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SHOULDER ARTHROPATHY SECONDARY TO SYRINGOMYELIA – NEW X-RAY CLASSIFICATION OF SHOULDER DEGENERATION

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ABSTRACT

Introduction

Shoulder neuroarthropathy is a rare joint degeneration mostly related to syringomyelia. X-ray is a basic tool to stage the advancement of shoulder destruction in shoulder neuroarthropathy.

Aim

We aimed to create and verify the reliability of our own radiographic classification of shoulder degeneration (NGH) and to correlate it with clinical features.

Material and Methods

The study included 39 cases (45 shoulders with neuroarthropathy secondary to syringomyelia) collected from a systematic literature review and our own pooled series of 10 cases. We found 34 papers, all being case reports or case series. Inclusion criteria were an X-ray in two projections and availability of clinical data. Our NGH classification was as follows: for glenoid (G) and head (H), three stages were distinguished: G0/H0, no X-ray changes; G1/H1, partial degeneration; and G2/H2, total degeneration.

Results

The statistical analysis showed almost perfect agreement between the evaluators for the humeral head and glenoid in both measurements and an almost perfect compatibility. The majority of patients had total head degeneration without correlation with range of motion. The condition of glenoid was various and also had no statistical influence on shoulder function. Both the G and H stages had an impact on the incidence of swelling and weakness.

Conclusions

These findings indicate that our NGH classification is highly reliable for staging shoulder degeneration related to syringomyelia. The classification seemed to partly correlate with the clinical condition. Under this system, patients with stages G0 and G1 can still qualify for reverse shoulder arthroplasty.

Keywords: syringomyelia, neuroarthropathy, shoulder degeneration, NGH classification

STRESZCZENIE

Wstęp

Neuroartropatia stawu ramiennego to rzadko występująca postać degeneracji stawu, najczęściej wywołana jamistością rdzenia kręgowego. Podstawowym narzędziem do oceny zaawansowania destrukcji stawu ramiennego w tym przypadku jest RTG.

Cel

Naszym celem było stworzenie oraz walidacja własnej klasyfikacji RTG oceniającej stopień destrukcji barku (NGH) oraz ocena korelacji z objawami klinicznymi.

Materiał i Metody
Badanie objęło 39 pacjentów (45 barków z neuroartropatią wtórną do jamistości rdzenia kręgowego) zebranych z przeglądu literatury i własnej grupy 10 przypadków. Wyodrębniliśmy 34 artykuły, wszystkie będące opisami przypadków lub seriami przypadków. Kryteriami włączenia były: zdjęcie rentgenowskie w dwóch projekcjach oraz dostępność danych klinicznych. Klasyfikacja NGH przedstawiała się następująco: dla panewki (G) i końca bliższego kości ramiennej (H) wyróżniono trzy stadia: G0/H0, brak zmian na zdjęciu rentgenowskim; G1/H1, częściowa degeneracja; oraz G2/H2, całkowita degeneracja.
Wyniki
Analiza statystyczna wykazała prawie idealną zgodność między oceniającymi zarówno dla końca bliższego kości ramiennej, jak i panewki w obu pomiarach (inter-rater) oraz prawie idealną zgodność (intra-rater). Większość pacjentów miała całkowite zwyrodnienie końca bliższego kości ramiennej bez korelacji z zakresem ruchu barku. Stan panewki był zróżnicowany i nie wpływał istotnie statystycznie na funkcję barku. Oba pomiary G i H miały wpływ na występowanie obrzęku i osłabienia barku.
Wnioski
Wyniki te wskazują, że nasza klasyfikacja NGH jest wysoce wiarygodna w określaniu stopnia zaawansowania zwyrodnienia barku związanego z jamistością rdzenia. Częściowo koreluje ona ze stanem klinicznym. Pacjenci w stadiach G0 i G1 mogą nadal kwalifikować się do odwróconej endoprotezoplastyki stawu ramiennego, jednak w stopniu zaawansowania H1 należy rozważyć endoprotezę anatomiczną.

Słowa kluczowe: syringomyelia, neuroartropatia, destrukcja stawu ramiennego, klasyfikacja NGH

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Introduction

Shoulder neuroarthropathy (Charcot joint) is a rare joint condition with a degeneration secondary to neurological diseases. (Schoch et al. 2016) Syringomyelia is the most common

cause of Charcot joint in the upper extremity. (Rickert et al. 2019; Atalar et al. 2010) Typical clinical symptoms include pain, swelling, range of motion limitation, weakness, and sensation disorders. (Wawrzyniak et al. 2021; Wang et al. 2018) Shoulder pain is mostly the first reason why patients seek medical attention. This usually leads to a later diagnosis of syringomyelia, while inevitably the disease progresses. (Snoddy et al. 2016) This complex neurological disease is the reason for the significant deterioration of the quality of life in patients.

Neurogenic arthropathy of the shoulder secondary to syringomyelia consists of the gradual degeneration and fragmentation of the proximal humerus and the glenoid with remodelling of bone fragments. (Schoch et al. 2016; Wang et al. 2018; Ruette et al. 2007) Conventional radiography (X-ray) is a basic tool to stage the advancement of shoulder destruction, and may have an impact on the clinical status of the disease. Radiographic staging might also help in deciding on treatment method. In cases of doubts about the degree of joint destruction and soft tissue condition or surgical planning, computed tomography (CT) and/or magnetic resonance (MR) of the shoulder might be necessary. (Ueblacker et al. 2007; Matsuhashi 2011)

Until now, the Eichenholtz classification has been the tool mostly used to stage the advancement of neuropathic joint degeneration. It attempts to reflect the pathophysiologic progression of joint disease, combining radiological signs with accompanying clinical symptoms. (Rosenbaum et al. 2010; Chantelau et al. 2014) However, to our knowledge, the Eichenholtz classification has never been validated neither for its correlation with clinical picture nor in terms of its reliability. (Rosenbaum et al. 2010) Another issue is that, as with every joint, the shoulder has its anatomic and functional specificity. Degeneration affects both the proximal humerus as well as the glenoid. Progression in both parts may not run at the same pace, making appropriate staging questionable. Recent advances in surgical treatment and, most of all, the development of reverse shoulder arthroplasty (RSA) have brought new possibilities but also new demands. In the case of degeneration with the preservation of the humerus tuberosities, an anatomical endoprosthesis can be considered. However, the loss of function with the tuberosities destruction is a basic indication of RSA. (Schoch et al. 2016; Wawrzyniak et al. 2021; Ueblacker et al. 2007) The main technical condition for inserting an implant is a well-preserved glenoid bone stock. Current classifications cannot support these predictions and planning. We found the need for a more precise, yet simple, classification of neuropathic shoulder arthropathy.

Aim

Therefore, we developed a new radiographic classification of neuropathic gleno-humeral degeneration (NGH Classification) that is specific for the neuroarthropathic shoulder and considers the degree of degeneration of the proximal humerus and glenoid separately. We subsequently validated this new classification system and correlated it with clinical features.

Material and methods

The radiographic evaluation material was based on X-rays of the cases collected from our own pooled series of 10 cases and a systematic literature review. Our material of this very rare condition was initially qualified from the patients of our institutions (seven patients). Data from three patients were obtained from inquiries among members of the Polish Shoulder and Elbow Society. Further along the project, we extended the material to X-rays obtained from a systematic review, which is part of a larger project. PubMed 1966-2021, Embase 1980-2021, the Cochrane Database of Systematic Reviews, and the Cochrane Central Register of Controlled Trial databases were searched for neuropathic shoulder cases with syringomyelia. Using the PICO method, we found 34 papers in the literature (only case reports and case series). (Linares-Espinós et al. 2018; Eriksen et al. 2018) Data for the

evaluation were included based on the diagnosis of shoulder neuroarthropathy secondary to syringomyelia, the availability of at least two X-ray views, and the availability of descriptive clinical data.

A systematic review revealed a description of 42 patients (47 shoulders with neuroarthropathy). Based on the inclusion criteria, 29 patients (34 shoulders) qualified for the evaluation. Altogether (including our own group), 39 patients (45 shoulders) were evaluated.

Classification of neuropathic gleno-humeral degeneration (NGH)

Our basic assumption was to create a simple classification to address the staging of both the proximal humerus and glenoid. (Fig.1) Therefore, the staging in the NGH classification describes the severity of degenerative changes in two shoulder components based on the X-ray examination in two radiographic views: AP and axial. The first component (G) describes the degree of degeneration within the glenoid of the shoulder joint, whereas the second component (H) describes the degree of severity of degeneration within the proximal humerus (including humeral head and tuberosities preservation). (Table 1) For both components (G and H), three stages were distinguished. (Figs.2 and 3)

- No X-ray changes are defined as a normal bone structure of the proximal humerus and/or the glenoid shoulder joint
- Partial degeneration is determined when the bone structure of the proximal humerus and/or the glenoid is partially damaged and partly loses the shape of selected joint elements (<50% circumference), in H1 significant part of humeral head has been lost however tuberosities are still preserved
- Complete or near-total degeneration of the proximal humerus and/or the glenoid indicates complete or nearly complete (> 50%) destruction of the proximal humerus and/or the glenoid. At this stage, either the glenoid or the proximal humerus has lost its shape, and the elements of that shape cannot be identified, in H2 stage tuberosities are not preserved

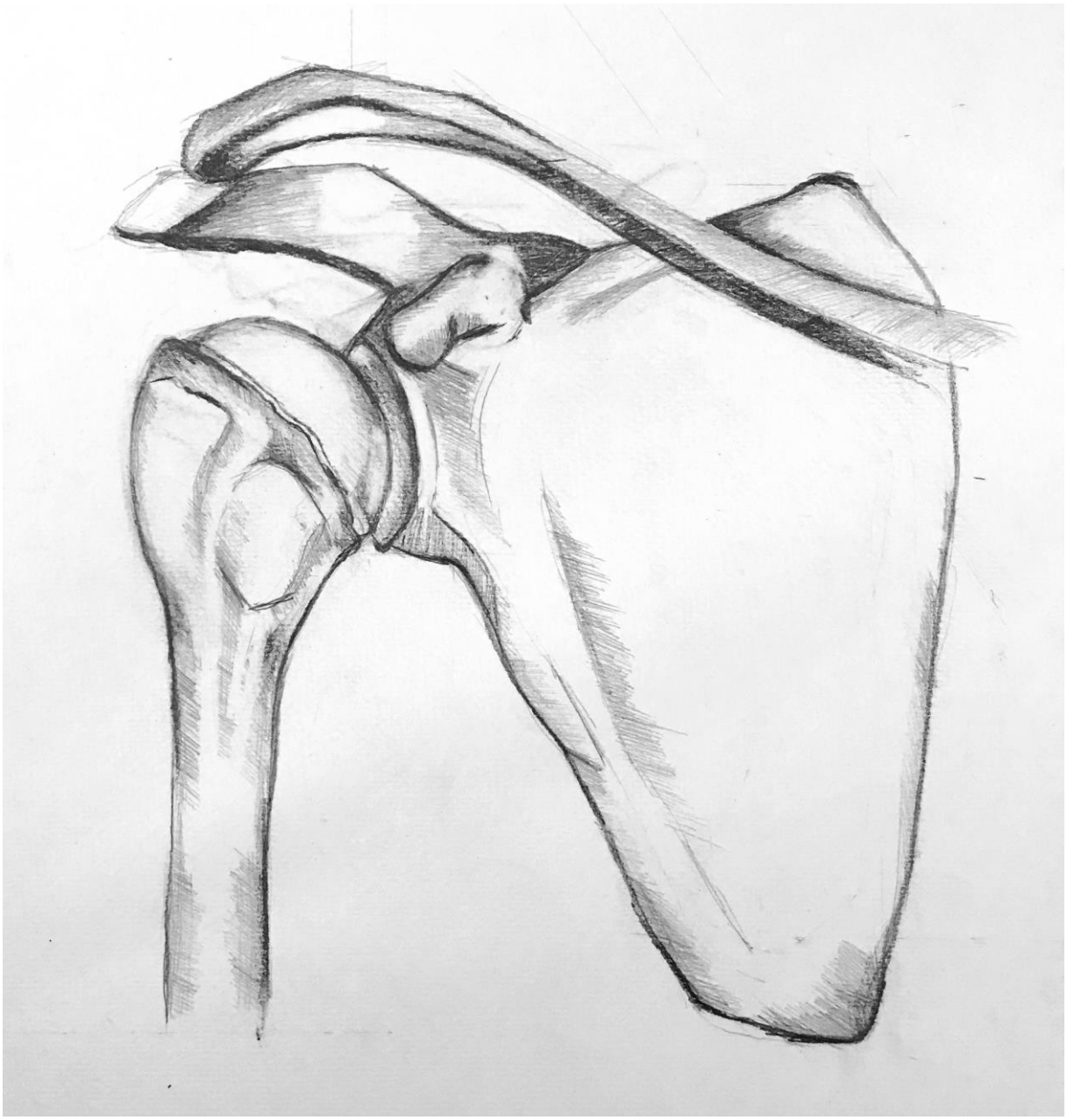

Figure 1. Anatomical conditions of the shoulder

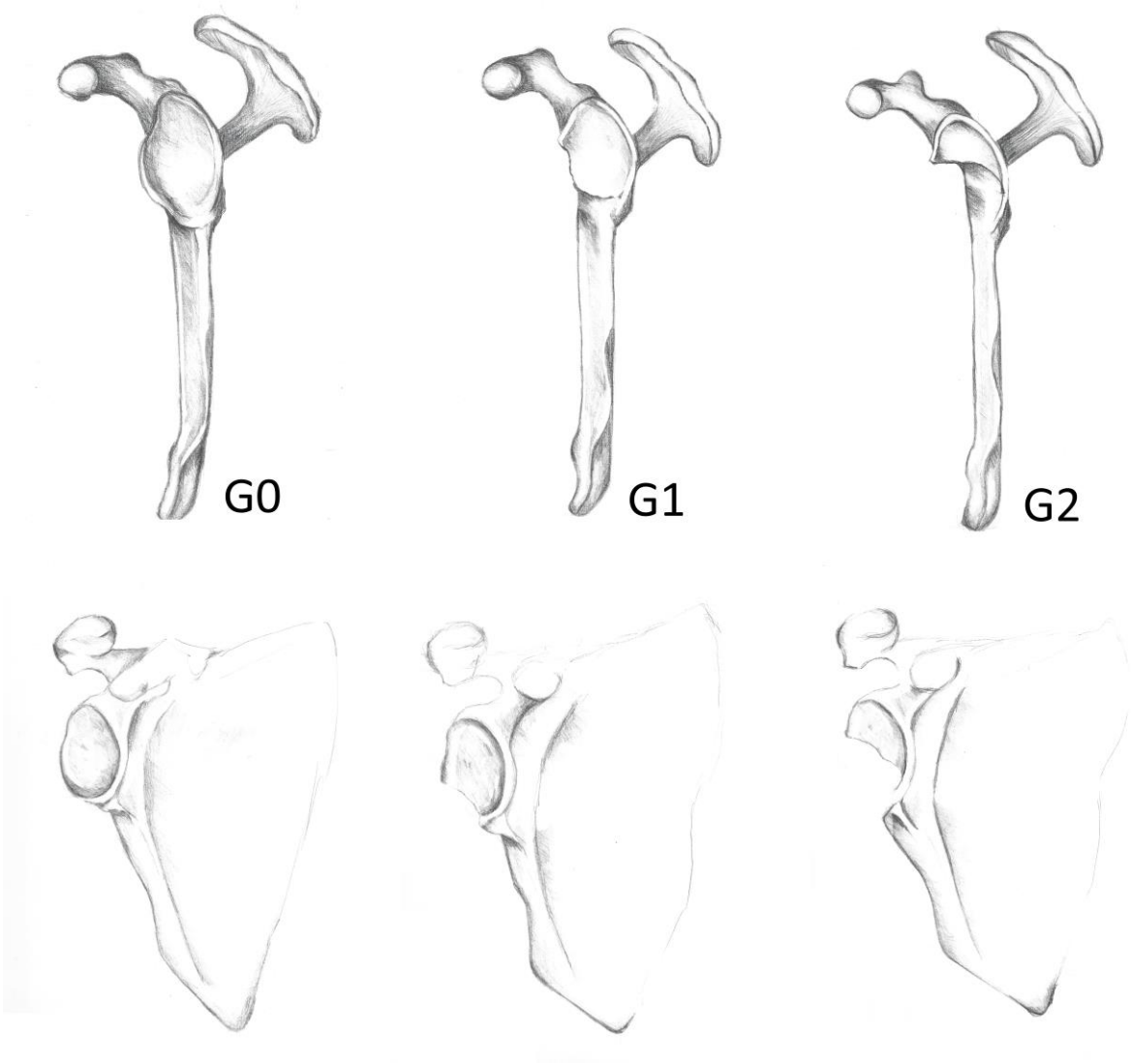

Figure 2. NGH classification – G stages

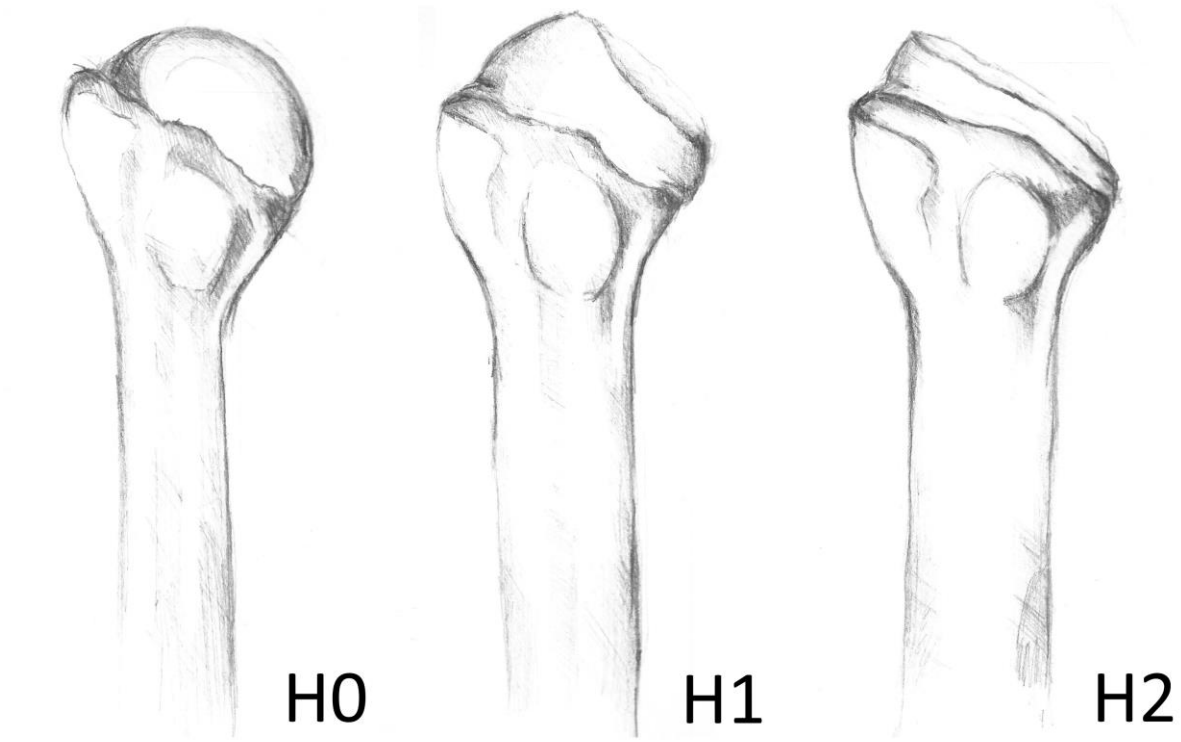

Figure 3. NGH classification – H stages

Table 1. NGH classification

G (glenoid)	**H (humeral head)**
0 – no X-ray changes of glenoid	0 – no X-ray changes of head
1 – partial glenoid degeneration	1 – partial head degeneration
2 – total glenoid degeneration	2 – total head degeneration

Reliability assessment

Both intra- and inter-reliability of the NGH classification were performed. Each of the available shoulder X-ray images of patients with arthropathy was assessed independently by two researchers. The intra-observer agreement assessment was made after 7 days. The degree of inter-rater reliability and intra-rater reliability of each evaluator were assessed on the basis of weighted Cohen's κ coefficient using SPSS statistics. The degree of compliance was assessed as follows: ≤ 0, no agreement; 0.01–0.20, no or slight agreement; 0.21–0.40, fair agreement; 0.41–0.60, moderate agreement; 0.61–0.80, substantial agreement; and 0.81–1,00, almost perfect agreement. (McHugh 2012)

Correlation between the NGH classifications and their clinical presentation

Fisher's exact test analysis was performed to compare the groups of patients with different degrees of H and G in terms of the occurrence of pain, swelling, weakness, and range of

motion (ROM) limitation. We compared patients with different degrees of H and G in terms of flexion, internal rotation, and external rotation per performing the Mann–Whitney U test for H and the Kruskal–Wallis for G. The significance level was $\alpha = 0.05$. Statistical analyses were performed using IBM SPSS Statistics 25.0.

Results

Reliability assessment

The statistical analysis showed almost perfect agreement between the evaluators for both the proximal humerus and glenoid in both measurements. For the compatibility of the scores within each evaluator, almost perfect agreement was observed in the grades of the proximal humerus and the glenoid for both judges. The results of the analyses are presented in Table 2.

Most of the patients (75%) presented with complete or nearly complete proximal humerus degeneration (H2), while the degree of glenoid preservation varied. (Table 3)

Table 2. Inter-rater and intra-rater reliability of NGH classification

	Inter-rater reliability	*Z*	*p*		Intra-rater reliability	*Z*	*p*
H1	0,928	6,24	<0,001	rater I for H	0,933	6,28	<0,001
H2	0,862	5,84	<0,001	rater II for H	1,000	6,71	<0,001
G1	0,924	7,37	<0,001	rater I for G	0,963	7,78	<0,001
G2	0,925	7,48	<0,001	rater II for G	0,961	7,68	<0,001

H – humeral head; G - glenoid; 1 – evaluation 1; 2 – evaluation 2

Table 3. Number of patients in each classification group

	H0	**H1**	**H2**
G0	0	3	1
G1	0	3	14
G2	0	5	18

Correlation

ROM limitations occurred in all patients regardless of grade, either G or H. Weakness was significantly more frequent in patients with the highest H grade (H2) than in patients with a lower grade (H1), with occurrence of 88% vs. 54% respectively ($p = 0.028$). Similarly for G grade, weakness occurred in all examined patients with the highest grade G (G2) and was significantly more frequent than in patients with G1 and G0 (occurrence of 100% vs. 65% and 25%, respectively; $p < 0.001$). Further, swelling was significantly more frequent in patients with G1 compared to patients with G0 (100% vs. 50%, $p = 0.021$). This was also observed in 82% of the G2 cases but did not reach statistical significance. Detailed results of the analyses are presented in Table 4 for grade H and Table 5 for grade G.

Table 4. Fisher's exact test analysis for the occurrence of pain, swelling and weakness depending on the degree of H.

		H				
		H1		H2		
		n	*%*	*n*	*%*	*p*
Pain	No	5	45,5	10	29,4	0,464
	Yes	6	54,5	24	70,6	
Swelling	No	2	18,2	4	11,8	0,624
	Yes	9	81,8	30	88,2	
Weakness	No	5_a	45,5	4_b	11,8	0,028
	Yes	6_a	54,5	30_b	88,2	

a, b - means that the proportions in the columns are different with $p < 0,05$ (Bonferroni's corrections)

Table 5. Fisher's exact test analysis for the occurrence of pain, swelling and weakness depending on the degree of G

		G						
		G0		G1		G2		
		n	*%*	*n*	*%*	*n*	*%*	*p*
Pain	No	1	25,0	6	35,3	8	34,8	1,000
	Yes	3	75,0	11	64,7	15	65,2	
Swelling	No	2_a	50,0	0_b	0	$4_{a,b}$	17,4	0,021
	Yes	2_a	50,0	17_b	100,0	$19_{a,b}$	82,6	
Weakness	No	3_a	75,0	6_a	35,3	0_b	0	<0,001
	Yes	1_a	25,0	11_a	64,7	23_b	100,0	

a, b - means that the proportions in the columns are different with $p < 0,05$ (Bonferroni's correction)

The Mann–Whitney U test was used to compare patients with H1 and H2. The analysis showed no significant differences in flexion, internal rotation, and external rotation among patients with grades 1 and 2. Differences between G grades of 0, 1, and 2 in terms of the analysed variables were also insignificant. However, clear trends were visible, as shown in the charts in Figs. 6 and 7.

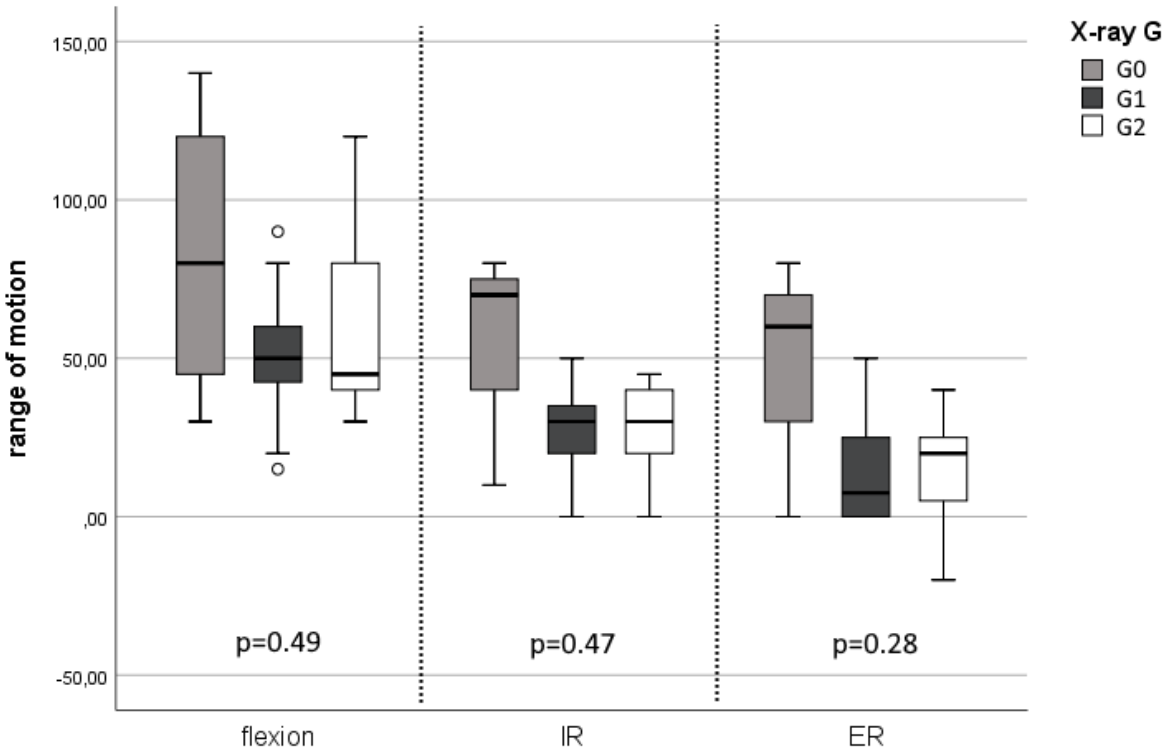

Figure 6. Correlation of H stages with range of motion of the shoulder

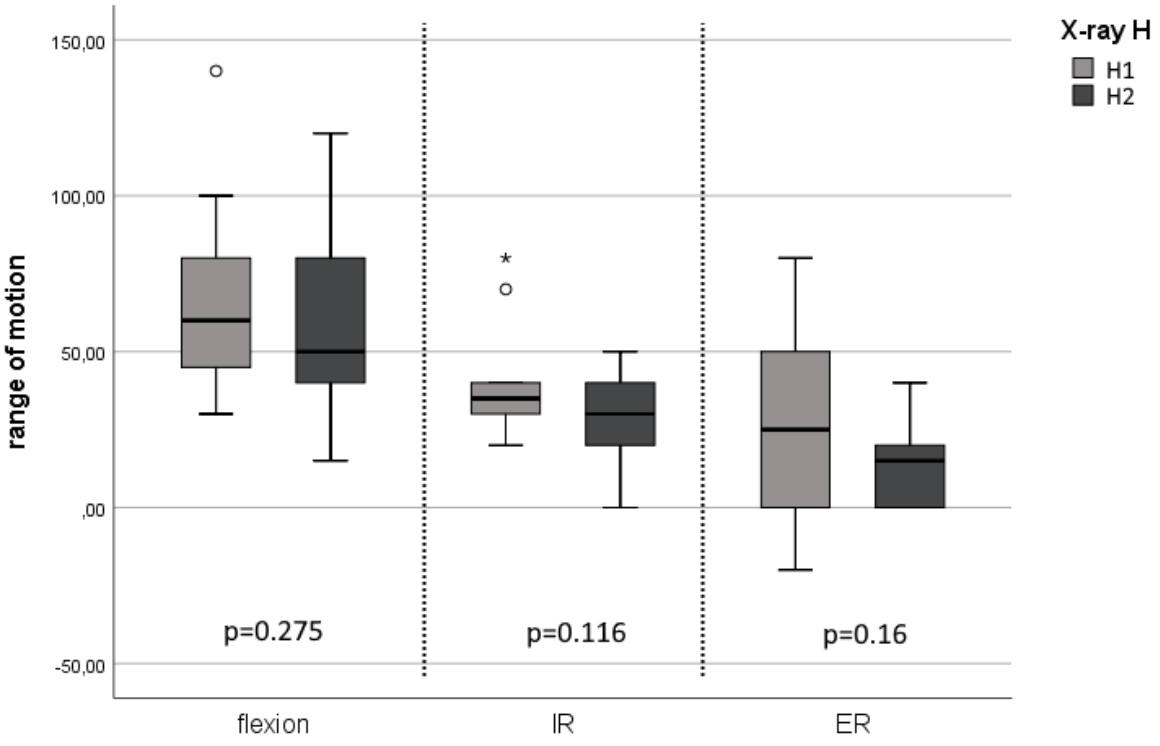

Figure 7. Correlation of G stages with range of motion of the shoulder

Discussion and conclusion

In this study, we designed the NGH classification that details the degree of proximal humerus and shoulder glenoid degeneration in shoulder neuroarthropathy based on X-ray views. Subsequently, we validated the classification, obtaining an almost perfect inter-rater

reliability level. We also proved that, to some extent, the NGH classification correlates with the clinical condition.

Typically, the advancement of changes in shoulder neuroarthropathy is evaluated on plain radiography performed in at least two perpendicular views (AP and axial). Gradual loss of contour, fragmentation of the glenoid and/or the proximal humerus, and, eventually, remodelling are the most common findings. (Wawrzyniak et al. 2021; Ruette et al. 2007)
The most widely and generally used classification of neurogenic arthropathy severity is the Eichenholtz classification. This scale contains both the severity of radiological changes and the clinical picture, and it is dedicated to all neuroarthropathic joints (related not only to syringomyelia). (Rosenbaum et al. 2010; Chantelau et al. 2014) The classification is based on the natural evolution of the Charcot joint and has been previously well accepted by researchers. The Eichenholtz classification is very universal; however, as applied in the assessment of the shoulder, it does not consider its anatomical and functional specificity, and therefore, it does not respond to our needs. Additionally, it has yet to be validated. (Rosenbaum et al. 2010; Rosskopf et al. 2019)

Currently, there is no available classification specific for arthropathic shoulder anatomic condition that is similar to the Sanders-Frykberg or Brodsky-Trepman classification for Charcot foot. Both classifications are very precise. They accurately assess the anatomical advancement of foot degeneration, and they are useful in therapeutic decisions. (Rosenbaum et al. 2010; Sekimpi et al. 2019) However, until recently, the validation of these classifications has not been assessed. Despite the lack of validation, both these classifications have been commonly used to define the severity of Charcot foot and to plan surgical treatment. Recently Wukich et al. showed that both classifications have high inter-rater and intra-rater ratios. (Wukich et al. 2021)

Currently, several classifications are used to assess the advancement of degenerative or necrotic changes in the shoulder joint. However, each of them relates to different clinical conditions. For example, the Walch classification (glenoid morphology in shoulder arthrosis) the Samilson and Prieto classification (validated classification for dislocation shoulder arthropathy), the Favard classification (arthrosis with massive rotator cuff tear), or the Hattrup and Coffield classification (humeral head degeneration in avasculalar necrosis). (Jawa et al. 2021; Ilg et al. 2001; Kappe et al. 2011) According to the literature, these classifications are very useful when choosing a treatment method. (Jawa et al. 2021; Hattrup et al. 1999) It is worth noting that Kohan et al. proved that radiographic severity of shoulder arthrosis does not correlate with patient-reported pain and function. (Kohan et al 2020) Nonetheless, we believe that determining the stage of shoulder arthropathy advancement in X-ray is a part of the full patient assessment and it is useful primarily for choosing the type of arthroplasty. Still, the decision on conservative or operative treatment should be made based on both the radiological stage in the NGH classification and the clinical assessment.

The classifications used for the severity of degenerative changes in the shoulder are the most applicable to our needs. We believe that it is important to appreciate changes in both parts of the joint, that is, the proximal humerus (both humeral head and tuberosities) and glenoid. First, the degeneration of each part may contribute differently to the pathophysiology of the shoulder, affecting the severity of symptoms and the loss of range of motion. Secondly, bone changes may imply choice of treatment. Preserved bones and relatively good ROM may be indications for a more conservative approach (G0H0). Lost and painful shoulder function with tuberosities preserved (H1) could be the indication for anatomical arthroplasty (partial or total – depending on the glenoid condition). However in the case of total proximal humerus degeneration (H2), with good glenoid bone stock could be the indication required for RSA (G0-12) for the symptomatic shoulder. (Wawrzyniak et al. 2021) Lastly, stage G2 may currently indicate no option of surgical approach and also limited perspective of restoring

function of the shoulder with rehabilitation. Moreover, plain radiographic evaluation is optimal for monitoring the progression of deformity to match the currently available treatment options for patients with neuropathic joints. Such an evaluation is easily accessible, inexpensive, and safe for the patient.

The purpose of using classifications in orthopaedics is to systematise radiological pictures, monitor the progression of disease, and decide on treatment method. Precise classifications could also enable the comparison of results between different departments. (Garbuzet al. 2002; Brox et al. 2003) However, a good and useful classification should meet certain conditions. The analysis of commonly used X-ray classifications shows that their most important features are reliability, ease of use (simple qualification for a particular grade), and clinical relevance.

In the current study, we present what we believe is an easy system of classification that helps us make proper therapeutic decisions (conservative, surgical, and palliative treatment) and monitor the progression of shoulder degeneration. We also showed that the classification is highly reliable. However, we still need to determine whether radiographic staging with our NGH classification corresponds to the clinical status of the patient (pain, function, disability).

In our opinion, a new classification with the separation of the glenoid and the proximal humerus condition is valuable, especially in terms of planning surgical treatment. Although there are reports on the high effectiveness of shoulder arthroplasty, different types according to proximal humerus and glenoid degeneration.

Correlation

The results presenting increasing weakness with progression of proximal humerus and glenoid destruction could be explained by rotator cuff destruction and a reduction of shoulder proprioception. (Wałecka et al. 2020) We observed that proximal humerus destruction also affected tuberosities, which are usually associated with a lack of function of the rotator cuff. (Matsuhashi et al. 2011) This process is also associated with a reduction in shoulder stability, which additionally results in a reduction in the joint proprioception. This is very similar to the avascular necrosis of the humeral head, where the gradual destruction of the humeral head is also associated with a reduction in the active range of motion. (Franceschi et al. 2017)

The severity of shoulder swelling can be explained by the increasing degeneration of the joint, its instability, and increased inflammation. However, our clinical observations confirmed that shoulder swelling in syringomyelia often wavers and depends on various factors.

Limitations

Due to the rarity of the condition, it is difficult to draw strong conclusions. The NGH classification is neither very detailed nor extremely precise, yet the reliability of the assessment is excellent. Regarding its simplicity, we are not entirely sure whether the radiological stages correlate with all aspects of the clinical condition. The numbers are still too low to perform an advanced statistical analysis due to the rarity of the condition and the sparsity of reports. Nevertheless, to some extent, we showed some link of degeneration with malfunction. Definitely, the staging of the proximal humerus and glenoid may be a strong indicator of the chance of an endoprosthesis reconstruction and the restoration of shoulder function. Whereas advanced collapse would encourage conservative approach. Any progression of degeneration is easily monitored with simple X-rays. This could also be the case for longer observation, which may take a long time due to the rarity.

We believe that a larger group of patients is needed to obtain statistically significant results. However, on the basis of the above results, we can attempt to predict, at least partially, the clinical course based on the radiological picture.

In conclusion, the NGH classification distinguishing proximal humerus and glenoid degeneration could be a useful and reliable tool for neuroarthropathic shoulder assessment. Radiological picture of shoulder in syringomyelia correlates with clinical symptoms like swelling and weakness, but does not correlate significantly with shoulder range of motion. Patients with preserved tuberosities (H1) can be qualified to anatomical arthroplasty and patients with stages H2G0 and H2G1 can still be qualified to reverse shoulder arthroplasty. Although shoulder syringomyelia is underreported, it should be a matter of continuing discussion.

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