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Preliminary CT study of C1–C2 rotational mobility in normal subjects

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Abstract A CT study of normal atlanto-axial (C1–C2) rotary mobility was carried out on ten normal immature subjects. In order to determine the limits of normality, the ten children underwent clinical and radiological examination. The clinical study included checking for objective signs of joint laxity and measurement of rotational neck mobility. The radiological study included standard lateral radiographs in neutral and maximal flexion positions and a CT scan taken in maximal left and right side rotation at the C1–C2 articular processes joint. The superpositioning of the images taken in every rotational direction showed, in all ten children, a wide contact loss between the C1–C2 corresponding facets, ranging from 74 to 85% of the total articular surface. The report on these images, carried out by three

independent radiologists, concluded that there was a rotary subluxation in all cases. In the ten children studied, there were no significant differences with regard to neck mobility or laxity signs in clinical or standard X-ray examination. Our results lead us to conclude that, except for complete C1–C2 rotational dislocation with facet interlocking, a CT scan showing a wide – but incomplete – rotational facet displacement is not sufficient to define a status of subluxation. This leads us to perceive that there is a risk of overdiagnosis when evaluating upper cervical spine rotational problems in children. The concept of both rotary C1–C2 fixation and subluxation should be revised.

Key words Cervical spine · Paediatric spine · Atlanto-axial subluxation

Introduction

Only in neutral position is the caudal surface of the atlas (C1) articular processes in full contact with its corresponding opposite in the axis (C2). Therefore, any rotational displacement of C1 on C2 should result in some degree of contact loss between the corresponding articular surfaces.

The classical concepts about atlanto-axial (C1–C2) mechanics give a 40° rotation to both the left and right [29, 30], so we can expect that a normal full rotation implies a wide articular surface contact loss. Nevertheless, not only is there no image representing this “expected fact” in

classical literature, but it is simply indicated by means of circular arrows [30] upon an anatomical schematic model representing a lateral view of C1 on C2 in neutral position. White and Panjabi [30] show a contact loss between the opposing surfaces of the C1 and C2 articular processes (with no mention of it) when trying to demonstrate the possibly troublesome lengthening that the vertebral artery undergoes when nearing its rotational limits. Furthermore, we have also found a paper showing images in which the previously mentioned surface contact loss can be seen when explaining another altogether different topic [16].

The paper by Fielding and Hawkins [8, 9] on atlanto-axial rotatory fixation has become a classic one and it is constantly referred to in almost all published papers relat-

Fig. 1 **a** Patient suffering from Grisel's syndrome, with torticollis that had lasted for 6 months. **b** CT scan of the same patient showing a Fielding and Hawkins type II rotary fixation

ing to C1–C2 mobility, instability or dislocation. Fielding and Hawkins described four degrees of contact loss between the corresponding C1 and C2 articular surfaces. The main value of their paper was imaging an axial view from a transoral and lateral cineradiography. Wortzman and Dewar [31] had previously proposed a parallel term, in which rotary fixation and subluxation were not that well differentiated and they correlated with Fielding and Hawkins' grade 1. Today, the vast majority of radiologists and cervical spine surgeons have very little clinical experience of C1–C2 rotational instability. Some therefore base their diagnostic and therapeutic decisions on these reports, while others rely on their personal experience of one or very few cases [1–3, 7, 10–12, 14, 17, 18, 21, 22, 24, 27]; sometimes a diagnosis (which may be an over-diagnosis) of instability, subluxation or rotary fixation can lead to a C1–C2 arthrodesis, especially in children.

In a recent clinical experience, we faced a controversial decision concerning a 6-year-old girl with a diagnosis of Grisel's syndrome. Her 6-month torticollis was completely corrected through manipulation under anaesthesia, which was followed by a 4-month halo-jacket immobilization. After removal of the halo, torticollis progressively appeared again; the patient was able to keep her head upright only on waking-up, torticollis appearing progressively throughout the day (Fig. 1a), while some degree of active rotational movement always existed. As a follow-up control, the patient underwent a galium scintigraphic study that showed hypercaptation in the retropharyngeal area corresponding to the C1–C2 level. A CT study was then performed and the radiologist reported type II [8] atlanto-axial rotational subluxation (Fig. 1b). Based on the clinical data, this diagnosis was rejected and antibiotic therapy was then prescribed. As a result, an immediate positive response was observed and the torticollis disappeared 5 months later, 1 year after the onset.

This experience throws up a number of questions, namely:

1. Can we still accept the classic diagnostic orientation given by Fielding and Hawkins [8]?
2. What is the rotational displacement limit that differentiates an atlanto-axial rotary fixation or subluxation from a wide but "normal" rotational attitude of a torticollis?
3. What does rotary fixation really mean?

The purpose of this paper is not to prove that the Fielding and Hawkins [8] statement may be inadequate, but to demonstrate the correlation between normal C1–C2 rotational mobility and its corresponding CT appearance, which has not yet been described.

If we were able to show objectively the normal range of C1–C2 rotational motion, we could probably establish a more appropriate basis to understand all the concepts related to rotary fixation.

Fig.2 Sequential CT study showing the whole set of structures used for performing measurements

Materials and methods

Our study was carried out on ten normal, healthy and asymptomatic 5- to 12-year-old children (average age: 9 years). We chose children because, in daily clinical practice, the diagnostic and decision-making difficulties referred to generally exist in infantile patients.

The clinical study included looking for objective signs of joint laxity in peripheral joints (elbow or knee extending beyond 0° , the thumb touching the forearm ventrally, the finger parallel to the forearm on extending the wrist and metacarpophalangeal joints, ankle dorsal flexion over 45° and flat foot) [32]. The laxity test was considered positive when three of the six signs searched for were found. Measurement of the active cervical rotational mobility was performed with a goniometer, recording the bilateral nose displacement with respect to the coronal plane from the top of the head. Measurement of the rotational mobility and laxity test were both recorded with the purpose of correlating them with the CT study.

The radiological study included standard lateral radiographs in neutral and maximal flexion positions to define the following:

1. Absence of normal cervical lordosis in neutral position
2. Increased distance between the odontoid process and anterior arch of the atlas

Fig.3 Angular measurements of C1–C2 rotation to the left (a) and right (b) sides. The images correspond to a 7-year-old girl

3. Absence of a uniform angle between adjacent vertebrae during neck flexion
4. C2–C3 pseudosubluxation
5. Congenital abnormalities

CT scans were taken in maximal left and right side rotation at the level of the C1–C2 articular processes joint (Fig.2); a superpositioning of the obtained images was carried out in every direction to determine the rotational movement related to C1 and C2 transverse axes (Fig.3) and the contact loss between C1 and C2, by measuring the area of C1 articular surface in contact with its corresponding C2 opposite, as shown in Fig.4.

All CT scans were studied by three independent radiologists, who were not informed that they belonged to normal people.

Results

The laxity test proved to be positive in six of the ten children.

After clinical examination, the average active rotational mobility was 68° (60° – 80°) in both left and right rotations (total: 136°).

Fig. 4 Schematic representation of the percentage measurement of the loss of contact between the C1 and C2 articular surfaces in left (a) and right (b) rotation. The images correspond to the same 7-year-old girl

Fig. 5 CT scan showing an osteoid osteoma of a C1 articular process that was accidentally found while searching for an abnormal C1–C2 position in a child with torticollis, who does not belong to the present study

Standard radiological examination was normal in all cases. Three children had absence of normal cervical lordosis in neutral position. The average distance between the odontoid process and anterior arch of C1 during flexion was 2.4 mm (range 2–3 mm). None had absence of a uniform angle between adjacent vertebrae with the neck in flexion; physiological C2–C3 pseudosubluxation was not observed.

The superpositioning of the CT scans taken in every rotational direction showed an average rotational mobility

of 36° (32°–44°) (Fig. 2) and a wide contact loss between the C1–C2 corresponding facets in the ten children; the contact loss ranged from 72 to 85% (average 78%) of the total articular surface. In the ten children under study, there were not significant differences related to neck mobility or laxity signs in clinical or standard X-ray examination.

The report given by the three independent radiologists who worked as blind referees was identical in all cases: rotary subluxation.

Discussion

Infection of the neck or pharynx, a surgical procedure on the pharynx, tumor or trauma can lead to neck fixation with rotated position of the atlanto-axial segment. The mechanism that causes this – shall we call “situation” – is not fully understood and may possibly be secondary to simple muscle spasm. This “situation” has appeared in specialized literature as rotary or rotatory dislocation or deformity [6], rotational subluxation [15, 31], rotary fixation and spontaneous hyperemic dislocation [28]. Fielding and Hawkins [8] preferred the term atlanto-axial rotatory fixation, since the fixation of the atlas on the axis may occur with subluxation, dislocation or when the relative position of the atlas and axis is still within the normal range of rotation, without specifying this point. We have not been able to find a book or paper on anatomy, biomechanics or diseases of the spine pointing out how C1 really rotates upon C2 or what the corresponding facets of C1 and C2 really look like during rotation.

Our results (36° of bilateral rotation, total: 72°) agree with the already established and accepted value of 40° for bilateral rotation. However, in addition, we report that 36° of rotation correlates with a very wide contact loss of the corresponding facets of C1 and C2 (80%, far more than was supposed). This normal contact loss, shall we call “facet stripping”, is actually unknown and does not correspond to the one shown by White and Panjabi in their book when explaining C1–C2 rotation by means of an “over the top” view [31].

Fielding and Hawkins [8] proposed a classification that included four types of rotational displacement on the seventeen patients in their series. Rotary fixation without anterior displacement of the atlas within a normal range of atlanto-axial rotation (type 1) was the most common deformity, whilst it is believed that the transverse ligament remains intact. They recommended posterior atlanto-axial arthrodesis to relieve the symptoms in patients with a late diagnosis (chronic cases) of rotary atlanto-axial subluxation, which is usually considered to be unreducible. Although these recommendations seem to be rational and, of course, widely accepted, no accurate indications exist to differentiate fixation in a normal rotation limit from a subluxation. Furthermore, the criterion to establish that a case is unreducible has yet to be defined. Consequently, spe-

cialists tend to assimilate both diagnostic possibilities (both considered as subluxations) and to accept that all chronic fixations are unreducible. A further difficulty is that there is no clear definition of chronicity, and the “failure of conservative treatment” could be a very vague concept when the treatment applied is a symptomatic one and the cause of the fixed rotation is unknown. The solved Grisel’s syndrome case we experienced and commented on, and other cases published by Schwartz [25, 26] or personally communicated by other authors, have been the generators of the present study on normal C1–C2 rotation. These “chronic” cases of rotary fixation, which healed in just a few months (not always spontaneously) constitute firm evidence that seems to hint that the classic concepts, which state chronic cases are unreducible and subsequently need surgery, are mistaken. We are not saying that a remedy for every lengthy case of “fixed rotation”, especially those with no etiological diagnosis, may be found in applying antibiotics and patience, but the cases we refer to above should lead us to reflection.

In daily practice, the chances of ever coming across torticollis with rotary fixation are real but infrequent. Owning to the fact that the experience of even a highly specialized physician is currently not very wide, when facing a prolonged torticollis they have to look for a reliable source of information. Therefore, since the Fielding and Hawkins [8] paper is the normal reference point, whatever the consulted bibliography may be, when radiological studies are performed, it is usually to “look for” an abnormal position of C1 articular processes with respect to their corresponding ones at the C2 level. The interpretation of the radiological study is often difficult and complex, especially with children. Cervical rotation resulting from patient attitude in the case of Grisel’s syndrome or other clinical conditions can cause radiographic changes that are indistinguishable from rotary atlanto-axial subluxation [8, 19, 20, 30]. Open-mouth radiographs of the upper cervical spine can be made by having the patient rotate their head to each side [15, 31], but they are also difficult to interpret [8, 15]. Unfortunately, a patient’s lack of co-operation or diminished active motion of the neck may prevent these special radiological studies [5, 7, 22]. Fielding and Hawkins recommended cineradiography, but patient co-operation may be inadequate due to pain and spasm [8]. Hence, considering the diagnostic prospects we have today, CT is proving to be widely accepted as mandatory and definitive. In our cases, the radiologists’ report of rotary subluxation in all cases should make us aware of the

prospect of making diagnoses of rotary subluxation in any case of rotation within the limits of normality. In such cases, rotation may be a fixed one if muscle spasm exists and, according to up-to-date knowledge, the most objective diagnosis will be rotary (in a rotated position) fixation (rigid) – just a clinical sign, not a disease.

Standard CT scans of the upper neck may only show the rotation of the atlas on the axis; this rotation is not currently accepted as being within the normal range of motion, because there are no previous reports of any CT studies on normal C1–C2 rotation. The possibility of diagnosing “subluxation” with a temporary neck-rotated position within the normal limits of C1–C2 motion – as our study strongly suggests – has not been sufficiently considered in any previous study.

Some misconceptions may reasonably exist about rotary fixation or subluxation, but everybody has real difficulties in approaching the truth based on evidence or confirmed knowledge. In his opening speech to all members of the Cervical Spine Research Society (European Section), Grob [13] urged us to change our “chip” from the asserted, widely accepted, consensus or experience-based knowledge to a firmer scientific line, based on evidence.

The results of our study lead us to think that, except for complete C1–C2 rotational dislocation with facet interlocking, CT views showing a wide, but incomplete, rotational facet displacement are not sufficient to define a status of subluxation. The concept of both rotary fixation and subluxation must be revised and changed. Otherwise, CT may not be very useful in torticollis assessment, except for finding (sometimes accidentally, Fig. 5) the causal condition, when it is not seen in standard radiographs.

Our results are of course preliminary and will need further validation or confirmation; we are already working on the possibility of studying a larger pool of normal children and adults. Although a similar study would also be interesting in abnormal subjects, the low incidence of rotary fixation and the great difficulty of carrying out dynamic studies in patients with painful muscle spasm render such a study – at the moment – quite utopian.

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