Case Report / Olgu Sunumu

Brachioradial Pruritus: Secondary to Cervical Ependymoma

Servikal Ependimoma Sekonder Gelişen Brakioradial Pruritus

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ABSTRACT

Brachioradial pruritus (BRP) is a localized neuropathic pruritus commonly affecting the dorsolateral aspect of the upper arm and forearm. It may also involve the shoulders, neck, and chest, and may extend across the upper back. A 58-year-old female presented herself to our clinic complaining of numbness, tingling, stinging, and itching with a burning sensation on her head, neck, shoulders, and upper limbs. The patient had suffered these symptoms for several years, which had intensified during the week prior to presentation. A biopsy was performed in the dermatology department for diagnosis. Cervical magnetic resonance imaging revealed an ependymoma.

Keywords: Pruritus, ependymoma, dermatitis

ÖZET

Brakioradial pruritus (BRP) klasik olarak kol ve ön kolun dorsolateral bölümünü etkileyen lokalize nöropatik pruritustur. Aynı zamanda omuz, boyun, göğüsü tutabilir ve sırta doğru yayılabilir. 58 yaşındaki kadın hasta boyun, baş, omuz ve kollarda yanıcı karakterde kaşınma, uyuşma, karıncalanma ve iğnelenme şikayetleriyle kliniğimize yatırıldı. Şikayetleri uzun yıllardır vardı ve son hafta daha da artmıştı. Teşhis amacıyla dermatoloji bölümü tarafından biyopsi yapıldı. Servikal manyetik rezonans görüntülemede epandimom saptandı.

Anahtar sözcükler: Pruritus, ependimom, dermatit

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Introduction

Brachioradial pruritus (BRP) is a localized neuropathic pruritus commonly affecting the dorsolateral aspect of the upper arm and forearm. It was considered for 20 years to be a photodermatosis before the neuropathic origin was identified. BRP may also involve the shoulders, neck, and chest, and it may extend across the upper back. In documented cases, BRP is more prevalent in fair-skinned people and in those aged 30–70 years. It is considered to be a neuropathic pruritus that can be worsened by UV irradiation but improved by extreme cold (ice-pack use). The pruritus may occur spontaneously, with intense itching and a sensation of burning, stinging, or pain at any time of the day or night, and it may be unilateral or bilateral, episodic or continuous. Even though BRP symptoms may be severe, a physical examination often proves benign (1). BRP was first reported in 1968 by Waisman (2), who termed it "solar pruritus" or "summer pruritus" of the elbows, describing its occurrence in patients in Florida who suffered from local itching of the skin on the dorsolateral aspect of the arm. BRP is a problematic condition attributed to either solar radiation or neuropathy, secondary to a cervical nerve root injury. It is often refractory to standard dermatological treatments.

However, successful treatment may be achieved using topical capsaicin, oral gabapentin, pregabalin, carbamazepine, lamotrigine, and amitriptyline; by physical treatment using cervical vertebra manipulation and acupuncture; or by a surgical approach involving a cervical rib, disc herniation, or spinal cord tumor (3–8).

Here, we present a patient who remained undiagnosed for several years because BRP was not considered. We aimed to take into consideration the patient's intractable pruritus. There may be numerous disorders underlying pruritus.

Case

A 58-year-old female presented to our clinic with a burning sensation, numbness, tingling, stinging, and itching of the head, neck, shoulders, and upper limbs. The patient had suffered these symptoms for several years, which had deteriorated during the week prior to presentation. Bilateral stinging of the hands had started at the thenar region approximately 15 years earlier, followed by a burning sensation, numbness, tingling, and itching of the forearm, arm, shoulders, and neck after 4–5 months. The patient had not experienced any trauma to the neck, and her family history was unremarkable. There was no history of prolonged sun exposure. The patient displayed dermatologic signs of intractable pruritus, which was unresponsive to oral or topical antihistamines and corticosteroids. The patient wore sleeveless dresses because the symptoms were intensified by dress contact. These symptoms were alleviated by cooling. A physical examination showed mild erythema and extensive atrophic plaques on both forearms, arms, shoulders, and the neck (Figure 1).

Hypoesthesia was determined on both upper extremities on neurological examination, but it was more severe on the right C6–8 dermatomes, with a reduced reflex of the right biceps. Previous magnetic resonance imaging (MRI) scans of the cervical spine showed a C4–T1 intradural intramedullary lesion. Cervical MRI was performed because of intermittent neck stiffness and pain. The most recent cervical MRI scans revealed an ependymoma (Figure 2). Periodic follow-up of the lesion by MRI was proposed. There was expansion of the central channel at the proximal side of the lesion, and hyperintense signal changes were observed. Electromyography demonstrated mild to middle chronic neurogenic involvement of the right C5–8 roots and innervated muscles together with mild chronic neurogenic involvement of the left C6–8 roots and innervated muscles. A punch biopsy of the right forearm to confirm the diagnosis was performed in the dermatology department, which proved consistent with the signs of chronic dermatitis. The results of blood tests, including an analysis of inflammatory markers, a full blood count, and fasting plasma glucose, renal, and liver function tests, were unremarkable. Pregabalin and physical therapy began to relieve the patient's symptoms.

Discussion

BRP is an intense itching sensation of the upper arms that usually appears between the shoulder and elbow of one or both arms. It is a problematic condition because of its disputed etiology; some authors propose that BRP is a photodermatosis, whereas others affirm that compression of the cervical nerve roots may trigger the occurrence of BRP lesions. Electromyography and nerve conduction studies may be considered in cases of suspected nerve root impingement. MRI of the spine is

Figure 1. Appearance of the patient's hand.

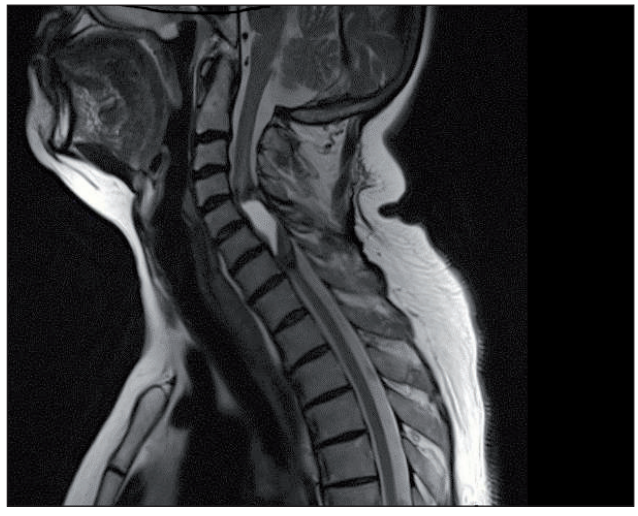

Figure 2. Image of the patient's cervical MRI.

recommended to locate a suspected nerve impingement, including in BRP and neuralgia paresthetica. Neuropathic itch is defined as an itch initiated or caused by a primary lesion or dysfunction at any point along the afferent pathway of the nervous system. Although it can be acute, most cases are chronic and persistent. Neuropathic itch is accompanied by sensory impairment experienced as paresthesia, hyperesthesia, or hypoesthesia in most cases; this can help the clinician diagnose the condition. It may also occur during the recovery period in isolated nerve injury, and may involve the peripheral and central sensitization of nerve fibers. This sensitization induces alloknesis, which is an itching phenomenon resulting from an innocuous stimulus that does not normally provoke itching. The characteristic feature of neuropathic itch that differentiates it from other forms of pruritus is an association with other sensory symptoms on a dermatome distribution and with other neurological sensory signs or neural injury, including motor and autonomic damage (9).

Neuropathic itch may be related to peripheral nervous system damage, such as in postherpetic neuropathy, BRP, and notalgia paresthetica. Goodkin et al. (10) detected cervical spine disorders in 11 of 22 patients with BRP from 1993–2000. They demonstrated cervical spine disorders in radiographs that could be correlated with the location of pruritus in each of these 11 patients, and in most of whom the disorder was prevalent between the C3 and C7 vertebrae. The changes included spondylosis, foraminal narrowing, spurs, and narrowing of the disk space and cervical ribs. BRP has been reported in association with ependymoma and secondary to cervical nerve compression (11). Heyl (12) reported that 4 of 14 patients demonstrated evidence of degenerative changes and osteoarthritis between C4 and C7. Marziniak et al. (13) reported that MRI revealed cervical spine changes in all of the patients examined; these changes led to nerve compression in 80.5% of the patients, and there were degenerative changes in 19.5% of the patients.

BRP caused by a spinal neoplasm is rare, but should be ruled out by cervical spine MRI. MRI changes were demonstrated in all of the BRP patients in several studies (13–17). Fisher (18) proposed that BRP is not primarily caused by the sun, rather it is related to cervical nerve root impingement, involving one or all of the C5–8 cervical nerve root segments. A case report described a patient in whom BRP was the presenting sign of a spinal-cord tumor compressing the spinal cord from C4–7 (18). Antipruritic therapies should target both local inflammation and spinal sensitization of itching processing. The treatment of BRP patients is difficult. Most authors agree that topical corticosteroids and oral antihistamines are of little, if any, value. Despite promising early reports, a capsaicin cream was no more effective than the placebo in a randomized controlled study (19,20). Interestingly, it was reported that the anticonvulsant gabapentin was effective in many patients. Anticonvulsants, including gabapentin, pregabalin carbamazepine, and lamotrigine, have demonstrated efficacy in case reports on the treatment of neuropathic itch (3–8).

The case presented here highlights the possibility that BRP can be initiated by an intramedullary tumor. Ependymoma normally seen in children, is thought to be associated with the presence of dysesthesia. Clinicians should consider BRP when the etiology for idiopathic pruritus is not detected and a rapid diagnosis is made by imaging examinations.

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http://www.textcheck.com/certificate/5lm7cl

References

1. Beuscher L, Reeves G. Brachioradial Pruritus: Approach to the patient with enigmatic forearm pruritus. The Journal for nurse practitioners 2012;8
2. Waisman M. Solar pruritus of the elbows (brachioradial summer pruritus). Arch Dermatol1968;98(5):481-4.
3. Kanitakis J. Brachioradial pruritus: Report of a new caseresponding to gabapentin. Eur J Dermatol 2006;16:311–2.
4. Winhoven SM, Coulson IH, Bottomley WW. Brachioradialpruritus: Response to treatment with gabapentin. Br J Dermatol 2004;150: 786–7.
5. Bueller HA, Bernhard JD, Dubroff LM. Gabapentin treatment for brachioradial pruritus. J EurAcadDermatolVenereol 1999;13: 227–30.
6. Tait CP, Grigg E, Quirk CJ. Brachioradial pruritus and cervical spine manipulation. Australas J Dermatol1998;39:168–70.
7. Rongioletti F. Pruritus as presenting sign of cervical rib. Lancet 1992;339:55.
8. Knight TE, Hayashi T. Solar (brachioradial) pruritusresponseto capsaicin cream. Int J Dermatol1994;33:206–9.
9. Yosipovitch G, Samuel SL. Neuropathic and psychogenic itch. DermatolTher 2008;21:32-41.
10. GoodkinR,Wingard E, Bernhard JD. Brachioradialpruritus: Cervical spine disease and neurogenic/neurogenic pruritus. J Am AcadDermatol 2003;48:521–4
11. Fleuret C, Dupre-Goetghebeur D, Person H et al. Brachioradial pruritus revealing an ependymoma. Ann DermatolVenereol 2009;136:435-7.

12. Heyl T. Brachioradial pruritus. Arch Dermatol.1983;119:115-6.

13. Marziniak M, Phan NQ, Siepmann D et al. Brachioradial pruritus as a result of cervical spinepathology: The results of a magnetic resonance tomography study.Journal of the American Academy ofDermatology 2011;65:756–62.

14. Schurmeyer-Horst F, Fischbach R, Nabavi D et al. Brachioradial pruritus: a rare, localized, neuropathicform of itching. Hautarzt 2006;57:523-7.

15. Mataix J, Silvestre JF, Climent JM, Pastor N, Lucas A. Brachioradial pruritus as a symptom of cervicalradiculopathy. ActasDermo-Sif 2008;99:719-22.

16. Binder A, Folster-Holst R, Sahan G et al. A case of neuropathic brachioradial pruritus caused bycervical disc herniation. Nat ClinPractNeurol 2008;4:338-42.

17. Kavak A, Dosoglu M. Can a spinal cord tumor cause brachioradial pruritus? J Am AcadDermatol2002;46:437-40.

18. Fisher DA. Brachioradial pruritus wanted: a sure cause (and cure) for brachioradial pruritus. Int J Dermatol 1997;36:817-8.

19. Knight TE, Hayashi T. Solar (brachioradial) pruritus–response to capsaicin cream. Int J Dermatol 1994;33:206-9.

20. Teixeira F, Miranda VA, Hojyo TT et al. Solar (brachioradial) pruritus—response to capsaicin cream [letter]. Int J Dermatol 1995;34:594-5.