



The Spectrum of Underlying Diseases in Children with Torticollis

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ABSTRACT

AIM: To investigate the underlying conditions in children with torticollis.

MATERIAL and METHODS: Between May 2016 and December 2019, 24 patients (10 girls and 14 boys; mean age, 8 years) presenting with twisted neck, neck pain, weakness of extremities, imbalance, and gait disorder were evaluated retrospectively.

RESULTS: Five of the patients had cranial pathologies (cerebellar anaplastic ependymoma and medulloblastoma, brain stem glioma, atypical teratoid rhabdoid tumor, and acute disseminated encephalomyelitis), and five of the patients had spinal pathologies (idiopathic intervertebral disc calcification, vertebral hemangiomas, compression fracture, multiple hereditary exostoses, and Langerhans cell histiocytosis at C4). Six of the patients had ocular pathologies (strabismus, Duane syndrome, and Brown syndrome each in two patients). Four patients had otorhinolaryngological infections (Sandifer syndrome, esophageal atresia, reflux, and spasmodic torticollis, with one patient each). Detailed clinical physical examination and necessary laboratory investigation were performed for all patients.

CONCLUSION: Torticollis is a sign that is not always innocent and may herald an underlying severe disease. Misdiagnosis can lead to wrong and unnecessary surgical procedures and treatments, and sometimes, the results can be damaging due to underlying severe conditions if diagnosed late. In addition, we first report a case of vertebral hemangiomas and temporomandibular joint ankylosis that presented with torticollis in the English medical literature.

KEYWORDS: Etiology, Physical examination, Radiology, Torticollis

ABBREVIATIONS: **ADEM:** Acute disseminated encephalomyelitis, **ATRT:** Atypical teratoid rhabdoid tumor, **CMV:** Cytomegalovirus, **CNS:** Central nervous system, **CRP:** C-reactive protein, **CT:** Computed tomography, **EBV:** Epstein-Barr virus, **EEG:** Electroencephalography, **ESR:** Erythrocyte sedimentation rate, **IVDC:** Idiopathic intervertebral disc calcification, **MHE:** Multiple hereditary exostoses, **MRI:** Magnetic resonance imaging

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INTRODUCTION

Torticollis, described as any asymmetrical posturing of the head and neck, is one of the most common congenital musculoskeletal abnormalities in infants. It is commonly called “wryneck,” an abnormal posturing of the head and neck that is generally caused by contraction of the sternocleidomastoid muscle due to birth trauma. Acquired torticollis can originate from many different benign or serious pathophysiologic processes. In the literature, underlying conditions causing torticollis are well described. Different etiologies of torticollis include congenital muscular torticollis, osseous pathologies, central nervous system/peripheral nervous system pathologies, increased intracranial pressure, ocular pathologies, nonmuscular soft tissue, benign paroxysmal torticollis, Sandifer syndrome, drug-induced torticollis, and conversion disorder (4,21,26,27).

Therefore, a better understanding of the spectrum of underlying diseases in torticollis is essential in making a correct diagnosis to be able to select the appropriate treatment method (6,9).

MATERIAL and METHODS

We present children whose conditions can be associated with head posture abnormalities. We aimed to investigate the spectrum of underlying disease of the children presenting with torticollis as an initial or main symptom. Between May 2016 and December 2019, we evaluated the medical records of 24 patients (10 girls and 14 boys; mean age, 8 years; range, 4.5 months–17 years) presenting with twisted neck, neck pain, weakness of extremities, and imbalance and gait disorder. The study excluded cases of congenital muscular torticollis.

A detailed clinical physical examination and necessary laboratory investigation were performed for all patients. Magnetic resonance imaging (MRI), cranial and/or spinal computed tomography (CT) imaging studies, and sternocleidomastoid ultrasound were performed to determine the etiology and surgical technique for those requiring surgeries.

The Ethics Committee of Erciyes University approved the review of all records (Date: 06.02.2019 and Number: 2019/84).

RESULTS

Five of the patients had cranial pathologies (cerebellar anaplastic ependymoma and medulloblastoma, brain stem glioma, atypical teratoid rhabdoid tumor [ATRT], and acute disseminated encephalomyelitis [ADEM]), and five of the patients had spinal pathologies (idiopathic intervertebral disc calcification [IVDC], vertebral hemangiomas, compression fracture, hereditary multiple exostoses [MHE], and Langerhans cell histiocytosis at C4). Six of the patients had ocular pathologies (strabismus, Duane syndrome, and Brown syndrome, with two patients each). Four patients had otorhinolaryngological situations (Sandifer syndrome, esophageal atresia, reflux, and spasmus nutans, with one patient each). Eleven patients underwent surgery, and two of them also received medical therapy; five patients received only medical therapy; three patients were treated

with physiotherapy, and one of them also received medical therapy; the patient with compression fracture was treated conservatively. The guardian of the patient with brain stem glioma refused treatment. The rest of the patients were regularly observed without any intervention. Table I shows detailed information, including clinical and radiological findings, diagnosis, and treatment.

Presentations of Some Selected Interesting Patients in Detail

Patient 3: A 16-year-old girl was admitted because of headache, neck stiffness, and twisted neck to the right. Complete blood count and biochemistry tests were normal. MRI showed non-specific focal signal changes in the bilateral periventricular white matter and centrum semiovale. The right part of the C1 vertebra was defective, and a 14 × 7-mm osseous lesion compatible with osteochondroma arising from the right lamina of the C2 was detected (Figure 1). The lesion was narrowing the spinal canal and compressing the spinal cord. The CT showed multiple osteochondromatosis lesions in the bilateral hipbone, bilateral thighbone, C2 vertebra, and two left costae. Lower abdomen CT revealed MHE. Laminectomy and mass resection were performed, and the histopathological diagnosis was MHE.

Patient 6: A 1.5-year-old boy was admitted because of inability to walk. He had cough, fever, and lethargy that started 10 days before admission. He was diagnosed with a viral infection. Symptomatic treatment (ibuprofen, pseudoephedrine with chlorpheniramine) was started in another medical center. Two days after treatment, bilateral strabismus, dystonia, vomiting, and syncope occurred, which was evaluated as seizure. The patient was referred to the authors' university clinic. Physical examination was significant for decreased strength of the lower extremity (4/5). Electroencephalography (EEG)

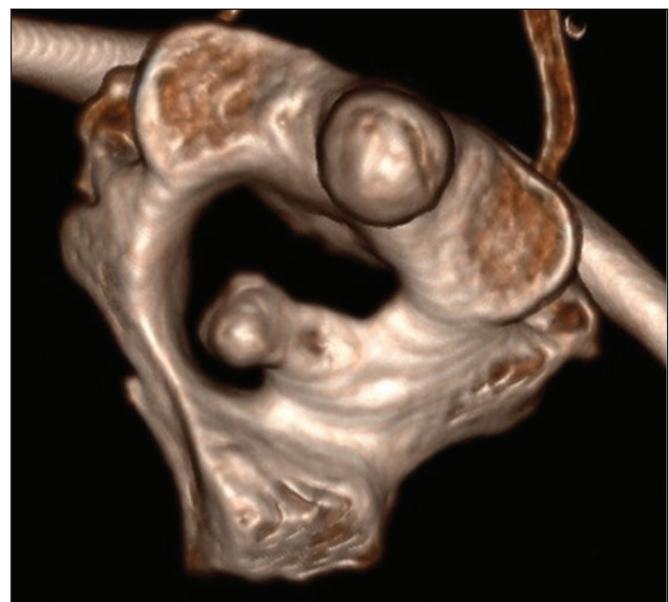


Figure 1: An osteochondroma arising from the left lamina of the C2, narrowing the spinal canal in 3D reconstruction of the CT image.

Table 1: Imaging Findings, Clinical Details and the Result of 24 Patients with Torticollis

Case No	Age / gender	Initial symptoms	Neurologic /systemic examination findings	Imaging findings	Diagnosis	Treatment
1	2 yo/boy	Twisted neck	nausea, vomiting, irritability, eyes shift to one side and walking disturbance	MRI: A mass compatible for medulloblastoma in infratentorial area, which caused a hydrocephaly	Medulloblastoma	Surgical
2	8 yo/boy	Twisted neck	Torticollis	Bilateral palatine tonsils were hypertrophic, bilateral cervical and parapharyngeal sub-centimetric benign reactive lymph nodes	Acute upper respiratory tract infection and related lymphadenopathy	Medical
3	16 yo/girl	Headache, neck stiffness, and twisted neck	headache, neck stiffness, and torticollis to right side, no neurological deficits	CT: C1 vertebral defective, osseous lesion compatible with osteochondroma, multiple osteochondromas in bilateral hipbone, bilateral femoral, C2 vertebral and posterior part of left 2 costae and osteoporosis	Multiple hereditary exocytosis	Surgical
4	17 yo/boy	Twisted neck	dizziness while walking and headache	Maxillary, sphenoid, and ethmoid sinusitis, psychometric test found to be compatible with age nine and IQ was 64	Pansinusitis and mental retardation	Medical and Physical therapy
5	9 mo/girl	Twisted neck to the left	Torticollis, strabismus in the right eye, reflux disease. History of esophageal atresia operation and jugular vein cannulation		Esophageal atresia	Physiotherapy
6	18 mo/boy	Incapability to walk, vomiting, seizure, twisted neck	torticollis, decreased strength of lower extremity (4/5)	MRI was compatible with post infectious encephalitis.	ADEM	Medical
7	3 yo/girl	Twisted neck to the right side, neck pain	Torticollis	A mass lesion in the posterior fossa	Anaplastic ependymoma	Surgical and medical
8	6 mo/girl	Twisted neck	Torticollis	Gastroesophageal reflux,	Sandifer Syndrome and gastroesophageal reflux	Medical
9	4.5 mo/boy	Twisted neck to the left	Torticollis, wide fontanelles and head circumference 90-97 percentile and no head control. It is also noticed that he had a twitch on the right hand and right foot	His thyroid function tests and B12 level were normal. His craniocervical MRI and neck USG, EEG, ophthalmologic evaluation, CBC, metabolic tests and electrolytes were normal.	Spasmus nutans	Discharged
10	6 yo/boy	Fever, sore throat, fatigue, vomiting and lump on left side of neck, twisted neck	Hyperemic tonsils and oropharynx, torticollis, left axillar and left cervical lymphadenopathy	WBC: 31,950 and neutrophil predominance, CRP:101 mg/dl and ESR:32 mm/h	Upper respiratory tract infection and lymphadenopathy	Medical

Table 1: Cont.

Case No	Age / gender	Initial symptoms	Neurologic /systemic examination findings	Imaging findings	Diagnosis	Treatment
11	17 yo/boy	Neck pain and twisted neck to the right side	Restricted jaw movements, history of operation for torticollis and growth hormone treatment and bleeding hemangioma in the right side of neck	MRI: wide vertebral hemangiomatosis, Neck USG: chronic fatty atrophic-fibrotic changes in the operation area CT:temporomandibular joint ankylosis	Vertebral hemangiomatosis, temporomandibular joint ankylosis	Physical therapy
12	5.5 yo/boy	Left eye gliding and right torticollis	Torticollis, strabismus	MRI: mastoiditis	Mastoiditis, strabismus	Surgical and medical
13	11 mo/girl	Strabismus and twisted neck	Torticollis, left gaze restriction to the lateral side	Radiology and laboratory tests were normal	Strabismus	Surgical
14	7 yo/boy	Twisted neck back pain	Physical examination was normal except torticollis	Thoracic 2-4 disc calcifications	Idiopathic intervertebral disc calcification	Conservative
15	7.5 yo/girl	Sliding eyes and Twisted neck	Torticollis, strabismus	Not done	Brown syndrome	Surgical
16	8.5 mo/boy	Twisted neck	Facial asymmetry and left endophthalmitis, Torticollis	Not done	Duane syndrome type 1	Surgical
17	9 yo/girl	Twisted neck, strabismus	Torticollis	Not done	Duane syndrome type 1	Surgical
18	12 yo/boy	Twisted neck, vomiting	Torticollis	Not done	Brown syndrome	Surgical
19	16 yo/boy	Twisted neck, hand pain, and restricted neck movement	Head rotation to the right side and lateral extension are restricted and L1-2 level tenderness with palpation	CT: C3, T1 and T2 mild depression compatible with a compression fracture	Compression fracture	Conservative
20	15 yo/boy	Twisted neck, neck pain	Normal findings except torticollis	CT showed bone destructive tumor involvement in C4	Langerhans cell histiocytosis	Surgical
21	5 yo/girl	Painful head rotation to both side and fever	Physical examination was normal except Torticollis and painful head rotation	Acute phase reactants were positive (CRP: 93 mg/dl, ESR: 91 mm/h) and leukocytosis was present, MRI showed a right peritonsillar abscess	Retropharyngeal abscess	Medical
22	15 yo/girl	Neck deviation to the right side and walking difficulty	Physical examination was normal except torticollis	3x3x3 cm in size cerebellar mass	ATRT	Surgical
23	10 yo/girl	Twisted neck walking, difficulty,	Brainstem glioma	Brainstem glioma	Brainstem glioma	Parents refused the treatment
24	7 yo/boy	Twisted neck to the right	Lymphadenopathy at the left neck and the difficulty of turning his head to the left	Idiopathic disk calcification	Idiopathic disk calcification	

ADEM: Acute Disseminated Encephalomyelitis, **ATRT:** Atypical Teratoid/Rhabdoid Tumor, **CRP:** C reactive protein, **ESR:** Erythrocyte sedimentation rate, **mo:** month-old, **yo:** year-old.

and ophthalmologic examinations were normal. Patient's serology for mycoplasma, rubella, cytomegalovirus (CMV), Epstein-Barr virus (EBV), adenovirus, parvovirus, and varicella were negative. MRI was compatible with postinfectious encephalitis (Figure 2A-D). The patient was diagnosed with ADEM. He received pulse methylprednisolone treatment, and he recovered. The patient's continuous control examinations were normal.

Patient 11: A 17-year-old boy was admitted because of neck pain and torticollis. He could rotate his head only 45° to the right and no constraint to the left. MRI showed multilevel vertebral hemangiomas (Figure 3A-D). An otorhinolaryngologist and neurosurgeons evaluated the patient. We assumed that the torticollis occurred because of the pain. He was treated conservatively. After administration of analgesic and anti-inflammatory drugs and bed rest, the complaints of the patient completely resolved.

Patient 14: A 7-year-old male patient was admitted to the pediatric neurology department because of back pain and torticollis. On physical examination, an approximately 2 cm lymphadenopathy at the left neck and difficulty of turning his head to the left were noted. MRI showed lytic lesions in three vertebrae. CT showed IVDC (Figure 4A-C). The patient

responded clinically to oral ibuprofen treatment. The presented case highlighted that clinicians must be vigilant about the exaggerated findings of IVDC on MRI.

Patient 21: A 5-year-old girl was admitted because of painful head rotation, irritability, and high-grade fever. Acute phase reactants were positive, and leukocytosis was present (C-reactive protein (CRP): 93 mg/dl, erythrocyte sedimentation rate: 91 mm/h). The patients used two different oral antibiotics but did not improve symptoms. She was hospitalized and evaluated for deep neck infection. Neck CT and MRI showed right peritonsillar abscess (11×8 mm) (Figure 5A, B). Empiric combination of antibiotic therapy (cefotaxime and clindamycin) was started after pediatric infectious disease consultation. She was also consulted with an otorhinolaryngologist who did not recommend surgery due to clinical improvement with antibiotic treatment. The patient's complaints regressed, and neck became supple after the 10th day of combination antibiotic treatment. After completion of antibiotic treatment, the patient's clinical control examination was normal.

Patient 22: A 15-month-old boy was admitted because of neck deviation to the right and walking difficulty. His physical examination was normal, except for torticollis. Cranial MRI revealed a 3×3×3 cm cerebellar mass (Figure 6A, B). The tumor

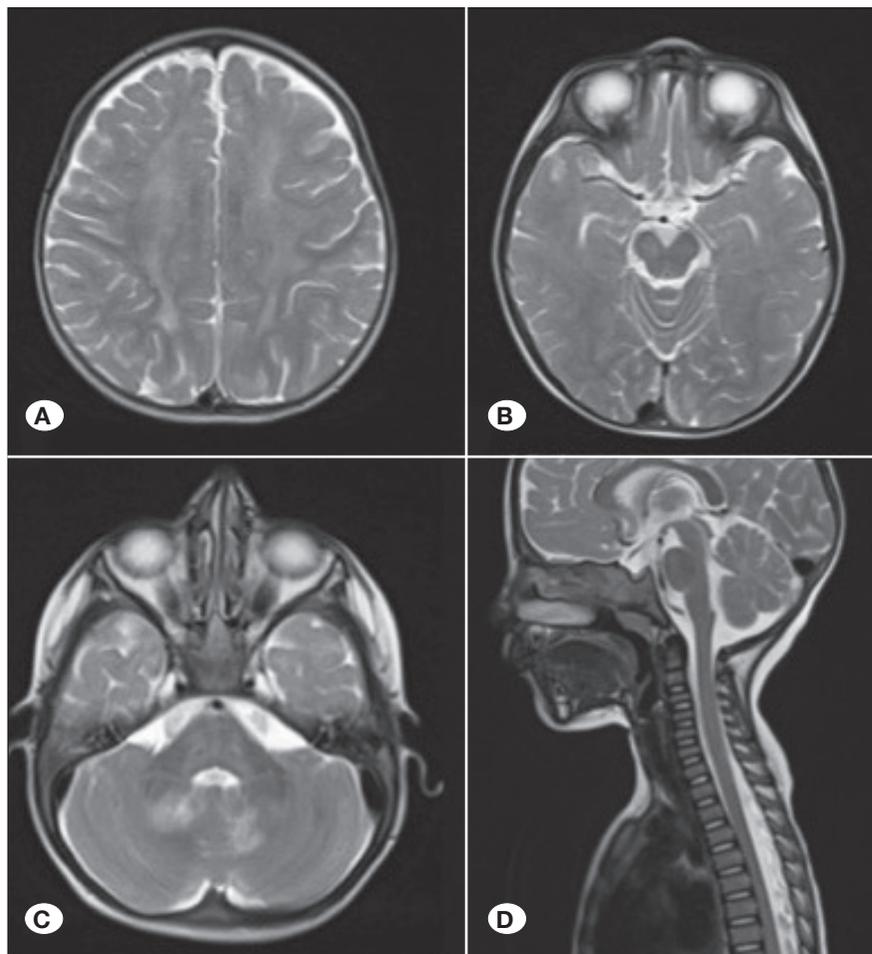


Figure 2: T2-weighted axial consecutive images show bilateral hyperintensities within the frontoparietal white matter (A), right cerebral pedicle (B), and dentate nuclei (C). Sagittal T2-W image shows the involvement of the cervical spinal cord (D).

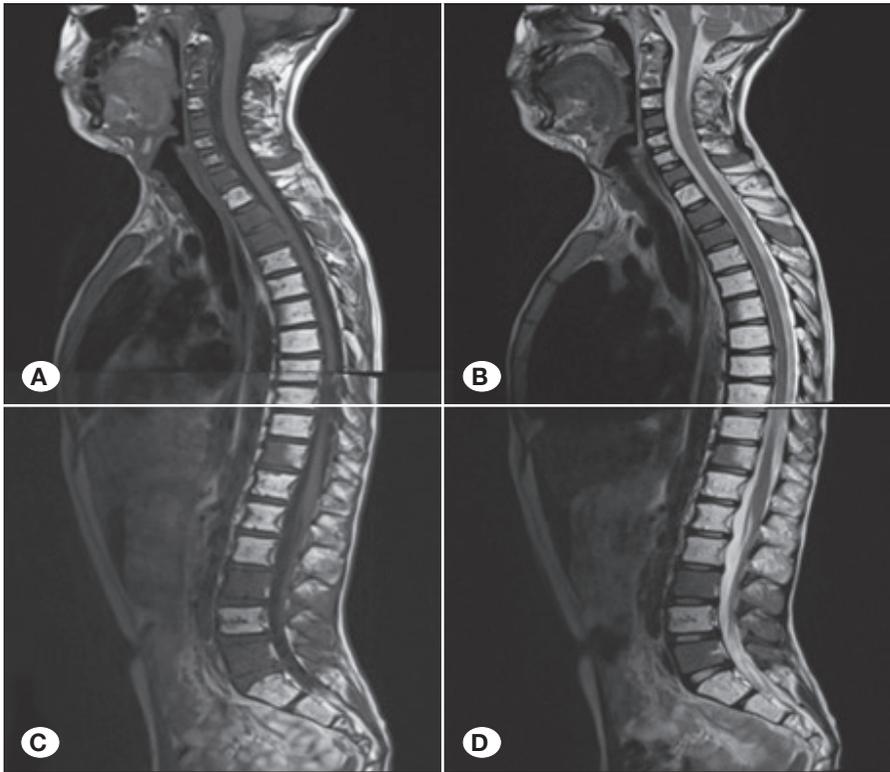


Figure 3: Consecutive sagittal T1- (A,C) and T2-weighted (B,D) images show vertebral hemangiomatosis.

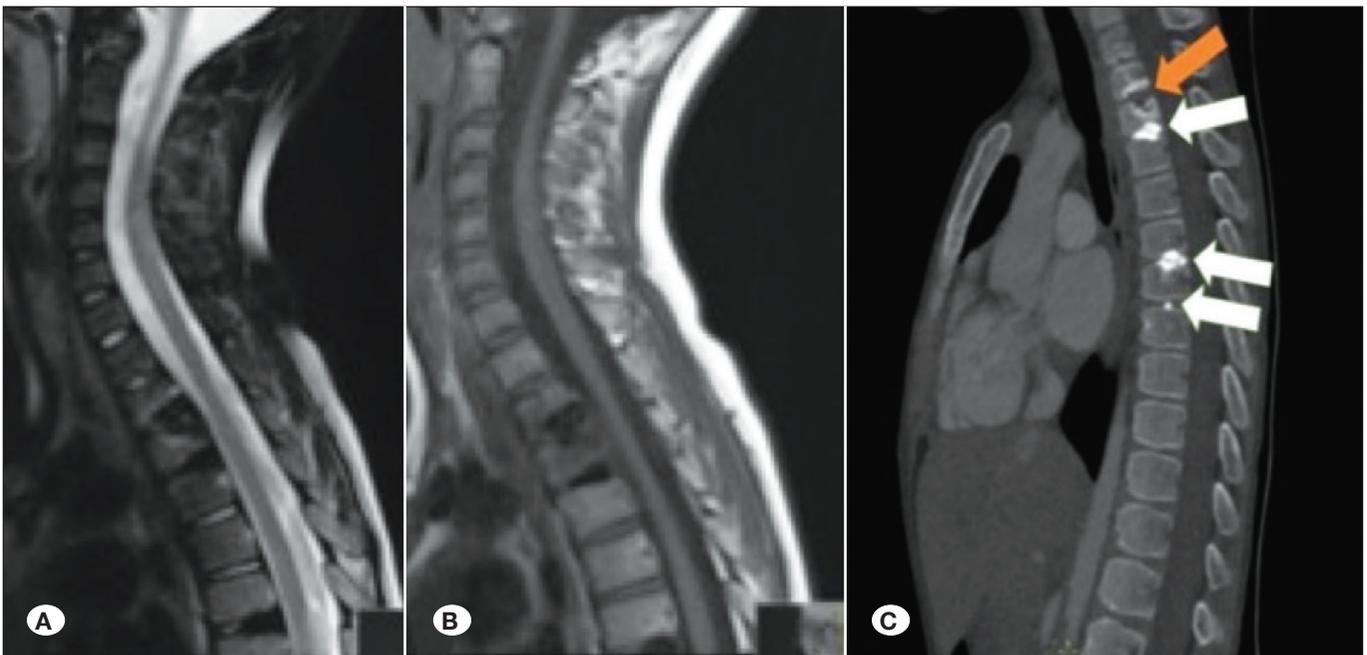


Figure 4: T1- and T2-weighted imaging show hypointensity at T2-3, T3-4, and T6-7 intervertebral disc spaces compatible with calcification. Degenerative changes are also seen at the surfaces of the vertebral body near the disc calcification (A, B). Sagittal reformat images of the thoracic vertebrae CT show multilevel intervertebral disc calcifications (white arrows) with loss of vertebral body height due to degenerative Schmorl nodule (orange arrow) (C).

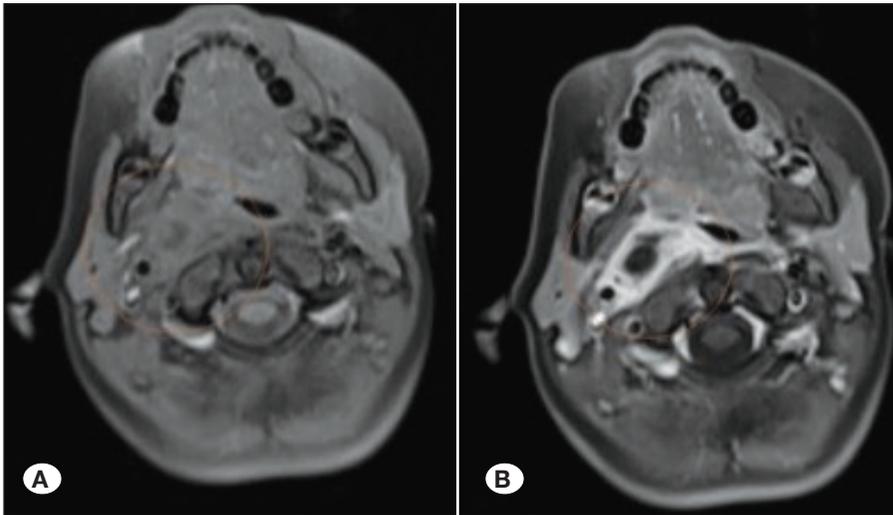


Figure 5: Pre- (A) and postcontrast (B) axial fat-saturated T1-weighted images through the cervical region show right parapharyngeal abscess formation and inflammation (circles) of the soft tissues.

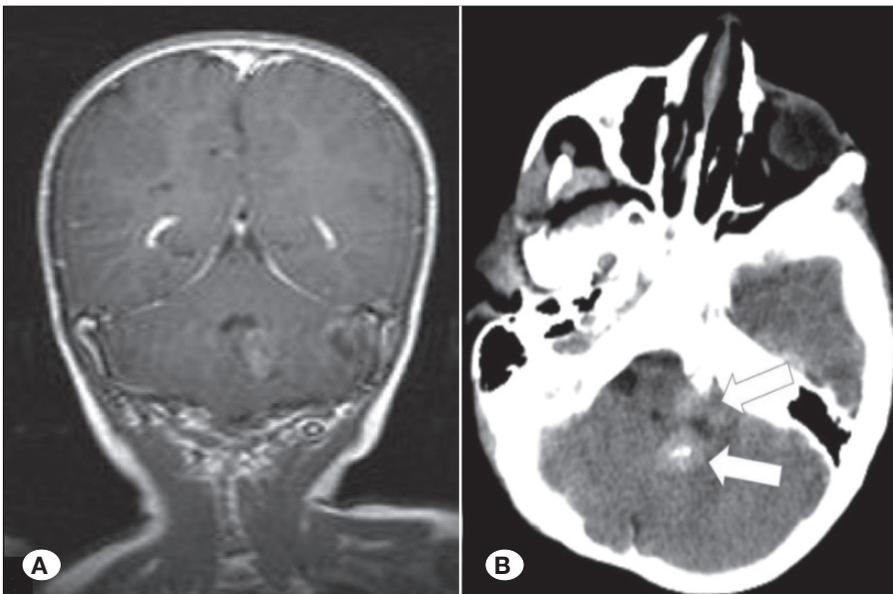


Figure 6: Postcontrast T1-W coronal image shows two masses in the left cerebellum and vermis with nodular and peripheral contrast enhancement (A). Axial brain CT image shows left cerebellar hyperdense lobulated mass lesion (B) (arrows).

was resected, and the pathology result was grade IV ATRT. He underwent chemotherapy (carboplatin, epirubicin, vincristine, ifosfamide, actinomycin, etoposide) and radiotherapy. Control MRI was positive for relapse.

■ DISCUSSION

Although the term torticollis is sometimes used as a diagnosis, it is not a specific diagnosis, but it may be a sign of a number of underlying diseases. Torticollis can originate from a wide variety of primary etiologies, such as muscular, ocular, osseous, and neurological abnormalities (5,20,26). The differential diagnosis of the causes of torticollis is critical for physicians. Hence, better understanding of the spectrum of underlying diseases in torticollis is essential for making the correct diagnosis. A correct diagnosis is essential in selecting the appropriate treatment method and to prevent disease progression and disease-related complications (21,27).

The physical examination for children presenting with acquired torticollis should be complete and must be focused on the vital signs, head, eyes, neck, throat, lungs, and neurological system. In addition, appropriate radiological examinations and adequate laboratory investigations are mandatory (20,21,27,28).

In this study, 24 cases of torticollis as the primary or initial signs of different etiologies are presented.

Although pediatric tumors may show a variety of clinical symptoms, torticollis is one of the critical signs of central nervous system tumors in children. It may be the primary or initial sign related to their location or size (9,17,21,27,29).

The most commonly proposed mechanisms of torticollis related to central nervous system (CNS) tumors include stretching and irritation of the dura and compression of the motor neurons of the external branch of the accessory cranial

nerve, causing pain and involuntary position of the head (9). Delayed diagnosis of CNS tumors can lead to an increase in tumor size, neurological deterioration, and even death (9,25). Although, the relationship between torticollis and intracranial, spinal cord, and vertebral column tumors is well known, some researchers speculated that the diagnosis may be delayed in cases where torticollis is the first or main symptom of CNS tumors (9,14,22,25), mainly because of delay in the indicated imaging (25).

The choice of imaging studies is based on the age of the patient, and use of necessary imaging modalities without any delay is critical in the differential diagnosis for torticollis.

In 10 (43%) of the presented cases, craniospinal pathologies were the underlying causes of torticollis. These were cerebellar anaplastic ependymoma, brain stem glioma, medulloblastoma, ATRT, ADEM, aneurysmal bone cyst of the T4 vertebra, IVDC, compression fracture, vertebral hemangiomas, hereditary MHE of the C2 vertebra, and C4 Langerhans cell histiocytosis.

Gross total resection was performed for ependymoma, medulloblastoma, and ATRT. The torticollis of the patients resolved postoperatively. The patient with ADEM was treated medically. The guardians of the patient with brain stem glioma refused treatment. The patient with compression fractures was treated conservatively with analgesic-anti-inflammatory drug, bed rest, and three-point brace.

Cervical spinal cord alterations may cause focal dystonia due to increased excitability of the spinal motor neuron because of disinhibitory path dysfunction (8,17).

MHE is an inherited genetic disease characterized by the presence of multiple osteochondromas. The most common clinical features include pain and functional impairment. Additionally, restricted motion of the joint is often encountered. Although rare, spinal cord compression and related myelopathy due to osteochondroma are severe complications (12,24). In addition, these lesions can lead to severe acute neurological deficits following minor trauma, including endotracheal intubation for surgery (1). Because of the high numbers of orthopedic surgery performed in these cases, and MHE is most frequently seen in the cervical spine, we think that a preoperative spinal column scan will be useful for early detection of the lesion. In addition, early decompression of the spinal cord must be performed to avoid possible spinal cord injury and related neurological deficits, and even death (1,23).

Eosinophilic granuloma is one of the three clinical entities of Langerhans cell histiocytosis and also known as a skeletal form of the disease. It is mostly seen in children and adolescents. However, Langerhans cell histiocytosis is a very rare cause of torticollis and not commonly considered in the initial differential diagnosis (11,16).

IVDC is a rare disease in childhood with an unclear etiology. The most common clinical symptoms of IVDC in children are torticollis, neck, and back pain (2,18). Patients may present with neck and back pain, torticollis, and spinal muscle contraction. The authors want to highlight that CT scan can

also add crucial diagnostic capacity of IVDC that cannot be ignored.

We did not encounter any torticollis case associated with vertebral hemangiomas in the English literature. We firstly report a case of a 17-year-old boy with torticollis to the right with vertebral hemangiomas in this study. His MRI showed multiple hemangiomas in the cervical, thoracic, and lumbar region without any spinal cord injury. However, Duran et al. reported a case with torticollis related to T1 vertebra hemangioma in a Spanish article. Although this case reported intractable pain not responding to any medication, our case showed resolution of pain after administration of muscle relaxant, analgesic, and physical therapy (8).

Although rare, parapharyngeal infections in children carry significant potential mortality and morbidity risk. These infections can present with torticollis (13). It is speculated that edema and neck muscles, such as the scalene group and *musculus longus* inflammation related to parapharyngeal abscess, cause torticollis (15). Relative infrequency, a wide variety of the symptoms of the disease, and difficulty of oropharyngeal examination in children make the diagnosis challenging. Thus, a misdiagnosis is also possible (27). When the diagnosis cannot be made clinically, CT and MRI are highly sensitive diagnostic modalities that should be performed (15). Five of the patients with torticollis had otorhinolaryngological infections (acute upper respiratory tract infections in two, pansinusitis in one, and mastoiditis in one). Of the five patients, three had acute upper respiratory tract infection and related lymphadenopathy. We did not detect atlantoaxial instability in these patients. Both were treated with appropriate antibiotics and non-steroidal anti-inflammatory drugs. We speculated that the torticollis of the patients was related to soft tissue inflammation.

One of the five patients had pansinusitis and mental retardation (IQ: 64). This patient was administered antibiotics for pansinusitis and physiotherapy for torticollis. In this case, we speculated that torticollis might be attributed to patients' psychological and mental status. The last patient in this group had mastoiditis and strabismus. Similar to the previous patient, we attributed the torticollis to the patients' strabismus. Moreover, this patient was treated with antibiotics for mastoiditis and surgery for strabismus. Torticollis completely resolved. All these patients were remarkable examples that show the importance of a careful otorhinolaryngological examination.

Many ophthalmic conditions may produce a compensatory change in head posture, and incomitant strabismus is the most common cause of ocular torticollis (30). The head and body are adapting to maintain the binocular single vision and improve visual acuity. The abnormal head position due to ophthalmological abnormalities may be related to pain relief and eye protection. That is why ocular torticollis is seen after achievement of head control by infants (19,30). Of the 24 patients in this study, six had ocular torticollis, two had strabismus (one with mental retardation, and this patient was mentioned above), two had Duane syndrome, and two had Brown syndrome. Strabismus is the ocular cause of abnormal head posture in this study.

In Duane syndrome, the 6th cranial nerve that controls the lateral rectus muscle does not develop properly. These patients often maintain an abnormal head posture. Zhang stated that abnormal head position accounts for 20.9% in their study, with 201 patients with Duane syndrome (31). Brown syndrome is the inability of upward movement of the eye related to superior oblique muscle malfunction. It is a mechanical problem wherein the movement of the superior oblique muscle tendon is restricted. Torticollis is the result of misalignment of the eyeballs, and children with Brown syndrome hold a head turn or chin-up head position to compensate for misalignment. Mitchell stated that 30% of patients with Brown syndrome in his study had torticollis (19). In this study, a 5.5-year-old boy with strabismus underwent surgery because of torticollis in another clinic. However, torticollis did not resolve postoperatively. Subsequently, the patient underwent surgery because of strabismus in the author's university ophthalmology clinic, and torticollis resolved. In this patient, misdiagnosis leads to wrong and unnecessary treatment. In the patient presenting with twisted neck, an ophthalmological examination must be performed to rule out underlying ophthalmological etiology.

Sandifer syndrome is characterized by abnormal head posturing and gastroesophageal reflux. The abnormal head posturing is presumed to be a response to pain and esophageal sensitivity related to gastric acid reflux (3,7). A 6-month-old girl was admitted because of twisted neck and vomiting, and neurological examination and imaging studies were normal. However, esophageal pH and impedance monitoring showed high-grade reflux. Torticollis was resolved after medical treatment.

■ CONCLUSION

Torticollis is a sign that is not always innocent, and there are many underlying benign or serious disease in the etiology. We firstly report a case of a vertebral hemangiomatosis and temporomandibular joint ankylosis presenting with torticollis in the English medical literature. Evaluation of children with torticollis should include thorough medical history, physical examination, neurological assessment, appropriate radiological examination, and laboratory investigations. It must be always remembered that misdiagnosis can lead to wrong and unnecessary treatment, and results can sometimes be damaging due to serious underlying conditions.

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