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Case Report

Carpal tunnel syndrome caused by the entrapment of a bifid Lanz IIIA Type anatomical variant of median nerve: A case report and systematic literature review

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ABSTRACT

Background: Carpal tunnel syndrome (CTS) is the most common entrapment peripheral neuropathy. Median nerve may present several anatomical variations such as a high division or bifid median nerve (BMN). A thorough knowledge of the normal anatomy and variations of the median nerve at the wrist are fundamental to reduce complications during carpal tunnel release.

Case Description: A 63-year-old man with CTS underwent preoperative ultrasound that showed the entrapment of the median nerve and disclosed a BMN Lanz IIIA Type anatomical variation at the carpal tunnel. During the surgery, the anatomical variant of a BMN at the wrist has been visualized. Both nervous rami entirely occupied the carpal canal and this may have predisposed to the development of the entrapment syndrome. Nor persistent median artery, or other associated abnormalities, have been identified. At the 6 months follow-up control, the patient referred a good surgical recovery with complete resolution of the preoperative symptoms of the median nerve entrapment.

Conclusion: A rare case of Lanz IIIA BMN Type at the wrist has been encountered in a patient with a CTS and a systematic review and practical considerations have been presented with the aim of raising awareness to the neurosurgical community of a such rare variant that could be encountered during carpal tunnel release procedures. CTS may be caused by the entrapment of a BMN Lanz IIIA Type anatomical variant of median nerve. Preoperative US would help to identify such patients to reduce risk of iatrogenic injuries.

Keywords: Anatomy, Bifid median nerve, Carpal tunnel syndrome, Median nerve, Peripheral nervous system diseases

INTRODUCTION

Carpal tunnel syndrome (CTS) is the most common entrapment peripheral neuropathy with a prevalence of 6%.[17] It is caused by a chronic compression of the median nerve within the carpal tunnel mainly resulting in pain and paresthesia in the palmar radial part of the affected hand.[45] Median nerve may present several anatomical variations that are surgically relevant.[26] Lanz described a classification of the median nerve identifying different types:[39]

- Group 0: Extraligamentous thenar branch (standard anatomy).
- Group I: Thenar branch variations in relation to the transverse carpal ligament (TCL).

- Group II: Distal accessory thenar branches.
- Group III: High division of the median nerve.
- Group IV: Proximal accessory thenar branches.

In 2009, Al-Qattan *et al*. further subclassified the high division of the median nerve, differentiating six subgroups, according to the associated anomalies:[1]

- Subgroup I: Bifid median nerve (BMN) without persistent median vessels or abnormalities
- Subgroup II: BMN with persistent median vessels without any pathology
- Subgroup III: BMN with persistent median vessels with aneurysm, thrombosis, or arteriovenous malformation
- Subgroup IV: BMN with each division passing separately in the carpal tunnel
- Subgroup V: BMN with aberrant muscle between its two divisions
- Subgroup VI: BMN with aberrant branches.

A thorough knowledge of the normal anatomy and variations of the median nerve at the wrist is fundamental to reduce complications during carpal tunnel release. In the present paper, we present a rare case of BMN, or Lanz IIIA Type, at the wrist in a patient with CTS and an associated systematic review to give awareness to the neurosurgical community of such scenario.

MATERIALS AND METHODS

The present case has been reported according to the international CARE guidelines.[49]

Literature review search strategy

A systematic literature review was performed in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-analyses statement guidelines, limited to the English language.[43] SCOPUS and PubMed databases were queried using individual keywords and MeSH terms. A purposely defined search string was performed for PubMed search: ("Bifid [All Fields] AND (median nerve [MeSH Terms] OR (median [All Fields] AND nerve [All Fields]) OR median nerve [All Fields])) AND (humans [MeSH Terms] AND English[lang]; and for SCOPUS search: (bifid AND median AND nerve) AND (LIMIT-TO [LANGUAGE, "English"]). The results were then limited to human subjects. After duplicate removal, title and abstracts were firstly screened and, for the papers deemed appropriate, full text was obtained and reviewed for appropriateness and extraction of data [Figure 1]. Article references list was also examined to identify any other relevant study.

Selection criteria

Only studies dealing with the presence of BMN within or proximal to the carpal tunnel were included, also considering anatomical and radiological researches. The articles without relevant data, review, visual vignette, or sonographic studies with a cohort already screened for only patients with BMN were excluded. Studies involving magnetic resonance imaging (MRI) and ultrasonography (US) as presurgical diagnostic test were included only if sufficient individual data on the prevalence of BMN and its clinical correlation could be obtained to meet the inclusion criteria.

Data extraction

Data from the included studies were extracted, organized, and analyzed using Microsoft Excel 2019 (Microsoft Corp., Redmond, WA, USA). Collected variables included first author, publication year, number of the total wrists considered, and number of those which presented BMN and other associated anatomical features (i.e., persistent median artery [PMA]/vein, anomalous tendon, neural loop, and aberrant muscle), Lanz and Al-Qattan classifications, presence of an associated CTS, and its presumed etiology.[1,39]

CASE REPORT

A 63-year-old man complained with a 6-month history of nocturnal paresthesia and intermittent burning pain of the right arm and hand, along with the distribution of the right median nerve. His clinical history was characterized by diabetes mellitus Type 2 and hypertension. He worked as a chef and his previous surgical history was negative. A first nonsurgical approach has been proposed to the patient but without beneficial response. The patient furtherly complained a worsening of his symptoms resulting in progressive motor and sensory deficits, such as loss of texture discrimination and fine motor hand skills. Electrophysiological diagnostics showed a marked alteration of the right median nerve, with an increased distal motor latency, reduction of nerve conduction velocity, and the presence of sporadic fibrillations to the abductor pollicis brevis muscle.

The patient came to our office and general physical examination was normal. Neurological examination disclosed an impairment of his right hand movements, specifically motor and sensory deficit attributable to the right median nerve, with mild thenar atrophy, positive Tinel's sign and Phalen's test. An open surgical decompression of the carpal tunnel was proposed. Preoperative ultrasound confirmed the entrapment of median nerve and disclosed a BMN anatomical variation Lanz IIIA Type, subgroup I, at the carpal tunnel [Figure 2].

The anatomical landmarks were recognized, and, under local anesthesia, a 4 cm skin incision has been made at the intersection of the proximal extension of the radial side of the ring finger with the Kaplan's cardinal line. The subcutaneous tissue with its fat component was incised until the exposure of the TCL. Afterwards, TCL has been incised and, underneath, the anatomical variant of a BMN at the wrist has been visualized [Figure 3].

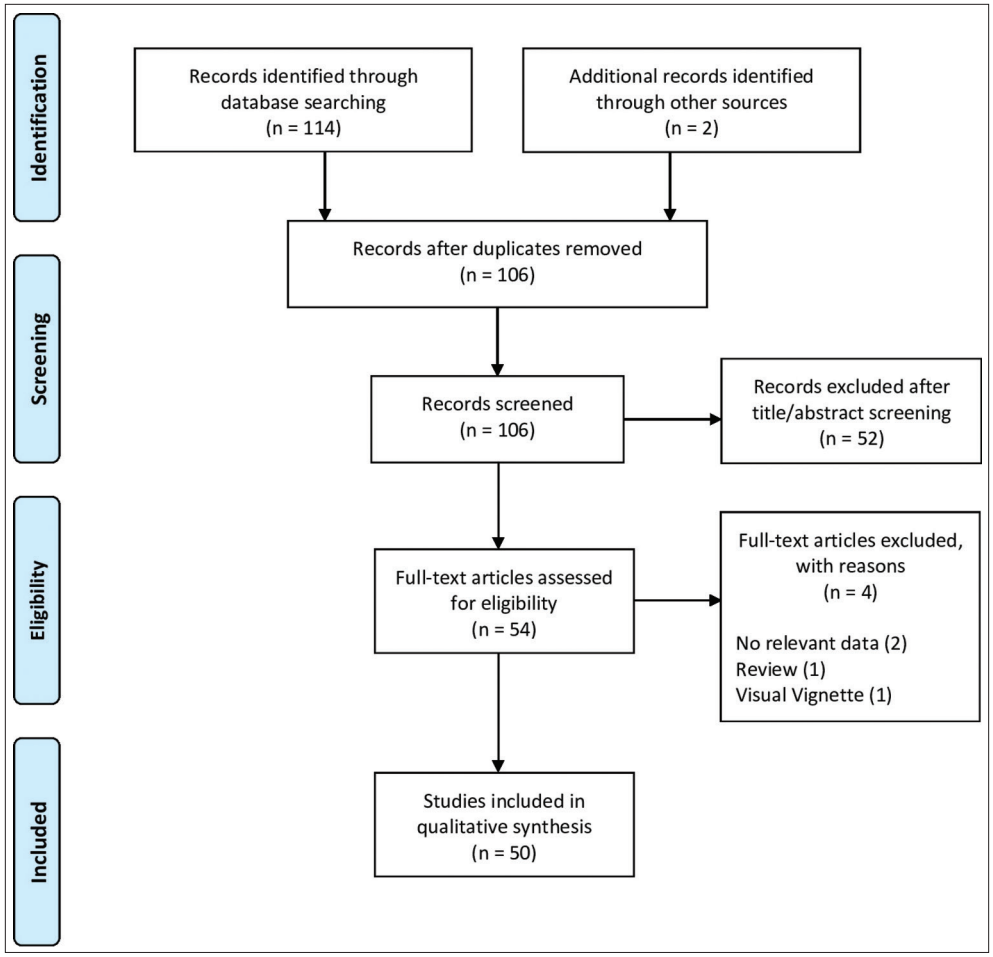

Figure 1: Flow diagram according to Preferred Reporting Items for Systematic Reviews and Meta-analyses guidelines.

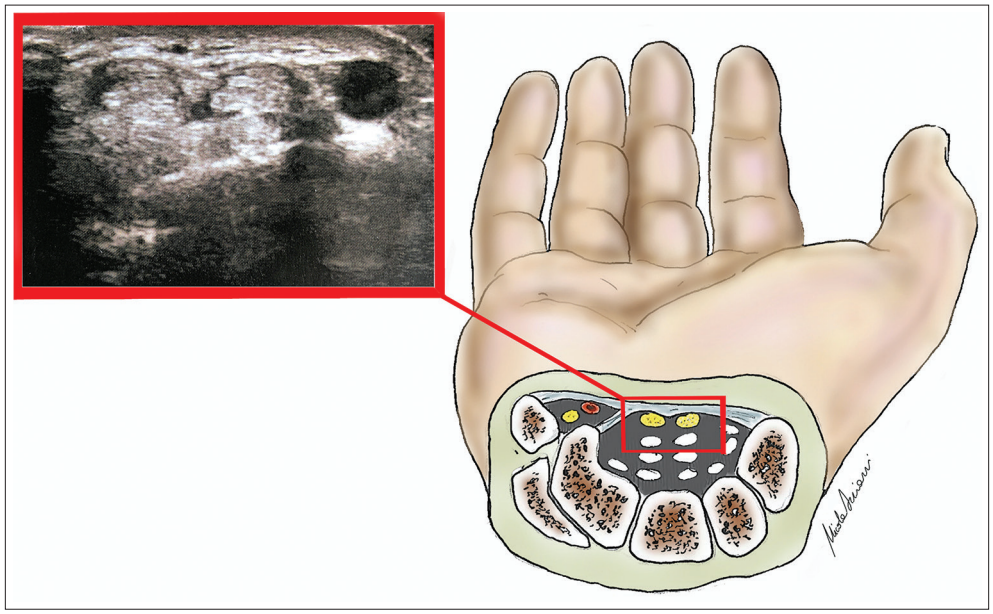

Figure 2: Preoperative ultrasonography showing the anatomical variation and an associated graphical representation of the bifid median nerve at the carpal tunnel.

The BMN macroscopically appeared as pale nerves indicating a chronic suffering. Both nervous rami entirely occupied the carpal canal and this may have predisposed to the development of the entrapment syndrome. No PMA nor other associated abnormalities have been identified. The TCL has been completely sectioned and BMN completely released [Figure 4].

Closure of subcutaneous and skin layers have done in a standard fashion. At the 6 months follow-up control, the patient referred a good surgical recovery with complete resolution of the preoperative symptoms of the median nerve entrapment.

RESULTS

The results of the systematic literature review are summarized in the [Table 1]. The anatomical variations of median nerve and, particularly, the definition of BMN have been widely described by Ulrich Lanz in 1977 who reported 7 (2.8%) high divisions of the median nerve in 246 hands and, among these, five nerves were also associated to a PMA.[39] Lanz collectively described such high divisions of median nerve variants as Group III; specifically, IIIA included ones without an artery; IIIB ones with a PMA; and IIIC ones with a lumbrical muscle [Figure 5].

Afterward, Amadio reported a prevalence of 3.3% in his anatomical series and described one case of a BMN with two separate compartments within the carpal tunnel. Moreover, he stated that these branches can also distally join together, or they may have a communicating bridge.[2,3] Szabo and Pettey analogously described an accessory compartment into the carpal tunnel, in which the radial branch of the BMN was localized.[54] Furthermore, Berry *et al.* reported a 4-year-old girl with BMN, after trauma exploration, where each branch was in a separate canal.[11] Poisel, in a study of 100 cadaveric hands, noted that 46% of variations were extraligamentous, 31% subligamentous, and 23% transligamentous.[6,39] Tountas *et al.*, in a study of 92 cadaveric hands, reported that 82% of variations were extraligamentous, 8.7% subligamentous, and 8.7% transligamentous.[56] According to our systematic literature review and the Lanz classification, 58% of the cases presented BMN proximal to the carpal tunnel (III group), 45% with also a PMA (IIIB group), 26.7% of the thenar branch variations were subligamentous (IA group), 4% transligamentous, and 3.3% presented distal accessory thenar branches (II group). Considering also the Al-Qattan classification, 5.4% of these cases presented anomalies of the PMA, such as aneurysm or arteriovenous malformation; 5.4% had aberrant branches of the median nerve (trifurcation or neuroma), 6.8% of the BMNs had the two branches passing into two separate compartments into the carpal tunnel, and 9.6% had also aberrant muscle between these two divisions (anomalous palmaris profundus muscle, as the most common).

In the surgical literature, different authors tried to establish a correlation between the presence of BMN and the development of CTS. Iannicelli *et al.* reported six patients with BMN in a population of 294 with CTS, underlying the importance of using US for recognizing such anatomical variations.[27] Tountas *et al.* identified 8 cases in a large series of 913 patients undergoing to carpal tunnel release (CTR).[56] After reviewing these large series, the incidence of BMN resulted among 0.8–2.8% in patients with CTS; although Bayrak *et al.* found an incidence much higher of BMN (18–19%) in their radiological study.[2,10,27,39,56] Granata *et al.* conducted a prospective study of 162 patients with CTS and a control group of 104 patients, in which there were respectively 30 and 16 BMNs and concluded there were not significant correlations between the presence

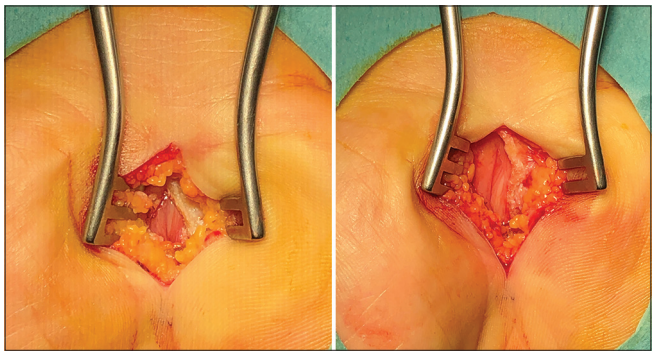

Figure 3: Intraoperative pictures showing the Type Lanz IIIA bifid median nerve released after transverse carpal ligament sectioning.

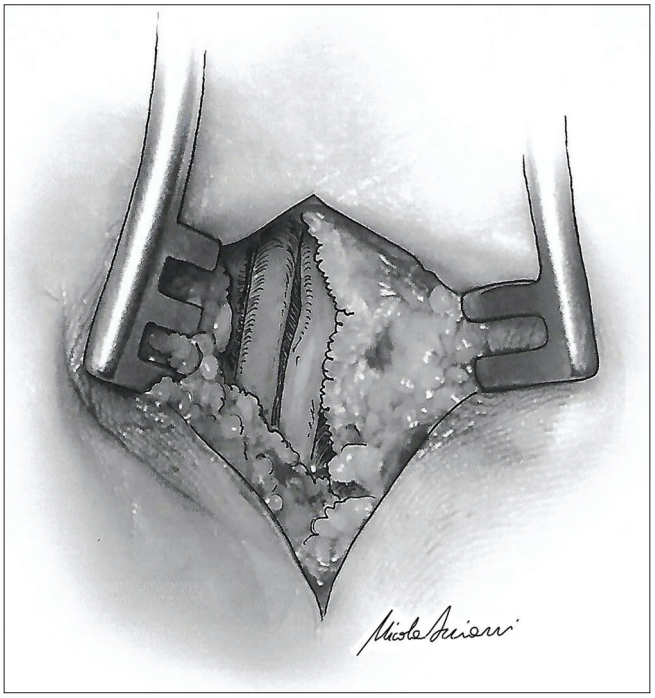

Figure 4: Surgical vignette representing the intraoperative view of a bifid median nerve during carpal tunnel release.

Table 1: Published studies mentioning the bifid median nerve anatomical variants.

Authors and year	Country	Type	Number of cases total	Bifid median *n*	Location	Lanz classification	Al-Qattan C.	CTS	Etiology
Lanz (1977)[39]	Germany	AS	246 Hands	7	Proximal to CT	N.A	N.A.	No	Idiopathic
Kornberg *et al.* (1983)[37]	Portsmouth (USA)	CR	1	1	Proximal to CT	3B (with three transligamentous motor branches)	6	No	Trauma
Amadio (1987)[2]	Rochester (USA)	CR	1	1	Proximal to CT	3A	4	N.A.	Idiopathic
Toranto (1989)[55]	Texas (USA)	CR	1	1	Proximal to CT	3B	3	Yes (recurrent)	Idiopathic
Fernandez-Garcia *et al.* (1994)[21]	Barcelona (Spain)	CR	1	1	Proximal to CT	3A	5 (anomalous FDS belly)	Yes	Trauma
Szabo and Pettey (1994) [54]	Sacramento (USA)	CR	1	1	Proximal to CT	3A	4	N.A.	Trauma
Artico *et al.* (1995)[6]	Italy	CR	2	2	Proximal to CT	1B	1	Yes	Idiopathic
Iannicelli *et al.* (2000)[27]	Italy	RS	294 Hands with CTS	6	Proximal to CT	3A	1	Yes	Idiopathic
Gutowski *et al.* (2000)[25]	NYC (USA)	CR	1	1	Proximal to CT	3B	3	N.A.	Idiopathic
Propeck *et al.* (2000)[48]	Michigan (USA)	AS/RS	1 Hand with CTS 10 cadavers	1 in CTS 2 in cadavers	One within CT two proximal to CT	3A (clinical case and cadaver two) 3B (cadaver one)	1 (clinical case and cadaver two) 2 (cadaver one)	Yes	Idiopathic
Yildirim *et al.* (2001)[61]	Turkey	CR	1	1	Proximal to CT	3A	1	N.A.	Idiopathic
Kele *et al.* (2002)[33]	Germany	CR	1	1	Proximal to CT	3B	3	Yes (acute)	Idiopathic
Jeon *et al.* (2002)[29]	Korea	CR	1	1	Proximal to CT	3A	1	N.A.	Idiopathic
Gassner *et al.* (2002)[22]	Austria	CS	2 hands with CTS 100 volunteers	2 in CTS 10 in volunteers	Two within CT 10 proximal to CT	3B	2	Yes (2)	Idiopathic
Berry *et al.* (2003)[11]	UK	CR	1	1	Proximal to CT	3A	4	Yes	Trauma (laceration)
Rossi *et al.* (2003)[51]	Italy	CR	1	1	Proximal to CT (distal third of forearm)	3A	1	Yes	Idiopathic
Wolter *et al.* (2003)[60]	Germany	CR	1	1	Proximal to CT	3A	4	N.A.	Idiopathic

(Contd...)

Table 1: (*Continued*).

Authors and year	Country	Type	Number of cases total	Bifid median *n*	Location	Lanz classification	Al-Qattan C.	CTS	Etiology
Lindley *et al.* (2004)[40]	Mississippi (USA)	AS	526	2	Proximal to CT (distal third of forearm)	3A	1 (one hand) 5 (one hand with anomalous FDS belly)	Yes	Idiopathic
Keramidas *et al.* (2004)[34]	UK	CR	2	2	Proximal to CT	3A (case one) 3B (case two)	4 (case one) 2 (case two)	N.A.	Trauma
Bataineh and Moqattash (2006)[9]	Jordan	CR/LR	1	1	Proximal to CT (proximal to flexor retinaculum)	3B	2	N.A.	Idiopathic
Jones (2006)[30]	New Zealand	CR	1	1	Proximal to CT (proximal to flexor retinaculum)	3A	5 (anomalous palmaris profundus)	Yes	Idiopathic
Bayrak *et al.* (2008)[10]	Turkey	AS	320 hands with CTS 240 hands	64 in CTS 22 in volunteers	12/240 proximal to CT	3A (12/240)	1 (12/240)	Yes (32)	Idiopathic
Sundaram *et al.* (2008)[53]	India	CR	1	1	Proximal to CT (proximal to flexor retinaculum)	3A	1	N.A.	Idiopathic
Pierre-Jerome *et al.* (2010)[47]	Atlanta (USA)	CS	194	48	36 within CT 12 proximal to CT	3A (12 proximal to CT) 3A (32 within CT) 3B (four within CT)	6 (one with trifurcation) 2 (four with median artery)	Yes (2)	Trauma (1) inflammatory arthropathy (1)
Al-Qattan *et al.* (2009)[1]	Saudi Arabia	CR	1	1	Proximal to CT	3B	3	N.A.	Idiopathic
Klauser *et al.* (2011)[35]	Austria	RS	684 hands with suspected CTS 551 hands	49 in CTS 27 in volunteers	N.A.	N.A.	N.A.	Yes (49)	Idiopathic
Granata *et al.* (2011)[23]	Italy	CS	162 hands with CTS 104 hands	30 in CTS 16 in volunteers	N.A.	N.A.	N.A.	Yes (30)	Idiopathic
Roll *et al.* (2011)[50]	Ohio (USA)	CR	1	1	Proximal to CT	N.A.	N.A.	Yes	Idiopathic
Checa and Hussain (2011)[14]	Pennsylvania (USA)	CS	6	3	Proximal to CT	3B (case one) 3A (case two, three)	2 (case one) 1 (case two, three)	Yes	Idiopathic
Granec *et al.* (2012)[24]	Croatia	CR/LR	1	1	Proximal to CT	3A	1	Yes	Idiopathic

(Contd...)

Table 1: (*Continued*).

Authors and year	Country	Type	Number of cases total	Bifid median *n*	Location	Lanz classification	Al-Qattan C.	CTS	Etiology
Duymus *et al.* (2013)[20]	Turkey	CR	2	2	Proximal to CT	3A	1 (right wrist) 6 (left trifurcation)	Yes (bilateral)	N.A.
McClelland *et al.* (2012)[41]	Baltimore (USA)	CR	1	1	Proximal to CT	3B	5 (anomalous palmaris profundus)	N.A.	N.A.
Walker *et al.* (2013)[59]	North Carolina (USA)	RS	1026	8.6%	Proximal to CT	N.A.	N.A.	N.A.	N.A.
Bagatur *et al.* (2013)[7]	Turkey	CS	6	4	Proximal to CT	3B	2	Yes	idiopathic
Kasius *et al.* (2014)[32]	Netherlands	CS	518 hands with CTS 108 hands	47 in CTS (15.8%) 10 in volunteers	N.A.	N.A.	N.A.	Yes (47)	idiopathic
De Franco *et al.* (2014)[16]	Italy	CS	2	2	Proximal to CT	3A	5 (reversed palmaris longus)	Yes	Idiopathic
Duymus *et al.* (2014)[19]	Turkey	CS	194 hands with CTS 73 hands	22 in CTS (11.3%) 15 in volunteers	Proximal to CT	N.A.	N.A.	Yes (22)	Idiopathic
Ibrahim *et al.* (2015)[28]	Japan	CR	1	1	Proximal to CT	3B	2	N.A.	N.A.
Depaoli *et al.* (2015)[18]	Switzerland	CR	1	1	Proximal to CT	3A	6 (neurinoma)	N.A.	N.A.
Karaahmet *et al.* (2016)[31]	Turkey	CR	1	1	Proximal to CT	3B	2	N.A.	N.A.
Ariyo and Shea (2016)[5]	Philadelphia (USA)	CR	1	1	Proximal to CT	3B	2	No	N.A.
Chen *et al.* (2017)[15]	China	Preliminary study	160	15 (9.4%)	Proximal to CT	N.A.	N.A.	Yes (3)	Idiopathic
Cartwright *et al.* (2017)[13]	NC (USA)	RS	20	4	Proximal to CT	N.A.	N.A.	No	N.A.
Negm *et al.* (2017)[44]	Egypt	CR	1	1	Proximal to CT	3B	2	Yes	N.A.
Petrover *et al.* (2017)[46]	France	AS	30	4	Proximal to CT	3A	1	N.A.	N.A.
Bale *et al.* (2018)[8]	Oregon (USA)	CR	1	1	Proximal to CT	3A	1	N.A.	N.A.
Bhat *et al.* (2018)[12]	India	CR	1	1	Proximal to CT	3A	1	Yes (bilateral)	Idiopathic

(Contd...)

Table 1: (*Continued*).

Authors and year	Country	Type	Number of cases total	Bifid median *n*	Location	Lanz classification	Al-Qattan C.	CTS	Etiology
Trachani *et al.* (2018)[57]	Greece	CS	100 hands with CTS 112 hands	19% in CTS 13,3% in volunteers	Proximal to CT	N.A.	N.A.	Yes (19)	Idiopathic
Kostoris *et al.* (2019)[38]	Italy	CR/LR	1	1	Proximal to CT	3A	5 (anomalous palmaris profundus)	No	Idiopathic
Shinagawa *et al.* (2019)[52]	Japan	Observational study	349	59 (16,9%)	Proximal to CT	N.A.	N.A.	No	Idiopathic
Present	Italy	CR/LR	1	1	Within CT	3A	1	Yes	Idiopathic

CTS: Carpal tunnel syndrome, N.A.: Not available, BMN: Bifid median nerve. *Type of article* CS: Case series, CR: Case report, LR: Literature review, RS: Radiological study, AS: Anatomical study. *Lanz classification* 0: Extraligamentous thenar branch (standard anatomy), 1: Thenar branch variations in relation to the TCL, 1A: Subligamentous, 1B: Transligamentous, 1C: Ulnarward, 1D: Supraligamentous, 2: Distal accessory thenar branches, 3: High division of the median nerve, 3A: Without an artery, 3B: With artery, 3C: With lumbrical muscle, 4: Proximal accessory thenar branches, 4A: Running directly in the thenar muscle, 4B: Joining another branch. *Al-Qattan classification* 1: BMN without persistent median vessels or abnormalities, 2: BMN with persistent median vessels without any pathology, 3: BMN with persistent median vessels with aneurysm, thrombosis or arteriovenous malformation, 4: BMN with each divisions passing separately in the carpal tunnel, 5: BMN with aberrant muscle between its two divisions, 6: BMN with aberrant branches

of this anatomical variation and the pathogenesis of CTS.[23] Moreover, vascular pathology related to the presence of BMN has been described in literature, such as the presence of a PMA, which gave in one case an arteriovenous malformation of the volar forearm, and a CTS caused by its thrombosis.[25,33] Other anomalies have been also correlated to the presence of BMN such as multiple motor branches and accessory muscles.[37,40]

DISCUSSION

A rare case of Lanz IIIA BMN Type at the wrist has been encountered in a patient with a CTS and systematic review and practical considerations have been presented with the aim of raising awareness to the neurosurgical community of such scenario that could be encountered during CTR procedures.

The median nerve originates from the spinal roots of the brachial plexus at the level of C5–T1, bringing both motor and sensory fibers. At the level of the wrist, it is the only nervous structure to run through the carpal tunnel, deep to the TCL, then it divides into medial and lateral branches. In most individuals, the median nerve is observed as a singular nerve structure proximal to and within the carpal tunnel,[42] however, the BMN is an anatomic variation which has been described in literature since 1977 by Lanz as Type III, occasionally divided either proximal to or within the tunnel.[39,42] Although, according to Al-Qattan classification, the I and II subgroups are usually asymptomatic and do not produce CTS. Our case corresponds to one of those rare exceptions, because it belongs to the I subgroup, presenting BMN without persistent vessels, but the patient was symptomatic at the corresponding wrist.[1]

At present, surgeons frequently rely only on clinical examination and nerve conduction studies to diagnose CTS, but they may require further tools to preferentially choose the tailored surgical approach in each patient. The use of both MRI and US to identify median nerve anomalies has been also reported.[22]

MRI with standard morphological sequences can be used to determine many peripheral nerve disorders; in particular, for the patients with CTS, it can reveal morphological changes, such as enlarged median nerve, nerve flattening, increased signal intensity of the nerve, and flexor retinaculum bowing.[36]

However, the sensitivity and specificity of conventional morphological MRI sequences are low.[4]

US at high frequencies easily recognizes the nerves as fascicular hypoechoic structures and also detects their corresponding relationship with vessels, such as the PMA. Its use as an adjunctive tool to study the cross-sectional

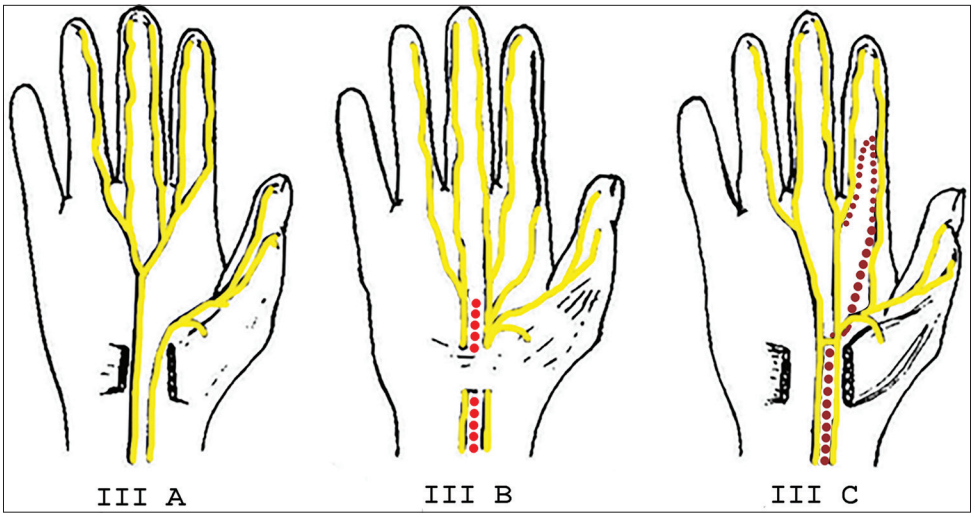

Figure 5: Lanz classification of high division of the median nerve anatomical variants. IIIA Type includes the bifid median nerve (yellow) without an artery or muscle; IIIB Type with a persistent median artery (red); and IIIC Type with a lumbrical muscle (brown).

area of the median nerve at the carpal tunnel is also appropriate to diagnose and grade the CTS. Some studies have expressed 99% of sensitivity and 100% of specificity for US to identify CTS with the additional advantage over the neurophysiological studies by showing the anatomy of the canal and its contents, as both BMN and PMA, that are common anatomical variants often associated with other median nerve anomalies.[10] In their presence, the ultrasound screening of patients before CTR surgery can help to identify those ones at increased risk of iatrogenic nerve damage and it can also allow for better surgical planning. In the present case, no iatrogenic nerve damage is expected since the bifurcation is proximal, but in rare cases, the bifurcation may occur within the carpal tunnel.[39,42] Although US can preoperatively determine the occurrence of such rare anatomical variation, its utility and indication to the surgical strategy should be verified in *ad hoc* studies.

Furthermore, the use of diffusion tensor imaging (DTI) has been considered for the efficacy in the diagnosis and management of CTS, resulting to be an important imaging technique which enables visualization and characterization of the median nerve, although the quality of images depends on field homogeneity, coil, as well as the gradient systems used, making it technically challenging.[58]

CTR surgery can be currently performed either with the endoscopic approach or with the conventional open approach, which might be recommended in those cases with the presence of BMN, or other structural abnormalities, to allow a proper view of the corresponding anatomical structures, preferring the access from the ulnar side to be able to cut under clear vision throughout the procedure.[39]

Despite a significant correlation between the presence of BMN and the pathogenesis of CTS has not been ascertained in the literature, however, when it is present, the risk of developing the compression of median nerve at the carpal tunnel increases, also because it is chaperoned by PMA in almost half of the reported cases.

CONCLUSION

CTS may be caused by the entrapment of a BMN Lanz IIIA Type anatomical variant of median nerve. Preoperative US would help to identify those patients presenting with anatomical variants at increased risk of iatrogenic injuries to better plan the surgery. Therefore, US use should be encouraged as pre-operative routine work-up in CTR surgery.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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