COMMUNICATION FROM THE COAST HOSPITAL APELVIKEN, SWEDEN. SENIOR SURGEON: ROBERT HANSON, M. D.

A CASE OF CERVICAL SPONDYLARTHRITIS AFTER TONSILLECTOMY

BY

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The following case might deserve attention as a rather rare complication after tonsillectomy:—

Stina S., born in 1914. No known heredity of or exposure to tuberculosis. The patient has had measles, whooping cough and rubella. Like her three sisters and her brother she has often had a cold with fever and tonsillitis. During the winter of 1929—30 she had a severe tonsillitis and tonsillectomy was recommended.

Tonsillectomy was performed by the writer under percain anæsthesia on Aug. 13th, 30; abrasio was *not* performed. The tonsils were large, with deep crypts, they were not irritated when operated upon. A few hours after the operation the patient got up from her bed, she fainted and fell on the floor with her head against the wall. She does not believe that she hurt herself on this occasion, immediately afterwards the movements of her head were unimpaired.

Already the night after the operation the patient states that she felt some pain in her neck, but she did not pay much attention to it. After Aug. 14th she was nursed at home; during the first week the temperature was about 38°, she felt some pain and stiffness in her neck. The pain and the stiffness increased during the first and second week after the operation but still no faulty position of the neck could be observed. The cervical lymph glands were tender and swollen for which reason the pain was considered to be due to inflammation of the

deep cervical glands. The fauces had healed completely on Aug. 31st, but the pain in the neck remained, localised to the left side. It was difficult for the patient to be out of bed, difficult to swallow, and she could not sleep without soporifics. As the condition did not improve she was admitted to the coast hospital Apelviken on Sept. 17th, 30, under the diagnosis of spondylitis cervicalis tbc. (?).

State of the patient on admission: Her general condition is rather bad, she is lean, pale, and her weight is 44 kgm. Pirquet's test is negative, Mantoux's test 0,1 mg. pos. Sedimentation test 32 mm/1 hour, 58 mm/2 hours. The interior organs show nothing abnormal. The fauces completely healed, pale, no fragments of the tonsils. The superficial lymph glands show nothing abnormal.

The head is held in a fixed position and bent forward, somewhat rotated to the left. There is a considerable scoliosis in the cervical part of the spinal column, with a left concavity. The patient avoids any unnecessary movements and supports her head with her left hand. Even the slightest movements of the head cause severe pain as does pressure on the scalp compressing the cervical column. The patient can only sit up in the erect posture, supporting her head with her hand, or lie down. On palpation the spinous process of the epistropheus, (axis), is felt to be more prominent than normal and tender.

The patient was treated according to the method usually employed at the coast hospital Apelviken. A short plaster-of-Paris bed enclosing head and neck is fixed on small wheels that run on metal bars. A weight of 1—2 kgm. is fixed to the upper end of the plaster-of-Paris bed, and the patient is placed in the latter. Thus the head is exposed to hyperextension and permanent traction, the body of the patient serving as counterbalance. (The method was originally stated by Rollier but has been modified by R. Hanson).

Already on Sept. 26th after one week's extension treatment the deformity and the pain had largely disappeared, and X-ray examination could be made without difficulty. On Nov. 25th the patient's pain had disappeared completely and she got a

plaster-of-Paris collar. The latter was used until Jan. 12th, 31, and the patient was now out of her bed without any fixation bandage.

Jan. 22nd, 31:— The general health is good. Weight: 51,2 kgm. The fauces: Pale cicatrices are seen at the site of the tonsils. The posterior wall of the fauces is of a normal shape, nothing pathologic can be palpated here. No faulty position of the cervical column, no direct or indirect tenderness on pressure. Bending of the head forwards and backwards is very slightly impaired, a very distinct crepitation is heard now and again when the head is bent backwards. There is a distinct limitation of the rotation of the head to either side. The patient was discharged on Jan. 31st, 31.

Since her discharge she has stayed at home and performed easy work; the condition has constantly improved. Only a few times, after sleeping with her head in an oblique position, she has had pain in the neck. Since Oct. 1st, 31, she has been employed as a housekeeper.

After-examination on Dec. 5th, 31:— The general health is good, Wassermann's test is negative. Sedimentation test 15 mm/1 hour, 44 mm/2 hours. No faulty position of the head or the neck, no contracture of the muscles. No deformity of the cervical column. In the neck the distinctly prominent spinous process of the epistropheus (C II) is palpated, now completely free from tenderness. The fauces shows nothing abnormal. The head can be bent forwards and backwards to the normal extent, rotation to the left is slightly restricted, rotation to the right is distinctly restricted. By fixation of the lower part of the cervical column this restriction of movement can be localised to the part above C II. On quick movements with the head crepitation is sometimes heard in the neck.

Skiagram on Sept. 12th, 30: Ventrodorsal picture (v-d-picture): The head inclines to the left, the cervical part of the spinal column is slightly rotated to the left with the spinous process to the right. Lateral view (comprising C II—IV): nothing pathological. No details.

Skiagram on Sept. 26th, 30: V-d-picture: The left lateral

joint between the atlas and the epistropheus shows moderate decalcification. The contours of the joint are fuzzy and rough The cartilaginous space is distinctly lowered, the left part of the atlas sloping towards the epistropheus. There is neither any lateral displacement nor any rotation between the vertebrae

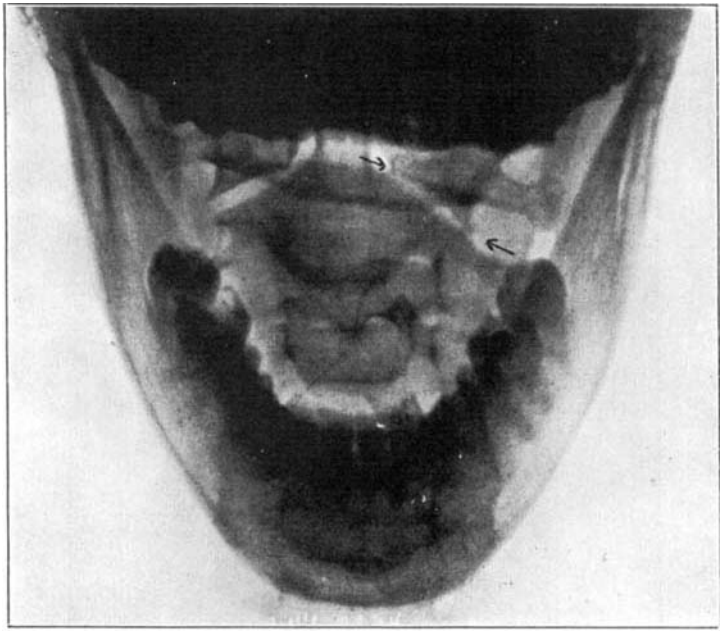


Fig. 1.

Stina S. Sept. 26th, 30. Acute arthritis of the left lateral intervertebral joint.

in question. The lateral view shows no displacement forwards of the atlas in proportion to the dens epistrophei.

Skiagram on Jan. 22nd, 31: V-d-picture: The atrophy of the left lateral intervertebral joint has nearly disappeared. The spongy tissue is now clearly defined, the contours of the joint are distinct. The joint space is narrower than on Sept. 26th, 30, on the borders of the articular surfaces thick deposits of bone can now be seen, most markedly on the lateral side of the

articular surface of the atlas. The right intervertebral joint shows nothing abnormal. Fig. 2.

Skiagram on Nov. 27th, 31: V-d-picture: No atrophy, the spongy tissue is clearly defined. The cartilaginous space of the left intervertebral joint is still narrower and scarcely visible in any picture despite varying direction of the rays, only a

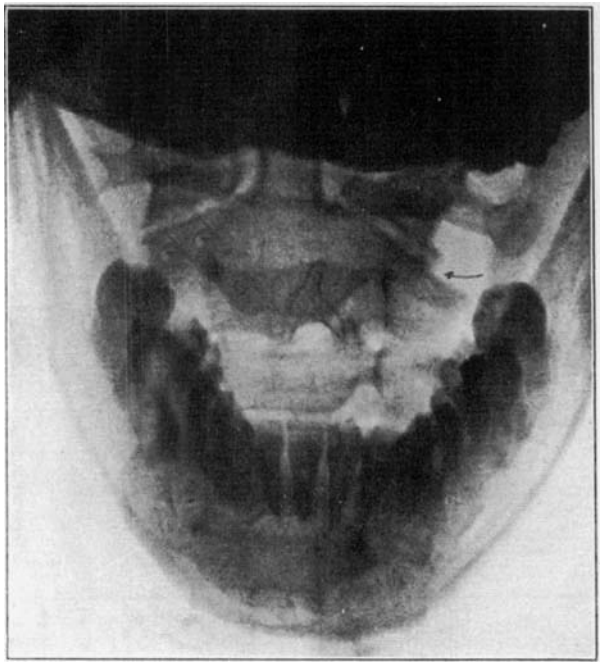


Fig. 2.

Stina S. Jan. 22nd. 31. Bony deposits on the articular surfaces of the left intervertebral joint. The cartilaginous space further diminished.

rareficated line marking its site. The deposits are unaltered since Jan. 22nd. The right intervertebral joint shows nothing abnormal. In lateral view nothing pathological is seen. Fig. 3. The atlantooccipital joints are seen in several pictures through the shadow of the occipital bone, they are of a normal shape and the joint spaces are of normal height.

The X-ray examinations have shown an acute osteoarthritis

in the left lateral intervertebral joint between the atlas and the epistropheus, which has healed under considerable osseous alterations.

In the present case an acute arthritis in the left lateral in-

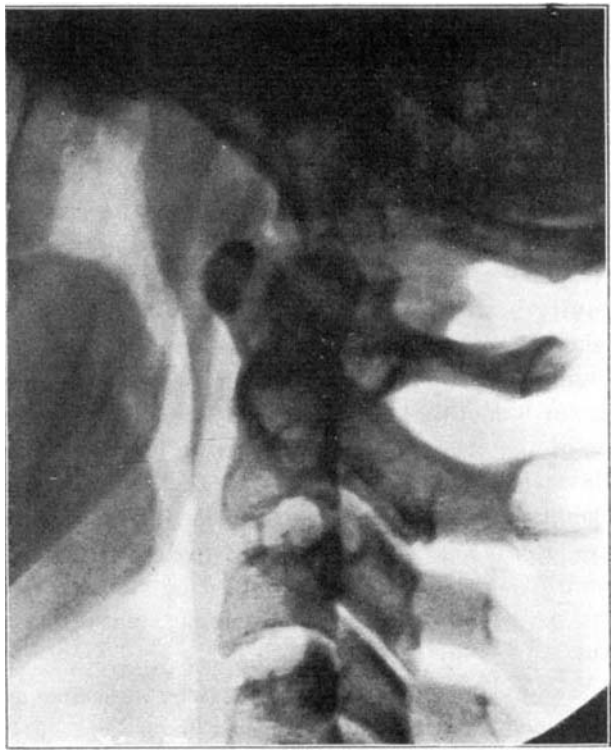


Fig. 3.
Stina S. Nov. 27th. 31. Lateral view, no pathological signs visible.

tervertebral joint between the atlas and the epistropheus occurs in a 16 year old girl, who was previously in good health, in direct association with tonsillectomy. At first the process was suspected to be a tuberculous malum suboccipitale. With the exception of a positive Mantoux's test for 0,1 mg »Alttuberkulin« (T. O.) no signs of tuberculosis are found. There

is no clue to lues; on Dec. 8th, 31, Wassermann's test is negative. Nor are there any clues to other specific infections.

In all probability the case is one of septic spondylarthritis, arising from a chronic infection of the tonsils, activated by the tonsillectomy and making its way through the retropharyngeal lymph vessels to the intervertebral joints. The fever and the swelling of the lymphatic glands in the neck as well as the quick subsiding of the process speak in favour of a septic etiology. The considerable bony deposits on the borders of the articular surfaces formed in the course of a few months speak above all in favour of a septic infection and against tuberculosis.

Numerous cases of this sort have been described in the literature. Swanberg reports the case of a soldier, aged 22 years, who during the night after a tonsillectomy got pain in the neck and a faulty position of the head with the chin hanging down on the chest. There was no trauma, the operation had been performed under local anæsthesia and its course had been normal. A skiagram 6 months after the operation showed a luxation forwards of the atlas in proportion to the epistropheus with a large diastasis between the dens and the anterior arc of the atlas. The massae laterales atlant. project before and somewhat below the lateral articular surfaces of the epistropheus. — The posterior wall of the fauces was bulged by the anterior arc of the atlas. An attempt at reposition under anæsthesia was unsuccessful.

Grisel quotes old French authors who describe an acute »torticolis occipito-atloidien« occurring in association with an infection of the fauces. Himself he reports two cases of unilateral subluxation of the atlas, one case after angina, the other after tonsillectomy. These cases showed a fixed faulty position of the head and neck. The ventrodorsal skiagram shows the following details: The shadow of the atlas is fairly symmetrical and both the lateral articular surfaces are of the same width. The right lateral articular surface of the epistropheus is considerably broader than the left one. The cartilaginous space of the right intervertebral joint shows the normal height, the lateral border of the articular surface of the atlas is placed

medial to the lateral border of the articular surface of the epistropheus. The articular surfaces on the left side cover each other partially so that the cartilaginous space is obliterated. The articular surface of the atlas projects here very much to the lateral side of the lateral border of the articular surface of the epistropheus. Fig. 4. The lateral view shows a moderate displacement of the atlas forwards with diastasis between the anterior arc of the atlas and the dens epistrophei.

Grisel believes that in his cases the facies is determined by a lymphogenous, retropharyngeal inflammation spreading to the prevertebral muscles. Through contracture of these muscles

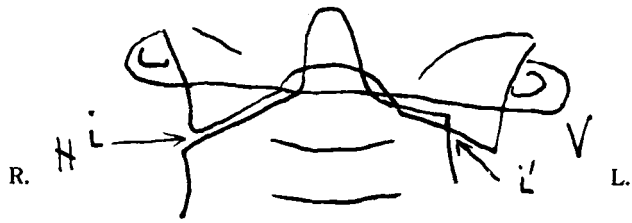


Fig. 4.
Ventrodorsal view according to Grisel.

the atlas is said to be dislocated forwards and rotated in proportion to the epistropheus. It was impossible to demonstrate bony destructions in his cases. He denies the possibility of a spondylarthritis as a cause of the facies. Other French authors accept Grisel's explanation and describe similar cases, most of them in the age of 8—10 years. Desfosses reports a case with the same clinical picture and the same ventrodorsal skiagram. The lateral view, however, does not show any dislocation forwards of the atlas in proportion to the epistropheus.

Berkheiser and Seidler report 5 similar cases in children from 8 to 11 years of age. All the cases occurred in association with an acute infection of the fauces. In one of the cases there was moreover polyarthitis and pericarditis. One of the patients had a maximal displacement forwards of the atlas with luxation forwards and distal of the lateral intervertebral joints. The anterior arc of the atlas was displaced anteriorly and down-

wards in relation to the dens, at the level of the corpus epistrophei. Like Grisel's cases the other patients had their heads fixed in rotation and inclined to one side. Berkheiser and Seidler regard this occurrence as a rotation luxation in one lateral intervertebral joint. In two cases they presuppose a unilateral (sub)-luxation forwards and in two other cases a subluxation backwards. Berkheiser and Seidler believe that the disappearance of one joint space between the atlas and the epistropheus in the ventrodorsal picture is characteristic of unilateral subluxation of the atlas (»... absence of the joint space between the lateral facets of the atlas and axis is pathognomonic of this condition«). Grisel is of the same opinion.

Berkheiser's and Seidler's cases with unilateral subluxation forwards show a distinct dislocation forwards of the anterior arc of the atlas in proportion to the dens when seen in lateral view. Both Grisel, Berkheiser and Seidler draw the attention to the clinical picture that is typical of the unilateral subluxation. In unilateral subluxation forwards the head is rotated towards the sound side and inclines towards the diseased side. The spinous process of the epistropheus can be palpated deviating towards the sound side. In the lower part of the cervical column a scoliosis arises that is convex towards the sound side. The movements of the head are impaired, especially rotation towards the diseased side. In the case of unilateral subluxation backwards Berkheiser and Seidler describe the reverse symptom picture with the head rotated towards the diseased side and inclined towards the sound side, whilst the cervical scoliosis is convex towards the diseased side. Berkheiser and Seidler believe that the condition described arises through a lymphogenous or hæmatogenous infection of the intervertebral joints of the cervical vertebrae originating from the inflammatory process in the fauces. When the wide and loose capsular ligaments of the upper intervertebral joints are distended with inflammatory exudate a faulty position may readily result. This faulty position may later on be fixed and aggravated through a contracture of the muscles caused by the pain.

All the patients were treated on Bradford's splint with the

neck in a position of hyperextension and with traction on the head. The treatment was commenced 3 weeks—6 months after the onset of the symptoms and led to a good result in all the

Fig. 5.
Healthy individual, M. H. Lateral view with left side towards the film, the head rotated 30° to the right. At p. the proc. lat. atlant. sin., at l. the anterior edge of the left articular surface of the atlas.

cases. Grisel's cases and others have even been treated by traction with a good result.

Buka and Dahmann describe septic spondylitis in the bodies of the upper cervical verterbrae, the former after phlegmonous angina, the latter after tonsillectomy under local anæsthesia.

A similar case, a septic spondylitis in C III—IV that came on after angina associated with glomerulonephritis, has been observed by the writer at the coast hospital Apelviken.

There is no doubt that there was a symmetrical luxation forwards of the atlas in Swanberg's case and in Berkheiser's and Seidler's case No. 1. It seems as if there is a unilateral

Fig. 6.

M. H. Ventrodorsal view with the head in rotation to the right. The cartilaginous space of the right intervertebral joint normal. The articular surfaces of the left intervertebral joint are beginning to cover each other.

anterior subluxation in Grisel's two cases and in Berkheiser's and Seidler's cases No. 2 and 5. In lateral view all these case show a distinct diastasis between the anterior arc of the atlas and the dens. A symmetrical posterior luxation of the atlas is prevented by the dens against which the anterior arc of the atlas is resting. A unilateral posterior subluxation would have to arise through rotation of the atlas round the dens. By disloca-

tion backwards of one lateral articular surface of the atlas the articular surface of the atlas on the opposite side must be dislocated forwards. Thus a subluxation backwards on one side implies a corresponding faulty position on the other side. Normally the atlas can rotate round the epistropheus 30° to either side. On examination on a skeleton it appears that already

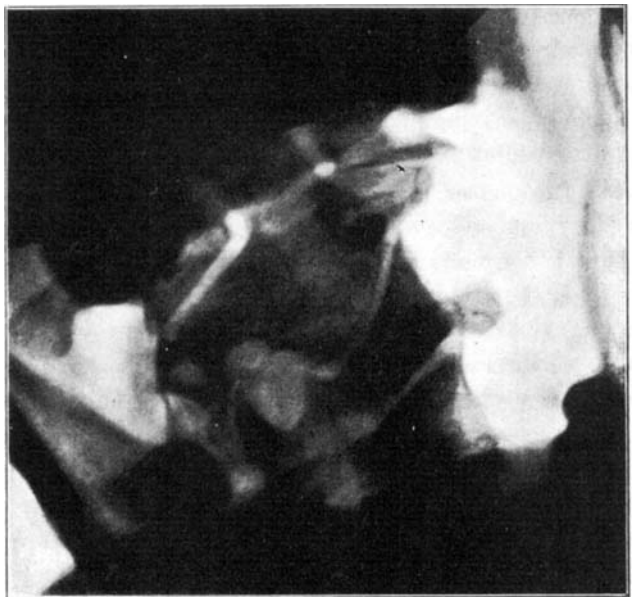
R.  L.

Fig. 7.

M. H. Ventrodorsal view with the head in maximal rotation to the right, the whole cervical column highly rotated to be right, the joint space of the right intervertebral joint visible, on the left side the articular surfaces cover each other. The epistropheus is highly rotated with the right articular surface broader than the left.

at a rotation of the atlas of about 25° to one side the processus lateralis atlantis of the opposite side will lie in front of the anterior arc of the atlas provided the visual direction is at a right angle to the sagittal diameter of the epistropheus. At a rotation of the head of about 30° to one side the processus lateralis atlantis will appear on the other side together with

the anterior portion of the articular surface of the atlas when seen in lateral view in a skiagram of the upper cervical vertebrae (Fig. 5). It should thus be easy to disclose a subluxation that has occurred through a mere rotation when it is seen in lateral view. A rotation of 50—55° is required to engender a *luxation* of the lateral intervertebral joints with »locking« between the borders of the articular surfaces. A *subluxation* backwards of one articular surface of the atlas cannot be imagined without a simultaneous subluxation forwards of the articular surface of the atlas on the opposite side, and a »rotation *luxation*« cannot be imagined without rupture or a high degree of tension of capsular ligaments and other ligaments.

Grisel, Berkheiser and Seidler state that disappearance of the cartilaginous space of one lateral intervertebral joint in the ventrodorsal picture is pathognomonic of a position of subluxation between the atlas and epistropheus. In the writer's opinion such a picture is only an evidence of a position of rotation of the vertebrae inter se. Both Grisel's and Desfosses' ventrodorsal pictures show a distinct oblique projection of the epistropheus, whilst the atlas is projected in the sagittal plane. All the details of the ventrodorsal pictures of these authors can be explained by the position of rotation between the atlas and epistropheus. The dislocation in the sagittal plane seen in lateral view shows that there exists a true subluxation in Grisel's case which cannot be said to be proved in Desfosses' case. In Berkheiser's and Seidler's picture No. 6 the epistropheus appears to be obliquely projected, the left lateral articular surface being broader than the right. As the reproduction is blurred this cannot be decided as a matter of course.

The writer has made ventrodorsal skiagrams of normal individuals with varying rotation of the head, which pictures show varying stages of lowering and disappearance of one or the other cartilaginous space of the lateral intervertebral joints (Fig. 6, 7). These pictures correspond to Grisel's, Defosses', Berkheiser's and Seidler's ventrodorsal pictures that have been quoted as evidence of subluxation.

In several of the cases of acutely occurring torticollis after

infection of the fauces reported in the literature there are signs of so severe a dislocation between the atlas and the epistropheus that the case can be regarded as one of luxation or subluxation. In other cases there is only a fixed position of rotation within the normal limits between the said vertebrae.

Grisel states that the facies morbi is caused by an infection in the retropharyngeal space with infiltration and contracture of the prevertebral muscles. Berkheiser and Seidler on the other hand presuppose an infectious arthritis in the intervertebral joints with secondary contracture of the muscles, fixing and sometimes aggravating the faulty position of the joints.

In the author's case the clinical picture corresponds in its main features with the cases of acute torticollis after infection of the fauces or after operation in the fauces described in the literature. Moreover the patient states that there is pain on pressure on the scalp with compression of the cervical part of the vertebral column. It appears distinctly from the skiagrams that there is an acute osteoarthritis of the left lateral intervertebral joint between the atlas and the epistropheus. It is not out of the question that several of the cases reported in the literature have a similar genesis, even if the arthritis in these cases has not engendered roentgenologic alterations of the articular surfaces.

SUMMARY

In a previously healthy girl, aged 16 years, an acute osteoarthritis occurs in the left lateral intervertebral joint between the atlas and the epistropheus with pain, faulty position of the head and impairment of mobility immediately after tonsillectomy under local anæsthesia. The clinical symptoms largely subside; considerably bony deposits are formed on the borders of the articular surfaces. The writer reports similar cases described in the literature. The opinion advocated by Grisel, Berkheiser and Seidler that the disappearance of one lateral cartilaginous space between the atlas and the epistrpheus should be

indicative of a subluxation between these vertebrae is wrong. This picture depends on a position of rotation between the vertebrae, which will appear from skiagrams of normal individuals. The unilateral posterior subluxation described by Berkheiser and Seidler may not exist. The writer puts the question whether several of the cases of acute torticollis after inflammation of the fauces or operation of the fauces described in the literature depend on acute arthritis in the intervertebral joints between the atlas and the epistropheus with secondary faulty position.

RÉSUMÉ

Chez une jeune fille de 16 ans, jusqu'alors saine, immédiatement à la suite de tonsillectomie en anesthésie locale une ostéoarthrite aiguë se declare dans l'articulation intervertébrale latérale gauche entre l'atlas et l'épistropheus. Les symptômes etaient des douleur, fausse position de la tête et mobilité diminuée. Les symptômes cliniques s'atténuent en quelque mesure; des dépôts osseux considérables se forment aux bords des surfaces articulaires.

L'auteur rapporte des cas analogues décrits dans la litterature. L'auteur regarde comme erronée l'opinion soutenus par M. M. Grisel, Berkheiser et Seidler, à savoir que la disparition d'un espace cartilagineuse entre l'atlas et l'épistropheus indique une subluxation de ces vertèbres. Le tableau se présente, si ces deux vertèbres sont en position de rotation, ce que montreront des radiogrammes pris sur des individus normaux. La subluxation unilatérale postérieure decrite par M. M. Berkheiser et Seidler n'existe peût-être pas.

L'auteur pose la question de savoir si un nombre des cas décrits dans la litterature comme des torticolis aigus après une pharyngite ou une intervention dans le pharynx, dependent plutôt d'une arthrite aiguë des articulation intervertébrales de l'atlas et de l'épistropheus, avec torticolis secondaire.

ZUSAMMENFASSUNG

Bei einem vorher ganz gesunden 16-jährigen Mädchen entsteht im unmittelbaren Anschluss an einer Tonsillectomie in Lokalbetäubung eine akute Osteoarthritis im linken lateralen Intervertebralgelenk zwischen Atlas und Epistropheus mit Schmerzen, Fehlstellung des Kopfes und Bewegungsbeschränkung. Die klinischen Symptome gehen grösstenteils zurück; an den Rändern der Gelenkflächen werden bedeutende Knochenablagerungen gebildet. Der Verfasser referiert ähnliche in der Literatur beschriebene Fälle. Die von Grisel samt Berkheiser und Seidler vertretene Ansicht, dass das Verschwinden der einen lateralen Knorpelspalte zwischen Atlas und Epistropheus ein Zeichen von einer Subluxation zwischen diesen Wirbeln sein sollte, ist falsch. Ein solches Gebilde beruht auf der Rotationsstellung zwischen den Wirbeln, welches aus Röntgenbildern von normalen Individuen hervorgeht. Die von Berkheiser und Seidler beschriebene einseitige Subluxation existiert wahrscheinlich nicht. Der Verfasser stellt die Frage auf, ob nicht mehrere von den in der Literatur beschriebenen Fällen von akutem Torticollis nach Schlundkopfinfektion oder -operation auf akuter Arthritis in den Intervertebralgelenken zwischen Atlas und Epistropheus mit sekundärer Fehlstellung beruhen.

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