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General Description of Pediatric Acute Wryneck Condition

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1. Introduction

At least 80 causes of torticollis have been documented in the literature [1]. Acutely developed torticollis may mask severe pathology requiring treatment including surgical one. First of all, it is necessary to rule out trauma as well as the destructive process of tumoral or inflammatory nature. General differential-and-diagnostic algorithms required for examination of children with torticollis have been proposed (Fig. 1).

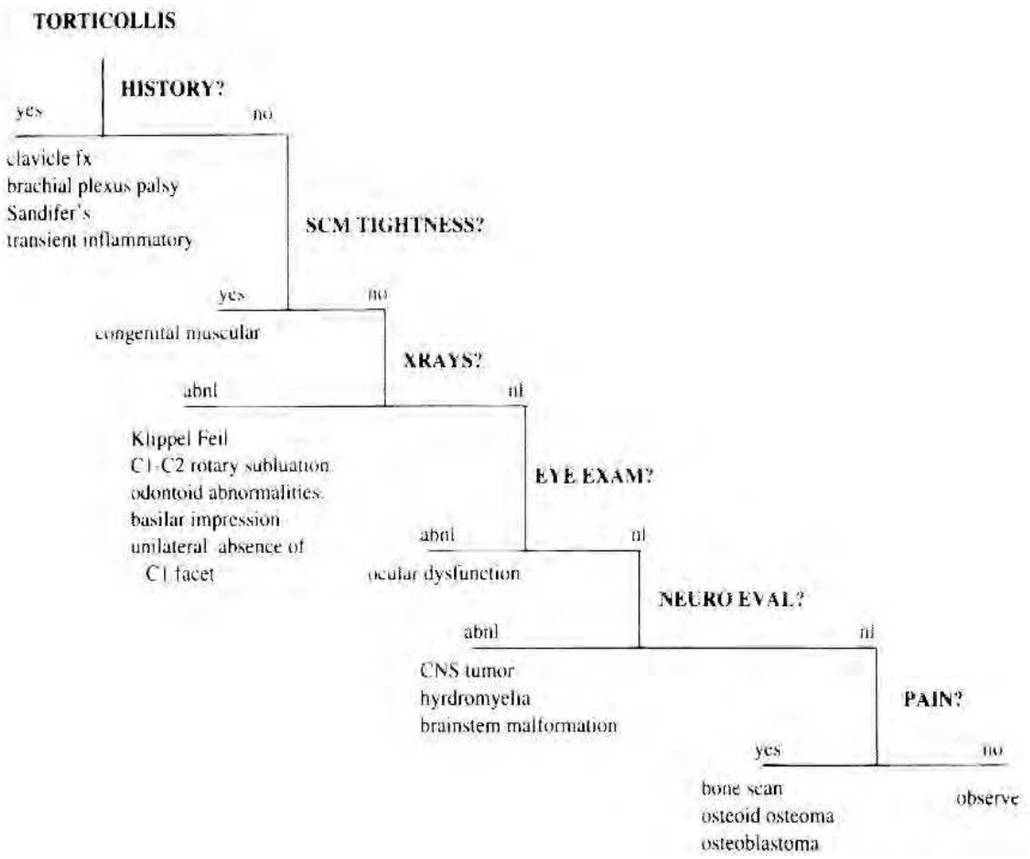


Fig. 1. Algorithm for examination of children with torticollis (cit. by [2]).

In case of acutely developed torticollis without traumatic effect the working diagnosis is made as acute torticollis or suspicion of rotatory subluxation, and in case of inflammation presence in nasopharynx it is made as the Grisel syndrome.

Such a child should be observed under out-patient conditions with prescription of immobilization and non-steroid anti-inflammatory medications. X-rays should not be made because torticollis prevents patient's proper positioning. In case of the pain syndrome and forced head position retention the patient should be hospitalized for examination and treatment. Thus, the acutely developed pathological head position and the pain syndrome in a child provided for ruling out traumatic and destructive causes is considered as a condition, the basis of which, according to most authors, is idiopathic atlantoaxial fixation or subluxation of CI.

Blankstein et al. (1997), who had analyzed the data of 33 patients over four years, found a clear seasonal trend – 58% cases accounted for the period from November to February, 33% of them – for the period from April to July [4]. Nemet et al. (2002) reported that 73% of cases in their group occurred in autumn and winter [5]. None of the authors mentioned could explain the phenomenon observed.

In our group (264 patients) the appearance of acute wryneck was the most characteristic for winter/spring period (70%). In the summer time mainly pre-school children were hospitalized with acute torticollis, while in the autumn children of the older school age prevail.

In majority of patients head side bending contra lateral to painful side has prevailed (Fig. 2). The «cock-robin position» with rotational motions block, classical for atlas-axial rotational subluxation description, is observed very rarely. Head side bending has varied from 10 to 45 degrees. The amount of rotational movements is restricted towards the painful side but had always prevailed over possibility for proper head positioning.

None of the authors tried to assess the pain syndrome intensity objectively. We found no attempts to connect the manifestation degree of the pathological head position with the characteristic features of x-ray picture. Despite the fact that some authors tried to characterize the range of motion in patients, it had no effect on the final diagnosis making or treatment character.

No relationship was found in the literature between the patient's age and the pain syndrome duration as well. In general, neck pain lasts from several hours to several days [5,6].

In our group minimal time needed to cut off pain syndrome is 24 hours; maximal one – is 10 days.

We've found the direct relationship between age of patients with acute torticollis and pain syndrome duration: the older a child, the longer is pain. So in children of babyhood the maximal duration of pain syndrome was 5 days and in older schoolchildren – 10 days.

With regard to neurological status, some authors consider complete neurological intactness [7] while others point to mild neurological symptoms as weakness in the limbs and headaches [4].



Fig. 2. Typical head position in a boy with wryneck.

2. Etiology and pathogenesis of acute torticollis in children

In N. Schwarz et al. opinion, the final decision on the direct pathogenetic cause of acute atlantoaxial rotatory subluxation (AARSL) in view of the current scope of scientific knowledge is difficult [8].

The inflammatory theory of CI subluxation was the principal until the middle of the XX century [9]. The Grisel syndrome was explained by the contracture of paravertebral muscles due to the pathological impulses from the focus of inflammation, or the inflammatory process dissemination to the lymph nodes behind the pharynx.

Most contemporary authors consider the Grisel syndrome cause as a direct inflammatory involvement of the soft-tissue structures of the atlantoaxial joint. The system of veins with frequent lymph-venous anastomoses between the periodontal venous plexus and the suboccipital epidural sinus may be a hematogenous intermediary to transfer the peripharyngeal inflammatory exudate to the zone of the atlantoaxial joint [10].

The theory of the entrapment of meniscoid bodies and torn ligaments in the cavity of the lateral atlantoaxial joints is more popular, and according to researchers it takes place for beyond-range rotatory motion of the neck with head tilt [11,12].

After performing a series of experiments on the anatomical preparations of atlantoaxial complexes with their subsequent freezing and making frontal and sagittal saw-cuts M.N. Nikitin (1965) put forward his theory [13,14]: «In case of uncoordinated movement of the head its lateral tilt occurs which leads to the expansion of the lateral atlantoaxial joint gap contralaterally, as a result of which the anterior and posterior parts of the joint capsule go

deeply into the joint cavity as folds by 2/3 of the joint sagittal length at the expense of negative pressure (normally this hollow amounts to ½ of the joint sagittal length). The tension of the joint capsule lateral part, which occurs at that, causes irritation of nerve endings, thereby leading to reflex protective contraction of the muscles around the atlantoaxial complex, with entrapment of the capsule folds deepened and development of the joint blockade». This theory explains frequent beginning of acute AARSL from a sharp spasm of muscles during movements.

Another theory consists in atlas lateral mass getting on the side of its forward rotation to axis underlying part with joint gap overlapping (intervertebral blocking) [15-21]. This manifests itself as the radiological symptom of «winking». The coupling of the adjacent articular facets of the lateral atlantoaxial joints takes place in case of their getting (Fig. 3).

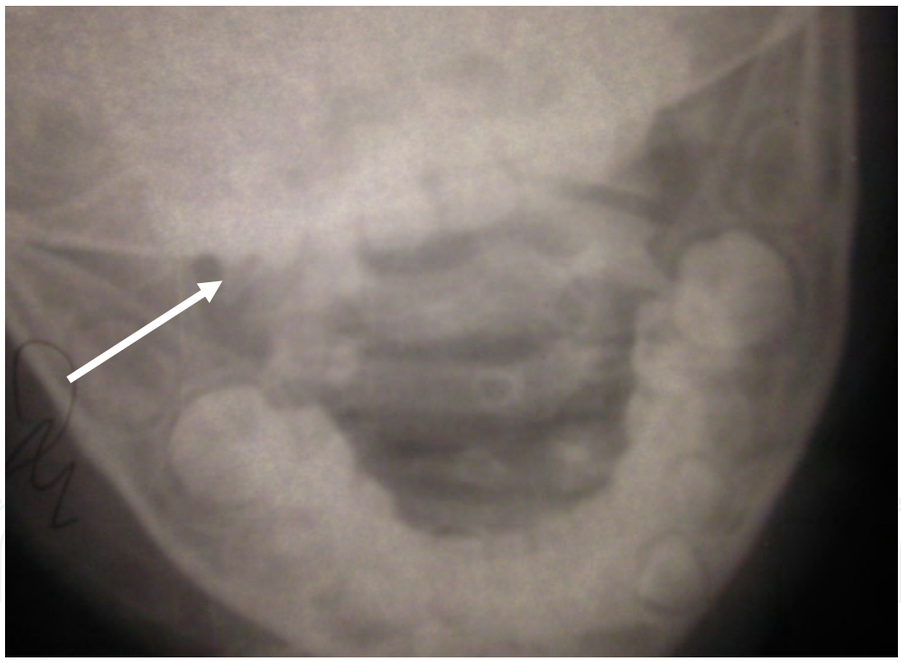


Fig. 3. «Winking» symptom in a child with AARSL. Marked CI-CII articulation locked (our case).

It should be noted that the analysis of the works demonstrating AARSL picture by dynamic 3-D CT confirms this point of view (Fig. 4).



Fig. 4. 3-D CT of a child with AARSL. The getting of CI left articular surface to CII is clearly determined (our case).

The theory of acute muscular torticollis is considered in the literature as well. It is assumed that the pathological displacement of cervical vertebrae occurs due to muscle spasm [22].

The literature analysis allows to note that all the existing theories dealing with acute torticollis and AARSL development aside from intervertebral blocking are not confirmed by modern research methods (CT, MRI) and are the result of the authors' assumptions only. The lack of clarity in determining the causal relationships of all the theories proposed is not unimportant. No assumption clearly answers the question, what is the starting point for suffering beginning. Is CI subluxation the cause of the problem or its consequence ?

Maigne et al. (2003) have performed MRI of cervical spine in a 15-year adolescent in the early hours from the onset of typical acute torticollis developed after night sleep [23]. The authors did not rule out the disorder of CI-CII relation, but the signs of signal intensification were not found there. A hyperintensive wedge-shaped signal was determined by them in the zone of CII-CIII uncovertebral articulation on the side of pains (Fig. 5). It disappeared after three weeks by control MRI data. Torticollis quickly resolved by conservative treatment. The authors explained the finding by acutely occurred rupture of the intervertebral disc.

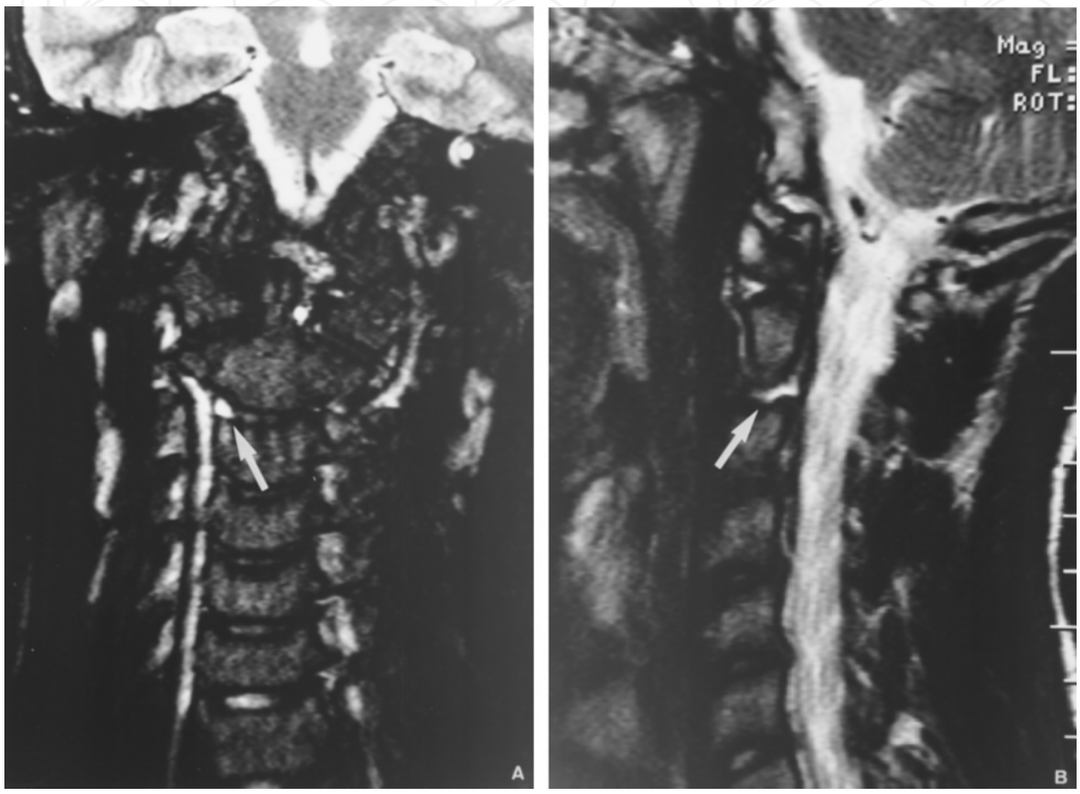


Fig. 5. A hyperintensive wedge-shaped signal in the zone of CII-CIII uncovertebral articulation (cit. [23]).

For identification of given pathological condition reasons we accepted a tactics of special MRI mode use in first hours after the patient's submission [24] .

We have made randomized sampling of patients with acute torticollis and atlas-axial block with a single selection criteria – first 12 hours after disease appearance. It has found to be reasonable for in 10 patients examined in succession, typical alteration were detected. They consisted in area of marked glowing of triangular or longitudinal shape in the area of external edge of C2-C3 or C3-C4 disk, and this glowing was always on the painful side (Fig. 6). The same findings that was presented by Maigne et al.

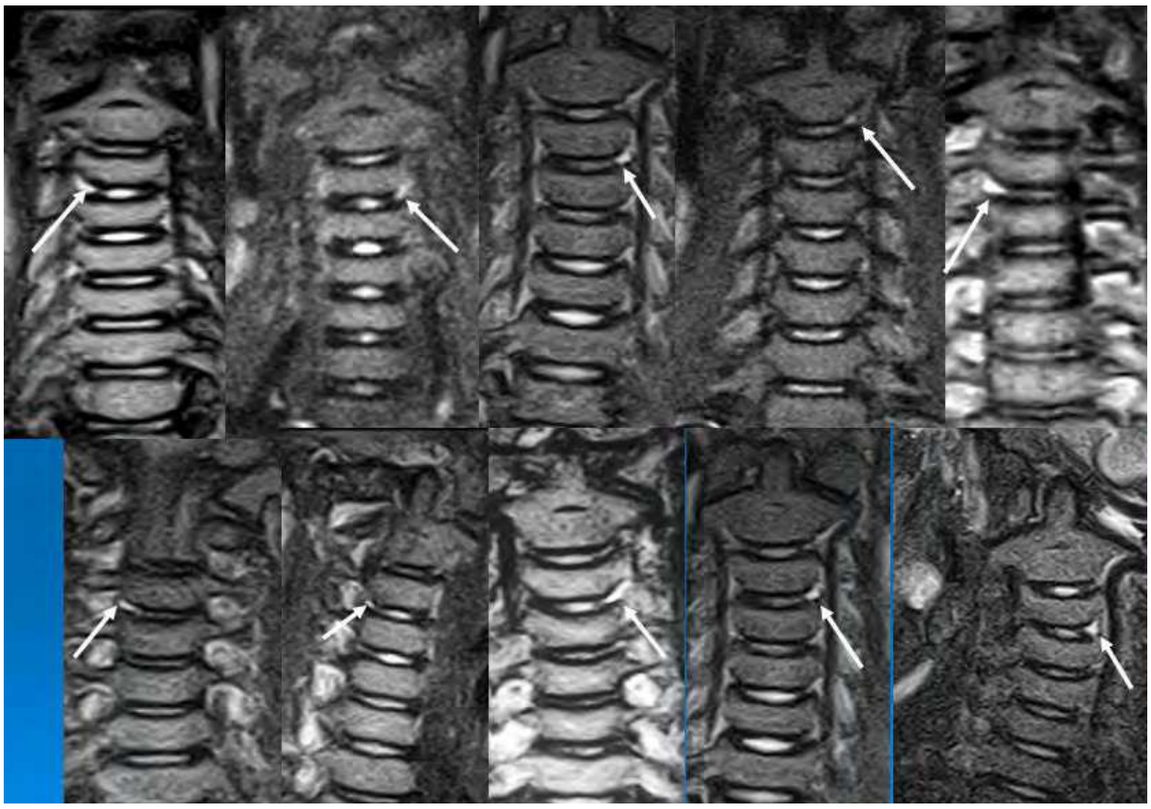


Fig. 6. MRI in the fat inhibition mode in 10 children with acute torticollis. The triangular area of hyperintensive signal is clearly seen in posterior-lateral parts of intervertebral space. The disk outline is separated from this area.

We propose the following mechanism of the syndrome development called "the uncovertebral wedge" [24,25]. Periosteal-facsial tissue in the area of uncovertebral joint is restricted by: hard borders of the disk fibrous ring from interiorly, posterior longitudinal ligament from posterior, hamus of caudal vertebra laterally and lamina of cranial vertebra from anterior. The reason of acute torticollis in children is in sharp or gradual compression of periosteal-facsial tissue in the uncovertebral fissure resulting from head movement or from long neck sidebending (sleep) with formation of a "wedge" from edematous tissue which irritates posterior longitudinal ligament. It leads to antalgic head position and, in some cases, to atlas-axial block. That's why traction reducing pressure in the uncovertebral fissure and contributing in venous drainage improvement and problem resolution is so efficient. Following arguments in favor of the given supposition seem to be equally important:

1. Uncovertebral joint is an exclusive anatomical neck specific, that's why similar conditions in children occur neither in lumbar nor in thoracic spine.
2. Pain appearance and its amplification in vertical posture, for here the pressure applied to intervertebral disk and correspondingly to uncovertebral "fissure" is increased.

3. Larger occurrence of acute torticollis in autumn/winter period may be explained by large amount of inflammatory alterations from the side of nasopharynx, that leads to venous drainage and adjacent tissue deterioration and edema complications.
4. Pathologically explainable becomes not only antalgic scoliosis (torticollis), but frequently observed kyphotic deformities in the cervical spine.

We suppose that age-related reduction and disappearance of acute torticollis and atlas-axial subluxations in adults is related to decrease of intervertebral disks resilience, to presence of powerful motion restrictors in the form of well-developed uncovertebral joints and to degenerative changes in Luschka joints.

3. Radiological findings in children with acute wryneck.

The vast majority of works deals with studying the atlantoaxial articulation X-rays. Some authors believe that the sign of asymmetric axis odontoid process location relative to atlas lateral masses is quite sufficient to make the diagnosis [26,27]. Others have doubts about the reliability of the most x-ray signs observed connecting them with the inability to achieve proper positioning in the process of x-ray study [28,29] or consider them as a variant of the norm [30]. While performing plane radiography Nicholson et al. (1999) reported that acute AARSL was not diagnosed in 67% of cases in their group of patients, and hyperdiagnosis had place in 29% of the cases [31]. That is to say, the radiological method has been recognized to be questionable in terms of assessing CI-CII relation for acute torticollis! Nevertheless, the most widely used classification of acute atlantoaxial rotatory subluxation has been proposed based on the clinical picture and radiological technique (Fig. 7). There are no Types III and IV in children.

The methods of dynamic radiography and cineradiography have not become popular because of the high level of radiation exposure [33].

The appearance of computer tomography in the 70-s of the XX century allowed researchers to define the details of atlas displacement more accurately [34-36]

At present dynamic and 3-D CT are mainly used [37]. Li et Pang (1995) have developed the criteria of 3-D dynamic CT using to define the diagnosis more exactly and develop the tactics for treating the atlantoaxial fixation [38]. They have refined the classification of Fielding J.W. et Hawkins R.J:

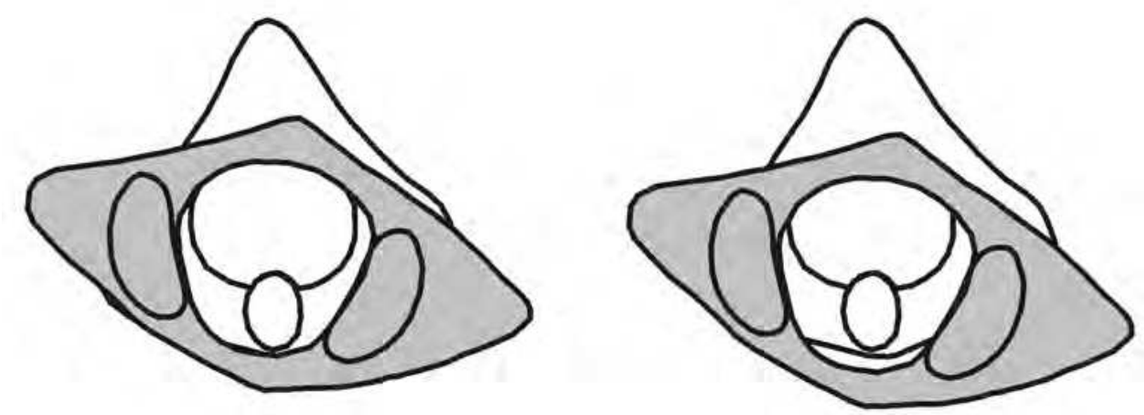
Type I – CI-CII is blocked with corrective rotation of the head;

Type II – CI-CII relation is improved with corrective rotation of the head:

Type IIA – CI-CII relation does not reach 0^o with corrective rotation of the head;

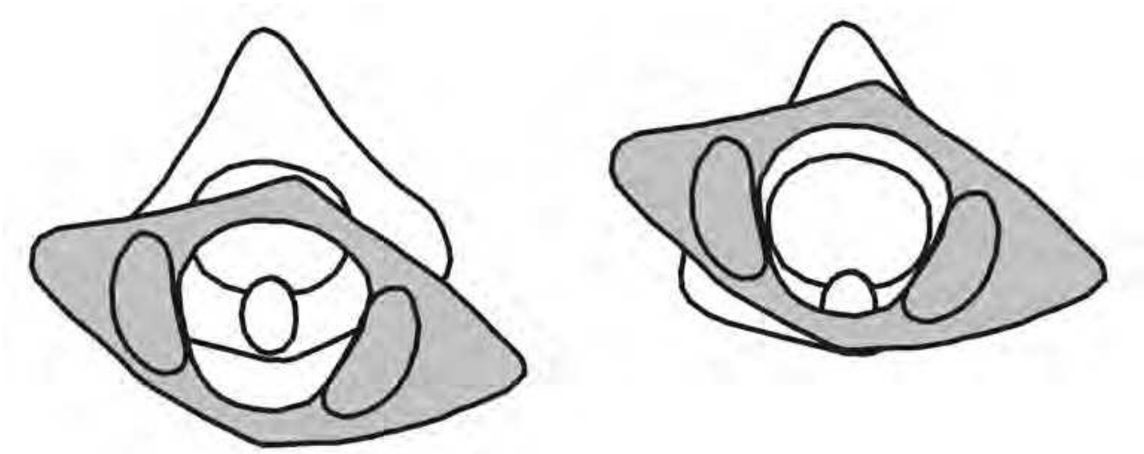
Type IIB – CI-CII relation reaches 10^o of rotation not more in the direction opposite blocking.

The more perfect tomography methods became and the more material collected, the more frequent were the data of the absence of visualization of the pathology in the atlantoaxial segment for the typical clinical picture of acute torticollis. Thus, Alanay et al. (2002) examined 15 girls and 21 boys at the age of 4-16 years with acute torticollis using dynamic CT, and they did not find the difference in CI-CII relations between them and normal children subjected to the similar study [39].



Type I

Type II



Type III

Type IV

Type of acute AARSL	CI sagittal displacement	Size of Cruveilhier joint gap
Type I	Absent	3 mm
Type II	Anterior	3-5 mm
Type III	Anterior	› 5 mm
Type IV	Posterior	-

Fig. 7. Acute AARSL classification (cit. by [32]).

In the authors' opinion, there is no need to use dynamic CT in case of the fundamentally good-quality condition with spontaneous recovery which is represented by acute torticollis. They propose to use tomography in the cases of prolonged (more than one week) pain syndrome only. They have confirmed their first work by the second one with mathematical analysis and come to the same conclusions [40]. Other researches also had difficulties in the interpretation of CT picture for acute torticollis in children [41]. The main answer is absent in all the CT observations found in the literature: whether CI-CII relation disorder is a primary problem or a secondary positioning of the head. First of all, its use is justified for chronic AARSL [42].

Mainly MRI for torticollis is used to rule out a traumatic or destructive process [43].

4. Management of acute wryneck

Some authors within the end of the XIX century-first half of the XX century recommended to perform acute manual reduction for the purpose of subluxation elimination. The technique developed by Heister and Richet-Hueter was used for this purpose [44]. However, by the 60-s of the XX century this technique has lost its popularity, and most of the authors has recommended to perform loop traction.

The standard treatment regimen for patients with acute torticollis (suspicion of acute AARSL) has been reflected in most guides to Orthopedics (cit. by [3]):

1. Below one week: immobilization with soft collar, analgesics, bed rest; in case of recovery absence: hospitalization, traction;
2. Above one week, but below one month: hospitalization, loop traction, cervical collar for 4-6 weeks;
3. Above one month: hospitalization, skeletal traction, cervical collar for 4-6 weeks.

5. Conclusion

Children with sudden pain in the neck and constrained head position are frequent pediatric patients. It is reasonable to make a syndrome-related diagnosis on the stage of initial examination. The main task for a doctor is to separate those from the patients whose condition demands more profound examination, observation and treatment.

It is impossible to provide each patient with acute torticollis with full radiological examination and long observation in the hospital. That's why it is necessary to single out "danger levels" to provide this patients' group with adequate management [45]. We propose to single out 3 levels built by exclusion method (Fig. 8).

First level represents main flow of patients with multietiological and, in majority of cases, with "innocent" damages of cervical spine.

Second level represents patients with true atlas-axial subluxations demanding obligatory traction treatment to prevent pathology transition to chronic stage.

Third level includes patients with risk of mechanical and neurological instability demanding, as a rule, operative treatment. Besides usual injury it includes children with cervical developmental defects manifestation.

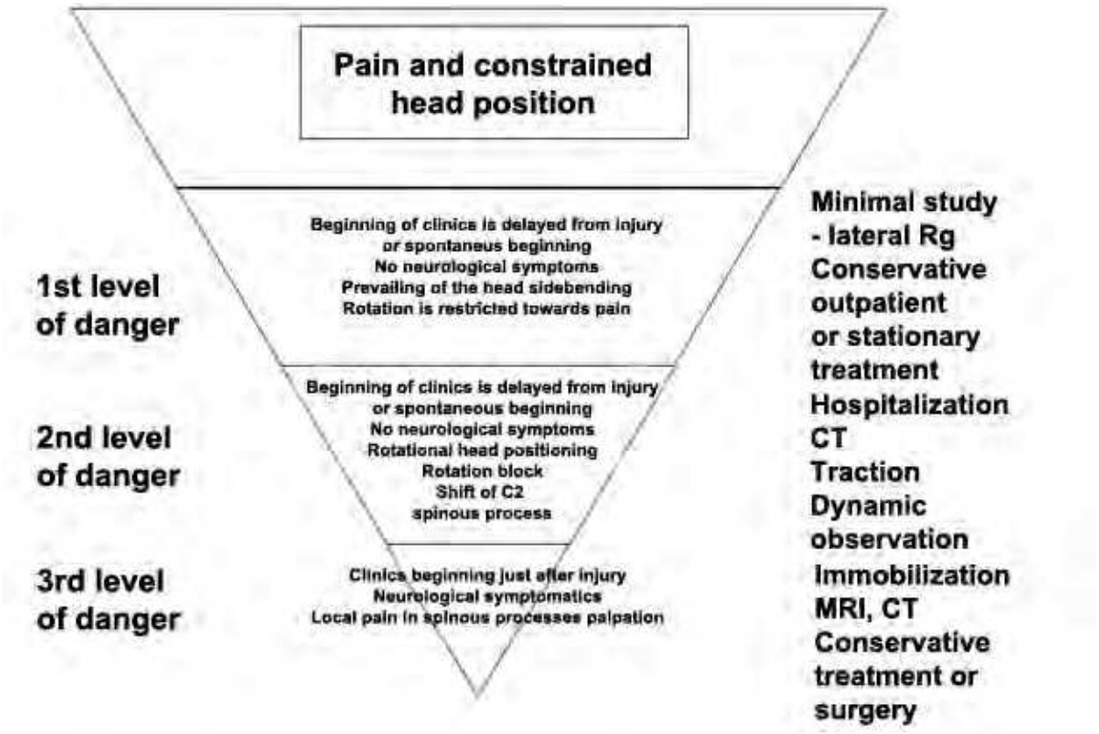


Fig. 8. Algorithm of surgeon's actions in case of admission of patients with acute pain syndrome and constrained head position. 3 incremental "danger levels". The pyramid narrowing to the 3rd level symbolically reflects amount of patients.

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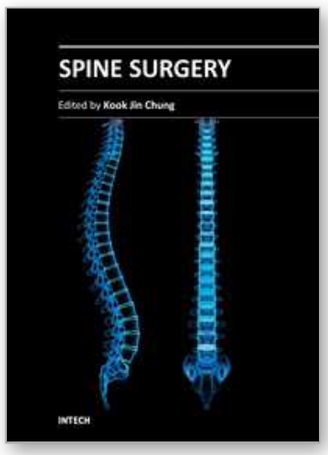
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