Contents lists available at ScienceDirect

Clinical Neurophysiology Practice

journal homepage: www.elsevier.com/locate/cnp

Case report

Clinical utility of contact heat evoked potentials (CHEPs) in a case of mentalis nerve lesion



^a EMG and Motor Control Unit, Hospital Clínic, University of Barcelona, Barcelona, Spain ^b Maxillofacial Unit, Plastic and Maxillofacial Surgery Department, Hospital Clínic, Barcelona, Spain

ARTICLE INFO	ABSTRACT
Article history: Received 29 August 2017 Received in revised form 21 March 2018 Accepted 25 March 2018 Available online 7 April 2018	<i>Objective:</i> Nociceptive evoked potentials are still infrequently used in electrodiagnostic studies of single patients. We report a case in which the results of contact heat evoked potentials (CHEPs) provided unique information for the diagnosis. <i>Methods:</i> After biopsy for a local cementoma, a 21-year-old woman presented with neuropathic pain in the distribution of her left mentalis nerve. A CT scan showed a well circumscribed lesion near the mentalis nerve
<i>Keywords:</i> Contact heat evoked potentials Facial pain Brainstem reflexes Small fiber neuropathy	groove. We examined brainstem reflexes and evoked potentials conveyed through the mentalis nerve. <i>Results:</i> Blink reflex responses recorded from the orbicularis oculi, jaw jerk and masseteric silent period recorded from the masseter muscles and long latency evoked potentials recorded from Cz to electrical stim- ulation of the mentalis nerve were all within normal values, with no differences between sides. However, CHEPs, recorded from Cz to thermoalgesic stimulation of the left mentalis area were decreased to approx- imately 1/3 their size in comparison to stimulation to the unaffected side.
	<i>Conclusion:</i> While the patient reported symptoms and had neuroimaging signs of mentalis neuropathy, the sole electrophysiological abnormality identified was that of CHEPs, which specifically test small, unmyeli- nated fibers.
	Significance: Nociceptive evoked potentials can provide unique information on damage of small nerve fibers in specific cases.
	© 2018 International Federation of Clinical Neurophysiology. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

Mononeuropathies solely affecting small fibers are difficult to confirm using conventional clinical neurophysiology, which generally assesses large myelinated fibers.

While evidence of nerve lesion supported by conventional nerve conduction studies are usually considered indubitable, it is infrequent to base the evidence of a lesion in results from the study of small fibers. Contact heat evoked potentials (CHEPS) can be useful in documenting possible nociceptive pathways conduction disturbances in patients with neuropathic pain. We present a clinical case demonstrating the utility of CHEPs for the evaluation of a focal lesion in the mentalis nerve that could not be otherwise documented.

Case report:

The patient was a 21-year-old woman with signs of neuropathic pain in her left chin. Five years earlier, she had been diagnosed

with an asymptomatic cementoma between the dental roots of the first and second left bicuspid teeth. Under the suspicion of growth, she was recommended to have a biopsy of the lesion and a piece of bony material was extracted in an apparently uneventful procedure. However, as soon as the anesthetics lost their effect, the patient began to feel numbness of the chin in her left side, combined with shots of lancinating pain. A computed tomography showed a region with loss of substance close to the cementoma and in close communication with the mentalis grove (Fig. 1). She was, then, referred for neurophysiological examination with the request to confirm the suspicion of iatrogenic left mentalis nerve damage.

When the patient was first seen in our department, she described pain of neuropathic characteristics in her chin, like stabbing but no numbness or decreased sensation. Severe tactile allodynia was present in the left side of her chin while other facial sites were normal. There were no abnormalities in other cranial nerves, with normal strength in facial and trigeminalinnervated muscles. We performed a neurophysiological testing of brainstem reflexes and trigeminal evoked potentials to characterize the dysfunction.

https://doi.org/10.1016/j.cnp.2018.03.004

2467-981X/© 2018 International Federation of Clinical Neurophysiology. Published by Elsevier B.V.

This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).





^{*} Corresponding author. E-mail address: jvalls@clinic.ub.es (J. Valls-Sole).



Fig. 1. Images of the cementoma and the biopsy-related lesion. The image in the left is an orthopantomography showing the cementoma before biopsy. A: Anterior; P: Posterior. The image on the right side of the figure is an axial TC showing the relationship between the biopsy lesion and the mental nerve grove.

2. Methods

2.1. Brainstem reflexes

We recorded the blink reflex from the orbicularis oculi, and the exteroceptive suppression of EMG activity during voluntary contraction from the masseter muscles, to mentalis nerve electrical stimulation to the affected and the unaffected sides, following standard methods (Aramideh et al., 2002). We measured onset latency and area (amplitude times duration) of the responses recorded in both sides.

2.2. Evoked potentials

We examined cerebral evoked potentials by recording epochs of stimulus time-locked EEG activity from Cz referenced to Fpz. The impedance of the recording was set below 5 kOhm before starting the recording. Two types of stimuli were applied: electrical and thermoalgesic. They were applied to two areas in each side: the mentalis region, where the patient referred her symptoms in the left side, and the infraorbital region, chosen as control site. Electrical stimuli were of 0.5 ms duration at intensity twice the sensory threshold. For thermal stimulation, we used a thermofoil thermode for contact heat stimulation (Pathway, Medoc, Israel) set to a rapid increase of temperature from 32 °C to 52 °C, at a speed of 70 °C/s. We analyzed the potentials evoked in Cz to repeated single stimuli, i.e., the long latency evoked potentials (LLEPs) for electrical stimuli and the contact heat evoked potentials (CHEPs) for thermoalgesic stimuli. We applied 10-12 stimuli of each type to the two sites in each side and made an off-line averaging of the responses after eliminating possible artifacts of blinking or electronic interference (Truini et al., 2007; Granovsky et al., 2016). On the averaged waveform of LLEPs and CHEPs, we measured peak latency (N2) and peak to peak amplitude (N2-P2) for each stimulation site.

Data were analyzed by taking into consideration reference values of our own department, published in pertinent literature (Valls-Solé, 2005; Valls-Solé and Deuschl, 2006; Cabib et al., 2014; Granovsky et al., 2016). Side-to-side maximum differences in healthy subjects were available for comparison for the blink reflex R2 latency (5.0 ms, respectively, according to Valls-Solé and Deuschl, 2006), and for the CHEPs N2 latency (88.7 ms) and N2/P2 amplitude (15.0 μ V), according to Granovsky et al. (2016). For the LLEPs, we simply compared latency and size of the responses between sides.

3. Results

Clinical exam was relevant for allodynia to superficial touch in the left side of the patient's chin. Pressure was better tolerated after the first unavoidable defensive reaction. Therefore, we were careful with the application of the stimulator to the affected region and waited a few seconds before applying the stimulus while maintaining steady the stimulator in place. The patient tolerated well the rest of the neurophysiological exam.

Data on blink reflex and exteroceptive suppression of masseter EMG activity are reported in Table 1. No abnormalities were observed in absolute values or between-sides differences. Results of recording the LLEPs and CHEPs are reported in Table 2. Latency and amplitude of the LLEPs to mentalis and infraorbital nerves were within normal limits, with no relevant side-to-side differences. In contrast, CHEPs were clearly asymmetric, showing reduced amplitude and delayed latency to left chin with respect to right chin stimulation (Fig. 2). Even though the N2/P2 amplitude value was still within normal limits for left side stimulation, the side-to side amplitude difference was larger than the maximum inter-side difference considered in healthy subjects (Granovsky et al., 2016).

4. Discussion

The case presented here shares clinical features with the wellknown numb-chin syndrome (Smith et al., 2015). Our patient had a clear iatrogenic cause of her symptoms, in relation to dental procedures, as in other reported cases (Elahi et al., 2014; Mishima et al., 2016; Brooks et al., 2017). Although numbness is the main symptom in many instances, allodynia may also be present, which develops slowly after an acute episode of pain (Elahi et al., 2014).

Table 1

Blink reflex and masseteric exteroceptive suppression responses in both sides.

	Right side stimulation		Left side stimulation	
	Right	Left	Right	Left
Orbicularis oculi R2	32.1	31.9	31.8	32.2
Masseter ES1	11.1	11.3	11.2	11.3
Masseter ES2	25.8	25.6	25.7	25.7

Data are latency given in ms of the R2 responses recorded in the orbicularis oculi, and of the two phases of exteroceptive suppression (ES1 and ES2) recorded in the masseter muscle, to mentalis nerve stimulation in both sides.

Table 2				
Results of	recording	LLEPS	and	CHEPs.

Stimulus type	Measurements	Cheek	Cheek		Chin	
		Right	Left	Right	Left	
Electrical (LLEPs)	Peak latency (ms)	122	125	120	118	
	Amplitude (µV)	57	54	48	45	
Thermal (CHEPs)	Peak latency (ms)	316	323	320	371	
	Amplitude (µV)	55	48	52	22*	





Fig. 2. Responses to mentalis nerve stimulation. On the left side of the figure there are blink reflex responses recorded in the orbicularis oculi (upper graphs), and exteroceptive suppression of the EMG activity recorded from the masseter muscles, to electrical mentalis nerve stimulation (lower graphs). On the right side of the figure there are CHEPs to left and right stimulation of the chin and cheek. Note the smaller CHEP to stimulation to the left side of the chin in comparison to stimulation of the right side or stimulation of right and left sides in the cheek.

Allodynia to touch suggests direct nerve injury and may mark a difference with respect to symptoms derived from nerve compression, which would be expected to express as numbness or hyposthesia, as it should mainly involve thick myelinated fibers. In direct nerve lesions, pain may result from direct axonal damage and subsequent ectopic discharges.

In our case, the clinical presentation and the results of electrophysiological tests indicated that the lesion occurred in small fibers. Indeed, we found preservation of normal conduction in medium size and large cutaneous afferent fibers (A β) mediating blink reflex responses. We think that A β fibers were also responsible for LLEPs, obtained in our patient with a conventional stimulation electrode. However, it must be taken into account that some forms of electrical stimulation, using specifically prepared electrodes, may activate small fibers, as shown by others (Katsarava et al., 2006; Üçeyler et al., 2013). In contrast, CHEPs were abnormally reduced in the affected side, beyond reference values for side-to-side differences (Granovsky et al., 2016). CHEPs are, indeed, comparable to laser stimuli in the electrophysiological diagnosis of small fiber polyneuropathy (Atherton et al. 2007; Casanova-Molla et al., 2011).

Our results indicate that CHEPs are a useful tool for the documentation of nerve damage in patients with neuropathic pain and suspected involvement of small fibers, as already shown by Truini et al. (2007). In fact, nerve damage rarely affects only small fibers and, even if no abnormalities were observed in our patient with cranial nerve testing or LLEPs, these results cannot be taken as an absolute indication of no damage to larger size mentalis nerve fibers. However, abnormalities may only be documented, as in our case, using tests for nociceptive pathways. We should, therefore, consider the use of CHEPs or other tools for the specific assessment of small fibers function, as an essential part of the armamentarium for neurophysiological testing in patients complaining of neuropathic pain.

Conflict of interest statement

All authors declare no conflict of interest.

References

Aramideh, M., Valls–Sole, J., Cruccu, G., Ongerboer de Visser, B.W., 2002. Cranial reflexes: electrodiagnostic technique, physiology, and normative data. In: Brown, W.F., Bolton, C.F., Aminoff, M.J. (Eds.), Neuromuscular Function and Disease. Basic, Clinical and Electrodiagnostic Aspects. WB Saunders, Philadelphia, pp. 443–453.

- Atherton, D.D., Facer, P., Roberts, K.M., Misra, V.P., Chizh, B.A., Bountra, C., et al., 2007. Use of the novel Contact Heat Evoked Potential Stimulator (CHEPS) for the assessment of small fibre neuropathy: correlations with skin flare responses and intra-epidermal nerve fibre counts. BMC Neurol. 7, 21.
- Brooks, J.K., Schwartz, K.G., Ro, A.S., Lin, C.D., 2017. Numb chin syndrome associated with vertical root fracture and odontogenic infection. Quintessence Int. 48, 225–230.
- Cabib, C., Llufriu, S., Martinez-Heras, E., Saiz, A., Valls-Solé, J., 2014. Abnormal control of orbicularis oculi reflex excitability in multiple sclerosis. PLoS One 9, e103897.
- Casanova-Molla, J., Grau-Junyent, J.M., Morales, M., Valls-Solé, J., 2011. On the relationship between nociceptive evoked potentials and intraepidermal nerve fiber density in painful sensory polyneuropathies. Pain 152, 410–418.
- Elahi, F., Manolitsis, N., Ranganath, Y.S., Reddy, C., 2014. Mental nerve neuropathy following dental extraction. Pain Physician. 17, E375–E380.
- Granovsky, Y., Anand, P., Nakae, A., Nascimento, O., Smith, B., Sprecher, E., et al., 2016. Normative data for Aδ contact heat evoked potentials in adult population: a multicenter study. Pain 157, 1156–1163.
- Katsarava, Z., Ayzenberg, I., Sack, F., Limmroth, V., Diener, H.C., Kaube, H., 2006. A novel method of eliciting pain-related potentials by transcutaneous electrical stimulation. Headache 46, 1511–1517.

- Mishima, S., Takahashi, K., Tomioka, T., Bessho, K., 2016. Numb chin syndrome as initial manifestation of bisphosphonate-related osteomyelitis of the jaw and methotrexate-associated lymphoproliferative disorders: a rare case. Br. J. Oral. Maxillofac. Surg. 54, 114–115.
- Smith, R.M., Hassan, A., Robertson, C.E., 2015. Numb chin syndrome. Curr. Pain. Headache Rep. 19, 44.
- Truini, A., Galeotti, F., Pennisi, E., House, F., Biasiotta, A., Cruccu, G., 2007. Trigeminal small-fiber function assessed with contact heat evoked potentials in humans. Pain 132, 102–107.
- Üçeyler, N., Kahn, A.K., Kramer, D., Zeller, D., Casanova-Molla, J., Wanner, C., et al., 2013. Impaired small fiber conduction in patients with Fabry disease: a neurophysiological case-control study. BMC Neurol. 13, 47.
- Valls-Solé, J., 2005. Neurophysiological assessment of trigeminal nerve reflexes in disorders of central and peripheral nervous system. Clin. Neurophysiol. 116, 2255–2265.
- Valls-Solé, J., Deuschl, G., 2006. Reflex responses, silent period and long latency reflexes. In: Kimura, J. (Ed.), Peripheral Nerve Diseases. Handbook of Clinical Neurophysiology. Elsevier, Amsterdam, pp. 237–262.