The Double Crush Syndrome: A Common Occurrence in Cyclists With Ulnar Nerve Neuropathy—A Case-Control Study

Tanya M. Smith, ScD, PT, Steven F. Sawyer, PhD, PT, Phillip S. Sizer, PhD, PT, and Jean-Michel Brismée, ScD, PT

Objective: To evaluate the incidence of double crush syndrome in the upper limbs of cyclists with clinical diagnosis of ulnar nerve neuropathy.

Design: Case-control study.

Setting: Outpatient clinics and university setting.

Participants: Consecutive sampling of 70 cyclists (140 upper limbs) with a mean age of 36 years (±11.3). Seventy-two upper limbs were excluded, leaving 40 upper limbs with a clinical diagnosis of ulnar nerve neuropathy [ULNN (+)] and 28 without symptoms of ulnar nerve neuropathy [ULNN (−)].

Assessment: Cyclists were examined clinically for the presence of proximal dysfunction using the following testing (independent variables): (1) thoracic outlet syndrome provocation testing: elevated arm stress test and modified Cyriax release test; (2) presence of an elevated first rib: cervical rotation lateral flexion test; and (3) presence of proximal symptoms: reports of neck pain and shoulder pain.

Main Outcome Measurements: The upper limbs of cyclists were categorized into 2 groups (dependent variable)—ULNN (+) and ULNN (−)—based on history, symptoms, motor, sensory, and provocative clinical testing.

Results: A significantly greater number of upper limbs of cyclists with ULNN (+) presented with positive provocative testing for thoracic outlet syndrome (elevated arm stress test $P = 0.005$; modified Cyriax release test $P = 0.002$) than did the upper limbs of cyclists with ULNN (−). The likelihood for the presence of neck pain, shoulder pain, and an elevated first rib was 3, 5, and 12 times greater, respectively, in the ULNN (+) than the ULNN (−) group.

Conclusion: A statistically significant greater number of the upper limbs of cyclists with clinical diagnosis of ulnar nerve neuropathy presented with proximal dysfunctions suggestive of double crush syndrome.

Key Words: ulnar nerve neuropathy, thoracic outlet syndrome, double crush syndrome, bicycling

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INTRODUCTION

Ulnar nerve neuropathy, a term used to describe symptoms related to compression and/or tension loading of the ulnar nerve at numerous locations along the length of its course, has a greater incidence in cyclists than in noncyclists and is referred to as "cyclist's palsy."[1]

Several mechanical causes of ulnar nerve neuropathy have been identified. The cubital tunnel is the most common site of ulnar nerve neuropathy at the elbow.[2,3] The ulnar nerve is also vulnerable to compression at the wrist resulting in ulnar tunnel syndrome, also known as Guyon's canal syndrome.[4] The recognition of ulnar nerve compression of cyclists in Guyon's canal has been commonly described in the literature for decades.[1,5–11]

A noteworthy predisposing factor for ulnar nerve neuropathy is mechanical nerve perturbation in the thoracic outlet, including thoracic outlet syndrome, which is most often caused by compression or tension loading of the brachial plexus as it passes through the thoracic outlet.[12,13] This mechanical irritation produces a double crush phenomenon, in which the ulnar nerve is susceptible to altered axonal transport at the cubital tunnel and/or Guyon's canal when the compression and/or tension of the C8-T1 nerve roots compromise the nerve's ability to withstand additional pressure.[14] Symptoms of thoracic outlet syndrome may not match the established patterns of typical peripheral entrapment neuropathies.[15] For example, neurologic thoracic outlet syndrome may lack definitive findings in electrodiagnostic studies and has been referred to as "disputed neurogenic" thoracic outlet syndrome.[16] As a consequence, many patients with thoracic outlet syndrome experience minimal sensory deficits and lack demonstrable muscle weakness.[16] Thus, examining the thoracic outlet remains challenging because of the lack of electrodiagnostic findings and specificity in provocative diagnostic examinations.

Although numerous studies demonstrate that cyclists have a high incidence of ulnar nerve neuropathy,[1,5–11] no study to date has evaluated the incidence of double crush phenomenon in cyclists with a clinical diagnosis of ulnar nerve

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From the Center of Rehabilitation Research, Texas Tech University Health Sciences Center, Lubbock, Texas.
Reprints: Jean-Michel Brismée, ScD, PT, Texas Tech University Health Sciences Center, Department of Rehabilitation Sciences, Center of Rehabilitation Research, 3601 4th Street, Mail Stop 6223, Lubbock, Texas 79430 (e-mail: jm.brismee@ttuhsc.edu).

neuropathy. The purpose of this study was to compare the incidence of symptoms and clinical testing suggestive of thoracic outlet perturbations and double crush in the upper limbs of cyclists with a clinical diagnosis of ulnar nerve neuropathy to those upper limbs of cyclists without symptoms of ulnar nerve neuropathy.

METHODOLOGY

Subjects

After obtaining approval from the local university Institutional Review Board, cyclists were recruited in the states of Arizona, Colorado, and Wyoming for consecutive sampling from advertisements in bike magazines, recruitment flyers, and mass electronic mailing to bike clubs and teams. Prior to testing, all subjects viewed an instructional video about the testing protocol and informed consent was obtained from all participants. A questionnaire including information pertaining to amount and type of cycling; medical history related to the exclusion criteria (see later); history of neck, shoulder, and/or arm pain; and personal demographic information was completed. Inclusion criteria were as follows: (1) subjects between age 18 and 65 years; and (2) riding the bicycle a minimum of 45 minutes 3 times a week (road and/or mountain bike). Exclusion criteria included: (1) being pregnant; (2) riding a recumbent bicycle; (3) having a history of thoracic outlet syndrome–related surgery or first rib resection; and (4) having a history of surgery for the distal upper limb (carpal tunnel release, cubital tunnel, and/or Guyon's canal release).

Classification of Subjects

All the upper limbs of subjects were categorized into either a symptomatic ulnar nerve neuropathy group [ULNN (+)] or an asymptomatic group [ULNN (−)] at the completion of data collection. To be included in the ULNN (+) group, the presence of the following criteria was required: subjective complaints of paresthesia and/or pain in the 4th and 5th digits of the hand following the ulnar nerve distribution in the previous 6 months[17] and at least 1 of the following: (1) diminished sensation in the ulnar nerve territory[18] using the Semmes-Weinstein monofilaments on the 5th digit, with values greater or equal to 3.61 mg/force[19,20]; (2) diminished strength of the intrinsic muscles of the abductor digiti minimi measured as being less than 2 standard deviations (SD) below normative data of subjects hand dominance and gender matched, were measured using the Rotterdam Intrinsic Hand Myometer[18,21,22]; and (3) symptom reproduction in the ulnar nerve distribution using the elbow flexion pressure provocation test at 60 seconds.[3]

To be included in the ULNN (−) group, the subject was required to report an absence of a history of paresthesia and/or pain in the ulnar nerve distribution.

The upper limbs of the subjects not meeting either criterion for ULNN (+) or ULNN (−) were excluded from data analysis. For example, if subjects reported subjective complaints of paresthesia without 1 of the 3 objective ulnar nerve tests being positive, that limb was excluded from data analysis.

Finally, to ensure independence of the data, if cyclists met the criteria for ULNN (+) in both upper limbs, only 1 limb was randomly included, whereas the contralateral limb was excluded from the data analysis. Similarly, upper limbs of cyclists were considered ULNN (−) when both upper limbs met the criteria for ULNN (−). Also in this case, only 1 of both limbs of each cyclist was randomly included and the contralateral limb was excluded from the data analysis.

CLINICAL TESTING

Simple random assignment was performed with the flip of a coin for clinical testing order of upper limbs (right or left limb tested first) for the tests described in the following section.

Clinical Testing for Ulnar Nerve Neuropathy

1. Sensory Testing: Semmes-Weinstein Monofilament Test (North Coast Medical, Morgan Hill, CA)

Subjects unable to detect filaments in the ulnar nerve distribution greater or equal to 3.61 were considered to have impaired somatosensation.[19,23] These values have yielded 79% sensitivity and 91% specificity for the diagnosis of carpal tunnel syndrome when testing was performed in the sensory distribution of the median nerve.[19]

2. Strength Testing of Fifth-Digit Abduction

Each subject was asked to resist an adduction force exerted on the 5th finger using the Rotterdam Intrinsic Hand Myometer (AH & RH Den Ouden, Rotterdam, the Netherlands) (Fig. 1).[22] Three consecutive trials were performed with 1-minute rest intervals in between trials. The same procedure was repeated on the contralateral hand. The mean peak force of 3 trials was calculated for each subject. The symptomatic subjects with strength measures that were less than 2 standard deviations from the mean of a similar asymptomatic gender and hand-dominant–side matched population were considered positive for strength loss.[21]

FIGURE 1. Rotterdam Intrinsic Hand Myometer used for the 5th-digit abduction strength test.

3. Elbow Flexion with Pressure Provocation Testing

Testing was performed with the elbow passively flexed and fully supinated, while the investigator applied pressure on the ulnar nerve, just proximal to the cubital tunnel. The test was considered positive if there was a reproduction of the subject's pain, numbness, and/or tingling in the arm or hand at the end of 60 seconds.[3] The elbow flexion with pressure provocation test has a sensitivity of 98% and specificity of 95% at 60 seconds of testing in the diagnosis of ulnar nerve neuropathy in the cubital tunnel.[3]

Clinical Provocation Testing for Thoracic Outlet Perturbation

1. Elevated Arm Stress Test

The subject was seated in a chair with feet on the floor and the head and neck in a neutral position, the scapulae in retraction, the shoulders in 90 degrees abduction elevation and 90 degrees external rotation, and the elbows flexed to 90 degrees. In this position the subject slowly and rhythmically opened and closed his or her hands for a 90-second trial. An elevated arm stress test was considered positive with production of pain, numbness, and/or paresthesia in the upper limb and/or the cessation of testing secondary to pain prior to 90 seconds (Fig. 2).[24] Plewa and Delinger[24] reported a rate of false-positive findings of 21% (specificity = 79%) at 90 seconds of testing when pain was produced and 36% (specificity = 64%) when paresthesia was produced during elevated arm stress testing. The authors did not report sensitivity values of the testing because their sample included asymptomatic subjects only. Howard et al[25] reported sensitivity for the elevated arm stress test of 82% and specificity of 100% to classify subjects into severe or not severe thoracic outlet syndrome.

FIGURE 2. Elevated arm stress test, front view, used for the provocative testing of thoracic outlet syndrome.

2. Modified Cyriax Release Test

While seated, the subjects' shoulder girdles were supported in submaximal elevation. The hands, wrists, and forearms were placed in neutral positions.[12] Testing position was held for 3 minutes (Fig. 3). The investigator asked the subjects to report pain, numbness/tingling, burning, and fatigue at 1, 2, and 3 minutes. A modified Cyriax release test was considered positive for thoracic outlet perturbation when the subject reported symptoms of pain, numbness, and/or paresthesia ("tingling") in the upper limbs.[12] Specificity values of 88% at 3 minutes and 97% at 1 minute (rate of false-positive results of 12% and 3%, respectively) of testing have been previously reported.[11]

Clinical Testing for the Presence of an Elevated First Rib

The cervical rotation lateral flexion test procedure included rotating the neck, passively and maximally, away from the side to be tested.[26] Once this maximally rotated position was assumed, the neck was flexed forward to end range, moving the ear toward the ventral chest (Fig. 4A and B). The test result was considered positive if the forward flexion part of the movement was notably decreased on either side with a hard end feel. The intertester reliability of this testing has been found to be excellent with a kappa statistic of 1.0.[26]

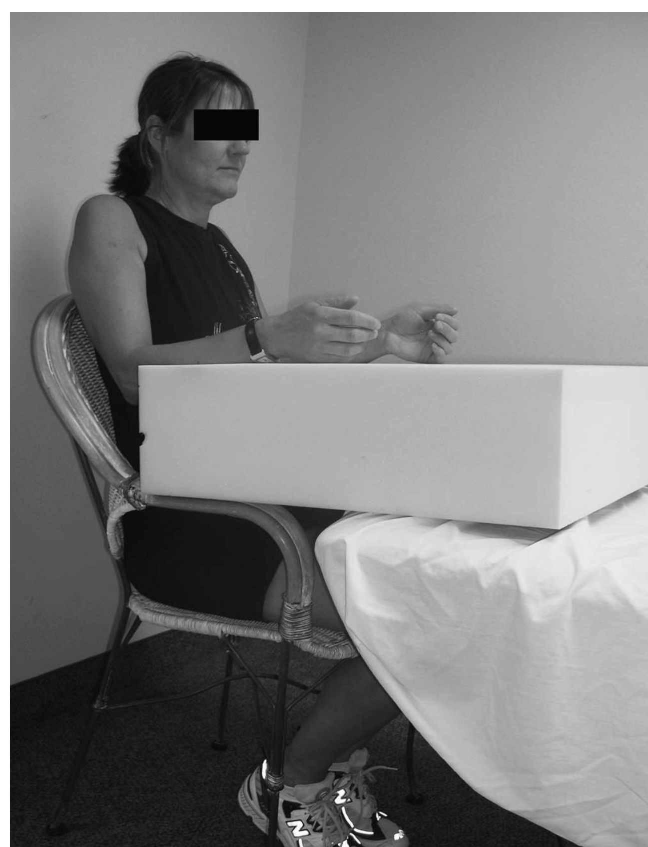

FIGURE 3. Modified Cyriax release test, side view, showing shoulder girdle passive elevation held for 3 minutes for the provocative testing of thoracic outlet syndrome.

FIGURE 4. Cervical rotation lateral flexion test, front view, showing end position of testing for an elevated position of the left first rib. A, Test shown is positive for an elevated left first rib; B, Test shown is negative for an elevated left first rib.

The sensitivity of the test for the presence of an elevated first rib has been reported as 93% with a specificity of 94%.[26]

Testing Sequence

The following procedures were completed in the same order for each subject tested: (1) sensory testing using the Semmes-Weinstein Monofilaments; (2) strength testing using the Rotterdam Intrinsic Hand Myometer; (3) elbow flexion pressure provocation test; (4) modified Cyriax release test; (5) cervical rotation lateral flexion test; and (6) elevated arm stress test. One investigator with 12 years of orthopaedic clinical practice experience performed all of the tests for each subject. The investigator performed a pilot study for reliability evaluation of all clinical tests prior to commencement of this study. To avoid bias, the investigator was not aware of the group [ULNN (+)] or [ULNN (−)] to which each subject belonged until data collection was completed.

Statistical Analyses

Reliability Pilot Testing

Intrarater reliability testing was performed using Statistical Program for the Social Sciences (SPSS) version 12.0 to calculate Intraclass Correlation Coefficient (ICC model 3,1) using a 2-way mixed model for sensory testing and strength testing of the abductor minimi. Kappa scores were calculated for dichotomous data of the elbow flexion pressure provocation testing, the modified Cyriax release test, the cervical rotation lateral flexion test, and the elevated arm stress test.

Ulnar Nerve Neuropathy and Double Crush Testing

Descriptive statistics were used to analyze the subjects' characteristics. Frequency counts were calculated for the elevated arm stress test, modified Cyriax release test, and cervical rotation lateral flexion test for subjects with a history and/or current neck and shoulder pain and symptoms reported by the subjects (paresthesia, numbness, and other symptoms) ipsilateral to the tested upper limb. The Fisher exact test was used in lieu of the more traditional Chi-square test because of the small counts in some contingency table cells. Statistical analyses were conducted using SPSS 12.0 and GraphPad InStat version 3.06 for Windows with an experimentwise alpha level of 0.05 to evaluate if there were significant differences in the frequency of symptoms (neck pain, shoulder pain) and clinical testing suggestive of perturbations in the thoracic outlet (elevated arm stress test, modified Cyriax release test, and cervical rotation lateral flexion test) in the upper limbs of cyclists with ULNN (+) as compared with the upper limbs of cyclists with ULNN (−). Alpha adjustments were made using the multiplicative correction for 5 pairwise comparisons to maintain a familywise alpha of 0.01 (Bonferroni correction).

RESULTS

Reliability Pilot Testing

Forty upper limbs of 20 subjects including 8 men and 12 women with a mean age of 44 years (±14) were tested by 1 investigator to determine intrarater reliability. Reliability scores are reported in Table 1.

Ulnar Nerve Neuropathy and Double Crush Testing

Following pilot testing, 140 upper limbs of 70 cyclists including 47 men and 23 women with a mean age of 36 years (±11.3) were evaluated for symptoms of ulnar nerve neuropathy and thoracic outlet syndrome. Nineteen of the 140 upper limbs were excluded because they did not meet

TABLE 1. Reliability Scores of Clinical Pilot Testing

Cohen's kappa	
Modified Cyriax release test	0.92
Elevated arm stress test	1.00
Cervical rotation lateral flexion test	1.00
Elbow flexion pressure provocation test	0.66
Interclass correlation coefficient	
Semmes-Weinstein monofilament test	0.47
5th-digit abduction test	0.86

inclusion criteria for ULNN (+) or ULNN (−) groups. To ensure independence of the data, another 72 upper limbs were excluded from the data analysis (see classification of subjects described previously). Demographic data are presented in Table 2. A total of 32% and 43% of the upper limbs of cyclists with ULNN (+) presented with a positive elevated arm stress test and Cyriax release test, respectively, whereas only 4% and 7% of the upper limbs of cyclists with ULNN (−) presented with a positive elevated arm stress test and Cyriax release test, respectively. These numbers were found statistically significant, with a greater number of the upper limbs of cyclists with ULNN (+) presenting with positive provocative testing for thoracic outlet syndrome than the upper limbs of cyclists with ULNN (−) (elevated arm stress test $P = 0.0048$; modified Cyriax release test $P = 0.002$). Results of Fisher exact test and odds ratios are presented in Table 3.

DISCUSSION

This is the first study to suggest a high prevalence of signs indicative of proximal thoracic outlet syndrome dysfunction suggestive of double crush syndrome in the upper limbs of cyclists with a clinical diagnosis of ulnar nerve neuropathy. The clinical diagnosis of ulnar nerve neuropathy was based on the subjective complaints of paresthesia and/or pain in the 4th and 5th digits of the hand in the ulnar nerve distribution and the presence of at least 1 objective sensory, motor, or provocation testing. The 3 objective tests used for the clinical diagnosis of ulnar nerve neuropathy have been validated[3,17,21] and their intratester reliability was established

TABLE 2. Demographic Data for Upper Limbs of Cyclists with ULNN (+) and ULNN (−)

Demographic Variables	ULNN (+)	ULNN (−)
Upper limbs (n)	40	28
Males/females	28/12	19/9
Age (years ± SD)	35.6 ± 11.4	35.5 + 11.0
Weight (kg)	69.1	68.6
Height (cm)	175	174
Body mass index (BMI)	23.6	22.7
Smoker	1	0
Road bike (%)	68.2	66.4
Mountain bike (%)	31.8	33.6
Padded gloves	28	23
Padded bars	29	19

in the pilot testing. We opted for a clinical diagnosis based on examination findings instead of electrodiagnostic recordings in view of the low sensitivity and rate of false-negative results (22%) reported in the literature for electrodiagnostic testing of ulnar nerve neuropathy.[27]

The presence of thoracic outlet syndrome dysfunction was evaluated based on the elevated arm stress test and the modified Cyriax release test, which were found to be highly reliable with kappa scores of 1.0 and 0.92, respectively. Thirteen upper limbs of cyclists (32%) presented with a positive elevated arm stress test in the ULNN (+) group as compared with 1 only (4%) in the ULNN (−) group, whereas 17 upper limbs of cyclists (43%) presented with a positive modified Cyriax release test in the ULNN (+) group as compared with 2 (7%) only in the ULNN (−) group. These results indicate that perturbation in the thoracic outlet is significantly more prevalent in cyclists with ULNN (+). The position of the cyclists with neck and shoulder girdle protraction, wrist extension, forearm pronation or neutral and elbow flexion, and the potential elevation of the first rib increase the strain of the ulnar nerve, brachial plexus, and nerve roots into the upper limb resulting in a greater potential for the development of ulnar nerve neuropathy.[28–30] The cycling position exposes the ulnar nerve to a continual low-grade loading associated with an entrapment neuropathy, resulting in the distal aspect of the nerve being easily damaged by chronic nerve compression and the proximal part of the same nerve being more susceptible to become symptomatic, thus producing the double crush syndrome.[31,32] More research investigating the location of ulnar nerve entrapment neuropathy at the cubital tunnel or Guyon's canal would be valuable.

Perturbation in the thoracic outlet region was also evaluated in the present study using the cervical rotation lateral flexion test for the presence of an elevated first rib. This test has been previously validated for the presence of an elevated first rib,[26] and its reliability was found to be high ($\kappa = 1.0$) in the pilot testing. Although not statistically significant, 7 upper limbs of cyclists (18%) with ULNN (+) neuropathy compared with none (0%) in the ULNN (−) group presented with a positive cervical rotation lateral flexion test. These results are similar to those reported in asymptomatic subjects (1.2%) by Sizer et al.[32] The odds ratio suggested that the presence of an elevated first rib was 13 times more likely to occur in the upper limbs of cyclists with ULNN (+) neuropathy than in asymptomatic limbs. This could be another factor contributing to the greater frequency of peripheral upper limb neuropathies observed in cyclists.[1]

Although the incidence of neck and shoulder pain was not significantly different in the ULNN (+) group as compared with the ULNN (−) group ($P = 0.054$ and 0.012, respectively), the odds ratios revealed that cyclists with ULNN (+) had a probability of reporting a history of neck and shoulder pain more than 3 and 5 times greater, respectively, than cyclists with ULNN (−). Neck pain has been found to be a factor associated with peripheral neuropathies of the upper limb. Chow et al[5] reported that patients with nonmedian nerve hand numbness had a high prevalence of more than 33% of neck pain. Similar results were found in the present study with 38% of subjects in the ULNN (+) group reporting a history of neck pain, whereas

TABLE 3. Fisher Exact Test to Evaluate Frequency of Occurrence of Thoracic Outlet Perturbation, Neck, and Shoulder Pain in the Upper Limbs of Cyclists with ULNN (+) Versus ULNN (−)

Variables	ULNN (+) (N = 40)	ULNN (−) (N = 28)	Fisher Exact Test *P*-Value	Odds Ratio (95% Confidence Interval)
(+) Modified Cyriax release test	17	2	0.0020*	9.61 (2.00 to 46.2)
(+) Elevated arm stress test	13	1	0.0048*	13.0 (1.59 to 106.5)
(+) Cervical rotation lateral flexion test	7	0	0.0360	12.8 (0.70 to 233.48)†
History of shoulder pain	16	3	0.0124	5.56 (1.43 to 21.53)
History of neck pain	15	4	0.0539	3.60 (1.05 to 12.4)

*Indicates statistical significance at the alpha-adjusted level of 0.01 (Bonferroni correction applied for the 5 tests, eg, familywise alpha = 0.05/ 5 tests = 0.01).

†Note that the cervical rotation lateral flexion test variable has 1 value of 0 in the Fisher exact test contingency table. To make confidence interval calculations possible for this variable, 0.5 was added to each value (but not for calculations of the *P* value).

only 14% of subjects in the ULNN (−) reported neck pain. Several authors have reported cervical dysfunctions associated with upper limb peripheral neuropathy.[33–35] Likewise, 40% of cyclists with ULNN (+) neuropathy reported a history of shoulder pain, whereas only 11% of cyclists with ULNN (−) neuropathy reported the same complaint. Shoulder pain could be related to proximal neural sensitivity because typical presentations of double crush syndrome often include pain in the neck, shoulder, and upper limb.[36] The possibility also exists that cyclists with intrinsic shoulder pathology and pain would change their shoulder girdle position while cycling to offload the shoulder, resulting in an increased nerve tension in the brachial plexus. The lack of significance in the present study for these 2 variables could be related to a lack of power. More research is needed using a larger sample size to evaluate if neck dysfunctions and shoulder pathologies are associated with ulnar nerve neuropathies.

Information about the high prevalence of proximal dysfunctions in cyclists with ULNN (+) could be used to educate cyclists regarding frequent changes in the position of the neck, shoulder girdle, and upper limb while on the bicycle as a preventive measure to lower the negative effect of a continual load on the neural tissue and potentially improve symptoms. Clinicians evaluating the upper limbs of cyclists with symptoms of ulnar nerve neuropathy could include in the clinical examination the evaluation of the cervical spine, shoulder girdle, first rib, and brachial plexus as an adjunct for identifying the presence of double crush.

CONCLUSION

A significantly greater number of upper limbs of cyclists with clinical diagnosis of ulnar nerve neuropathy presented with symptoms associated with proximal dysfunctions, suggestive of double crush syndrome. Evaluation of proximal structures involved with thoracic outlet perturbations in cyclists with ulnar nerve neuropathy should not be neglected.

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