C2-fractures: part I. Quantitative morphology of the C2 vertebra is a prerequisite for the radiographic assessment of posttraumatic C2-alignment and the investigation of clinical outcomes

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Abstract Pertinent literature exists concerning indications, techniques, complications of treatment, and risk factors for nonunion in axis and odontoid fractures; however, there are scarce data regarding the incidence and definition of malunion in these fractures. As a prerequisite for the study of anatomical alignment following surgical and nonsurgical treatment of C2-fractures, an understanding of normal C2 anatomy is essential. Therefore, the authors intended to evaluate morphometrical dimensions of the C2 vertebra. The purpose was to provide normalized quantitative data to enable assessment of malalignment following the treatment of C2-fractures within a classification system. Using digitized cervical spine lateral and transoral odontoid radiographs of 100 consecutive patients without any evidence of traumatic or neoplastic disorders, the authors performed measurements on distinct anatomical

structures and investigated morphometrical dimensions of the normal axis vertebra. The incidence of atlantoaxial arthritis was also evaluated. In addition, with the assessment of twenty arbitrarily chosen sets of radiographs by three different observers we calculated the interobserver reliability in terms of intraclass correlation coefficients for each parameter. With calculation of SD and 95% confidence limits, pathological cut-offs were reconstructed from measurements performed resembling non-physiological and pathological limits. Distinct parameters were selected to form a new classification system for radiographical follow-up that focuses on the quantitative C1–2 vertebral alignment. The measurement process resulted in 2,400 data points. Distinct morphometrical parameters, such as a quantitative characterization of the sagittal atlantoaxial congruency, the lateral mass inclination and the type of degenerative changes at the atlantoaxial joint could be demonstrated to be valuable and reliably used within a proposed classification for C2-malunions following C2-fractures. The current study offers a template including recommended radiological measurements for further research on the study of clinical outcome and posttraumatic alignment following C2-fractures.

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Introduction

Controversy remains over the appropriate classification and treatment of C2-fractures [23, 28, 33, 39, 45, 58, 63]. There is yet no single comprehensive treatment-related classification that stratifies C2-fractures according to their

propensity to heal with or without surgery and their propensity to heal anatomically or with distortion of atlantoaxial anatomy. Previous studies concentrated on indications, complications, risk factors for and incidence of nonunion as well as the technical and biomechanical advantages of surgeries to stabilize C2-fractures [2, 3, 6, 19–21, 23, 24, 29, 33, 39, 45, 49, 55, 58, 63, 64, 67]. Although a number of practice guidelines have been developed [33], outcome measures seldom included patient's satisfaction or validated outcome measures, the incidence of long-term disability [1, 9, 19, 20, 39, 63, 67] or the quality of anatomical restoration achieved [1, 8–10, 37]. Following nonsurgical treatment of C2-fractures, the fragments frequently yield fusion in a position of slight to severe malunion, a fact that is rarely recognized and underreported [26, 31, 46, 49]. Accordingly, data on the incidence and definition of quality in achieving and maintaining reduction in C2-fractures are scarce and the literature lacks reports on the impact of the posttraumatic C2-alignment on the clinical outcome and the resulting C1–2 axial rotation [1, 2, 8, 9]. It remains a little-known clinical entity that, with nonsurgical treatment, a considerable number of distinct C2-fracture patterns (including multiple C2-fractures and combined C1–2 fractures, atypical hangman's fractures, comminuted odontoid type II fractures, coronal and oblique vertebral body and type III odontoid fractures, lateral mass split and burst fractures [18, 20, 29, 38–40, 43]) and particularly those involving the superior articular facets of C2 frequently heal with slight to severe distortion of the C2-anatomy resulting in atlantoaxial incongruity [18, 20]. Some of these malunions are symptomatic in regard to cervical pain and limited rotation of C1–2 [31], and can cause painful atlantoaxial osteoarthritis (AAOA) [20, 31].

To assess any distortion of the C2 anatomy and to define a 'C2-malunion', one needs data of normals for comparison. Therefore, as a prerequisite for the assessment of the posttreatment anatomical C2-alignment after C2-fractures, the authors intended to investigate the anatomical C2-dimensions and C1–2 relationships in a sample of 100 healthy patients. The current study yields a physiological standard for the main parameters that describe the *in vivo* C2 anatomy and the incidence of AAOA. In addition, based on this physiological standard our purpose was to describe a classification system that delineates the post-treatment alignment in C2-fractures. The adoption of the proposed classification might allow a more accurate assessment of posttraumatic C2-alignment. It might enable comparative studies including the investigation of C1–2 axial rotation and clinical outcome.

In part II of the current project on the outcome of C2-fractures, the classification of the C2-alignment is

applied on clinical cases and the impact of the posttreatment C2-alignment on the remaining C1–2 rotation and clinical outcome is evaluated.

Materials and methods

Our study sample consisted of 100 consecutive patients who had indications for cervical spine radiographic investigation. Only radiographs showing no evidence of cervical fracture, ligamentous instability, rheumatoid arthritis, ankylosing spondylitis, DISH or neoplastic disorder were included. Lateral and transoral radiographs were selected only if there were no characteristics of a misaligned X-ray beam defined by congruency of the atlantoaxial joints on transoral odontoid views and the absence of duplication of the C1 and C2 cortical boundaries on both lateral and transoral views. In total, 54 female and 46 male patients were enrolled for anatomical measurements. Their mean age was 49.8 years (range 15–94), 44.7 years in male and 54.0 years in female patients. The radiographs were taken on a digital X-ray system (Vertix 3D-III unit, Siemens, Germany) with the patient in sitting position and stored digitally (PACS Magic View VC 42, Siemens, Germany). Using the cursor, digital measurements on the radiographs (0.1 mm increments) were performed with a commercial software program (Escape Medical Viewer V3, Escape, Greece).

The C2 vertebra consists of a body, paired pedicles, lateral masses (superior articulating facets), odontoid, pars interarticularis, inferior articulating facets, lamina, and bifid spinous process. Each structure can be involved in C2-fractures [38, 40, 43], therefore different measurements on transoral and lateral radiographs were chosen to analyse anatomical structures of clinical interest that can be affected. Besides absolute numerical parameters, distinct ratios in the sagittal plane and side-related differences in the coronal plane were calculated from the single parameters measured. The anatomical structures evaluated, dimensions measured and ratios calculated, as well as the descriptions of measuring techniques are summarized in Tables 1, 2 and Figs. 1, 2. One measuring technique requires further explanation: in clockwise fashion, the Harris-Ring-C2 [22, 26, 27, 48, 52, 61] resembles the radiological projection of the anterior-superior and superior parts of the C2 pedicle, the posterior vertebral border of C2, the caudad part of the transverse foramen and process, and the anterior border of the C2 superior lateral mass. Radiologically, the Harris-Ring-C2 is elliptical in shape (Fig. 1), but can be recognized in almost any lateral cervical spine radiograph. Therefore, we determined the superior depth of the vertebral body (sVBD) on a line perpendicular to the tangent of the C2 posterior wall at the level of half the diameter of the Harris-Ring-C2. Radiologically,

Table 1 Description of anatomical landmarks and measurement techniques used on lateral cervical spine radiographs

Abbreviation	Measurement	Description and technique
aOL	Anterior odontoid line	Tangent line drawn along the anterior odontoid cortex
sPEP	Sagittal plane of endplate	Line drawn connecting the antero-inferior and postero-inferior corner of the axis body (constructed 'endplate of C2')
aVBL	Anterior vertebral body line	Tangent line drawn along the anterior cortex of the axis body
pVBL	Posterior vertebral body line	Line drawn along the posterior cortex of the axis body
iVBD	Inferior vertebral body depth	Distance between antero-inferior and postero-inferior corner of axis body
sVBD	Superior vertebral body depth	Distance between posterior and anterior vertebral body line at the square diameter of the Harris-ring-C2
sDSC1	Sagittal diameter spinal canal C1	Perpendicular distance between posterior vertebral body line and anterior cortex of the posterior arch of the atlas
sDSC2	Sagittal diameter spinal canal C2	Perpendicular distance between posterior vertebral body line and anterior cortex of the axis lamina
sVBH	Sagittal vertebral body height	Distance between intersection point of pVBL with sPEP and level of upper bow of the Harris-Ring-C2
sOTH	Sagittal odontoid tip height	Distance between intersection point of pVBL with sPEP and level of odontoid tip
sR1	Sagittal ratio 1	Ratio of sOTH to sVBH
sR2	Sagittal ratio 2	Ratio of iVBD to sVBD
sR3	Sagittal ratio 3	Ratio of sDSC1 and sDSC2
sA1	Sagittal angle 1	Sagittal angle formed between sPEP and aOL
sA2	Sagittal angle 2	Sagittal angle formed between sPEP and aVBL
sA3	Sagittal angle 3	Sagittal angle formed between pVBL and sPEP
sA4	Sagittal angle 4	Sagittal angle formed between aOL and pVBL

an increased sVBD with widening of the axis body is characterized as the 'C2 fat sign' [52, 61]. The superior vertebral body height (sVBH) was measured from the caudad posterior corner of the axis vertebra to the upper cortical merging of the C2 pedicle, resembling the superior bow of the Harris-Ring-C2.

To analyse degenerative changes of the atlantoaxial joints observed in elderly patients or following C2-fractures in younger patients [20, 31], all C1–2 joints were scored on transoral odontoid views according to Lakshamanan [44] (Table 3), and the median height of the C1–2 joints was measured. As the study showed difficulties in differentiating the subtypes 0 and 1, as well as type 2 and 3, statistical calculations were performed to stratify a group A (none or mild degenerative changes C1–2, type 0 and 1 [44]) and a group B (advanced degenerative changes C1–2, type 2 and 3 [44]). Hence, consistent with a previous statistical analysis [44], only moderate and severe changes were considered as resembling degenerated and arthritic joints, respectively.

Radiographic examples for each kind of degenerative grades at the C1–2 joints are illustrated in Fig. 3.

The first author performed all measurements in 100 sets of radiographs. However, the value of anatomical measurements strongly depends on their reproducibility within the use of different observers. Therefore, reliability was evaluated by interobserver testing between three of the authors who assessed 20 arbitrarily chosen sets of radiographs. Interobserver differences were evaluated using the intraclass correlation coefficient (ICC). An ICC score from 0 to 0.4 was rated poor, 0.4 to 0.75 fair or moderate, and >0.75 excellent [60].

To assess the severity of any pathological alignment following fracture and union of the C2 vertebra, a classification system was constructed based on the anatomical measurements performed. The classification should equilibrate morphometrical changes measured on radiographs and the descriptive assessment of the C1–2 alignment. Measured dimensions and ratios were selected for inclusion

Table 2 Description of anatomical landmarks and measuring techniques used on transoral odontoid views

Abbreviation	Measurement	Technique and description of measurement
cOML	Coronal odontoid mid-line	Bisector-line of odontoid
sLML	Superior lateral mass level	Line drawn between superior corner of left and right lateral mass
iLML	Inferior lateral mass level	Line drawn between inferior corner of left and right lateral mass
lLMP	Left lateral mass plane	Line drawn from left inferior to superior corner of lateral mass
rLMP	Right lateral mass plane	Line drawn from right inferior to superior corner of lateral mass
lLMH	Left lateral mass height	Line drawn between intersection point of perpendicular mid-sagittal line of iLML and superior corner of left lateral mass
iLMH	Right lateral mass height	Line drawn between intersection point of perpendicular mid-sagittal line of rLML and superior corner of right lateral mass
lAAJH	Left atlanto-axial joint height	Distance between inferior facet of left lateral mass of C1 and superior facet of C2 at center of lateral atlanto-axial joint
rAAJH	Right atlanto-axial joint height	Distance between inferior facet of right lateral mass of C1 and superior facet of C2 at center of lateral atlanto-axial joint
cA1	Coronal angle 1	Angle formed between iLML and lLMP
cA2	Coronal angle 2	Angle formed between iLML and rLMP
cA3	Coronal angle 3	Angle formed between lLMP and cOML
cA4	Coronal angle 4	Angle formed between rLMP and cOML
cA5	Coronal angle 5	Angle formed between lLMP and rLMP
cA6	Coronal angle 6	Angle formed between lLMH and iLML
cA7	Coronal angle 7	Angle formed between rLMH and iLML
cA8	Coronal angle 8	Angle formed between cOML and sLML
cA9	Coronal angle 9	Angle formed between cOML and iLML
cA10	Coronal angle 10	Angle formed between sLML and iLML ^a
Diff1	Difference cA6-cA7	Absolute algebraic difference between cA6 and cA7, left and right height of lateral mass
Diff2	Difference cA1-cA2	Absolute algebraic difference between cA1 and cA2, left and right inclination of lateral mass
Diff3	Difference cA3-cA4	Absolute algebraic difference between cA3 and cA4, right and left odontoid-facet angle

^a The cA10-angle was scaled positive if the angle inclined to the lateral mass, and negative if it declined to the opposite direction

into the classification, if standard deviations as well as upper and lower limits of the 95% ranges were shown to be small, and particularly if the ICC was shown to be ‘excellent’ with an ICC > 0.75.

Statistical analysis

In addition to overall and gender-related calculations of means, SD and ranges, upper and lower 95% ranges were calculated. Correlations among variables were analysed using Pearson’s correlation coefficient. Student t-tests were computed to analyse differences among subgroups. ICC with 95% confidence limits were used to evaluate the inter-rater reliability among the three investigators. A p-value less than 5% was considered statistically significant. All

analyses were performed with SPSS 11.0 (SPSS Inc, Chicago) and Statistica 6.1 (StatSoft Inc, Tulsa).

Results

Investigation of the in vivo C2 anatomy resulted in 2,400 data points. Means, standard deviations, ranges, and the lower and upper limits of the calculated 95% ranges are summarized in Tables 4 and 5.

Calculation of ICCs showed that all but 5 out of 22 parameters had an ICC > 0.75, judged as excellent agreement [60]. Moderate reliability was found for the assessments of the odontoid tilt angle in the coronal and sagittal planes (sA1 and sA4, cA8 and cA9).

Fig. 1 Artist's drawings depict landmarks at anatomical structures of interest for making the measurements. *Left* AP-view of axis vertebra. *Right* Sagittally sectioned axis vertebra at the level of the right C2 pedicle

Fig. 2 AP and lateral radiographs with drawings depict measurements performed. See Table 1 and 2 for description of abbreviations used. *Dashed lines* mark the accessory lines resembling the endpoints for measurement on distinct anatomical landmarks

Table 3 Grading of the severity of degenerative changes of the left and right lateral atlantoaxial joints

Classification of Lakshamanan			Modified classification	
Description	Type	Grade	Type	Description
Normal joint space with no osteophyte formation	0	None	A	Normal/mild degeneration
Narrowed joint space or normal joint space with osteophyte formation	1	Mild	A	
Obliterated joint space with or without osteophyte formation	2	Moderate	B	Advanced degeneration/ arthritic joint
Completely obliterated joint space with osteophyte formation, excrescences and/or fusion of the joint	3	Severe	B	

Our analysis showed that concerning isolated anatomical dimensions wide variations exist within the axis vertebra for 95% of the population. However, we emphasize that calculations for the sagittal 'ratios' (sR1 to sR3),

as well as for the differences between left and right sides in the coronal plane (Diff1–Diff3), showed small ranges and SDs. Inter-individually large differences were found within the numerical, but only once within the angular

Fig. 3 The applied atlantoaxial osteoarthritis grading system according to Lakshamanan et al. [44] and the modified classification (in brackets). *Left* bilaterally Grade 0 (Type A); *middle* bilaterally Grade

1 (Type A); *right* left atlantoaxial joint shows Grade 3 (Type B), right joint shows Grade 2 (Type B)

measurements with a few statistically significant differences between males and females. However, differences between left and right measurements in the coronal plane

(parameter ‘Diff1–Diff3’) calculated for gender were found to be not statistically significant. In sagittal plane the anatomical ratios calculated showed small ranges within

Table 4 Results of anatomical measurements on lateral cervical spine radiographs, $n = 100$ patients (in mm and degrees)

Parameter	Males			Females			All patients	All patients	All patients	$N = 20$ ICC**
	95% ranges			95% ranges			95% ranges			
	Means and SD range	Lower limit	Upper limit	Means and SD range	Lower limit	Upper limit	Means and SD range	Lower limit	Upper limit	
iVBD	19.2 ± 2.2* 13.9–23.5	15.1	22.8	16.7 ± 1.7 13.3–21.1	13.4	20.8	17.82 ± 2.28 13.3–23.48	13.8	22.81	0.98
sVBD	15.9 ± 1.7* 12.6–18.8	12.9	18.6	14.1 ± 1.6 9.7–18.2	10.0	16.7	14.90 ± 1.88 9.67–18.76	10.48	18.56	0.90
sDSC1	25.1 ± 3.0* 19.3–33.1	20.4	32.0	23.2 ± 2.1 19.0–29.7	19.2	27.1	24.11 ± 2.69 18.98–33.06	19.26	31.06	0.98
sDSC2	22.2 ± 2.5* 15.2–28.4	16.2	27.7	20.8 ± 3.1 14.6–38.5	15.8	24.3	21.49 ± 2.91 14.61–38.49	15.79	27.67	0.96
sVBH	26.3 ± 2.4* 21.8–33.2	22.7	31.3	23.1 ± 1.8 19.4–28.0	20.0	27.6	24.57 ± 2.61 19.41–33.18	20.34	30.12	0.91
sOTH	45.6 ± 4.4* 37.5–58.8	38.4	52.1	41.0 ± 4.1 20.5–48.2	34.2	47.3	43.11 ± 4.78 20.5–58.77	35.33	51.93	0.93
sA1	63.1 ± 6.0* 48.1–75.1	53.9	72.2	59.1 ± 6.0 44.0–71.6	48.7	70.8	60.93 ± 6.28 44.0–75.1	48.7	71.6	0.58
sA2	68.1 ± 5.5 55.0–81.8	56.2	78.8	66.8 ± 4.4 55.5–75.1	56.3	74.6	67.40 ± 4.95 55.0–81.8	56.2	77.2	0.56
sA3	79.5 ± 5.0 64.2–90.2	71.1	88.4	79.0 ± 5.0 70.4–98.4	71.8	87.1	79.22 ± 4.99 64.2–98.4	71.1	88.4	0.76
sA4	17.0 ± 5.7 6.3–37.5	7.10	27.10	18.8 ± 5.7 6.8–34.7	8.50	27.80	17.96 ± 5.72 6.3–37.5	7.1	27.8	0.37
sR1	1.7 ± 0.1 1.5–2.2	1.50	1.99	1.8 ± 0.1 0.9–2.0	1.57	2.00	1.76 ± 0.14 0.91–2.21	1.5	2.0	–
sR2	1.2 ± 0.1 1.1–1.5	1.09	1.36	1.2 ± 0.1 1.0–1.5	1.00	1.47	1.20 ± 0.10 1.00–1.52	1.04	1.47	–
sR3	1.1 ± 0.1 0.9–1.4	0.92	1.34	1.1 ± 0.1 0.6–1.7	1.00	1.30	1.13 ± 0.12 0.61–1.70	0.92	1.34	–

* Statistically significant differences among male and female patients, $P < 0.05$; ** ICC calculated from measurements on 20 arbitrarily chosen patients by three different observers; values in bold type denote excellent interobserver reliability

Table 5 Results of anatomical measurements on transoral odontoid views, $n = 100$ patients (in mm and degrees)

Parameter	Males			Females			All patients	All patients	All patients	<i>N</i> = 20 ICC**
	95% ranges			95% ranges			95% ranges			
	Means and SD range	Lower limit	Upper limit	Means and SD range	Lower limit	Upper limit	Means and SD range	Lower limit	Upper limit	
IAAJH	3.7 ± 0.8 1.4 to 5.9	1.79	5.06	3.2 ± 0.9 0.3 to 5.3	0.51	4.64	3.41 ± 0.88 0.25 to 5.92	1.43	5.06	0.8
rAAJH	3.8 ± 0.8 2.3 to 6.3	2.36	5.41	3.3 ± 1.0 0.0 to 5.5	1.47	5.07	3.53 ± 0.94 0.00 to 6.25	1.79	5.41	0.73
cA1	23.8 ± 2.9 18.8 to 31.7	18.5	29.1	23.1 ± 3.4 15.0 to 30.5	15.1	29.1	23.44 ± 3.21 15.0 to 31.7	18.1	29.1	0.96
cA2	23.9 ± 4.0 14.6 to 35.2	17.3	30.8	22.9 ± 3.8 15.5 to 31.1	15.8	29.3	23.35 ± 3.90 14.6 to 35.2	15.8	30.8	0.96
cA3	114.6 ± 3.9 106.4 to 124.0	107.1	123.0	113.8 ± 5.0 101.1 to 130.7	103.80	123.8	114.16 ± 4.52 101.1 to 130.7	105.0	123.8	0.96
cA4	112.9 ± 4.4 102.6 to 122.9	106.4	122.5	111.9 ± 4.8 99.1 to 121.6	99.5	120.8	112.39 ± 4.67 99.1 to 122.9	101.0	121.6	0.98
cA5	132.5 ± 5.9 120.7 to 147.4	121.9	143.1	134.0 ± 7.1 119.8 to 148.2	120.1	147.8	133.28 ± 6.57 119.8 to 148.2	120.7	147.40	0.98
cA6	37.8 ± 6.1* 26.0 to 52.7	26.4	49.0	34.2 ± 5.3 22.6 to 45.5	23.9	43.7	35.88 ± 5.93 22.6 to 52.7	24.8	47.4	0.88
cA7	37.9 ± 6.4* 22.7 to 52.2	26.2	51.6	34.8 ± 5.8 21.1 to 48.6	25.0	44.8	36.26 ± 6.28 21.1 to 52.2	25.0	51.0	0.77
cA8	89.7 ± 2.1 86.2 to 95.3	86.2	93.2	89.5 ± 2.9 76.1 to 95.6	82.8	95.5	89.62 ± 2.57 76.1 to 95.6	84.7	95.3	0.55
cA9	89.2 ± 2.0 85.3 to 94.2	85.9	92.4	89.6 ± 2.0 76.1 to 95.8	83.2	94.9	89.46 ± 2.55 76.1 to 95.8	83.6	94.2	0.59
cA10	0.4 ± 1.8 −2.5 to 7.9	−2.00	3.50	−0.1 ± 1.1 −2.2 to 2.1	−2.20	1.80	0.14 ± 1.44 −2.5 to 7.9	2.2	3.0	0.84
Diff1 (cA6-cA7)	0.0 ± 3.9 7.5 to 8.9	−6.60	7.20	−0.6 ± 3.7 −6.0 to 11.1	−5.90	6.60	0.34 ± 3.79 −7.5 to 11.0	6.0	7.2	−
Diff2 (cA1-cA2)	−0.1 ± 2.6 −6.5–6.7	−4.20	5.90	0.2 ± 2.0 −3.9 to 5.1	−3.90	3.80	0.1 ± 2.3 −6.5 to 6.7	3.9	5.1	−
Diff3 (cA3-cA4)	1.7 ± 5.0 −9.3 to 15.3	−7.09	14.70	1.5 ± 6.7 −13.2 to 31.2	−9.20	15.8	1.6 ± 5.94 13.2 to 31.2	9.2	15.3	−

*Statistically significant differences among male and female patients, $P < 0.05$; ** ICC calculated from measurements on 20 arbitrarily chosen patients by three different observers; values in bold type denote excellent interobserver reliability; negative values in calculating differences exist due to strict comparison of, i.e., left side to right side (i.e., 100° – $110^\circ = -10^\circ$)

the axis and the C1–2 vertebrae (parameter ‘sR1–sR3’). With the latter, slight gender-related differences were statistically non-significant.

Concerning age-related differences the statistical analysis showed a significant correlation between increasing age and the measurement of iVBD ($r = 0.26$; $P = 0.01$), sVBD ($r = 0.21$; $P = 0.04$), and sR1 ($r = 0.44$; $P < 0.001$); see Fig. 4.

Atlantoaxial joint morphology

With the use of a modified classification that characterizes atlantoaxial joints as ‘normal/moderately degenerated’ and ‘advanced degenerated/arthritis’ according to our definition of Type A and B (see Table 3), the incidence of AAOA was 5.0% ($n = 5$). Three patients had bilateral joints of Type B and 2 had unilateral Type B. As concerns

Fig. 4 Scatterplot with regression lines depict age-related increase of sR1

the influence of age on the median height of the atlantoaxial joint space (IAAJH/rAAJH), we observed that patients with Type A changes were significantly younger (left atlantoaxial joints: mean age 48.7; right atlantoaxial joints: 47.9 years) compared to those patients with Type B changes (left atlantoaxial joints: 69.8 years; right atlantoaxial joints: 84.6 years, $P = 0.045$; see Fig. 5). Statistical analysis revealed that at the left and right atlantoaxial joints, patients with Type A changes showed a mean IAAJH and rAAJH of 3.52 ± 0.7 and 3.63 ± 0.8 mm, respectively. In contrast, patients with Type B changes showed a mean IAAJH of 1.31 ± 0.9 mm and rAAJH of 1.60 ± 0.9 mm. The differences regarding the AAJH between Type A and B were strongly significant (IAAJH: $P < 0.0001$, rAAJH: $P < 0.0001$).

For the purpose of forming the classification system of posttreatment C2-alignment, the cut-off for Type A and B changes of the AAJ was arbitrarily set with a joint space height of ≤ 2.0 mm with the decision being strongly

influenced by the differences of AAJH in Type A and B joints. Our evaluation process demonstrated that judging different grades of AAOA with four subtypes according to the classification of Lakshamanan et al. [44] was difficult, both for the differentiation of type 1 and 2 as well as type 3 and 4. To simplify the assessment of AAOA, we applied a modified version that showed an ICC of 1.0 and 0.85 for IAAJH and rAAJH, respectively, denoting excellent agreement. The modified classification might ease the evaluation process of AAOA, making it sufficiently accurate to assess posttraumatic degenerative changes.

Creating the classification system

The clinical relevance and reproducibility of morphological parameters measured as well as their application in a classification in a future clinical study was one of the decisive factors to perform the current study. The use of a classification without evidence of good interobserver reliability can result in inconsistent results [7]. Therefore, dimensions and ratios measured were selected for inclusion into the classification based on their SD and upper and lower limits of the 95% ranges if the ICC was shown to be 'excellent'. Two parameters showed fair ICCs (sA4 and sA1), but were included as both measurement assess the odontoid inclination in sagittal plane. Drawing tangents along the anterior odontoid cortex was shown to be less reproducible, but significant deviations from the physiological standard that can be detected using this measurement add information to the assessment of C2-alignment following, e.g., an odontoid fracture. The parameters and morphometrical cut-offs recommended for application in future studies addressing posttraumatic C2-alignment are listed in Table 6. The proposed classification system of posttraumatic C2-alignment (CPA-C2) includes an ordinal system to grade the C2 vertebral shape and C1–2 alignment with four

Fig. 5 Age-related decrease of rAAJH (left) and IAAJH (right), respectively

Table 6 Recommended parameters for the radiological assessment of alignment following C2-fractures

Sagittal plane	Parameter	Description of fracture sequelae assessed	Threshold values	Score (points)
OR	sR1	Loss of vertebral/odontoid height	≤ 1.5 or $2.0 \leq$	1
	sR2	Vertebral widening	≤ 1.0 or $1.5 \leq$	1
	sR3	Spinal stenosis/sagittal subluxation C1–2	≤ 0.9 or $1.3 \leq$	2
	sA1	Odontoid inclination	$\leq 50^\circ$ or $71 \leq$	1
	sA4	If sA1 is not applicable due to fracture at sPEP	$\leq 7^\circ$ or $28^\circ \leq$	
	sA3	Distortion of posterior/inferior vertebral body	$\leq 70^\circ$ or $90 \leq$	1
				Max.: 6 points
Coronal plane	Parameter	Description of fracture sequelae assessed	Threshold values	Score (points)
OR	cA1 and cA2	Loss of lateral mass height	$\leq 17^\circ$ and $30^\circ \leq$	1
	or Diff 2		$5^\circ \leq$	
	cA6 and cA7		$\leq 25^\circ$ or $48^\circ \leq$	
	or Diff 1	Loss of lateral mass height/change of odontoid tilt	$7^\circ \leq$	1
	cA3 and cA4		$\leq 103^\circ$ or $122^\circ \leq$	
	or Diff 3		$15^\circ \leq$	
	cA5	Incongruency of left & right lateral mass	$\leq 121^\circ$ or $147^\circ \leq$	1
IAAJH or rAAJH Type A and B			Change to < 2.0 mm/Type B ^a	2
				Max.: 5 points

^a Any change in measurement from time at injury to follow-up at one or both C1–2 joints extending threshold values listed, or any change from Type A to Type B

Table 7 Classification of alignment following treatment of C2-fractures

Total score	Type	Grade
0	I	No malalignment
1–2	II	Mild malalignment
3–4	III	Moderate malalignment
≥ 5	IV	Severe malalignment

types (Table 7). With this classification, nine groups of morphometrical parameters are included and the evidence of any ‘pathological’ value within each group is awarded 1 or 2 points. The total point score is accumulated by grading the overall C2-alignment in the sagittal and coronal planes from a minimum of 0 points, delineating no malalignment, to 5 or more points, delineating severe malalignment. Because C1–2 spinal stenosis, which is described as ‘sR3’, and posttraumatic arthritis C1–2, described as IAAJH/rAAJH and Type B changes are supposed to represent serious sequelae following a C2-fracture, each is assigned 2 points in case of ‘pathological’ values. With this classification, different parameters are grouped, i.e., sA1 and sA4, as both assess the odontoid tilt angle. However, if the inferior endplate of C2 is altered due to a distinct fracture pattern, sA1 cannot be applied because of the distortion of its

reference plane (sPEP). The same is true for distinct groups of parameters in coronal plane: loss of height of the lateral mass in the coronal plane due to collapse or fracture can be indicated by changes in several parameters (cA1 and cA2, Diff 2, cA6 and cA7, Diff 1, as well as cA5). But, for example, if cA1 and cA2 are within normal limits and only cA3 or cA4 shows pathological values, there is evidence of an isolated ‘pathological’ odontoid tilt towards the left or right side. Figure 6 illustrates the clinical application of the proposed classification in a case of C2-malunion.

There was a statistically significant correlation of increasing age and sR1 ($r = 0.44$; $P < 0.001$). With the proposed classification, measurements of sR1 adjusted for age according to Fig. 4 might be used to guide accurate assessment of the C2-alignment. For clinical application of the classification, the small differences associated with age-corrected values of sR1, i.e., 0.075 between a 40- and 80-year-old patient, will have marginal impact. Distinct upper and lower cut-offs were therefore selected for the sR1 parameter. Gender was shown to statistically affect the numerical dimensions of some parameters (see Tables 4, 5), however, calculation of the anatomical ‘ratios’ and side-related ‘differences’ indicated no significant dependence upon gender. Therefore, with the classification separation of values according to gender was not performed.

Fig. 6 Illustrative case example for the application of the posttreatment C2-alignment, see text for explanation

Illustrative case example

The classification system will be explained on a case illustrated in Fig. 6. A 45-year-old patient with an odontoid type III fracture affecting both superior articular facets of C2 was treated by means of a Minerva cast for 3 months. Radiographic follow-up at 25 months depicted osseous malunion in the sagittal plane. The patient showed motion-induced pain and his total ROM for rotation in a flexed position was 20°. The measurement of parameters and the calculations performed on the lateral radiograph (Fig. 6, left) showed that sR3 (Ratio sDSC1 : sDSC2) was 0.83, sR2 (Ratio iVBD:sVBD) was 0.88, sA1 was 72.9° and sA4 was 6°. Hence, measurements exceeded the physiological 95% ranges of a normal population. In the coronal plane, all measurements and calculations performed were within normal 95% ranges (rAAJH = 3.94 mm and lAAJH = 4.18 mm) with no changes compared to time of injury (cA6 = 40.5°; cA7 = 38.8°; Diff1 = 1.7°; cA1 = 21.4°; cA2 = 20.9°; Diff2 = 0.5°; cA3 = 108.5°; cA4 = 105.1°; Diff3 = 3.4°). There were no signs of advanced degenerative changes at the atlantoaxial joints at final follow-up compared to time at injury (radiographs not shown). Hence, the C1–2 joints were graded as Type A.

In the sagittal plane, according to our proposed classification system, 2 points are assigned for malalignment in terms of C1–2 subluxation with the atlas protruding anteriorly (sR3 value), 1 point for widening of vertebral body with deformity (sR2 value) and 1 for odontoid malalignment (sA1 and sA4 value both assess odontoid tilt angle). In the coronal plane all measurements were within normal limits. The total score summed from single score values

was 4, denoting Type 3 ‘moderate malalignment’ of the axis vertebra.

Discussion

The majority of anatomical *in vivo* dimensions investigated in the current study have not been studied previously as a whole, any isolated dimensions in *ex vivo* studies, or used in the comparison of differing measuring techniques [11, 41, 65]. The authors are not aware of any study that included interobserver calculations for the purpose of assessing the reproducibility of the C2 measurements performed.

In an anatomical study of Kandiziora et al. [34], based on 50 axis vertebrae, the radiologic average of posterior body height was 18.2 mm, though this measurement was 24.6 mm in the current study using a different measuring technique. Similarly, the C2 superior body depth was 12.5 mm [34], but was 14.9 mm in our study using the Harris-Ring-C2 as the reference landmark. Monu et al. [47] also measured the odontoid tilt angle with a technique identical to ours in 175 patients with a mean age of 32.2 years. All patients had some degree of posterior tilting of the odontoid with a mean of 17.4° ± 6.1°, which showed a mean of 18.0° ± 5.7° in the current study. In another cadaver-based anatomical study, Xu et al. [70] measured the superior facet angle of C2 formed between the C2 midsagittal line and facet plane. The mean angular measurement was 66.3°/68.4° in males/females. Calculating our data according to their measurement technique gave similar results (65.9° left sides/68.6° right sides). In

the study of Kandiziora et al. [34], the angle between the horizontal plane and the superior facet of C2, which was 23.4° in the current study, was 23.9° on average.

Zapletal et al. [71] evaluated the incidence of AAOA radiologically in 355 patients and considered the C1–2 joints degenerative when severe narrowing or obliteration of the joint space, subchondral sclerosis, and/or osteophytes were present. The authors found the incidence of AAOA to be 4.8%. We modified the classification of Lakshamanan et al. [44], applied it to 100 patients, and the incidence of AAOA was found to be 5.0%. In summary, the reviewed data reported in literature were similar to those of the current study, offering additional validation for the measurements we performed. Therefore, the results found in the current measuring process should be reproducible in clinical applications.

In a radiological study, Sgabanti et al. [59] evaluated the mean height of the atlantoaxial joint space in 50 males and 52 females on both sides at the medial, lateral and median levels. No statistically significant differences were found in the mean values when the genders were compared, consistent with the results of the current study. Measurement of the mean width at the median level was 2.4 mm each in males and females [59]. The same averaged measurements in our study for the left and right sides were 3.75 and 3.25 mm, respectively. Sgabanti observed that the articular space width, at the three levels, had a linear relationship with the patients' age with progressive decrease in elderly patients. The current study confirmed these observations. However, age-related differences in joint space height are not included in the proposed classification because the acquired degenerative changes in C1–2 that appear during the clinical course following C2-fractures are supposed to be the decisive factors in clinical outcome rather than any absolute morphometrical value of joint space height.

Implications behind the assessment of posttreatment C2-alignment

Previously, outcome surveys of C2-fractures rarely included CT-assessment as endpoint anchors and held no information on the incidence and severity of malalignment and its impact on the clinical outcome [19, 20, 31, 39, 49, 58, 63]. In a study of Seybold et al. [58] the long-term outcome in patients treated with the halo-vest or cervical orthosis was not significantly worse than that after surgery, suggesting that other factors than the rate of union achieved might influence the outcome. Correspondingly, concurrent clinical results regarding similar treatment concepts for C2-fractures exist [1, 9, 42, 49, 58, 67]. There is no consensus on the appropriate classification and treatment of C2-fractures because many previous studies had heterogeneous samples, incomplete follow-up data, absence of validated

outcome vehicles, simplified description of fracture morphology and, thus, many different C2-fracture patterns, e.g., those including the C2-facets, a burst lateral mass or a comminuted odontoid base, were stored together in groups such as 'odontoid II or III fractures', 'vertebral body fractures' and 'hangman's fractures' [31, 38–40]. Frequently, apples and pines were compared. Therefore, to allow for a more detailed characterization of how a C2-fracture has healed we developed a physiological standard of the morphometric dimensions that sets cut-offs for pathological limits. So, this study offers a template to assess the C2 and C1–2 alignment after fractures of the axis vertebra. Although the measurement and classification process for a single, fused C2-fracture takes less time than that of a thoracolumbar scoliosis case using the Lenke classification, the clinical application of the current classification might seem strenuous. However, evaluation of the anatomical alignment following the treatment of C2-fractures will be one of the decisive factors when assessing clinical outcome and atlantoaxial rotation in comparative studies. Why? Because literature serves evidence that malaligned C2-anatomy can alter the complex C1–2 motional characteristics and damage to the articular surfaces of C2 can confer cervicocephalic pain and cause painful AAOA: The C1–2 joints and vertebrae are responsible for a complex kinematic arch of rotational motions [15, 50, 51] and contribute approximately 60% of the total neck rotation [50] that is rotation of 30°–43° to each side [15, 53, 56]. Accordingly, this was used as a strong argument in favour of motion-preserving surgical techniques, such as anterior odontoid screw fixation (AOSF) [2, 12, 19, 25, 28, 45, 54, 63]. In contrast, Jeanneret et al. [31], evaluating the postoperative C1–2 rotation following AOSF using functional CT-examination, observed a mean ROM to each side of about 25°, only 38% of patients having normal [16] C1–2 rotation. The authors emphasized that distinct fracture patterns such as an intraarticular fracture or arthritic changes at C1–2 contributed to decreased C1–2 rotation. Notably, the authors could document that intraarticular comminution of the C1–2 joints can produce painful posttraumatic malunion and AAOA. As concerns the decreased rotational ability of the C1–2 joints following non-surgical and surgical treatment of C2-fractures, several authors shared the observations of Jeanneret et al. [4, 49, 62, 67]. Müller et al. [49] reporting on 27 odontoid fractures undergoing AOSF, observed motion related pain in 29%. Forty-one percent of patients demanded pain medication. Radiographic follow-up indicated that there was no anatomic realignment in 18%. Seven patients had significant loss of ROM whereas one patient showed advanced AAOA. The authors supposed that 'as it is with long bone surgery' anatomical realignment should be a primary goal in upper cervical spine

surgery, but it was presumed difficult in axis fractures. Tuite et al. [66] reported that almost 50% of hangman's fractures (Hmfx) showed substantial sequelae following surgical stabilization by means of ACDF C2-3 if osseous union had occurred in a position of subluxation at C2-3. As early as 1993, Heller et al. [29] reported the surgical treatment of a malunited odontoid fracture with fixed atlantoaxial subluxation requiring odontoid resection and posterior C1–2 fusion. The authors observed that odontoid malunion had received little attention in previous series, either not having occurred in the past, or to have been unrecognized. Indeed, most malunions of the axis are neglected. In a current literature review [33], the incidence of malunion reported was low in type II and III odontoid fractures following halo treatment (5 and 10%, respectively). However, with the latter the articular facets of C2 are frequently affected and symptomatic malunion or segmental C1–2 deformation after nonsurgical treatment has been observed several times [5, 10, 13, 14, 20, 24, 26, 29, 33, 35, 36, 42, 55, 57]. Clark [10] already stretched that type III odontoid fractures were 'not benign as previously reported'. Ferrer emphasized that type III lesions involvement of the vertebral body or collapse of the lateral masses of C2 are prone to a high rate of non-anatomical osseous union [18], that is a malunion. Several reports confirmed that C2-fractures affecting the upper facets can show symptomatic atlantoaxial incongruity at follow-up [20, 32, 43] and there is evidence of AAOA following cervical trauma [17, 30, 31]. AAOA can be a distracting source of pain [17, 25, 30], and one of the sequelae following C1–2 fractures [20, 26, 31]. In a normal healthy population, AAOA was shown to be as high as 4.8% [71]. The incidence of AAOA was 5.0% in the current study in a group of healthy patients with a mean age of 49.8 years. However, the incidence of AAOA related to C2-fractures is not known. AAOA following C2-fractures remains a neglected entity [25] and demands further investigation.

A meaningful number of distinct subtypes of C2-fractures exists, in which outcome is not favourable [1, 8, 9, 29, 37, 62, 69]. However, differences in functional and clinical outcomes related to the posttreatment C2-alignment have not been studied. In this context, it is noteworthy that some of the painful C2-malunions indicate secondary anterior or posterior C1–2 fusion [20, 41, 62, 68]. Malunion in C2-fractures can produce significant sequelae and those subtypes of C2-fractures that are prone to malunion with nonsurgical therapy or motion-preserving surgery should be defined and a further stratification performed. With the current classification, the posttreatment C2-alignment can be assessed and related to the fracture morphology, the treatment applied, the C1–2 rotation and clinical outcome at follow-up. The main goal of the proposed classification system is to provide taxonomy, a tool for the assessment of

malalignment following C2-fractures, and to enable comparison of like cases in follow-up studies. Further ongoing work includes its application to clinical cases. Refining the system by assessing the impact of various treatments on the clinical and morphometric outcome will be required for complete validation of the classification as a guidance used for further stratification of distinct C2-fractures that are more or less prone to clinically symptomatic malunion.

The brief review of literature shows that various C2-fracture characteristics can affect outcome. With application of the proposed classification in clinical outcome studies, guidance on when to do a C1–2 fusion can be substantiated: If there is a predictive malunion in some distinct C2-fracture pattern following the use of current motion preserving techniques or conservative treatment, then primary fusion C1–2 might be considered a sound treatment.

The authors formed an anatomical template to assess the amount of anatomical distortion of the C2 vertebra and C1–2 alignment using reproducible parameters. Whether there will be distinct relationships between degrees of malunion, a decreased C1–2 rotation and worse clinical outcome has yet to be answered and will be discussed in part II of this study project.

Limitations

In the current study the authors used plain radiographs instead of CT-scans to investigate the C2 anatomy because they represent the most commonly used diagnostic tool for radiological follow-up in C2-fractures. In a study of Kandiziora et al. [34], only slight differences were observed between measurements performed using direct caliper technique, radiographs and CT-scans. To simplify the application of our radiographic results to further studies, including CT- and MRI-scans, we calculated anatomical ratios and side-related differences scaled in 'millimeters'. With the combination of angular measurements performed, the recommended parameters included in the classification should be reproducible with CT-scans. Nonetheless, research on the reliability of parameters measured on radiographs compared to CT-scans is to be performed.

Although interobserver calculations were performed, we did not test consistency over time and intraobserver reliability. However, at first inspection, interobserver reliability was shown to be excellent for most parameters. All but two of the parameters that were included for construction of the classification demonstrated excellent reliability, with ICCs > 0.75.

All patients included in the current study were of European origin. Because differences in patients from

Asian origin might be expected, the author cautions against applying our data indiscriminately in these patients.

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