Isolated Lateral Chronic Exertional Compartment Syndrome of the Leg

A New Entity?

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Background: Chronic exertional compartment syndrome (CECS) mostly occurs in the anterior or deep posterior compartments (ant-CECS and dp-CECS, respectively) of the leg. It is generally accepted that CECS of the third or lateral compartment (lat-CECS) always occurs together with ant-CECS. However, whether exertional leg pain (ELP) can be caused by an isolated form of lat-CECS is unknown.

Purpose: To determine the existence of isolated lat-CECS and study whether history taking and a physical examination aid in discriminating between different subtypes of CECS.

Study Design: Case series; Level of evidence, 4.

Methods: Patients were eligible for this single-center study, conducted between January 2013 and February 2018, if they reported anterolateral ELP and completed a questionnaire scoring the frequency and intensity of pain, tightness, cramps, muscle weakness, and paresthesia during rest and exercise. They were asked to mark areas of altered foot skin sensation, if present, on a drawing. All patients underwent a dynamic intracompartmental pressure (ICP) measurement of the anterior and lateral compartments simultaneously. The diagnosis of CECS was confirmed by elevated ICP (Pedowitz criteria). There were 3 patient groups: (1) isolated ant-CECS with elevated ICP in the anterior compartment and normal ICP in the lateral compartment, (2) isolated Iat-CECS with elevated ICP in the lateral compartment but normal ICP in the anterior compartment, and (3) ant-/lat-CECS with elevated ICP in both the anterior and lateral compartments.

Results: A total of 73 patients with anterolateral ELP fulfilled study criteria (isolated ant-CECS: n = 26; isolated lat-CECS: n = 5; ant-/lat-CECS: n = 42). Group differences were not observed regarding age (isolated ant-CECS: median, 26 years [range, 13-68 years]; isolated lat-CECS: median, 20 years [range, 17-63 years]; ant-/lat-CECS: median, 28 years [range, 17-57 years]; χ^2 (2) = 0.466; P = .79), sex (isolated ant-CECS: 50% male; isolated lat-CECS: 40% male; ant-/lat-CECS: 62% male; P = .49), or bilateral symptoms (isolated ant-CECS: 54%; isolated lat-CECS: 80%; ant-/lat-CECS: 69%; P = .40). However, cramps at rest were present in a portion of the patients with isolated ant-CECS (38%) and ant-/lat-CECS (57%) but not in those with isolated lat-CECS (P = .032). Patient drawings of altered foot skin sensation did not contribute to the diagnosis (P = .92). ICP values after provocation were all lower in patients with isolated ant-CECS and isolated lat-CECS compared with those with ant-/lat-CECS (P < .05).

Conclusion: Seven percent of patients with CECS and anterolateral ELP who had symptoms due to isolated lat-CECS in the presence of normal muscle pressure in the anterior compartment.

Keywords: CECS; chronic exertional compartment syndrome; lateral compartment; peroneal compartment

The differential diagnosis of exertional leg pain (ELP) includes a range of entities such as medial tibial stress syndrome, stress fractures, neurovascular entrapment, and chronic exertional compartment syndrome (CECS).¹¹ The pathophysiology of CECS is poorly understood, but crucial

is elevated intracompartmental pressure (ICP) that may cause exercise-induced pain, tightness, and cramps.⁹ In over 90% of patients with CECS, tibialis anterior or deep posterior (flexor) muscle compartments are affected (ant-CECS and dp-CECS, respectively). An early study found that 1 in 7 patients who were evaluated for ELP had ant-CECS of the leg.¹⁰

Ant-CECS or dp-CECS each has a characteristic patient history and physical examination and are considered

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2 distinct syndromes.^{4,17} Ant-CECS or dp-CECS usually present in an isolated form, although combinations of both do occur. The status of the third or lateral (peroneal) compartment is unclear. Some are convinced that lat-CECS will only occur in combination with ant-CECS.^{5,13} Others have suggested that lat-CECS can occasionally present independent from ant-CECS.^{2,8,15} ICP of the tibialis anterior muscle is routinely measured when a patient with anterolateral ELP is suspected of CECS. ICP of the lateral compartment, however, is often not obtained simultaneously.

With this study, we aimed to determine whether isolated lat-CECS can exist in patients with anterolateral ELP. Moreover, we wanted to determine whether history taking and a physical examination aid in discriminating between different subtypes of CECS. If so, tailored management in patients with CECS may be justified.

METHODS

One focus of our department of sports medicine is diagnosing the causes of ELP. Yearly, approximately 500 patients with ELP, the vast majority (>95%) engaged in sport activities, are evaluated, and over 200 of these undergo dynamic ICP measurements.

This study included patients who were analyzed between January 2013 and February 2018 in a Dutch center of referral. Inclusion criteria were a history and physical examination suggesting CECS of the anterolateral leg, an ICP measurement performed in both the anterior and lateral compartments simultaneously, and complete data sets of questionnaires and patient drawings. Patient history was consistent with anterolateral CECS if both the anterior and lateral compartments were subjectively painful or felt tense during exercise or some hours thereafter. During the physical examination, patients often reported discomfort after deep palpation of the tibialis anterior or peroneal muscle. Patients with ELP and deep flexor CECS, peripheral neurovascular disease, a history of lower leg trauma, recurrent CECS, or previous leg surgery were excluded.

All procedures in the current study complied with the Declaration of Helsinki and its amendments. The rules laid down in the Medical Research Involving Human Subjects Act did not apply to this research, as confirmed by the local medical ethical review board.

Symptoms Questionnaire

Patients were asked in the 15-minute waiting period before an ICP measurement to complete a questionnaire investigating the severity and frequency of pain, tightness, cramps, muscle weakness, and paresthesia using a 5-point verbal rating scale. These verbal rating scale scores of symptom severity and frequency were converted into a numerical scale to allow for statistical analyses. Severity was quantified as nonexistent (1 point), mild (2 points), moderate (3 points), severe (4 points), and very severe (5 points). Frequency was expressed as never (1 point), sometimes (2 points), half of the time (3 points), most of the time (4 points), and all the time (5 points). A severity score was multiplied by a frequency score for each symptom to calculate a symptom score. For instance, when a patient experiences severe pain (4 points) half of the time (3 points), the symptom score for pain in this patient is $4 \times 3 = 12$ points. A symptom score was calculated for all 5 symptoms associated with CECS (pain, tightness, cramps, muscle weakness, and paresthesia). These 5 scores were summed to create an aggregate total symptom score, representing an approximation of the total discomfort that is experienced by a patient because of CECS (minimal: 5 points [never any of the 5 symptoms]; maximal: 125 points [always very severe discomfort due to all of the 5 symptoms]).

Pain Drawings

It is our experience that some patients with ELP spontaneously report altered foot skin sensation, possibly reflecting compromised leg peripheral nerves in the presence of CECS.⁵ A recent study indicated that patient pain drawings may offer a potential instrument contributing to identifying the cause of ELP.¹² Therefore, when patients experienced altered foot skin sensation and/or tingling, they were asked to mark this area on a drawing (Figure 1).

Dynamic ICP of 2 Compartments Simultaneously

The diagnostic gold standard of CECS is currently provided by a dynamic ICP measurement using the Pedowitz cutoff points (ICP \geq 15 mm Hg at rest, \geq 30 mm Hg after 1 minute, or \geq 20 mm Hg at 5 minutes after exercise).³ Before a provocative treadmill test, 2 slit catheters (Indwelling Slit Catheter Set; Stryker) were inserted in the most symptomatic leg: 1 in the anterior compartment and 1 in the lateral compartment. Both slit catheters were alternatingly connected to an arterial line manometer and a display (pressure monitor device 783547; Hewlett-Packard). In both compartments, ICP was measured at rest and after a provocative treadmill running test with a speed of 8 km/h and an 8% inclination. Running was aborted if patients experienced familiar leg symptoms. They were then asked to lie

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Figure 1. Areas of the foot and their associated nerves: (1) saphenous nerve, (2) deep peroneal nerve, (3) superficial peroneal nerve, (4) medial plantar nerve, (5) lateral plantar nerve, (6) calcaneal nerve, and (7) sural nerve.

down on the stretcher, and ICP was again measured in both compartments immediately after cessation and 1 and 5 minutes thereafter. Line flushing was always performed between the 2 measurements. Specifics on the dynamic ICP measurement procedure of each of the 2 compartments have been published previously.^{3,15}

3 Groups of Anterolateral CECS

Study patients were grouped on the basis of ICP results. The first ELP group (ant-CECS) had elevated ICP in the anterior compartment and normal ICP in the lateral compartment, the second group (lat-CECS) had elevated ICP in the lateral compartment and normal pressure in the anterior compartment, and the third group (ant-/lat-CECS) had elevated ICP in both the anterior and lateral compartments.

Statistical Analysis

Statistical analysis was performed using SPSS Statistics for Windows (version 25; IBM). The Shapiro-Wilk test was used to assess the normality of distribution of continuous data. When distributed normally, data were expressed as mean \pm standard deviation. If data were not distributed normally, they were expressed as median and range. The Kruskal-Wallis H test determined possible group differences in age and the (total) symptom score. The percentage of patients reporting altered foot skin sensation suggesting superficial or deep peroneal nerve compression (possibly reflecting CECS of the lateral or anterior compartment, respectively) was compared with the percentage of patients reporting altered skin sensation corresponding to other peripheral nerves (sural, saphenous, and tibial) using the Fisher exact test. This test was also used to determine possible group differences in sex, bilaterality, and presence of symptoms. Repeated-measures analysis of variance was used to assess potential interactions between ICP in the anterior and lateral compartments and the number of affected compartments at the different ICP measurement points (ie, ICP in patients with isolated ant- or lat-CECS compared with patients with ant-/lat-CECS). Because the assumption of sphericity was violated, we used the Greenhouse-Geisser correction. The Mann-Whitney U test was used to determine if there were between-group differences in ICP at rest as well as immediately, 1 minute, and 5 minutes after exercise. Statistical significance was set at the P < .05 level.

RESULTS

During the 5-year period of observation, 747 patients underwent ICP measurements for possible CECS. A total of 73 patients fulfilled study inclusion criteria (isolated ant-CECS: n = 26; isolated lat-CECS: n = 5; ant-/lat-CECS: n = 42). The other 674 patients did not meet study criteria for a variety of reasons (tendon and/or muscle injury or not specified: 36%; elevated ICP in the anterior or lateral compartment but not in both compartments: 25%; elevated ICP in the deep posterior compartment: 20%; medial tibial stress syndrome: 14%; nerve injury: 4%; vascular injury: 1%).

ICP at 5 minutes after exercise was above the Pedowitz cutoff point ($\geq 20 \text{ mm Hg}$ at 5 minutes after exercise) in all patients with isolated lat-CECS. In contrast, just 40% of these patients had elevated ICP beyond the cutoff point at 1 minute after exercise ($\geq 30 \text{ mm Hg}$) (Figure 2).

Group differences were not observed regarding age (isolated ant-CECS: median, 26 years [range, 13-68 years]; isolated lat-CECS: median, 20 years [range, 17-63 years]; ant-/lat-CECS: median, 28 years [range, 17-57 years]; χ^2 (2) = 0.466; P = .79), sex (isolated ant-CECS: 50% male; isolated lat-CECS: 40% male; ant-/lat-CECS: 62% male; P = .49), or bilaterality (isolated ant-CECS: 54%; isolated lat-CECS: 80%; ant-/lat-CECS: 69%; P = .40).

Symptoms are depicted in Table 1. During exercise, all patients reported pain, and almost all (72/73; 99%) experienced tightness. Contrary to current beliefs, the majority also reported pain and tightness at rest. Surprisingly, half of the patients also experienced symptoms during the night. However, none of the patients with isolated lat-CECS had cramps at rest, while 38% of the patients with isolated ant-CECS and 57% of the patients with ant-/lat-CECS did (P = .032).

One-third of all patients (25/73) reported alterations in foot skin sensation in areas that may suggest compression of the deep or superficial peroneal nerve due to pathologically elevated ICP in the anterior or lateral compartment. In contrast, almost 66% (48/73) felt sensation only in skin areas that are not innervated by the deep or superficial peroneal nerve (Table 2). These findings suggest that the patient-reported location of neurological disorders may not aid in diagnosing anterolateral CECS.

There was a statistically significant interaction between ICP and the number of affected compartments (isolated vs combined CECS) in both the anterior and lateral compartments (F(3,168) = 11.235; P < .001; partial $\eta^2 = 0.167$; $\varepsilon = 0.609$ and F(3,111) = 3.497; P = .048; partial $\eta^2 = 0.086$; $\varepsilon = 0.517$, respectively). ICP after exercise was significantly higher in patients with ant-/lat-CECS than in patients with isolated ant-CECS immediately and 1 and



Figure 2. Percentage of patients with isolated lateral chronic exertional compartment syndrome (lat-CECS; n = 5), isolated anterior CECS (ant-CECS; n = 26), and anterior and lateral CECS (ant-/lat-CECS; n = 42) with positive or negative intracompartmental pressure (ICP) according to the Pedowitz criteria at rest and 1 and 5 minutes after exercise.

	Ant-CECS $(n = 26)$	Lat-CECS $(n = 5)$	Ant-/Lat-CECS $(n = 42)$	P Value
Symptom at rest, n (%)				
Pain	22 (85)	2 (40)	34 (81)	.103
Tightness	21 (81)	5 (100)	37 (88)	.549
Muscle weakness	12 (46)	1 (20)	14 (33)	.437
Cramps	10 (38)	0 (0)	24 (57)	$.032^{b}$
Paresthesia	12 (46)	2 (40)	25 (60)	.461
Symptom score at rest, m	edian (range)			
Pain	12 (1-25)	4 (1-6)	4 (1-20)	.057
Tightness	9 (1-25)	9 (6-12)	12 (1-25)	.538
Muscle weakness	2 (1-16)	1 (1-6)	1 (1-25)	.377
Cramps	1 (1-16)	1 (1-1)	4 (1-16)	.082
Paresthesia	2 (1-16)	1 (1-9)	4 (1-20)	.715
Total	34 (5-85)	18 (13-24)	29 (5-89)	.408
Symptom during exertion	, n (%)			
Pain	26 (100)	5 (100)	42 (100)	>.999
Tightness	25 (96)	5 (100)	42 (100)	.425
Muscle weakness	16 (62)	3 (60)	25 (60)	>.999
Cramps	14 (54)	2 (40)	29 (69)	.275
Paresthesia	15 (58)	3 (60)	31 (74)	.360
Symptom score during ex	ertion, median (range)			
Pain	20 (9-25)	20 (9-25)	20 (6-25)	.799
Tightness	20 (1-25)	16 (16-25)	16 (8-25)	.962
Muscle weakness	8 (1-25)	12 (1-25)	4 (1-25)	.332
Cramps	6 (1-25)	1 (1-25)	5 (1-25)	.820
Paresthesia	14 (1-20)	4 (1-25)	4 (1-25)	.601
Total	63 (16-110)	57 (43-125)	57 (18-94)	.564
Symptom at night, n (%)				
Pain	17 (65)	3 (60)	23 (55)	.804
Tightness	16 (62)	3 (60)	28 (67)	.928
Cramps	13 (50)	1 (20)	22 (52)	.496
Paresthesia	11 (42)	2 (40)	20 (48)	.932

TABLE 1 Symptoms and Symptom Scores During Rest, During Exertion, and at Night^a

^aSee Methods section for calculation of the symptom score (minimum, 5; maximum, 125). Ant-CECS, isolated anterior chronic exertional compartment syndrome; ant-/lat-CECS, anterior and lateral chronic exertional compartment syndrome; lat-CECS, isolated lateral chronic exertional compartment syndrome.

 ${}^{b}P < .05.$

 TABLE 2

 Patients Reporting Altered Skin Sensation in Areas of the Foot^a

		$\begin{array}{c} Lat\text{-}CECS\\ (n=5) \end{array}$		Р
Paresthesia				
1. Saphenous nerve	7	1	7	
2. Deep peroneal nerve	4	1	6	
3. Superficial peroneal nerve	4	0	10	
4. Medial plantar nerve	5	2	7	
5. Lateral plantar nerve	4	0	6	
6. Calcaneal nerve	3	0	4	
7. Sural nerve	6	0	6	
Paresthesia corresponding to superficial or deep peroneal nerve				
Yes (2 and/or 3)	8	1	16	
No (1, 4, 5, and/or 6 or not)	18	4	26	

^aData are reported as No. Ant-CECS, isolated anterior chronic exertional compartment syndrome; ant-/lat-CECS, anterior and lateral chronic exertional compartment syndrome; lat-CECS, isolated lateral chronic exertional compartment syndrome.

5 minutes after exercise (U = 752, z = 3.635, P < .001; U = 778, z = 3.994, P < .001; and U = 685, z = 4.216, P < .001, respectively) and in patients with isolated lat-CECS 1 minute after exercise (U = 122; z = 1.975; P = .048) (Figure 3).

DISCUSSION

If CECS is present in the leg, it is mostly ant-CECS or dp-CECS.³ However, sporadic reports suggest that the lateral (peroneal) compartment can also develop CECS (lat-CECS).^{7,8} Ant-CECS and dp-CECS are clearly separate entities with characteristic features, but the status of lat-CECS is unclear. Interestingly, 7% (5/73) in our population of patients with CECS and anterolateral ELP was found to have such isolated lat-CECS. This percentage is probably an underestimation, as it is currently not a standard procedure to perform a dynamic ICP measurement of both the anterior and lateral compartments simultaneously in patients with ELP with anterolateral complaints.

A distinction between CECS of the tibialis anterior and peroneal muscles is seldom made in anterolateral ELP, but instead, the somewhat generic term of "anterolateral CECS" is preferred.¹³ If the type of CECS is not reported in a study, one assumes that the study participants have tibialis anterior muscle compartment syndrome.⁶ Most researchers are convinced that lat-CECS, if present, is obligatorily combined with ant-CECS.^{5,13} Others consider lat-CECS as an entity that is possibly separate from ant-CECS.^{7,8,15} The present study suggests that a small portion of patients with anterolateral ELP indeed suffer from isolated lat-CECS. We advise that subtypes of CECS are standardly reported in future studies, allowing for a better comparison of management and treatment results.

Certain clues in history taking and a physical examination point toward the presence of a certain type of leg CECS. For instance, pain halfway up the calf combined with tender deep palpation of distal deep flexor muscles is associated with dp-CECS.¹⁷ In contrast, differentiating between isolated anterior and isolated lateral CECS based on a combination of history taking and a physical examination is difficult, if not impossible. Although group size is limited, the present study demonstrates that symptoms in patients with isolated ant-CECS, isolated lat-CECS, or both (ant-/lat-CECS) are rather alike. History taking and a physical examination may therefore not allow for a discrimination of these anterolateral subtypes. Therefore, a



Figure 3. Median intracompartmental pressure (ICP) at rest, immediately, and 1 and 5 minutes after provocative exercise in the (A) anterior compartment and (B) lateral compartement in patients with anterior and lateral chronic exertional compartment syndrome (ant-/lat-CECS), isolated anterior CECS (ant-CECS), or isolated lateral CECS (lat-CECS). Error bars represent standard error of the mean. *Statistically significantly different: P < .05.

simultaneous dynamic ICP measurement of both the anterior and lateral compartments is justified in patients with ELP having anterolateral pain and tightness that are potentially caused by CECS. Future research should establish whether these 3 subtypes require a tailored treatment.

As history taking and a physical examination probably do not aid in diagnosing each of the 3 types of anterolateral CECS, we explored the use of simple tools such as patientreported alterations in skin sensation of the foot. A recent study indicated that patient pain drawings can be a valuable instrument in diagnosing various causes of ELP.¹² Patients with high muscle pressure in the anterior or lateral compartment occasionally experience awkward foot sensations in the dermatome of the deep peroneal nerve (web space between first and second toes) or the superficial peroneal nerve (dorsum of foot), respectively.⁵ Our patients were asked to indicate areas of altered skin sensation of the foot, if present, on a drawing. Approximately one-third of our patients reported alterations in skin sensation, possibly reflecting compression of either the deep or superficial peroneal nerve. In contrast, almost 66% reported altered sensation in territories of nerves that are located outside of the anterolateral compartments such as branches of the tibial nerve or saphenous or sural nerve (Table 2). Therefore, patient drawings of altered skin sensation of the foot did not contribute to diagnosing the type of anterolateral CECS.

A dynamic ICP measurement is still the gold standard diagnostic tool in CECS. ^{1,14} As some are convinced that the anterior and lateral compartments are always simultaneously affected in anterolateral CECS, they advise fasciotomy of both compartments in recalcitrant patients, even if elevated ICP is demonstrated in the anterior compartment only.^{5,13} The presence of elevated muscle pressure of both compartments would support their approach. The present study challenges their method, as just 58% (42/73) of all patients with CECS reporting anterolateral ELP who underwent ICP measurements of both the anterior and lateral compartments had elevated pressure in both. Therefore, it is likely that pressure in the anterior and lateral compartments may not always be related. Interestingly, the present study also found that ICP values immediately after a treadmill run were consistently higher in patients with CECS in both compartments compared with patients with either isolated ant-CECS or isolated lat-CECS. The clinical relevance of these significantly higher ICP values is yet to be determined. However, 1 study¹⁶ suggested that surgery was more successful in patients with higher ICP.

The present study has limitations, including a restricted number of patients. Moreover, it is not standard procedure in our department to measure both the anterior and lateral compartments of patients with anterolateral ELP, potentially introducing selection bias. In addition, patient selection is based on dynamic ICP measurements, a diagnostic tool that is under debate but still preferentially used in the absence of a generally accepted alternative.^{1,14} A correlation of the ICP measurement patterns with results of compartment release would have strengthened the conclusion that isolated lateral compartment syndrome exists. Finally, we used a symptom score that is not yet validated, which makes an analysis of the symptoms even more challenging.

In conclusion, this study is the first to demonstrate that 7% of a general population with CECS in the area of the anterolateral leg has symptoms due to elevated muscle pressure in the lateral compartment only. A physical examination and patient-reported symptoms are not always helpful in differentiating between lat-CECS and ant-CECS. Future research should establish whether these patients require individualized treatment.

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