

## **MORPHOFUNCTIONAL FEATURES OF PARADOXICAL NEUROGENIC TORTICOLLIS**

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## **МОРФОФУНКЦІОНАЛЬНІ ОСОБЛИВОСТІ ПАРАДОКСАЛЬНОЇ НЕЙРОГЕННОЇ КРИВОШИЇ**

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### **Summary/Резюме**

Torticollis is an abnormal head position characterized by tilting of the head to one side. It is not an independent disease but a symptom associated with a wide range of congenital or acquired disorders. Congenital torticollis is most often caused by intrauterine developmental abnormalities, including structural defects of the cervical vertebrae, underdevelopment or shortening of the sternocleidomastoid muscle, or disorders of the central nervous system leading to asymmetric muscle tone. Neurogenic torticollis occurs when neurological disorders lead to changes in the tone or function of the cervical muscles. The aim of this study was to highlight the clinical features of paradoxical neurogenic torticollis and its differential diagnosis based on clinical examination and instrumental imaging methods, including ultrasound, computed tomography, and magnetic resonance imaging. A clinical observation of a four-year-old boy with torticollis of unknown origin was conducted over a one-year period. Initially, the child demonstrated a persistent tilt of the head to the right without signs of muscle shortening or trauma. Clinical examination revealed decreased tone of the left sternocleidomastoid muscle while functional tests of active and passive head correction remained within normal limits. Over time, the child developed progressive neurological symptoms, including headaches, vomiting, gait instability, and signs of intracranial hypertension. MRI examination revealed a cystic-solid tumour in the left hemisphere of the cerebellum measuring 60 Ч 54 Ч 52 mm with a pronounced mass effect on adjacent brain structures and signs of occlusive hydrocephalus. The clinical diagnosis included a tumour of the left cerebellar hemisphere, occlusive hydrocephalus, and paradoxical right-sided neurogenic torticollis. The patient underwent suboccipital craniectomy with maximal removal of the tumour. Histological examination confirmed diffuse astrocytoma (Grade II). After surgery, the abnormal head position disappeared and the child's neurological condition significantly improved. Follow-up MRI confirmed postoperative changes without tumour progression. Six years of observation demonstrated normal development and absence of neurological deficits.

**Key words:** torticollis, neurogenic torticollis, cerebellar astrocytoma, posterior cranial fossa, hydrocephalus, MRI.

Кривошия - це аномальне положення голови, що характеризується нахилом голови в один бік. Це не самостійне захворювання, а симптом, пов'язаний з широким спектром вроджених або набутих розладів. Вроджена кривошия найчастіше спричинена внутрішньоутробними аномаліями розвитку, включаючи структурні дефекти шийних хребців, недорозвинення або вкорочення грудино-ключично-соскоподібного м'яза або розлади центральної нервової системи, що призводять до асиметричного м'язового тону. Нейрогенна кривошия виникає, коли неврологічні розлади призводять до змін тону або функції шийних м'язів. Метою цього дослідження було висвітлити клінічні особливості парадоксальної нейрогенної кривошиї та її диференціальну діагностику на основі клінічного обстеження та інструментальних методів візуалізації, включаючи ультразвукове дослідження, комп'ютерну томографію та магнітно-резонансну томографію. Протягом одного року проводилося клінічне спостереження за чотирирічним хлопчиком з кривошиєю невідомого походження. Спочатку дитина демонструвала постійний нахил голови вправо без ознак скорочення м'язів або травми. Клінічне обстеження виявило зниження тону лівого грудинно-ключично-соскоподібного м'яза, тоді як функціональні тести активної та пасивної корекції голови залишалися в межах норми. Згодом у дитини розвинулися прогресуючі неврологічні симптоми, включаючи головний біль, блювоту, нестійкість ходи та ознаки внутрішньочерепної гіпертензії. МРТ-дослідження виявило кістозно-тверду пухлину в лівій півкулі мозочка розміром 60 Ч 54 Ч 52 мм з вираженим масивним ефектом на сусідні структури мозку та ознаками оклюзійної гідроцефалії. Клінічний діагноз включав пухлину лівої півкулі мозочка, оклюзивну гідроцефалію та парадоксальну правобічну нейрогенну кривошию. Пацієнту було проведено субоципальну краніектомію з максимальним видаленням пухлини. Гістологічне дослідження підтвердило дифузну астроцитому (II ступінь). Після операції аномальне положення голови зникло, а неврологічний стан дитини значно покращився. Контрольна МРТ підтвердила післяопераційні зміни без прогресування пухлини. Шість років спостереження продемонстрували нормальний розвиток та відсутність неврологічних дефіцитів.

**Ключові слова:** кривошия, нейрогенна кривошия, астроцитوما мозочка, задня черепна ямка, гідроцефалія, МРТ.

Torticollis is an abnormal position of the head, tilted to one side. It is not a separate disease, but a symptom of a number of disorders that can be congenital or acquired [1]. Congenital torticollis is caused by intrauterine developmental defects: spasm of the sternocleidomastoid muscle (*musculus sternocleidomastoideus*) on one side; bony malformations of the cervical vertebrae (Klippel-Feil syndrome, Sprengel's disease)); underdevelopment, shortening of the sternocleidomastoid muscle; defects of the central nervous system causing excessive tone of the neck muscles on one side [2-4]. The main causes

of acquired torticollis are injuries (rotational subluxation of the first vertebra), fractures, dislocations, subluxations of the cervical vertebrae, visual disorders (strabismus, astigmatism), hearing disorders, inflammatory processes of the head (Grisel-Burge syndrome), chronic and acute myositis, tumours of the sternocleidomastoid muscle, and scarring after damage to the fascia and skin of the neck.

The main causes of neurogenic torticollis are spastic paralysis of the cervical muscles, flaccid paralysis of the muscles, and reflex torticollis in cases of inflammation

of the mastoid process, clavicle, or parotid gland. Given the morphofunctional features of the sternocleidomastoid muscle, when the right muscle contracts, the head tilts to the right, the face turns to the opposite side, and the head extends backward [5-8].

**The purpose of the research** is to highlighting the problem of paradoxical neurogenic torticollis, differential diagnosis, based on clinical, sonographic and magnetic resonance imaging data from examinations of the patient over a period of one year.

### Materials and Methods

A boy with torticollis of unknown origin was clinically observed for one year. A set of instrumental studies was performed to establish the aetiology of the pathology and assess the condition of the internal organs.

The brain and abdominal organs were examined using MEDISON SA-8000EX and Siemens Acuson X-300 ultrasound machines. Computed tomography was performed on a Philips Brilliance CT 64 machine to clarify structural changes.

Magnetic resonance imaging (MRI) allowed for detailed visualisation of all parts of the brain, which made it possible to detect the neoplasm, determine its size, and assess the relationship of the tumour to adjacent anatomical structures.

### Results and Discussion

A 4-year-old boy was born from the first pregnancy, which proceeded without complications, as a result of physiological childbirth. The child's early psychomotor development corresponded to age norms. According to his mother, recently, a head position disorder had been noticed: the child could not hold his head in the correct position, and his head was tilted mainly to the right. There was no history of traumatic injury. During the clinical examination, it was established that the head was tilted to the right. A decrease in the tone of the left sternocleidomastoid muscle was detected. There were no signs of its shortening or thickening. The results of functional tests with active and passive correction of the head position remained within the normal

range.

A year later, the child began to complain of nausea, loss of appetite, and general lethargy. An ultrasound of the abdominal cavity revealed dilation of the bile ducts. Giardiasis and gallbladder dysfunction were diagnosed. He received appropriate therapeutic treatment. The boy's general condition deteriorated. His headaches intensified, he vomited periodically, and he developed unsteadiness when walking. He was examined by a neurosurgeon and a neurologist, who found pronounced signs of intracranial hypertension, cerebellar ataxia, and large-scale SpHNg. An ophthalmologist found that his vision was preserved, but the fundus showed initial signs of optic disc oedema. An otoneurologist found pronounced static and coordination disorders.

Magnetic resonance imaging visualised all parts of the brain, revealing the tumour, its size and its relationship to neighbouring structures. In the posterior cranial fossa in the left hemisphere of the cerebellum, there is a cystic-solid formation measuring 60 x 54 x 52 mm, with clear bumpy contours. The solid part has a moderate hyperintense signal on T2, while the cystic component is located more on the periphery of the tumour. Moderate perifocal oedema. Pronounced mass effect on adjacent sections of the cerebellum, deformed, pushed forward and slightly to the right, medulla oblongata, pons, midbrain. The cerebellar tonsils are displaced downward, wedged into the foramen magnum and penetrated the spinal canal by 5 mm. The aqueduct is dilated in the form of a funnel, especially in the upper part. The lateral ventricles are asymmetrical, the walls are edematous. The chiasmal cistern is displaced by 5 mm, the pituitary gland is pushed to the bottom of the sella turcica by 2 mm. Conclusion. Formation in the posterior cranial fossa, in the projection of the left hemisphere of the cerebellum (astrocytoma?) with a pronounced mass effect on adjacent structures, periventricular oedema. Intracranial prolapse of the chiasmal

cistern (most likely due to increased intracranial pressure). Clinical diagnosis established. Tumour of the left hemisphere of the cerebellum. Occlusive hydrocephalus. Paradoxical right-sided neurogenic torticollis.

The operation performed was a suboccipital craniectomy with maximal total removal of the tumour in the left hemisphere of the cerebellum. Pathological histological examination, haematoxylin-eosin staining, eyepiece x 300. Histological conclusion - diffuse astrocytoma (Gr-II).

Three months after the operation, a repeat MRI of the brain was performed before and after intravenous administration of a contrast agent. A series of tomograms showed a bone defect in the left occipital bone covered with a bone flap. A hygroma above the left hemisphere of the cerebellum, 2.1 cm wide, and a postoperative hygroma of the anterior part of the left hemisphere of the cerebellum, 2.7 x 2.3 x 2.6 cm. The sulci of the left hemisphere of the cerebellum are widened and deepened. The optic nerves, chiasm, pituitary gland, and right hemisphere of the cerebellum are normal. The cerebellar tonsil is normally positioned.

MRI conclusion. Condition after surgical removal of an intracerebral tumour (diffuse astrocytoma) of the left hemisphere of the cerebellum.

After the operation, the head assumed the correct position and the torticollis disappeared. Mild static-coordination disorders persisted, but the congestive phenomena of the optic discs in the fundus of the eye significantly decreased and the general condition improved significantly.

Astrocytoma is a tumour that grows from astrocytes, which are cells of the nervous system. Most cerebellar astrocytomas (80%) are pilocytic (malignancy grade I) and are considered benign. The average time to diagnosis is 7 years. Fibrillary astrocytomas account for 15% of all cerebellar astrocytomas (malignancy grade II). This is a primary

intracerebral tumour that mainly affects young people. The tumour may contain many cysts that grow slowly and compress adjacent parts of the brain, causing intracranial hypertension with corresponding clinical signs [9-11].

According to scientific literature, after surgical removal of tumours in the posterior cranial fossa, a serious postoperative complication may develop - posterior cranial fossa syndrome (cerebral mutism) [12]. This condition occurs after surgery for tumours located in the posterior cranial fossa and is characterised by a complex of neurological disorders. The most common symptoms are mutism (loss of speech), dysarthria, dysphagia, apraxia, ataxia, hemiparesis, and damage to the VI and VII pairs of cranial nerves. The abducens nerve (VI pair) is motor and innervates the lateral rectus muscle of the eye, ensuring the outward movement of the eyeball. The facial nerve (VII pair) is mixed, innervates the mimic muscles, provides secretory innervation of the salivary glands and taste sensitivity of the anterior two-thirds of the tongue, and is functionally associated with the vestibulocochlear nerve (VIII pair), which is responsible for hearing and balance. In addition, children may experience behavioural and emotional disturbances. Posterior cranial fossa syndrome usually occurs 1-10 days after tumour removal surgery. Tumours of this location account for more than half of all brain tumours in children. After medulloblastoma removal, the syndrome develops in approximately 25% of cases, while after astrocytoma resection, it occurs much less frequently, as was observed in our clinical case.

Six years of dispensary observation revealed no abnormalities in the child's development.

The uniqueness of this case lies in the fact that the first signs of a cerebellar tumour appeared a year earlier than the general brain symptoms. The first alarming 'warning signs' of the tumour were masked by torticollis. Classic muscular torticollis has clear symptoms. The main cause of such

torticollis is a change in the sternocleidomastoid muscle. It is one of the strongest muscles in the anterior-lateral part of the neck. If the right and left muscles contract simultaneously, the cervical spine bends and the head extends (tilts backward). If one muscle contracts, it turns the head to the opposite side and tilts it toward itself [6]. This muscle is innervated by the XI accessory cranial nerve (n. accessorius). The nuclei of the XI pair of nerves have bilateral cortical innervation, therefore central paralysis of the innervated muscles is possible with bilateral damage to the cortical-nuclear pathways. With unilateral damage to the nucleus, root, or trunk of the XI nerve, peripheral paralysis of the muscles occurs, making it difficult to turn the head to the healthy side, the shoulder on the affected side is lowered, the shoulder blades do not adhere to the chest (wing-like), and raising the arm above the horizontal line is limited. The main signs of such paralysis are flexia, atrophy, and atony of the affected muscles [10]. The nucleus of this nerve is located in the grey matter of the medulla oblongata at the C1-C2 level. The roots of the spinal cord emerge on the lateral surfaces of the cervical brain, merge into a common trunk, rise upward and through the large occipital foramen (foramen occipitalis magnum) penetrate the cranial cavity, then join the bulbar part of the nerve and leave it through the jugular foramen (foramen jugulare), innervating the sternocleidomastoideus muscle (m. sternocleidomastoideus) and trapezius muscle (m. trapezius). The functions of these muscles are: tilting the head to one side while rotating it in the opposite direction, raising the shoulders, pulling the shoulder girdle back, and bringing the shoulder blades toward the spine.

In this case, the decrease in the tone of the left sternocleidomastoid muscle was caused by compression of the excessively enlarged left amygdala, which wedged into the foramen magnum (mild peripheral paralysis). The imbalance in the tone of these muscles caused the head to tilt to the

right with the face turning in the opposite direction - paradoxical, neurogenic torticollis.

The slow and gradual growth of the cerebellar tumour contributed to increased pressure in the cranial cavity. The cerebrospinal fluid dynamics of the brain were unbalanced. Normally, the circulation of cerebrospinal fluid has the following main direction of fluid movement: lateral ventricles - Monroe's (interventricular) openings - III ventricle - cerebral aqueduct - IV ventricle - Luschka and Magendie openings - large cistern and external subarachnoid space of the brain - subarachnoid space of the spinal cord. The fluid is produced by the vascular plexus of the lateral ventricles of the brain. The volume of cerebrospinal fluid is 120-150 ml. It is completely replaced 4-5 times a day. Cerebrospinal fluid is absorbed by the pachyoeuronal granulations of the soft meninges. Blockage (occlusion) of these pathways leads to the development of occlusive hydrocephalus, which was observed in our case.

Cerebellar ataxia was caused by a tumour in the left hemisphere of the cerebellum, in the form of a cystic-solid formation measuring 60 x 54 x 52 mm. The cerebellum is located in the posterior cranial fossa under the occipital lobes of the large hemispheres, from which it is separated by the cerebellar tentorium, and is an extremely complex and important coordination centre for motor function. It controls balance, stabilises the centre of gravity, regulates the work of antagonist muscles, coordinates movements that require the precision of a large group of muscles, and influences the psyche and emotions. Six years after the operation, the signs of cerebellar ataxia have disappeared. The child is practically healthy. Annual monitoring of the child's health is carried out.

### Conclusions

The peculiarities of the topographic and anatomical structures of the posterior cranial fossa, cerebellum, cerebrospinal fluid circulation, structure of the accessory nerve,

and cerebellar astrocytoma were the cause of the unique clinical picture of paradoxical neurogenic torticollis. Torticollis is not an independent disease, but a symptom of a whole range of disorders that can be congenital or acquired. Surgical removal of the tumour guaranteed a positive outcome of recovery.

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