biologic drugs to control psoriasis. Only 1 case has been reported, of a 44-year-old man with Crohn disease and recipient of a liver transplant due to cryptogenic cirrhosis, who was treated with ustekinumab to control the inflammatory bowel disease, with a good response and no adverse effects after 12 months of follow-up.[5]

The literature contains little evidence of the need to modify the immunosuppressant regimen in these patients when the biologic drug is introduced. Many of the reported cases do not mention any modification of the immunosuppressant treatment. In our case, we decided to suspend the mofetil mycophenolate and maintain the tacrolimus at 3 mg/12 h, together with the etanercept. Furthermore, anti-TNF drugs are being used off-label as immunosuppressants in recipients of solid-organ transplants.[6,7]

In conclusion, with this new case, we provide additional information on the use of biologic drugs in transplant patients. Biologic drugs may be used in this type of patient provided that the patients are carefully selected and closely observed for potential infections, although we believe that more cases and studies are needed to corroborate this.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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Trigeminal Trophic Syndrome Secondary to Meningioma Recurrence☆

Síndrome trófico trigeminal secundario a recidiva demeningioma

Dear Editor:

Trigeminal trophic syndrome (TTS), also known as trigeminal neurotrophic ulceration, is a rare condition that occurs when the trigeminal nerve is damaged. It presents with anesthesia or unilateral dysesthesia that typically affects the ala nasi, and with ulcerations that are self-inflicted, usually unconsciously, by the patient.[1]

A 38-year-old woman was examined for pruriginous erosive and mildly painful lesions that had appeared 2 months earlier in the left frontoparietal region. The patient had undergone surgery for a meningioma measuring 46 × 40 × 38 mm in the left cavernous sinus a year and a half earlier by means of frontotemporal cutaneous incision and left pterional craniotomy. Since the surgery, the patient had presented sequelae including paresis of cranial nerves III and IV in the left eye, neurotrophic keratitis in the left eye, and panhypopituitarism with secondary hypogonadism and hypothyroidism. The last follow-up imaging study performed a month before visiting our department and approximately a month after onset of the skin condition, showed a recurrence of the meningioma in the left cavernous sinus. The patient had been examined a month earlier at the ophthalmology department for an episode of herpes zoster that involved the V1 branch of the trigeminal nerve, with no ophthalmologic involvement. Without microbiologic confir-

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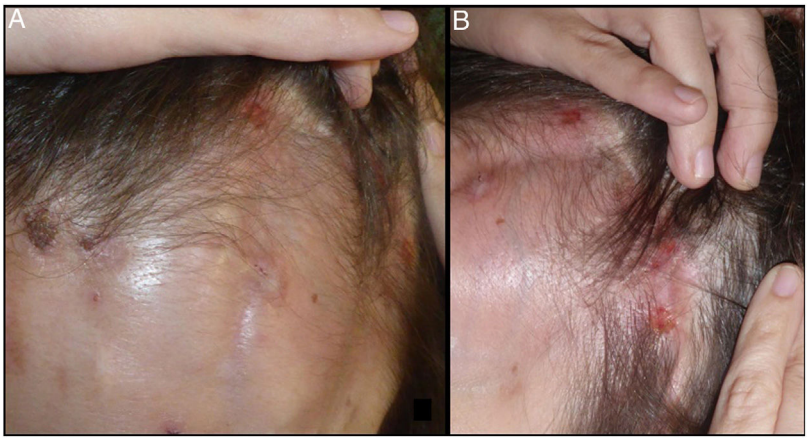

Figure 1 A and B, Crusted erosive lesions on an atrophic background in the region innervated by the V1 branch of the trigeminal nerve.

mation, she received treatment with valaciclovir and was sent to our department due to the persistence of the lesions when the treatment had been completed. When she visited our department, she presented several crusted erosive lesions of varying morphology on a background of hypersensitive atrophic skin compared to the contralateral region (Fig. 1). We reached a diagnosis of TTS, with ulcers secondary to dysesthesia and anesthesia in the left V1 region, probably linked to the recurrence of meningioma. Treatment was instated at that time with tacrolimus cream, 0.01% and covered with a hydrocolloid dressing. The patient was also offered the option of starting treatment with oral gabapentin, which se rejected. After 2 months of treatment, the lesions improved considerably.

TTS is characterized by the presence of unilateral facial ulcers caused by persistent scratching as a result of the dysesthesia (described as itching, or a burning or tingling sensation) and anesthesia in the sites innervated by the previously damaged trigeminal nerve or one of its branches. Patients often scratch to alleviate the feeling of discomfort and, due to the anesthesia, cause persistent ulceration. A systematic review of the literature in by Sawada et al. in 2014,[2] which included 61 cases of TTS published to that date, describes the principal etiologies of this syndrome. The most commonly reported causes are ablation of the trigeminal nerve (30%), cerebrovascular accident (30%), and surgical complications, including a history of craniotomy (21%). Another, less frequently reported cause is herpes zoster, which is also mentioned by other authors, such as Dolohanty et al.,[3] who describe development of TTS in the V1 region 2 months after an episode of herpes zoster in the same dermatome, which was treated with valaciclovir. In our case, we believe that the initial clinical presentation, diagnosed as herpes zoster, was probably the onset of TTS.

Furthermore, damage to the trigeminal nerve may occur in up to 11% of patients who undergo surgery for meningioma.[4] The first case of TTS associated with meningioma was described in 1982.[5] Three further publications link TTS and meningioma surgery, although in one of them, the TTS appeared after surgery for a recurrence of the meningioma 9 years after the primary tumor.[6] Thus, although our patient potentially presents 3 of the etiologic factors most frequently associated with development of TTS (meningioma, craniotomy and a doubtful episode of herpes zoster), we believe that the sequence of appearance of the symptoms and the coincidence with the recurrence of the meningioma make the meningioma the most probable cause.

In terms of treatment of TTS, improvement of cutaneous symptoms has been reported with tacrolimus cream, 0.01%, gabapentin, and hydrocolloid dressing.[7] Improvement of the lesions has also been observed after use of high doses of carbamazepine (200 mg/3 times daily).[8] Other drugs, such as amitriptyline, pregabalin, and alprazolam have been used with little or no clinical benefit.

Conflicts of interest

The authors declare that they have no conflicts of interest.

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Painful Cutaneous Syncytial Myoepithelioma: From Nonspecific Symptoms to Histopathologic Diagnosis☆

Mioepitelioma sincitial cutáneo doloroso: desde la clínica inespecífica al diagnóstico histopatológico

Dear Editor:

Tumors with myoepithelial differentiation include mixed tumor, myoepithelioma, and myoepithelial carcinoma. They tend to involve the salivary glands, but they may also be located in the sinonasal area, in the larynx and lungs, and on the skin. Skin tumors with myoepithelial differentiation include chondroid syringoma (mixed tumor), cutaneous myoepithelioma, malignant chondroid syringoma, and myoepithelial carcinoma.[1–3] The syncytial variety of cutaneous myoepithelioma has been described in recent years.[4]

A 33-year-old man with no relevant past personal history visited our department with a papule measuring 0.2 cm in diameter. The papule had appeared 2 years earlier on the right flank and was slightly erythematous and painful. It was not associated with signs of bleeding or growth. A clinical diagnosis of angioleiomyoma and angiolipoma was considered and an excision biopsy of the lesion was performed.

Histology revealed an elevated sessile lesion due to the presence of a solid, well circumscribed, nodular tumor with lobular edges, located on the superficial third of the reticular dermis (Fig. 1A). The lesion consisted of fusiform and epithelioid cells with an oval or rounded nucleus, with no significant atypia, variable quantities of cytoplasm, poorly defined edges, and practically no interstitial component (Figs. 1B-D). A maximum of 4 mitotic figures for every 10 fields at a magnification of 40×. No tumor necrosis was observed.

The cancerous population showed diffuse and generalized positive immune staining for S100 and EMA (Fig. 2) and was focal for smooth muscle actin (Figs. 3C and 3D) and caldesmon. No immune staining was observed for cytokeratins AE1-AE3 (Figs. 3A and 3B), Melan-A, desmin, glial fibrillary acidic protein, p63, or claudin-1.

An RT-PCR and EWSR1 rearrangement (EWSR1-POUF5F1, EWSR1-ZNF444 y EWSR1-PBX1) sequencing study was performed[5] and the results were negative.

Syncytial myoepithelioma is a rare tumor that presents clinically as a papule or nodule on the limbs of middle-aged men. Histologically, it is a solid tumor with fusiform cells or histiocytes with a pale eosinophilic cytoplasm and vesicular nucleus, with sparse stroma.[5,6]

It does not usually present mitosis, necrosis or lymphovascular invasion, but in rare cases, up to 4 mitotic figures per 10 fields at 40× have been reported.[7] The diagnostic criteria for cutaneous myoepithelial carcinoma are not well established, but tumors with marked cytologic atypia, a high mitotic index, and necrosis show more aggressive behavior, with increased probability of recurrence and distant metastasis.[5,7–9]

Fifty percent of cutaneous syncytial myoepitheliomas present rearrangement of the EWSR1 gene.[4,5,7]

Myoepithelial tumors usually express cytokeratins and the S100 protein. Myoepithelioma, however, presents positive immune staining for EMA and S100 protein, and most cases are negative for cytokeratins.[5]

The differential diagnosis includes epithelioid benign fibrous histiocytoma, juvenile xanthogranuloma, melanocytic lesions, and epithelioid sarcoma.[5,9]

Epithelioid benign fibrous histiocytoma presents as a dermal nodule of epithelioid cells, frequently with binucleation, positive for EMA, with a fibrovascular stroma. This lesion, however, does not present a syncytial architecture or positive immune staining for S100 protein, or for GFAP or p63, as is the case with syncytial myoepithelioma.[4,7,9]

In the early stages, juvenile xanthogranuloma presents as an exophytic lesion with eosinophilic histiocytes that does not usually present mononucleated or multinucleated lipidized cells (Touton cells).[9] However, it tends to affect children, immune staining is positive for CD163, CD68, and is negative for EMA and S100 proteins.[4]

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