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# The incidence of Wartenberg's syndrome: a systematic review of the current literature.

Author: Endris Carlos Rosales Campos<sup>a</sup>

<sup>a</sup> Estudiante de Medicina, 3er año. Alumno ayudante de Cirugía Plástica y Reconstructiva.

La Habana, Cuba

#### 1. Abstract

Introduction: Wartenberg's syndrome is a rare condition, explained as a compression neuropathy of the superficial radial nerve. Little is known about the incidence of this neuropathy.

Objective: To Identify the incidence of Wartenberg's syndrome in the general population according to the current literature

Methods: A systematic review of English written articles retrieved from the MEDLINE database. Search terms included: Radial Neuropathy/epidemiology [Mesh], Radial Neuropathy [Mesh], Wartenberg's syndrome, superficial radial neuropathy and cheiralgia paresthetica.

Results: Obtained incidences of specific populations were 0.0004%, 0.002% and 0.82%. Observations of Wartenberg's syndrome in the six included studies were, respectively: two in one year, seven in 15.5 years, 50 in 4.5 years, three in 14 years, 12 in one year and 21 in eight years.

Conclusions: The results from this study confirmed that Wartenberg's syndrome is very rare, but a specific incidence could not be retrieved from the articles. Further research is required before an incidence for the general population can be found for this syndrome.

Keywords: Radial Neuropathy, Epidemiology

#### 2. Introduction

Wartenberg's syndrome, also described as cheiralgia paresthetica or superficial radial neuropathy (SRN), is a compression neuropathy of the superficial radial nerve<sup>1</sup>. The superficial radial nerve is branched off from the radial nerve at circa two thirds of the forearm<sup>2</sup>. The nerve is sensory cutaneous nerve innervating the skin on the dorsum of the hand, including skin between the thumb and index finger, and providing articular branches to joints in the hand<sup>3</sup>. Figure 1 shows the distribution of the terminal branches of the superficial branch of the radial nerve.



Superficial radial nerve

Figure 1; The distribution of the terminal branches of the superficial branch of the radial nerve.

Symptoms are described as unpleasant and are negatively present during the whole day. Due to anatomical variation, symptoms may vary per person. However, symptoms such as vague pain, tingling sensation or numbness, all located on the dorsal radial part of the hand or forearm, as shown in figure 2, are the most common<sup>1</sup>.

Multiple types of trauma can cause this rare condition. A variety of cases resulting in Wartenberg's syndrome have been described in current literature. For instance, trauma caused by tight watchbands, acupuncture, intravenous infusion, direct blunt trauma to the superficial radial nerve, overtightened handcuffs, fractures or surgery<sup>4-10</sup>.

Different types of treatment used for Wartenberg's syndrome are described in literature. Most of these treatments are based on local surgery of the affected area and aim to be an instant relief of symptoms. For instance, endoscopically assisted decompression is a remedy used in the majority of cases. In this procedure, a

longitudinal incision is made at the Tinel's sign, which is the location where a tingling sensation is felt by lightly tapping on the nerve, followed by the removal of the entrapment<sup>11</sup>.



Figure 2; The area of paresthetica usually described by the patient.

Wartenberg's syndrome is a very rare condition<sup>3</sup> and little is known about the specific occurrence of this neuropathy. To this day, an incidence remains unknown. To the knowledge, a systematic review has been performed to study the incidence. It is expected to deduce and conclude an incidence out of the current literature, mainly to provide a basis for further research regarding Wartenberg's syndrome, but also to create more awareness for this burdensome condition. Therefore, the primary aim of this study is to deduce the incidence of Wartenberg's syndrome in the general population.

#### 3. Methods

#### 3.1 Search strategy

A systematic search was performed on January 2022 in the MEDLINE database. In the search, was used the following search term to find studies relating to the primary study aim: (("Radial Neuropathy/epidemiology"[Mesh] OR "Radial Neuropathy"[Mesh]) AND ("superficial radial" OR "superficial branch" OR "wartenberg" OR "cheiralgia paresthetica")) OR "wartenberg's syndrome" OR "superficial radial neuropathy" OR (("radial neuropathy" OR "cheiralgia paresthetica")

To clarify the search term, the terms 'radial neuropathy' and 'cheiralgia paresthetica' occur twice in the search term. Firstly, were combined these terms with Mesh-terms.

Secondly, they were combined separately with a time interval between 2018 and 2022. This was done to provide as many studies as possible that may be useful, including studies published in the past four years that did not get a Mesh-term yet.

#### 3.2 Inclusion and exclusion criteria

To be included, studies had to contain information about the observation of at least one of the following terms: Wartenberg's syndrome, cheiralgia paresthetica or SRN. Studies written in any language other than English were excluded, and studies done on animals. Systematic reviews and meta-analyses were also excluded. In addition, studies without an incidence or necessary data about the observation of SRN were excluded.

This is shown in the flow-chart; see figure 3.

#### 3.3 Quality assessment

To determine the quality of the included studies, the NHLBI quality assessment tool<sup>12</sup> was used. To quantify the results, two points were given if a question was answered with a YES, zero points if a question was answered with a NO and one point if a question was answered with an OTHER, meaning not applicable or cannot be determined. The questionnaire consisted of nine questions, with a maximum of 18 points. An article must score at least 60 percent, or 11 points, of the total points to be considered of good quality. The results of this quality assessment were not used as exclusion criteria.

#### 3.4 Statistical analysis

In case the incidence of Wartenberg's syndrome had not been indicated in the articles, the formula of incidence (number of cases / total population) x 100 (for a percentage) was used.

#### 4. Results

#### 4.1 Study selection

The search of the MEDLINE database produced 192 studies. After applying the exclusion criteria, six studies were included; see figure 3. In figure 3, the steps and reasons for the exclusion are described.



Figure 3; flow-chart study selection

#### 4.2 Study characteristics

All six studies were published in English between 1975 and 2018. An overview of the methods used in the included studies is provided in table 1. Five out of six studies<sup>13-17</sup> were performed in Western countries; one was done in Japan<sup>18</sup>. All included studies collected their data retrospective. The number of patients varied between 8 and 91. Four studies<sup>13,14,16,18</sup> were set in medical centers, of which three were hospitals

<sup>13,16,18</sup>. One study used electrodiagnostic studies performed in the Electromyography Laboratory at a hospital<sup>15</sup> and another study used medical records from two electromyography services<sup>17</sup>. In two studies<sup>15,17</sup>, the neuropathy of the superficial radial nerve originated from bone damage.

#### 4.3 Outcome measures

An overview of the outcome measures of the included studies is provided in table 2. Two studies<sup>15,17</sup> used abnormal electrophysiological data for the outcome measures. The relief of pain and/or paresthesia was used to measure the results by Braidwood<sup>13</sup> and Lanzetta et al.<sup>16</sup>.

Lanzetta et al.<sup>16</sup> used this information as a criterion to differentiate between excellent, good, fair and poor results of the treatment given in this study. The article does not further explain what this means.

In addition to studying patients, Matsubara et al.<sup>18</sup> was the only study to use imageand operative findings as outcome measure. Only Anderson et al.<sup>14</sup> used patients' charts with information on the type of fixator used and complications. The information was collected during a follow-up. The follow-up period lasted until the patients were discharged from care.

#### 4.4 Results

An overview of the results and calculated incidences of the included studies is provided in table 2.

Anderson et al.<sup>14</sup> researched the complications of treating distal radius fractures with external fixation. One of these complications was SRN, and the observation thereof in this study was two out of 24 patients. In 2001, *640.000 cases* of distal radial fractures were reported in the United States (US) alone<sup>19</sup>. The treatment thereof with external fixation was *2.1%* in 2014 <sup>20</sup> and the average US population between 2001 and 2014 was 301.6 million<sup>21</sup>. This results in an incidence of 0.0004% for SRN after external fixation as a treatment for distal radius fractures in the US. Karakis et al.<sup>15</sup> analysed the clinical and electrophysiologic patterns of nerve injury in pediatric patients - children and adolescents - with radial neuropathy. Seven patients

with SRN were found in 15.5 years. They reviewed electrodiagnostic studies performed in the Electromyography Laboratory at the pediatric hospital. Hence the cases they collected were patients from that hospital. Therefore, with an annual admission rate of 25.000<sup>22</sup>, the calculated hospital incidence was 0.002%.

Lanzetta et al.<sup>16</sup> reported 50 cases of Wartenberg's syndrome in 4,5 years in their hand surgery hospital. They indicated that this was 0.82% of the total patients referred to their unit during this period. So, the hospital incidence of Wartenberg's syndrome was 0.82%.

Matsubara et al.<sup>18</sup>, who studied eight cases of radial nerve palsy due to a spaceoccupying lesion (SOL) at the elbow, reported three cases of SRN. Braidwood<sup>13</sup> reported 12 patients with Wartenberg's syndrome in one year in their division and Mondelli et al.<sup>17</sup> reported 21 cases of SRN in eight years while researching clinical and neurographic findings of radial nerve mononeuropathies.

#### 4.5 Quality assessment

To determine the quality of the studies the NHLBI quality assessment tool<sup>12</sup> was used. The main points scored of the six included studies were 12.83. Five studies<sup>13,15-18</sup> were case series, and one was a chart review<sup>14</sup>. There was no relation between the quality score and the study design. The results of the assessment are shown in table 3 in the appendix.

#### 5. Discussion/Conclusions

#### 5.1 Summary

The main aim for this systematic review was to measure the incidence of Wartenberg's syndrome in the general population. This is the first systematic review about this subject. Little research is done about Wartenberg's syndrome; therefore, few studies contain information about this subject. The six studies included mentioned observations about Wartenberg's syndrome. Three studies<sup>14-16</sup> showed an incidence of SRN in a specific population.

#### 5.2 Limitations

The six studies analysed were very heterogeneous. For instance, because of relatively small study population sizes, due to the syndrome being so rare and poorly documented. This meant that it was unable to perform a meta-analysis, which means this article is of lower level of evidence. The age, sex and conditions under which the participants developed the syndrome were different in the six studies. Karakis et al.<sup>15</sup> analysed collected data from patients with SRN in a pediatric hospital, meaning that the average age of the study population was below 18. Lanzetta et al.<sup>16</sup> analysed data from their own hospital, specialised in hand surgery, meaning that they encountered more cases of Wartenberg's syndrome than other studies. The heterogeneity in all studies meant that performing a meta-analysis was not possible.

The incidence of SRN could be calculated for two studies<sup>14,15</sup>, in which it could be found the necessary information. However, for the calculations of Anderson et al.<sup>14</sup>, numbers of the general population were compared to the numbers of this small and specific study population. This incidence is therefore only an estimate of the true incidence for the general population. Karakis et al.<sup>15</sup> retrieved their data from electrodiagnostic studies performed in the Electromyography Laboratory at their hospital from a period of 15.5 years. Calculations for SRN incidence were done with the yearly mean hospital admissions. Therefore, It was assumed that all patients with SRN in that hospital during that period of 15.5 years, were also included in those electrodiagnostic studies.

Another limitation was that for both Matsubara et al.<sup>18</sup> and Braidwood<sup>13</sup> was not found the necessary numbers to calculate a hospital incidence of SRN. The results found in Mondelli et al.<sup>17</sup> were not applicable to calculate a specific incidence for SRN, because they got their cases from two unidentified electromyography (EMG) services.

### 6. Conclusions

The results of the systematic review did confirm that Wartenberg's syndrome is very rare in the general population.

The study provided an overview of the current observations of Wartenberg's syndrome in specific populations.

This study serves as a starting point for a further research on the incidence of Wartenberg's syndrome in the general population.

#### 7. Recommendations

For future studies, it is recommended to perform a retrospective study design of all the cases found in a national database over a long period of time in order to find a scientifically strongly substantiated incidence for Wartenberg's syndrome.

#### References

(1) Anthony JH, Hadeed A, Hoffler Charles E. Cheiralgia Paresthetica.: StatPearls Publishing LLC; 2021.

(2) Robson AJ, See MS, Ellis H. Applied anatomy of the superficial branch of the radial nerve. Clinical anatomy (New York, N.Y.) 2008 Jan;21(1):38-45.

 (3) Ariyasinghe, C., Hacking, C. Superficial radial nerve. Reference article, Radiopaedia.org. [online] Available at: <<u>https://radiopaedia.org/articles/superficial-</u> <u>radial-nerve</u>> [Accessed on 09 Jan 2022]

(4) Bierman HR. Nerve Compression Due to a Tight Watchband. The New England journal of medicine 1959 Jul 30,;261(5):237-238.

(5) Capkin S, Kaleli T. Superficial radial nerve compression due to fibroma of the brachioradialis tendon sheath: A case report. Acta orthopaedica et traumatologica turcica
 2019
 Sep;53(5):394-396.

(6) Grant AC, Cook AA. A prospective study of handcuff neuropathies. Muscle & nerve 2000 Jun;23(6):933-938.

(7) Hu S, Choi J, Son B. Cheiralgia Paresthetica: An Isolated Neuropathy of the Superficial Branch of the Radial Nerve. The Nerve 1970 Jan 1,;1(1):1-5.

(8) Marchant MH, MD, Gambardella RA, MD, Podesta L, MD. Superficial radial nerve injury after avulsion fracture of the brachioradialis muscle origin in a professional lacrosse player: A case report. Journal of shoulder and elbow surgery 2009;18(6):e9-e12.

(9) Politylo J, Decina PA, Lopes AA. Superficial radial neuropathy secondary to intravenous infusion at the wrist: a case report. Journal of the Canadian Chiropractic Association 1993 Jun;37(2):92-96.

(10) Xu S, Wang L, Cooper E, Zhang M, Manheimer E, Berman B, et al. Adverse Events of Acupuncture: A Systematic Review of Case Reports. Evidence-based complementary and alternative medicine 2013 Mar 20,;2013:581203-15.

(11) Spieß CK, Müller LP, Oppermann J, Neiss WF, Hahn P, Unglaub F. Surgical decompression of the superficial radial nerve: Wartenberg syndrome. Operative Orthopädie und Traumatologie 2016 Apr;28(2):145-152.

(12) Nhlbi.nih.gov. 2021. Study Quality Assessment Tools | NHLBI, NIH. [online] Available at: <a href="https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools">https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools</a>> [Accessed 4 January 2022].

(13) Braidwood AS. SUPERFICIAL RADIAL NEUROPATHY. Journal of bone and joint surgery. British volume 1975 Aug;57(3):380-383.

(14) Anderson JT, Lucas GL, Buhr BR. Complications of treating distal radius fractures with external fixation: a community experience. The Iowa orthopaedic journal 2004;24:53-59.

(15) Karakis I, Georghiou S, Jones HR, Darras BT, Kang PB. Electrophysiologic Features of Radial Neuropathy in Childhood and Adolescence. Pediatric neurology 2018 Apr;81:14-18.

(16) LANZETTA M, FOUCHER G. Entrapment of the superficial branch of the radial nerve (Wartenberg's syndrome). A report of 52 cases. International orthopaedics 1993;17(6):342-345.

(17) Mondelli M, Morana P, Ballerini M, Rossi S, Giannini F. Mononeuropathies of the radial nerve: Clinical and neurographic findings in 91 consecutive cases. Journal of electromyography and kinesiology 2005;15(4):377-383.

(18) Matsubara Y, Miyasaka Y, Nobuta S, Hasegawa K. Radial Nerve Palsy at the Elbow. Upsala journal of medical sciences 2006;111(3):315-320.

(19) Chung KC, Spilson SV. The frequency and epidemiology of hand and forearm

fractures in the United States. The Journal of hand surgery (American ed.) 2001 Sep;26(5):908-915.

(20) Kakar S, Noureldin M, Van Houten HK, Mwangi R, Sangaralingham LR. Trends in the Incidence and Treatment of Distal Radius Fractures in the United States in Privately Insured and Medicare Advantage Enrollees. Hand (New York, N.Y.) 2020 Jun 7,:155894472092847-1558944720928475.

(21) Worldometer.info. 2022. United States Population. [online] Available at:<a href="https://www.worldometers.info/world-population/us-population/">https://www.worldometers.info/world-population/us-population/</a>> [Accessed on 06 Jan 2022]

(22) A brief introduction to Boston Children's Hospital. Pediatric investigation 2017 Dec;1(1):7-8.

# Table 1: Method characteristics

Authors	Year	Country, Clty	Study design	Data collection	Patient population (n)	Specific population	Setting
Anderson et al.	2004	US, Wichita	Chart review	Retrospective	24	Patients with SRN after distal radius fracture treated with external fixation	Two community medical centers
Karakis et al.	2018	US, Boston	Case series	Retrospective	19	Sensory loss in superficial radial nerve distribution, in children and adolescents with radial neuropathy	Electrodiagnostic studies performed in the Electromyography Laboratory at a pediatric hospital
Lanzetta et al.	1993	France, Strasbourg	Case series	Retrospective	50	Patients with Wartenberg syndrome	Hand surgery hospital
Matsubara et al.	2006	Japan, Ishinomaki and Sendai	Case series	Retrospective	ø	Patients where surgery was performed for radial nerve palsy due to a space-occupying lesion of the elbow	Three hospitals
Braidwood	1975	Scotland, Glasgow	Case series	Retrospective	12	Patients with SRN	Teaching hospital
Mondelli et al.	2005	Italy, Siena	Case series	Retrospective	91	Cases of radial nerve mononeuropathy	Medical records in two electromyography services
Abbreviations	s: SRN, si	uperficial radial neur	ropathy;				

## Annexes

Authors	Year	Country, City	Study design	Outcome measures	Results	Quality assessment (points scored/ percent)	Calculated incidence
Anderson et al.	2004	US, Wichita	Chart review	Patients' charts during a follow-up	2/ 24 (8.33%) - 1 year	13 (72%)	0.0004% in the US
Karakis et al.	2018	US, Boston	Case series	Collected electrophysiologic data	7/ 12 (58%) - 15.5 years	14 (78%)	0.002% in referred hospital
Lanzetta et al.	1993	France, Strasbourg	Case series	Using recordings of relief of pain as a criterion to differentiate between ex- cellent, good, fair and poor results of the given treatment	50 cases - 4.5 years	13 (72%)	0.82% in referred hospital
Matsubara et al.	2006	Japan, Ishinomaki and Sendai	Case series	Patient studying, image findings and operative findings.	3/ 8 - 14 years	13 (72%)	-
Braidwood	1975	Scotland, Glasgow	Case series	The relief of the burning pain and paraesthesia	12 cases - 1 year	12 (67%)	
Mondelli et al.	2005	Italy, Siena	Case series	A electrophysiological severity scale	21/ 91 - 8 years	14 (78%)	-

Table 2: Outcome measures, results and calculated incidences

	Anderson et al.	Karakis et al.	Lanzetta et al.	Matsubara et al.	Braidwood	Mondelli et al.
<ol> <li>Was the study question or objective clearly stated?</li> </ol>	Yes (2)	Yes (2)	No (0)	Yes(2)	Yes (2)	Yes (2)
<ol> <li>Was the study population clearly and fully described, including a case definition?</li> </ol>	Yes (2)	Yes (2)	Yes (2)	Yes (2)	Yes (2)	Yes (2)
3. Were the cases consecutive?	Other (1)	Other (1)	Other (1)	Other (1)	Other (1)	Other (1)
<ol><li>Were the subjects comparable?</li></ol>	No(0)	No (0)	Yes (2)	No (0)	No (0)	No (0)
5. Was the intervention clearly described?	Yes (2)	Other (1)	Yes (2)	Yes (2)	Yes (2)	Yes (2)
6. Were the outcome measures clearly defined, valid, reliable, and implemented consistently across all study participants?	Yes (2)	Yes (2)	Yes (2)	Yes (2)	Yes (2)	Yes (2)
7. Was the length of follow-up adequate?	Other (1)	Yes (2)	Yes (2)	Other (1)	Other (1)	Other (1)
8. Were the statistical methods well-described?	Other (1)	Yes (2)	No (0)	Other (1)	No (0)	Yes (2)
9. Were the results well-described?	Yes (2)	Yes (2)	Yes (2)	Yes (2)	Yes (2)	Yes (2)
Total points given:	13	14	13	13	12	14

Table 3: Quality assessment results (Appendix)