astrocytoma was considered most likely. An urgent neurosurgery consultation was availed and a staged excision of the tumour was done. The histopathology showed astrocytes in a gliofibrillary background with focal multicystic areas with no mitosis or nuclear atypia consistent with Pilocytic astrocytoma (Figure 3). Following the surgery, the child developed secondary pseudomeningocoele which was diagnosed when she presented with fever and vomiting 2 weeks after the procedure. VP shunting was performed followed by an open sub occipital craniotomy and near total excision of the tumour.1 year after the surgery, the child is on physiotherapy and is doing well. She has no torticollis, her limb is gaining power and she has reacquired her age appropriate developmental milestones.

DISCUSSION

There are many causes of torticollis in infants. The most common is Congenital muscular torticollis. Acquired causes of torticollis include musculoskeletal like muscle spasm, infections including head and neck, spine infections including osteomyelitis, discitis, epidural abscess, CNS infections like meningitis, atlantoaxial rotatory fixation, trauma and ligamentous laxity (e.g. as part of underlying disorders)post head/neck surgery, Grisel syndrome, inflammatory causes like Juvenile idiopathic arthritis, neoplastic conditions including cervical cord tumours and bone tumours and also dystonic syndromes (idiopathic spasmodic torticollis, drug reactions) (Kumandaş, 2006).

Slowly evolving torticollis, without signs of infection, associated with motor weakness, should alert one to the rare possibility of spinal cord tumours. Isolated regression of gross motor milestones with preserved cognition, vision, speech and hearing without organomegaly goes against neurodegenerative white and grey matter diseases. The loss of motor milestones as in this case may be due to the weakness.

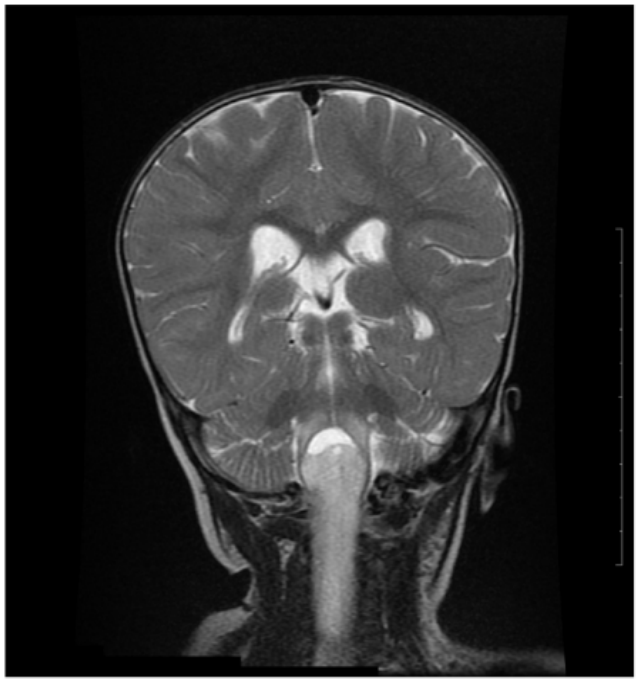

Figure 1- T2 Coronal MRI Cervical spine showing cervical cord intramedullary lesion extending from cervicomedullary junction down to c7 vertebral level

The most frequent spinal cord tumors are the pilocytic and anaplastic astrocytomas (60%) and ependymomas (30%). Spinal astrocytomas are the most common spinal cord tumors in children. Their primary location is more rostral than in adults; 50% are in the cervico-thoracic region and usually affect a small number of segments.

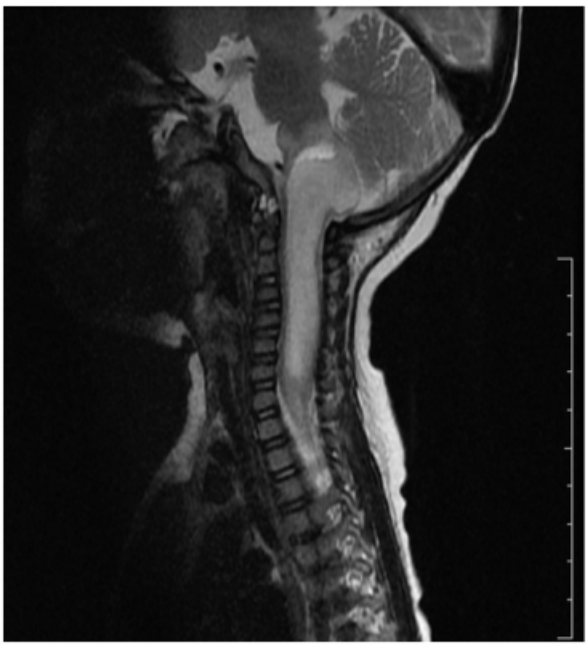

Figure 2- T2 Sagittal MRI Cervical spine showing intramedullary cervical cord tumor

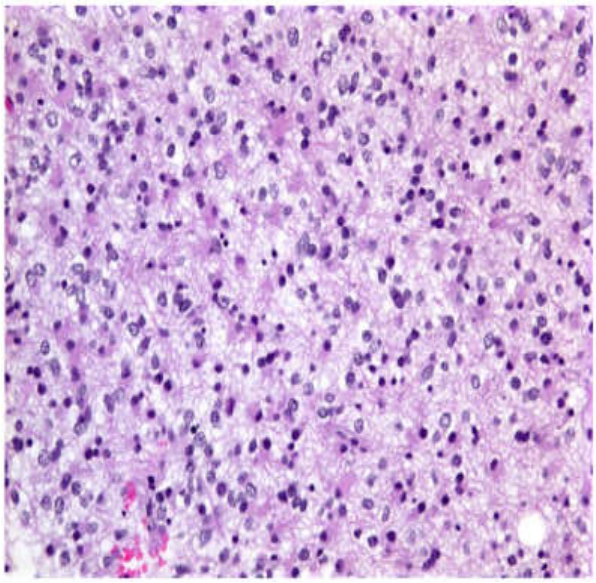

Figure 3- Histopathology showing spindle-shaped cells with fiber like processes (astrocytes)in gliofibrillary background with focal microcystic areas,no mitosis/necrosis/nuclear atypia-consistent with pilocytic astrocytoma

Astrocytomas most frequently present with pain and motor dysfunction, followed by gait disturbance, torticollis and scoliosis. Bowel and bladder dysfunction is uncommon due to their predominant cervical location. The majority are of low grade, WHO grade I and II (75–80%) including pilocytic and fibrillary type astrocytomas[5].On imaging the lesions are eccentric within the spinal cord and may show an asymmetric spinal cord expansion.

Lesions are typically T2-hyperintense, T1-iso- or hypointense and may show a mild to moderate contrast enhancement. On follow-up imaging, low grade spinal cord astrocytomas are typically stable or may show a slow progression. Surgery to remove all of a tumour is a likely first step.

The exception is gliomas in areas where surgery can be too risky. Surgery may be enough to cure grade 1 tumours. Surgery usually doesn't remove all of a higher-grade tumour. Radiation often follows in case parts of a tumour could not be removed. Chemotherapy is often used for glioblastoma and anaplastic astrocytoma. It can be used before or after radiation.

Conclusion

We aim to highlight two key points from this report. The first is that cervical cord tumours should be considered as a differential for torticollis in infants and children, especially in the presence of other clinical signs like neck and upper limb weakness.

Secondly developmental regression in a young child need not always be secondary to neurodegenerative disorders. Structural causes should also be considered in the same.

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Key Points

- There are many congenital and acquired causes of torticollis in infants
- Cervical cord tumors should be considered as one of the causes of infantile torticollis especially if associated with focal limb weakness.
- Structural causes should be considered in infants with a recent history of weakness and motor regression

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