# Supplementary file 1. Details on search strategy and process.

Supplementary table S1. Databases with corresponding search string.

Database	Search string
PubMed	(Sjögren's syndrome) OR Sjogren's syndrome [MeSH Terms]) AND Capillaroscopy) OR capillaroscopy [MeSH Terms])
Web of Science	ALL=(Sjögren Syndrome) AND ALL=(capillaroscopy)
Embase	('Sjoegren syndrome'/exp OR 'sjoegren syndrome') AND ('capillaroscopy'/exp OR 'capillaroscopy') AND [humans]/lim AND [em base]/lim

ALL: all fields; MeSH: Medical Subject Headings; exp: explosion; lim: limited.

Supplementary table S2. Inclusion and exclusion criteria for title, abstract and full text selection.

Inclusion criteria	Exclusion criteria
Sjögren Syndrome	Other diseases (e.g. rheumatoid arthritis, systemic lupus erythematosus,) or healthy subjects
Assessment of the microcirculation	Assessment of the macrocirculation
Structural assessment (i.e. qualitative, [semi-] quantitative) of the microcirculation at the nailfold	Functional assessment of the microcirculation (e.g. thermography, laser techniques)
Capillaroscopy as outcome measure	Other outcome measure such as fingers wounds, ulcers, etc
Original research	Other studies such as reviews, opinions, conference papers, etc
	Case reports or series under five patients
	Absence of abstract / full text
English written language	Non-English language

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## Supplementary file 2. Study design and methodology.

Supplementary table S3. Details on study design, methodology and population of the included nailfold capillaroscopy studies in SS.

Author, study design	Sample size	Classification criteria	Sex (M/F)	Age (years)	RP/ACA	Tool, magnification	N° of fingers/site
Ohtsuka <i>et al</i> . 1997 [1], case series	22	Daniels and Talal criteria [2]	0/22	52.1	NR	Microscope + videography	3 <sup>rd</sup> fingers
Tektonidou et al. 1999 [3], case-control	40	1993 Diagnostic Criteria from the European Community for pSS [4]	3/37	55	14 RP- 16 RP+ 10 ACA+	Stereomicroscope	4th and 5th fingers (capillary density), other parameters on 10 fingers
Nagy et al. 2004 [5], cross-sectional	7	NR	0/7	47.3	NR	Stereomicroscope, 100X	NR
Capobianco et al. 2005 [6], case-control study	61	1993 Diagnostic Criteria from the European Community for pSS [4]	2/59	49.2	31 RP- 30 RP+ 1 ACA+	Stereomicroscope, 6.5-65X	8 fingers (all except thumbs)
Aguiar et al. 2006 [7], case-control	15	2001 Manthorpe criteria [8]	1/14	45.1	7 RP- 8 RP+	Microscope + videograph, 310X	4th left finger
Szabo <i>et al.</i> 2008 [9], case-control	30	2002 AECG criteria [10]	1/29	57.2	14 RP- 16 RP+	NVC	4 <sup>th</sup> finger of non-dominant hand
Riccieri <i>et al</i> . 2009 [11], case series	66	2002 AECG criteria [10]	1/65	54.2	40 RP- 26 RP+	NVC	NR
Pavlov-Dolijanovic et al. 2012 [12], prospective cohort study	102	2002 AECG criteria [10]	NR	NR	102 RP+	Stereomicroscope, 16X-100X	8 fingers (all except thumbs)
Baldini et al. 2013 [13], case series	41	2002 AECG criteria [10]	0/41*	45	0 RP- 41 RP+/ ACA+	NR	NR
Çakmakçı Karadoğan et al. 2015 [14], cross-sectional	18	NR	2/16	56	18 RP-	NVC, 16-100X	8 fingers (all except thumbs)
Corominas et al. 2015 [15], cross-sectional	136	2002 AECG criteria [10]	6/144**	58	92 RP- 44 RP+	NVC, 200X	8 fingers (all except thumbs)
Shenavandeh <i>et al.</i> 2019 [16], cross-sectional	14	2016 ACR/EULAR criteria [17]	1/13	NR	NR	Stereomicroscope, 250X	NR
Van Roon et al. 2019, cross-sectional [18]	30	2002 AECG criteria [10]	NR	NR	NR	Stereomicroscope, 180X	3 <sup>rd</sup> and 4 <sup>th</sup> fingers of both hands
Bernardino et al. 2020, cross-sectional [19]	15	2016 ACR/EULAR criteria [17]	0/15	49.5	1 RP- 14 RP+	NVC, 200X	8 fingers (all except thumbs)

ACA: anticentromere antibodies; ACR/EULAR: American College of Rheumatology/European League Against Rheumatism; AECG: American and European Consensus Group criteria 2002; NVC: nailfold videocapillaroscopy; NR: not reported; pSS: primary Sjögren's syndrome RP: Raynaud's phenomenon; SS: Sjögren's syndrome.

Only the studies in italic were considered for general conclusion-making (quality check score  $\geq 5/7$ ).

<sup>\*</sup> All patients had overlap syndrome of SS and (early) systemic sclerosis (ACA+) \*\*A total of 150 SS patients underwent NC, 136 of which were diagnosed of SS according to the 2002 criteria of the American-European Consensus Group.

**Supplementary table S4.** Quality assessment of the included nailfold capillaroscopy studies.

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Total score
Ohtsuka <i>et al</i> . 1997 [1]	0.5	1	1	0	0	1	0	3.5
Tektonidou et al. 1999 [3]	1	1	1	0.5	1	1	1	6.5
Nagy et al. 2004 [5]	0	0	1	0	1	0	0	2
Capobianco et al. 2005 [6]	1	1	1	1	1	1	1	7
Aguiar et al. 2006 [7]	1	1	1	0	1	0.5	0	4.5
Szabo et al. 2008 [9]	1	0	1	0	1	1	0	4
Riccieri et al. 2009 [11]	1	0	0	0.5	1	1	1	4.5
Pavlov-Dolijanovic et al. 2012 [12]	0	0	1	1	1	1	1	5
Baldini et al. 2013 [13]	1	1	0	0	1	1	1	5
Çakmakçı Karadoğan et al. 2015 [14]	0	1	1	1	1	1	1	6
Corominas et al. 2015 [15]	1	0	1	1	1	1	0	5
Shenavandeh et al. 2019 [16]	1	0	1	0	1	1	0	4
Van Roon et al. 2019 [18]	0.5	0	1	0	1	1	1	4.5
Bernardino et al. 2020 [19]	1	0	1	1	1	1	1	6
Total score of all studies per quality criterion	10	6	12	6	13	12.	5	8

Only the studies in italic were considered for general conclusion-making (Quality check score  $\geq$ 5/7).

Q1: Was the target population well described (fulfilment to defined classification criteria, disease duration, disease activity)?; Q2: Was a (healthy or not) control group present?; Q3: Was the used tool well described, including the used magnification, and adequate to evaluate the capillaroscopic characteristics?; Q4: Has the capillaroscopy been performed in a standardized way, being the inclusion of 8 fingers (all except for the thumbs)?; Q5: Was a clear description given of the capillaroscopic characteristics?; Q6: Was the correct statistics used (continuous vs dichotomous/ordinal/nominal scores)?; Q7: Otherwise good methods (no other flaws in the methodology)?

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## Supplementary file 3. Tables with overview of nailfold capillaroscopic findings in SS patients.

Supplementary table S1. Summary on density in SS patients.

Author	Sample size	Description	Resul	t in SS p	oatients	Result in HC		Significance	e
Tektonidou et al. 1999	40 SS (14 RP-, 16 RP+, 10 ACA +),	Mean capillary density*	RP- 9.8±1.5	RP+ 8.4±2.0	ACA + 7.2±1.6	10.5±1.1	RP- p=0.09	RP+ p<0.00001	ACA+ p<0.00001
	40 HC	Avascularity*	vascularity* 0 ND NI		ND	ND		ND	
Capobianco et al. 2005	61 SS (30 RP+, 31 RP-),	Deletion score	0.0 (0.0-0.3)		0.0 (0.0-0.0)		p<0.001		
	21 HC	(median)**	RP+ 0.1 (0.0-0	.4)	<i>RP</i> - 0.0 (0.0-0.1)	0.0 (0.0-0.0)		0.050 ( <i>p</i> =0.0) were consider	09 if only dered vs HC)
Çakmakçı Karadoğan et al. 2015	8 SS-ILD/ 10 SS (RP-)	Mean capillary density§		SS-ILD SS 9.75 (9-12) 11.2 (9-13		NA	NA)		
		Avascularity§	0 0						
Bernardino et al. 2020	15 SS (14 RP+, 1 RP-)	Mean capillary density <sup>^</sup>	8.8		NA		NA		

ACA+: positivity for anticentromere autoantibodies; HC: healthy controls; NA: not applicable; ND: not defined; RP-: absence of Raynaud's phenomenon; RP+: presence of Raynaud's phenomenon; SS: Sjögren's syndrome; SS-ILD: SS associated interstitial lung disease.

#### Supplementary table S2. Summary on dimension in SS patients.

Author	Sample size	e size Description Result in SS patients		tients	Result in HC	Significance		
Tektonidou et al. 1999	40 SS (14 RP-, 16 RP+,	Presence of enlarged	RP-	RP+	ACA+			
	10 ACA+), 40 HC	capillaries*	0%	12.5%	80%	ND	ND	
Capobianco et al. 2005	61 SS (30 RP+, 31 RP-),	Mean number of ectasias**		0.9		0.5	p=0.301	
	21 HC	Mean number of megacapillaries**		0.0		0.0	p=0.107	
		Presence of megacapillaries**		11.5%		0%	p=0.182	
Çakmakçı Karadoğan et al.	8 SS-ILD/ 10 SS (RP-)	Presence of ectasia§	SS-ILD SS					
2015			25% 0%		NA	NA		
		Megacapillaries§		0		0	NA	
Corominas et al. 2015	136 SS (44 RP+, 92 RP-)	Presence of dilated capillaries <sup>ç</sup>	RP+	I	RP-			
			40%			NA	p=0.008 in RP+ vs. RP- patients	
		Presence of giants 9	ND	]	ND	NA	NA	
Bernardino et al. 2020	rnardino et al. 2020 15 SS (14 RP+, 1 RP-) Presence of enlarged capillaries <sup>6</sup>		73.3%			NA	NA	
		Presence of giant capillaries <sup>^</sup>	33.3%					

ACA+: positivity for anticentromere autoantibodies; HC: healthy controls; NA: not applicable; ND: not defined; RP-: absence of Raynaud's phenomenon; RP+: presence of Raynaud's phenomenon; SS: Sjögren's syndrome; SS-ILD: SS associated interstitial lung disease.

<sup>\*</sup>The classification was made according to Maricq with some modifications. The capillary density was estimated as the mean number of capillary loops/mm in the distal capillary row of the fourth and fifth finger on both hands (1, 2).

<sup>\*\*</sup>Deletion score according to Lee's scale (3): for each digit assessed in 10x, a deletion degree from 0 to 3 is established according to Lee's scale (0- absence of capillary deletion, 1- one or two discrete deletion areas, 2- more than two discrete deletion areas, 3- confluent areas of capillary deletion). Deletion area is defined as the loss of more than two successive capillaries in the distal row of the periungual region. The degrees assigned to each digit are added up and divided by the number of digits assessed, resulting in a deletion score > 0.3 was deemed abnormal.

<sup>§</sup> Avascular areas were defined by referring to Lambova et al. as a distance between two adjacent capillary loops from the distal rows greater than 0.5 mm or greater than 0.3 mm in the proximal area (4).

<sup>^</sup> Density was calculated as the mean capillary number in the distal row by mm<sup>2</sup>.

<sup>\*</sup>The nailfold capillaries were classified as normal, borderline, definitely enlarged and extremely enlarged, based on the descriptions of Maricq *et al.* (1, 2). Of note, the description of enlarged capillaries in this study is prone to interpretation bias as no specification was given on exact dimensions.

<sup>\*\*</sup>Ectasias were defined as capillaries about 4x larger than the normal calibre; megacapillaries were defined as capillaries about 10x larger than normal ones (1). The number of each of the capillary characteristics represents the total count divided by the number of digits examined.

<sup>§ &</sup>quot;Enlarged capillary" or "ectasia" is used when the magnitude of a capillary is wider than 0,05 μm. Though this definition seems not correct and should be interpret with caution. No definition was given for megacapillaries.

 $<sup>\</sup>varsigma$  Dilated capillaries: 20–50  $\mu m;$  Megacapillaries: >50  $\mu m.$ 

<sup>^</sup> Enlarged capillaries: >30 μm; Giant capillaries: >50 μm.

## Supplementary table S3. Summary on morphology in SS patients.

Author	Sample size	Descr	ription	Result	in SS p	atients	Result in HC	Significance		ce		
Tektonidou et al. 1999	40 SS (14 RP-,	Normal*	Crossing	RP-	RP+	ACA+	12.5%	RP-	RP+	ACA+		
	16 RP+, 10 ACA+),			21.4 %	43.7%	10%		p>0.05	p<0.05	p>0.05		
	40 HC		Tortuous	ous 35.7 % 37.5% 10% 32.5%		p>0.05						
		Abnormal*	Bizarre	0	6.2%	10%	0		p>0.05			
Capobianco et al. 2005	61 SS (30 RP+, 31 RP-), 21 HC	Abnormal**	Median number of meandering		0.5		0.3		p=0.200			
			Median number of bushy	0.0			p=0.556					
			Median number of bizarre		0.0		0.0		p=0.404			
Çakmakçı Karadoğan	8 SS-ILD /10 SS (RP-)		Normal	SS-I	LD	SS	NA		ND			
et al. 2015				N	D	ND						
		Abnormal§	Multiple crossing	62.:	5%	60%	NA					
			Meandering	25	%	0%						
			Bizarre	37.5	5%	20%						
			Bushy	12.:	5%	0%						
Bernardino et al. 2020	15 SS (14 RP+, 1 RP-)	Normal <sup>^</sup>	Crossing		100%		NA NA		NA			
			Tortuous		100% 27%							
		Abnormal <sup>^</sup>	Meandering				27%		27%			
			Neoangiogenesis		0%							

ACA+: positivity for anticentromere autoantibodies; HC: healthy controls; NA: not applicable; ND: not defined; RP-: absence of Raynaud's phenomenon; RP+: presence of Raynaud's phenomenon; SS: Sjögren's syndrome; SS-ILD: SS associated interstitial lung disease.

<sup>\*</sup>Abnormal morphology, including bushy capillaries and bizarre capillaries according to Andrade *et al.* (1, 2). Tortuous and crossings were considered as variations of a normal morphology such as described by Andrade *et al.* (1, 2) and as proposed by the EULAR Study Group on Microcirculation in Rheumatic Diseases (5-7).

<sup>\*\*</sup>Abnormal morphology, including meandering capillaries (novel shaped), bushy capillaries (resembling arboreal formations), bizarre capillaries (those not fitting any particular description) according to Andrade *et al.* (1, 2). The number of each of the capillary characteristics represents the total count divided by the number of digits examined. The median of all subjects was reported.

<sup>§</sup>Abnormal morphology included bushy (Loops with limbs that originate from multiple small buds), meandering (limbs upon themselves or that cross another several times), bizarre (capillaries with striking atypical morphology, not conforming to the previously defined categories) according to Andrade *et al.* (1, 2). Tortuosity is reached with the presence of 2 or more cross-capillaries in a 1 mm area. Of note, none of the patients were suffering from Raynaud's phenomenon

<sup>^</sup>Normal (tortuositas and crossing) and abnormal (branched and meandering) were interpreted as consented by the EULAR Study Group on Microcirculation in Rheumatic Diseases (5-7).

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Supplementary table S4. Summary on haemorrhages in SS patients.

Author	Sample size	Description	Resul	lt in SS pa	atients	Result in HC	Significance				
Tektonidou et al. 1999	40 SS (14 RP-,	Haemorrhages1*	RP-	RP+	ACA +	10 %	RP-	RP+	ACA+		
	16 RP+, 10 ACA+),		7.1%	25%	10%			p>0.05			
	40 HC	Haemorrhages2*	7.1%	31.2%	50%	0	p >0.05	p<0.05	p<0.05		
		Pericapillary haemorrhages*	0	18.7%	40%	0	p >0.05	p<0.05	p<0.05		
		Thrombotic capillaries*	0	6.2%	20%	0	p >0.05	p>0.05	p<0.05		
Capobianco et al. 2005	61 SS (30 RP+,	Mean number of haemorrhages**		0.0		0.0		0.0		p>0.05	
	31 RP-), 21 HC	Presence of haemorrhages**		36%		19%		p>0.05			
Çakmakçı Karadoğan	8 SS-ILD/10 SS (RP-)	Micro-haemorrhages§	SS-	S-ILD SS		NA		ND			
et al. 2015			12	.5%	0%						
Corominas et al. 2015	136 SS (44 RP+, 92 RP-)	Pericapillary haemorrhages	RI	P+	RP-	NA		ND			
			15	1%	% 10%		10%				
		Thrombosis	13	1%	7%						
Bernardino et al. 2020	15 SS (14 RP+, 1 RP-)	Haemorrhages <sup>^</sup>	46.67%		NA	NA					

ACA+: positivity for anticentromere autoantibodies; HC: healthy controls; NA: not applicable; ND: not defined; RP-: absence of Raynaud's phenomenon; RP+: presence of Raynaud's phenomenon; SS: Sjögren's syndrome; SS-ILD: SS associated interstitial lung disease.

### **Supplementary table S5.** Summary of the semi-quantitative assessment in SS patients.

Author	Sample size	Description	Result in SS patients		Result in HC	Significance
Çakmakçı Karadoğan et al. 2015	8 SS-ILD/10 SS	Normal*	SS-ILD	SS	NA	p>0.05
			0%	40%		(SS vs. SS-ILD)
		Minor abnormalities*	37.5%	40%		
		Major abnormalities*	62.5%	20%		
Bernardino et al. 2020	15 SS (14 RP+, 1 RP-)	NFC-score**	1.67	NA	NA	

NA: not applicable; NFC-score: nailfold capillaroscopic score; RP-: absence of Raynaud's phenomenon; RP+: presence of Raynaud's phenomenon; SS: Sjögren's syndrome; SS-ILD: SS associated interstitial lung disease.

Of note, the term tortuosity was not conform to the EULAR Study Group on Microcirculation in Rheumatic Diseases consented definition (5-7). The authors defined tortuosity as the presence of two or more cross-capillaries in a 1mm area.

\*\* NFC-score was calculated as follows: each capillaroscopic finding rated from 0-3 according to variations in density, dimension, morphology or haemorrhages. Score 1: normal density, 10% longer capillaries, 50% of "morphological changes", absence of haemorrhages; Score 2: normal or decreased density, 10% of elongated capillaries, 50% of "morphological changes", presence of haemorrhages; Score 3: decreased density, 10% of elongated capillaries, 75% of "morphological changes", presence of haemorrhages.

Of note, the term "morphological change" was not defined, as such it remains unclear if variation of normal, such as 'tortuous' and 'crossed' capillaries should be considered as a "morphological change".

## References

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<sup>\*</sup>Pathological haemorrhages were defined as "more than two punctate haemorrhages per finger" (haemorrhages2), "confluent areas" of haemorrhages or "pericapillary" haemorrhages. This in contrast with non-pathological haemorrhages which were defined as "less than two punctate haemorrhages per finger" (haemorrhages 1).

<sup>\*\*</sup>Haemorrhages were counted in each digit, summed and divided by the number of fingers evaluated. The mean of all patients was reported.

<sup>§</sup>Micro-haemorrhages were defined as the presence of 2 or more bleeding areas in at least 2 fingers.

<sup>^</sup>No specified description/definition of haemorrhages was given.

<sup>\*</sup>The overall capillaroscopic patterns were defined as follows: • Normal: 7-10 capillaries/mm, normal morphology (hairpin-shaped loops but absence of normal variations such as "tortuous", "crosses"), normal distribution (arranged in parallel rows), absence of haemorrhages, absence of abnormal morphology ("neoangiogenesis" aspect); • "Minor abnormalities": 7-10 capillaries/mm, less than 50% "tortuous" loops (a type of normal morphology), arranged in parallel rows, absence of haemorrhages, absence of abnormal morphology ("neoangiogenesis"); • "Major abnormalities": decreased capillary density, more than 50% "tortuous" loops (a type of normal morphology), dilated capillaries ("enlarged"-no clear definition), and/or disarranged loops, with haemorrhages; more than 50% abnormal morphology ("neoangiogenesis"), dilated capillaries ("enlarged"-no clear definition), and/or haemorrhages.

**Supplementary table S6.** Summary of the qualitative assessment in SS patients.

Author	Sample size	Description	Result in SS patients			Result in HC	Significance	
Tektonidou et al. 1999	40 SS (14 without RP, 16	Normal pattern*	RP-	RP+	ACA +	67.5%	p>0.05	
	with RP, 10 ACA +), 40 HC			57.1%	12.5%	0		
		Non-specific abnormalities	Non-specific abnormalities*		65.8%	40%	32.5%	p>0.05
		Scleroderma pattern*		0	12.5%	80%	0	p>0.05
Capobianco et al. 2005	61 SS (30 RP+, 31 RP-),	Normal pattern**		59%		85.7%	ND	
	21 HC	Non-specific abnormalities*	**		29.5%	14.3%		
		Scleroderma pattern**		11.5	% (6RP+,	0%		
Pavlov-Dolijanovic et al. 2012	vlov-Dolijanovic <i>et al.</i> 2012 102 SS (RP+) Normal pattern^				84 (82%)	NA	NA	
		Non-specific abnormalities^		7 (7%)				
		Scleroderma pattern^		11 (11%)				
Baldini et al. 2013	41 OS (RP+)	Non-specific abnormalities	0	32%			NA	p=0.05
		Scleroderma pattern°	Early	29%			NA	ND
			Active	18%			NA	p=0.03
			Late		21%		NA	ND
Corominas et al. 2015	136 SS (44 RP+, 92 RP-)	Normal pattern <sup>ç</sup>			41.1%		NA	NA
		Non-specific abnormalites	ç		27.2%	_		
	Scleroderma			10.2%		_		
Bernardino et al. 2020§	Bernardino <i>et al.</i> 2020 <sup>§</sup> 15 SS (14 RP+, 1 RP-)				0.0%		NA	NA
		Non-specific abnormalities	§		66.7%			
		Scleroderma pattern§		33.3%				

ACA+: positivity for anticentromere autoantibodies; HC: healthy controls; NA: not applicable; ND: not defined; NS: not significant; OS: overlap syndrome, indicating patients fulfilling both the criteria for "early" systemic sclerosis (8) and SS (9); RP-: absence of Raynaud's phenomenon; RP+: presence of Raynaud's phenomenon; SS: Sjögren's syndrome; SS-ILD: SS associated interstitial lung disease.

\*Adapted from Maricq et al. 1976 the authors distinguished (10): 1) Normal pattern. 2) Non-specific findings: variations of normal morphology (tortuous and crossed), abnormal morphology ("bizarre") and haemorrhages (less than 2 punctuate haemorrhages per finger) 3) 'Other findings': more than 2 punctuate haemorrhages per finger, pericapillary haemorrhages and/or thrombotic capillaries. 4) Scleroderma pattern: giants ("definitely or extremely enlarged capillaries"), lowered density, abnormal shapes ("bushy") and haemorrhages ("capillary telangiectasias"). The authors sub-specify on an "active pattern", a "slow pattern" and an "overlap pattern", which we did not consider in this table as it is a non-standardised definition and we could not identify the abnormalities with commonly used definitions.

\*\*The scleroderma pattern was defined as the occurrence of avascular areas and the presence of enlarged capillaries, according to Maricq's description (11). Non-specific findings were poorly described, though included abnormal morphologies ("bushy", "bizarre" and "meandering).

^Normal pattern was defined as normal morphology (hair pin) or minor changes in dimension and distribution. "Non-specific abnormalities" were defined as normal and abnormal morphology ("meandering"-no definition given, and crossed-no definition given), focal haemorrhages, capillary thinning, capillary spasm, nonhomogeneous distribution/dimension (including width and length) of loops, prominent venous plexus. A scleroderma patterns was described according to Cutolo *et al.*'s definition (12). Of note ACA-status was unknown for most of the patients (n=81), though from the 21 with known ACA status 11 were ACA+ and had Raynaud.

°No specified definition of the qualitative assessment was given.

<sup>e</sup>The capillaroscopic patterns were defined as "normal" (no abnormalities are observed), non-specific abnormalities (when several capillaroscopic changes are observed although they do not fulfil the scleroderma pattern) and a scleroderma pattern according to Cutolo *et al.* (13).

§The capillaroscopic patterns were defined as "normal" (no abnormalities are observed), non-specific abnormalities (when several capillaroscopic changes are observed although they do not fulfil the scleroderma pattern) and a scleroderma pattern according to Cutolo *et al.* (13).

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