

CLINICAL SCIENCE LETTERS

Widespread Idiopathic Comedones and Lichen Ruber Planus: Clinical and Histological Association

E. Rosón, C. Posada, C. de La Torre, and M. Cruces

Servicio de Dermatología, Complejo Hospitalario de Pontevedra, Pontevedra, Spain

To the Editor:

The presence of numerous acquired comedones on the trunk and limbs of an adult patient, with no other family member affected and with no triggering factors to explain their presence, requires us to rule out those conditions in which comedones are the predominant clinical feature. The fact that these lesions appear in association with areas of lichen ruber planus presents the dilemma of whether the lesions develop simultaneously by chance or whether there could be some pathogenic relationship. In the case we present below, we observed that, from a clinical point of view, the 2 types of lesion (comedones and lichen) were frequently closely related, with the comedones presenting a lichenoid halo, but they were sometimes separate. However, in all samples analyzed, histological study showed lichenoid infiltration of the comedones.

A 72-year-old woman came to our outpatient clinic for mildly pruritic lesions that had been present on the trunk and limbs for approximately 2 years. The patient had a history of hypertension and dislipidemia, for which she had been on treatment with felodipine and pravastatin for at least 7 years. There was no family history of similar skin lesions. On physical examination, we observed small, shiny, erythematous-violaceous plaques and papules with a smooth surface, suggestive of lichen ruber planus (Figure 1). In addition, numerous comedones were identified on the trunk, arms, and thighs; some of these had an erythematous halo of lichenoid inflammatory appearance (Figure 2). A whitish, reticulated pattern was observed on the mucosa of both cheeks. There were no alterations of the hair or nails. The patient was unable to say whether the 2 types of lesion appeared simultaneously or in succession, and there was no history of the use of comedogenic products.

We performed biopsies of the comedones, with the interesting finding of a lichenoid pattern around the infundibular microcysts of the comedones (Figure 3); this infiltrate was observed irrespective of whether there was a visible erythematous halo around the comedone or not. The biopsy from the violaceous plaques showed a linear lymphoid infiltrate in the superficial dermis and the presence of necrotic keratinocytes, also confirming the clinical impression of lichenoid lesions. There were no relevant pathological findings in the routine laboratory

tests and serological tests for viral hepatitis were negative. The patient was treated with oral isotretinoin (0.5 mg/kg) for 4 months, with resolution of the lichenoid lesions and a marked reduction in the number of comedones.

The presence of numerous comedones with a generalized distribution on the trunk and limbs, appearing in adult life, with no triggering factors and no family history of similar lesions is very rare. Recently, Zhang and Zhu¹ reported a similar case in a boy who, like our patient, presented disseminated idiopathic comedones. The histological



Figure 1. Lichenoid plaques on the patient's trunk. Occasional comedones can be seen between the plaques.



Figure 2. Although not always present, many comedones were surrounded by an erythematous halo of lichenoid appearance.

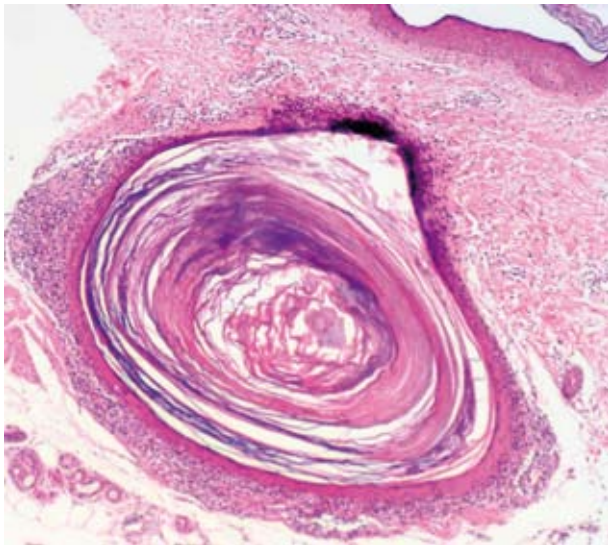


Figure 3. A lymphoid infiltrate with a clearly lichenoid pattern was observed around the infundibular microcysts of the comedones. Hematoxylin-eosin, $\times 10$.

findings, the distribution of the lesions, the age at presentation, and the absence of triggering factors led us to rule out similar conditions known at the present time, such as familial dyskeratotic comedones, comedo nevus, follicular keratosis, physically or chemically induced acne, and follicular mycosis fungoides.

Interestingly, our patient presented a lichenoid dermatitis of the lichen planus type; the cause was unknown as she did not report any associated systemic symptoms and the complementary tests performed were normal or negative. We cannot totally exclude a relationship between these lesions and the treatment that was being taken; in fact, 2 cases of lichenoid reaction related to pravastatin have been reported in the literature.^{2,3} However, in our case, this seems very unlikely due to the absence of any temporal relationship and due to the presence of lesions on the mucosas, which are uncommon in drug-induced lichenoid reactions.

Up to now, the relationship between lichen planus and comedones has been limited to cases of lichen planus follicularis tumidus with cysts and comedones, clinically characterized by edematous plaques with cysts and comedone-type lesions, usually in a retroauricular position.⁴⁻⁶ The involvement of the pilosebaceous unit in lichen planus is observed in cases of lichen planus follicularis, which typically affects the scalp, although and there has been 1 case report in which the trunk was affected.⁷ However, the clinical presentation in those

cases, so different from what is described here, makes us think that there could be no relationship. Lucke et al⁸ described the development of milia-like cysts during the course of lichen planus. Like those authors, we considered the possibility that the comedones formed due to inflammation or degeneration of the basal layer caused by the lichen; however this hypothesis appeared unlikely in our case, as many of the comedones were found in skin with no previous lichenoid lesion. Independently of the temporal relationship between the 2 types of lesion, we believe it is more likely that the lichenoid infiltration of the comedones was secondary, and it could even be a typical occurrence in lichen planus. In the literature, there have been sporadic reports of lichenoid infiltration of similar structures, such as of an epidermal cyst in a patient with lichen planus.⁹ Although the excision of benign lesions is not common practice in an inflammatory disorder, histological study of this type of lesion in patients with lichen planus could corroborate this hypothesis.

Correspondence:

Elena Rosón.
Servicio de Dermatología.
Hospital Provincial de Pontevedra.
C./ Loureiro Crespo, 2.
36001 Pontevedra Spain.
eroson64@yahoo.es

Conflicts of Interest

The authors declare no conflicts of interest.

References

1. Zhang RZ, Zhu W-Y. Idiopathic disseminated comedones in a child. *Pediatric Dermatol.* 2006;23:163-6.
2. Keough GC, Richardson TT, Grabski WJ. Pravastatin-induced lichenoid drug eruption. *Cutis.* 1998;61:98-100.
3. Pua VS, Scolyer RA, Barnetson RS. Pravastatin-induced lichenoid drug eruption. *Australas J Dermatol.* 2006;47:57-9.
4. Belaich S, Bonvalet D, Civatte J. Lichen planus folliculaire tumidus retroauriculaire. *Ann Dermatol Venerol.* 1977;104:147-50.
5. Rongioletti F, Ghigliotti G, Gambini C, Rebora A. Agminate lichen follicularis with cysts and comedones. *Br J Dermatol.* 1990;122:844-5.
6. Vazquez-García J, Pérez-Oliva N, Pereiro-Ferreirós MM, Toribio J. Lichen planus follicularis tumidus with cysts and comedones. *Clin Exp Dermatol.* 1992;17:346-8.
7. Baker K, Pehr K. Linear lichen planopilaris of the trunk: first report of a case. *J Cutan Med Surg.* 2006;10:136-8.
8. Lucke T, Fallowfield M, Burden D. Lichen planus associated with milia. *Clin Exp Dermatol.* 1999;24:266-9.
9. Bork K, Hoede N. Lichen planus: histologic involvement of an epidermoid cyst. *Dermatologica.* 1983;166:319-21.

A Case of Transient Rectangular Alopecia After Aneurysm Embolization

G.M. Garnacho, M.V. Amorrich, R. Salido, and J. Espejo

Servicio de Dermatología, Hospital Universitario Reina Sofía, Córdoba, Spain

To the Editor:

Endovascular procedures are now widespread in vascular interventional neuroradiology, but there are very few reports of their cutaneous adverse effects in the literature; such effects are typically poorly recognized and are underdiagnosed. Transient rectangular alopecia, considered to be a subtype of radiodermatitis, is a condition that has been known to dermatologists for only about 10 years, and scarcely 10 cases have been published.

We report the case of a 39-year-old man, with no past history of interest, who presented with oppressive, left frontal headaches that had started 2 months earlier, were refractory to multiple treatments, and were associated with ipsilateral, retro-ocular symptoms; the rest of the neurological examination was normal. Magnetic resonance imaging revealed a giant fusiform aneurysm of the middle cerebral artery (Figure 1), and it was therefore decided to perform embolization using a 3-dimensional Guglielmi detachable coil (platinum spiral) (Matrix2). The angiographic procedure was performed in 2 sessions separated by 5 days, taking a series of 12 digital subtraction images in the first session and 20 in the second. Total fluoroscopy time was of 90 to 100 minutes and the approximate total dose was of 3 Gy. The principal planes used were the posteroanterior and left-right oblique. On completion of the procedure, a clot developed around the spiral, blocking the lumen of the aneurysm (Figures 2 and 3). The patient's headaches improved, but 2 weeks later he came to the dermatology outpatient clinic for a rectangular, left parieto-occipital plaque of alopecia with no signs of inflammation (Figure 4). There were no exclamation mark hairs and the hair pull test was strongly positive. The condition progressed to complete alopecia in that area. The superficial arterial pulses were present bilaterally and the blood tests were normal. As the plaque was situated in the area of maximum radiation, we related it to the endovascular procedure and therefore prescribed no treatment, performing follow-up on a monthly basis to evaluate the clinical course. Complete repopulation had occurred after 5 months.

Alopecia after radiotherapy to the head and neck is reported extensively in the literature; however, there are very few reports of alopecia after diagnostic and therapeutic endovascular procedures and it is underdiagnosed. It is considered to be a subtype of radiodermatitis caused by the marked radiosensitivity of hair follicles in anagen.¹

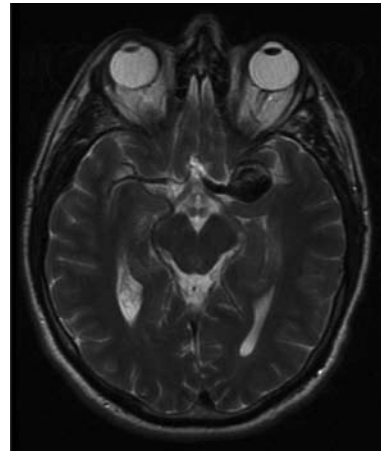


Figure 1. Absence of signal on the magnetic resonance image in the area of the left middle cerebral artery. Giant fusiform aneurysm (2.5 cm).

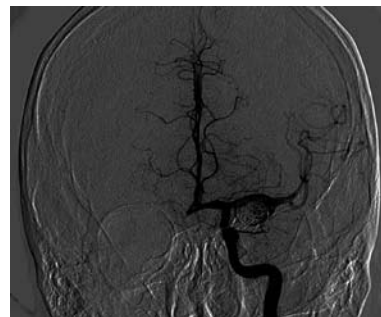


Figure 2. Formation of a coagulum around the spiral, leading to occlusion of the lumen of the aneurysm.

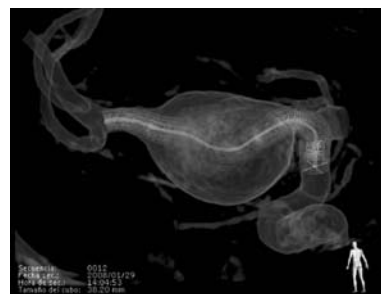


Figure 3. Three-dimensional image showing the Enterprise endovascular prosthesis and the Matrix2 spiral, which produced occlusion of the aneurysm by the formation of a coagulum and fibrosis.

Onset usually occurs 2 to 3 weeks after embolization, and it is most common in cases of large, complex vascular disorders,² or ruptured lesions.³ It may be the only manifestation of the adverse effects of the radiation, or it may be associated with other symptoms, which are usually neurological.⁴ There is a series of factors associated with a



Figure 4. Rectangular plaque of alopecia in the left parieto-occipital area.

higher probability of the onset of this type of alopecia,⁵ including total fluoroscopy time (more than 100 minutes), total dose received (cicatricial alopecia if the dose is over 7 Gy, transient with doses between 3 and 5 Gy), predominant area of radiation, and biologic factors (age, oxygen level, hair density, hormonal status, genetic factors, ethnic differences, etc).

The main differential diagnoses are pressure alopecia,⁶ due to hypoxia and associated with long periods of general anesthesia (more than 4 hours), and alopecia areata.⁷

Postembolization alopecia does not require treatment, as repopulation occurs spontaneously after a mean time of 12 to 14 weeks. The only method of prevention is to monitor the radiation dose during angiography. A recently published study investigated this condition in 103 patients.⁸ It was found that the percentage of adverse effect was undetectable if the different parameters evaluated—cumulative cutaneous dose during endovascular embolization, total fluoroscopy time, total cumulative dose, and number of series of digital subtraction images—did not exceed a certain threshold. It is therefore very important for interventional radiology departments to establish a series measures to avoid these undesirable effects.

This type of alopecia was first described in 1994 in a radiology journal,⁹ and it was not until 1998 that it was first published in a dermatology journal, *Hautarzt*, by the group of the German investigator Krasovec.¹⁰ A year later, Tosti et al⁷ published a further 3 cases. Since then, only about 10 cases have been published in journals of dermatologic interest, the last in 2008,¹¹ making it a condition that appears to be known better by neuroradiologists and neurosurgeons than by dermatologists.

We can affirm that transient alopecia after embolization is a rare and little-known phenomenon. However, due to the increase in minimally invasive endovascular procedures for the diagnosis and treatment of vascular disorders, its

incidence may rise. Because of this, patients should be informed and the disorder should be included in the list of possible complications of this technique, together with the local, systemic, and cerebral adverse effects already listed.

Correspondence:
Gloria Garnacho Saucedo.
C/ Cantueso, 10.
14012 Córdoba, Spain.
glorina_81@hotmail.com

Conflicts of Interest

The authors declare no conflicts of interest.

References

1. D'incan M, Roger H, Gabrillargues J, Mansard S, Parent S, Chazal J, et al. [Radiation-induced temporary hair loss after endovascular embolization of the cerebral arteries: six cases]. *Ann Dermatol Venerol*. 2002;129:703-6.
2. Wen CS, Lin SM, Chen Y, Chen JC, Wang YH, Tseng SH. Radiation-induced temporary alopecia after embolization of cerebral arteriovenous malformations. *Clin Neurol Neurosurg*. 2003;105:215-7.
3. Nannapaneni R, Behari S, Mendelow D, Gholkar A. Temporary alopecia after subarachnoid haemorrhage. *J Clin Neurosci*. 2007;14:157-61.
4. Thorat JD, Hwang PY. Peculiar geometric alopecia and trigeminal nerve dysfunction in a patient after Guglielmi detachable coil embolization of a ruptured aneurysm. *J Stroke Cerebrovasc Dis*. 2007;16:40-2.
5. Mooney RB, McKinstry CS, Kamel HA. Absorbed dose and deterministic effects to patients from interventional neuroradiology. *Br J Radiol*. 2000;73:745-51.
6. Lee WS, Lee SW, Lee S, Lee JW. Postoperative alopecia in five patients after treatment of aneurysm rupture with a Guglielmi detachable coil: pressure alopecia, radiation induced, or both? *J Dermatol*. 2004;31:848-51.
7. Tosti A, Piraccini BM, Alagna G. Temporary hair loss simulating alopecia areata after endovascular surgery of cerebral arteriovenous malformations: a report of 3 cases. *Arch Dermatol*. 1999;135:1555-6.
8. Suzuki S, Furui S, Matsumaru Y, Nobuyuki S, Ebara M, Abe T, et al. Patient skin dose during neuroembolization by multiple-point measurement using a radiosensitive indicator. *Am J Neuroradiol*. 2008;29:1076-81.
9. Huda W, Peters KR. Radiation-induced temporary epilation after a neuroradiologically guided embolization procedure. *Radiology*. 1994;193:642-4.
10. Krasovec M, Trüeb RM. Temporary roentgen epilation after embolization of a cerebral arteriovenous malformation. *Hautarzt*. 1998;49:307-9.
11. Martí N, López V, Pereda C, Martín JM, Montesinos E, Jordá E. Radiation-induced temporary alopecia after embolization of cerebral aneurysms. *Dermatol Online J*. 2008;15:14-9.

Generalized Linear Porokeratosis Limited to One Side of the Body

A. Martorell-Calatayud,^a M. Blanes,^b C. Requena,^a R. Botella-Estrada,^a and C. Guillén-Barona^a

^aServicio de Dermatología, Instituto Valenciano de Oncología, Valencia, Spain

^bServicio de Dermatología, Hospital Marina Baixa, Villajoyosa, Alicante, Spain

To the Editor:

Poroqueratosis (PQ) refers to a group of disorders of keratinization with the common histological feature of the presence of cornoid lamellae, narrow columns of parakeratosis on a focally disorganized epidermis.¹

We present 2 patients with lesions compatible with linear PQ, with extensive involvement of one side of the body.

The first patient was a 23-year-old man with no past history of interest. He was seen for the presence of multiple lesions that had been present for 20 years. The lesions had started to appear in the left inguinocrural region in the first year of life, affecting a progressively larger area, following a metamer pattern on the left lower limb and medial region of the left foot. At 4 years of age, when it appeared that the disorder had stabilized, he began to develop new lesions in the left scapular region and on the left upper limb, following a blaschkoid distribution. The

patient noticed that no new lesions had appeared after 18 years of age.

On physical examination the patient presented multiple annular lesions that became confluent, forming polycyclic and tortuous patterns. The basic lesion consisted of a central area of atrophy, occasionally verrucous, that was clearly demarcated by a peripheral hyperkeratotic border. The lesions were not infiltrated, were not adherent to the deep planes, and varied in size between 1 and 5 cm in their largest diameter. The lesions were present only on the left side of the patient's body, following a metamer or blaschkoid linear distribution, affecting the upper and lower limbs, trunk, and genital region (Figure 1), but were not found on the face.

A 5 mm biopsy that included the hyperkeratotic border and the central atrophic area was taken from a lesion on the left ankle. Central epidermal atrophy surrounded by a peripheral parakeratotic column was visible at low magnification (Figure 2). Higher magnification revealed



Figure 1. Linear porokeratosis. A. Right lower limb. B. Right scapular region. C. Right upper limb. D. Right side of the foreskin.

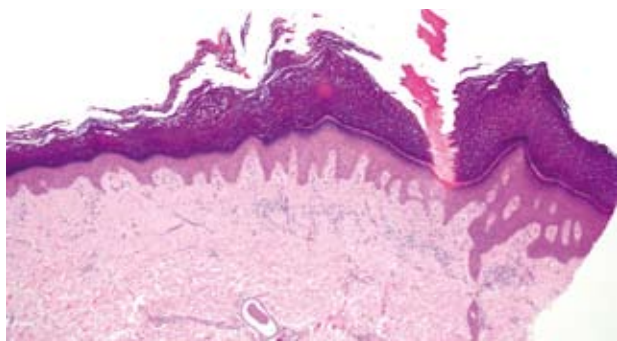


Figure 2. Histological image stained with hematoxylin and eosin. Presence of orthokeratotic hyperkeratosis surrounding an intensely eosinophilic area of parakeratosis ($\times 40$).

the presence of a column of parakeratotic hyperkeratosis, known as a cornoid lamella, associated with hypogranulosis and a discrete subjacent acanthosis.

The second case was of a 58-year-old woman, with no past history of interest. She was referred for evaluation and treatment of skin lesions situated in the right infraclavicular region and that had been present for more than 20 years.

Physical examination revealed the presence of multiple linear lesions of brownish color, with a well-defined, elevated border. The lesions were distributed over the upper third of the right side of the trunk, following a blaschkoid pattern (Figures 3A and 3B). There was a smaller number of similar lesions on the right upper and lower limbs.

With a suspicion of PQ, a 6 mm punch biopsy was performed, which showed findings compatible with this diagnosis.

The linear form of PQ, currently considered a mosaicism of classic PQ, is a rare variant that accounts for between 3.5% and 15% of all cases of PQ.^{2,3} It usually occurs



Figure 3. Linear porokeratosis affecting: A. Left scapular region. B. Left thoracic region. C and D, Left lower limb.

sporadically, and presents at birth or in early adolescence as a verrucous rash that can follow the Blaschko or metameric lines.⁴ The lesions are typically asymptomatic, and pruritus is very rare.⁵ There are currently 2 subtypes that have been described, one localized and the other generalized.^{4,6} The localized form is more common; the lesions are distributed unilaterally and are limited to a single limb, usually distally. The generalized form is rarer. It presents with very numerous lesions on more than 1 anatomic region, usually symmetrically, and can affect the trunk.^{4,6} The presence of erosions or ulcers, underlying bone abnormalities, and onychodystrophy have been reported very rarely in association with this variant of PQ.⁷

The patients we describe presented lesions that were clinically and histologically compatible with PQ, with a characteristically unilateral pattern of distribution, affecting the upper and lower limbs and the trunk, but not the face. This clinical pattern could therefore be considered to be a clinical variant within the group of generalized PQ.

The most important complication of PQ is malignant transformation of the lesions (mainly to squamous cell carcinoma and, to a lesser degree, to basal cell carcinoma),⁸ with frequencies that vary between 7.5% and 19%⁹ of cases on long-term follow-up. The lesions that present the highest risk of malignant transformation are large, long-standing lesions of the linear variant.²

It has been suggested that allelic loss or overexpression of tumor suppressor gene p53 detected in linear PQ could explain this higher risk of neoplastic transformation.^{7,10}

The therapeutic arsenal in PQ is extensive but lacking in efficacy, and the most important aspect in these patients is periodic follow-up for the early detection of malignant change in any of the lesions. Neither of our

patients developed a malignant tumor after long-term follow-up.

Correspondence:
Antonio Martorell Calatayud.
C/ San José de la Montaña, puerta 11.
46008 Valencia, Spain.
antmarto@hotmail.com

Conflicts of Interest

The authors declare no conflicts of interest.

References

1. Derviş E, Demirkessen C. Generalized linear porokeratosis. *Int J Dermatol*. 2006;45:1077-9.
2. Chen HH, Liao YH. Onychodystrophy in congenital linear porokeratosis. *Br J Dermatol*. 2002;147:1272-3.
3. Valdivielso-Ramos M. Poroqueratosis genital. *Actas Dermosifiliogr*. 2008;99:217-20.
4. Kaur S, Thami GP, Mohan H, Kanwar AJ. Co-existence of variants of porokeratosis: a case report and a review of the literature. *J Dermatol*. 2002;29:305-9.
5. Erkek E, Bozdoğan O, Sanlı C, Ozoguz P. Clinicopathologic challenge: linear brown macules on the chest and arm. Linear porokeratosis. *Int J Dermatol*. 2008;47:539-41.
6. Malhotra SK, Puri KJPS, Goyal T, Chahal KS. Linear porokeratosis. *Dermatol Online J*. 2007;13:15.
7. Tseng SS, Levit EK, Ilarda I, Garzon MC, Grossman ME. Linear porokeratosis with underlying bony abnormalities. *Cutis*. 2002;69:309-12.
8. Suh DH, Lee HS, Kim SD, Cho KH, Kim KH, Park KC. Coexistence of disseminated superficial actinic porokeratosis in childhood with congenital linear porokeratosis. *Pediatr Dermatol*. 2000;17:466-8.
9. Palit A, Inamadar AC, Yelikar BR. Unilateral linear annular lesions in a child. *Pediatr Dermatol*. 2004;21:682-3.
10. Happle R. Cancer proneness of linear porokeratosis may be explained by allelic loss. *Dermatology*. 1997;195:20-5.

Hair Transplantation in Temporal Triangular Alopecia

F. Jiménez-Acosta and I. Ponce

Clínica Dermatológica Dr. Jiménez Acosta, Las Palmas de Gran Canaria, Spain

To the Editor:

Temporal triangular alopecia (TTA) or congenital triangular alopecia is a rare form of alopecia circumscripta. First described by Saboraud,¹ its most common clinical presentation is that of a round or oval patch of noninflammatory and nonscarring alopecia, typically located in the anterior temporal area. The patch may

increase in size with the growth of the child, generally reaching a diameter of 2 to 4 cm. Inspection through a magnifying glass shows the presence of fine vellus hair and an absence of terminal hair.

TTA can be present at birth or can develop between the first few months of life and the age of 6 years. There are rare cases of adult onset,² and the current tendency is therefore to



Figure 1. Patient with a bald patch in the left temporal area.



Figure 2. Result 2 years after hair transplantation.

consider it an acquired rather than a congenital condition.^{3,4} Although TTA is usually unilateral, about 20% of cases are bilateral.⁴ Some rare familial cases have also been reported.⁵

The pathogenesis of TTA is unknown. The fact that TTA develops in the first years of life in an area of the scalp that had previously appeared normal seems to indicate the presence of a localized process of miniaturization of the hair follicles, which produces regression to vellus hair. The stimulus responsible for this irreversible regression is unknown.⁶ Histologically, TTA shows hypoplasia of the follicles in a clinically normal scalp. The number of hair follicles is normal, but all are of the vellus (miniaturized) type.⁶

The diagnosis of TTA is clinical: its typical clinical manifestation and location, along with its childhood onset, are sufficient to establish the diagnosis. In doubtful cases, histology makes it possible to distinguish it from other types of alopecia circumscripta, such as epidermal nevi, aplasia cutis congenita,⁷ pseudopelade of Brocq, or alopecia areata.

We report a typical case of TTA treated by follicular unit hair transplantation with a satisfactory cosmetic result.

The patient was an 18-year-old woman who came to our clinic for treatment of TTA. She had had a bald patch in the left temporal area since childhood, although she was unable to specify at what age it had appeared. The patch measured 8 cm² and was oval in shape (Figure 1).

We performed follicular unit hair transplantation on the bald patch.

The procedure consisted of removing a small strip of skin 4 cm long by 1 cm wide from the occipital scalp (donor area). The surgical wound was closed with 4-0 Ethilon sutures.

From this donor strip a total of 250 follicular units, each containing from 1 to 4 hairs, were dissected using a Zeiss Stemi DV4 stereomicroscope.

These 250 follicular units were implanted into the bald patch under local anesthesia. The 250 incisions (30 incisions per cm²) made using surgical blades (Personna prep blades) were 0.7 mm in diameter for 1-hair follicular units and 1 mm in diameter for units of 2 to 4 hairs. The 250 follicular units were inserted one by one into these incisions using jeweler-type forceps. The patient was discharged and the stitches in the donor area removed 1 week later. The transplanted grafts began to grow after 3 to 4 months and the patient was extremely satisfied with the cosmetic result (Figure 2).

TTA only requires treatment for cosmetic reasons, as the alopecia is stable and asymptomatic. Medical treatment of TTA with minoxidil or topical or intralesional corticosteroids has not proven effective and surgical treatment is the only therapeutic alternative.⁶ Surgery with complete removal of the bald patch is an option to consider, but the closing of surgical wounds 3 to 4 cm in diameter on the scalp may require the use of flaps or tissues expanders. Follicular unit hair transplantation, on the other hand, is a technique that has been used with satisfactory results, not only in male pattern alopecia, but also in other types of cosmetic reconstructions of the scalp.^{8,9}

It is interesting to note that there have been hardly any studies published on the usefulness of hair transplantation in TTA, despite the fact that TTA is an indication that is well-known to surgeons specializing in this field. The only case we found was published in 1984 by Bragman,¹⁰ who described the case of a 36-year-old patient with TTA treated with hair grafts obtained using a 4 mm punch. For this reason, we believe it is important for dermatologists to know about the usefulness of hair transplantation in TTA, performed as described in this article, with a satisfactory cosmetic result.

Correspondence:

Francisco Jiménez Acosta.

Clínica Dermatológica Dr. Jiménez Acosta.

Angel Guimerá, 2.

35003 Las Palmas de Gran Canaria, Spain.

fjimenez@clinicadelpelo.com

Conflicts of Interest

The authors declare no conflicts of interest.

References

1. Saboraud R. Manuel élémentaire de dermatologie topographique régionale. Paris: Masson and Cie; 1905; p. 197.
2. Trakimas CA, Sperling LC. Temporal triangular alopecia acquired in adulthood. *J Am Acad Dermatol.* 1999;40:842-4.
3. Elmer KB, George RM. Congenital triangular alopecia: a case report and review. *Cutis.* 2002;69:255-6.
4. Akan IM, Yildirim S, Avci G, Aköz T, Karadayi N. Bilateral temporal triangular alopecia acquired in adulthood. *Plast Reconstr Surg.* 2001;107:1616-7.
5. Ruggieri M, Rizzo R, Pavone P, Baieli S, Sorge G, Happle R. Temporal triangular alopecia in association with mental retardation and epilepsy in a mother and daughter. *Arch Dermatol.* 2000;136:426-7.
6. Trakimas C, Sperling LC, Skelton HG III, Smith KJ, Buker JL. Clinical and histologic findings in temporal triangular alopecia. *J Am Acad Dermatol.* 1994;31:205-9.
7. Kenner JR, Sperling LC. Pathological case of the month. Temporal triangular alopecia and aplasia cutis congenita. *Arch Pediatr Adolesc Med.* 1998;152:1241-2.
8. Barrera A. The use of micrografts and minigrafts in the aesthetic reconstruction of the face and scalp. *Plast Reconstr Surg.* 2003;112:883-90.
9. Haber RS, Stough DB. Trasplante de pelo. Madrid: Elsevier; 2007.
10. Bargman H. Congenital temporal triangular alopecia. *Can Med Assoc J.* 1984;131:1253-4.

Lichen Planus and Lichen Striatus: Opposite Ends of the Same Spectrum?

F. Pulgar,^a R. Rivera,^a J.L. Rodríguez-Peralto,^b and F. Vanaclocha^a

^aServicio de Dermatología. Servicio de Anatomía Patológica, Hospital Universitario 12 de Octubre, Madrid, Spain

To the Editor:

Lichen planus is an inflammatory dermatosis with an immunological pathogenesis that is not yet fully understood. It is characterized by an eruption consisting of pruritic violaceous polygonal papules that can affect the trunk or limbs and that may often have whitish linear striae, called Wickham striae, on their surface. Involvement of the mucosae is common.¹

Lichen striatus is an asymptomatic dermatosis that generally occurs in childhood. It consists of small, scaly, erythematous papules of 1 to 2 mm in diameter, occasionally with a vesicular component, distributed in a linear band along the Blaschko lines. The lesions are usually distributed unilaterally on a limb and, unlike those of lichen planus, usually leave a residual hypopigmentation.¹ Bilateral or multiple lesions are very rare.^{2,3}

We present a case that started as a linear eruption and subsequently became generalized.

The patient was a 7-year-old girl weighing 20 kg and with a history of celiac disease. She was seen for a pruritic linear eruption that had appeared 3 months earlier on the left upper limb, with involvement of the ipsilateral thumb nail. There was no fever or other clinical symptoms. The condition was diagnosed as lichen striatus, and we prescribed treatment with topical corticosteroids (0.1% methylprednisolone aceponate in a once-daily application).

After a month of topical treatment the lesions were no longer linear and localized, but had spread irregularly over the body. No mucosal lesions were detected. The lesions were shiny violaceous erythematous papules with a lichenoid appearance (Figures 1 and 2) that coalesced to form plaques spread randomly over the trunk, legs, and lumbar region. Some, such as those affecting the antecubital fossae, showed a degree of desquamation.

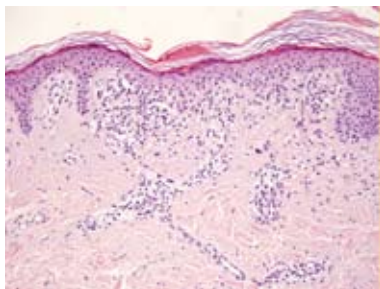


Figure 1. Erythematous violaceous papules with a lichenoid appearance.

Figure 2. Nail dystrophy on the left thumb.



Figure 3. Focal parakeratosis and lymphocytic infiltrate involving the glands and the dermal-epidermal junction. Hematoxylin-eosin, original magnification $\times 10$.



The cubital side of the left thumb nail was dystrophic and had longitudinal striations (Figure 2). The face and popliteal fossae were affected, as were the soles of both feet. There was also extremely intense erythema and scaling on the scalp. A skin biopsy taken from the arm showed a lymphocytic infiltrate at the dermal-epidermal junction with vacuolar degeneration of the basal layer and a deep perifollicular and glandular infiltrate (Figure 3). In this first biopsy, the lesions were both clinically and histologically consistent with lichen striatus. In a second skin biopsy from the lumbar region, orthokeratotic hyperkeratosis, wedge-shaped hypergranulosis, acanthosis, papillomatosis, a bandlike lymphocytic infiltrate at the dermal-epidermal junction, pigmentary incontinence, and colloid bodies were observed. The infiltrate also extended towards deep glandular structures. High-dose (1 mg/kg/d) prednisone therapy was initiated, with no response after 3 weeks. In view of the inefficacy of corticosteroid therapy and the severe scalp involvement (with the consequent risk of scarring alopecia), we decided to initiate treatment with cyclosporine at 2.5 mg/kg/d and obtained a clear improvement of all the lesions, including the nail dystrophy, in 6 weeks.

Lichen planus and lichen striatus are entities that are well-defined clinically, although there may be cases in which the differential diagnosis is not really clear.

Generally, lichen striatus shows a linear distribution along the Blaschko lines,³⁻⁵ while lichen planus usually presents as papules or plaques that only occasionally

exhibit a linear distribution.⁶ The differential diagnosis between linear lichen planus and lichen striatus involves very subtle clinical distinctions. For this reason histology is necessary, although given the similarity between the 2 conditions, it is not always diagnostic.⁶ The absence of pruritus, a residual hypopigmentation, and a tendency to resolve spontaneously support the diagnosis of lichen striatus.¹ In our patient the initially linear eruption became generalized, though with no apparent trigger; there was pruritus, residual pigmentation, and no clear evidence of spontaneous resolution.

Histologically, lichen planus usually shows orthokeratotic hyperkeratosis, hypergranulosis, irregular sawtooth acanthosis, vacuolar degeneration of the basal layer of the epidermis, a bandlike infiltrate, apoptosis, pigmentary incontinence, and Civatte bodies.¹ The histology in lichen striatus varies somewhat depending on the stage of the disease. A dense and occasionally bandlike lymphohistiocytic perivascular infiltrate can usually be seen. Epidermal hyperkeratosis with focal parakeratosis is a typical finding in lichen striatus but rare in lichen planus. The infiltrate can extend as far down as the eccrine glands and ducts. Such a finding can help differentiate lichen striatus from lichen planus. However, the histologic variability of lichen striatus is considerable and the findings may sometimes be indistinguishable from those of lichen planus.¹

Cases of overlap between these 2 entities have been reported in the literature.

Herd et al⁶ described the case of a 37-year-old woman with an asymptomatic and spontaneously resolving linear eruption. Histologically, the condition was consistent with lichen planus and, in addition, showed occasional plasma cells in the papillary dermis. Rubio et al⁷ described the case of a 29-year-old woman with an asymptomatic and spontaneously resolving linear eruption that left residual hyperpigmentation. Histology showed acanthosis, hypergranulosis, basal layer degeneration, colloid bodies, and a bandlike lymphomononuclear infiltrate, more consistent with the microscopic diagnosis of lichen planus. Numata et al⁸ reported the case of a 21-year-old man who developed symptoms of linear lichen planus, but with histologic involvement of the eccrine sweat glands, following the intramuscular injection of triamcinolone acetonide.

In our patient, the first biopsy (obtained from the initial lineal eruption) was consistent with lichen striatus, given the presence of parakeratosis and a deep glandular infiltrate. The second biopsy (taken from one of the generalized lesions), however, showed epidermal changes that were more characteristic of lichen planus.

In view of the intense involvement of the scalp and the lack of response to oral corticosteroid therapy, we decided to initiate treatment with cyclosporine. The result was spectacular and we were thus able to prevent scarring alopecia.

We present a case that had clinical and histologic characteristics both of lichen planus and of lichen striatus. This is exceptionally common, and supports the hypothesis that these 2 diseases represent the opposite ends of the same disease spectrum.

Correspondence:
Fernando Pulgar Martín.
C/ Norias, 47.
28220 Majadahonda. Madrid. Spain.
ferpulmar@hotmail.com

Conflicts of Interest

The authors declare no conflicts of interest.

References

1. Tilly JJ, Drolet BA, Esterly NB. Lichenoid eruptions in children. *J Am Acad Dermatol*. 2004;51:606-24.
2. Aloï F, Solaroli C, Pippione M. Diffuse and bilateral lichen striatus. *Pediatr Dermatol*. 1997;14:36-8.
3. Mittal R, Khaitan BK, Ramam M, Verma KK, Manchanda M. Multiple lichen striatus—An unusual presentation. *Indian J Dermatol Venereol Leprol*. 2001;67:204.
4. Peramiquel L, Baselga E, Dalmau J, Roé E, del Mar Campos M, Alomar A. Lichen striatus: clinical and epidemiological review of 23 cases. *Eur J Pediatr*. 2006;165:267-9.
5. Taniguchi Abagge K, Parolin Marinoni L, Giraldi S, Carvalho VO, de Oliveira Santini C, Favre H. Lichen striatus: description of 89 cases in children. *Pediatr Dermatol*. 2004;21:440-3.
6. Herd RM, McLaren KM, Aldridge RD. Linear lichen planus and lichen striatus—opposite ends of a spectrum. *Clin Exp Dermatol*. 1993;18:335-7.
7. Rubio FA, Robayna G, Herranz P, de Lucas R, Hernández-Cano N, Contreras F, et al. Linear lichen planus and lichen striatus: is there an intermediate form between these conditions? *Clin Exp Dermatol*. 1997;22:61-2.
8. Numata Y, Okuyama R, Tagami H, Aiba S. Linear lichen planus distributed in the lines of Blaschko developing during intramuscular triamcinolone acetonide therapy for alopecia areata multiplex. *J Eur Acad Dermatol Venereol*. 2008;22:1370-1.

Unilateral Allergic Contact Dermatitis of the Eyelid Caused by Iopimax

M.T Bordel-Gómez, J. Sánchez-Estella, and J.C. Santos-Durán

Servicio de Dermatología, Complejo Asistencial Virgen de la Concha, Zamora. Spain

To the Editor:

Apraclonidine is a drug belonging to the group of α -2 adrenergic agonists; it is widely used as short-term additional treatment for chronic glaucoma in patients receiving medical treatment and who require additional reduction of intraocular pressure in order to delay treatment with laser or surgery. The drug is also widely used to control pressure within the eye after glaucoma surgery.

We report the case of a 63-year-old woman with a history of hypertension on treatment with Ameride (amiloride plus hydrochlorothiazide); the woman suffered from long-term glaucoma in the left eye for which she had been receiving treatment for several years with Xalatan ophthalmic solution (latanoprost, 50 mg). Despite this treatment, the last ophthalmic check-up, performed in early 2007, revealed high intraocular pressure and treatment with Iopimax (apraclonidine, 0.5%) ophthalmic solution was therefore added. The main objective of this additional treatment was to control intraocular pressure, as well as to delay surgery because the patient had already undergone surgery for glaucoma in the right eye. The patient consulted for an 8-month history of progressive and recurrent appearance of intensely pruritic, eczematous

lesions on the upper and lower eyelids of the left eye; the lesions had developed after starting treatment with Iopimax and had improved substantially following application of different topical corticosteroids, although they recurred when corticosteroid treatment was suspended.

Examination of the skin revealed a small erythematous plaque with fine desquamation, located on each eyelid; desquamation was more accentuated on the internal surface (Figure 1). Laboratory tests (general analysis, immunologic study, and thyroid hormones) revealed no



Figure 1. Eczematous lesions on the upper and lower left eyelids.

relevant pathology data; we found only a slight increase in total cholesterol levels. Skin prick testing was performed using a standard battery of the Spanish Skin Research and Allergy Group (GEIDAC) and the standard technique recommended by the International Contact Dermatitis Research Group (ICDRG). The allergens were applied to normal skin on the upper back in vertical strips and were fixed to the skin by means of a hypoallergenic adhesive tape. The allergen patches were removed after 48 hours and the sites were examined; a second examination was performed at 96 hours. Patch testing was also performed with the following ophthalmic products: Timofтол (timolol, 0.25% and 0.5%), Trusopt (dorzolamide, 1%), Xalatan (latanoprost, 0.005%), and Iopimax (apraclonidine, 0.5% and 1%), as well as a 1% aqueous solution of atropine sulfate, 10% aqueous solution of phenylephrine hydrochloride, 1% aqueous solution of pilocarpine hydrochloride, and 0.1% aqueous solution of benzalkonium chloride, supplied by Trolab Hermal (Reinbek, Germany). Results for all of these products were negative on examination after both 48 hours and 96 hours, except for Iopimax, 0.5% (3+ at 96 h).

Treatment was started with moderate-potency topical corticosteroids and we recommended withdrawal of Iopimax and a new assessment by the ophthalmologist; treatment for the glaucoma was changed to Combigan ophthalmic solution (brimonidine and timolol). The clinical course was satisfactory and no new episodes of eczematous lesions developed.

Allergic contact dermatitis is a recognized adverse effect of several drugs used to treat glaucoma. A recent review identified 10 drugs that, when used topically to treat this eye disorder, may cause a contact allergic reaction. These agents include β -blockers (timolol, befunolol, betaxolol, carteolol, and metipranolol), carbonic anhydrase inhibitors (dorzolamide), a parasympathomimetic drug (pilocarpine), and sympathomimetic drugs (dipivefrin and apraclonidine).¹

Apraclonidine, the active ingredient in Iopimax, is an α_2 -selective drug that blocks α_2 presynaptic receptors, acting indirectly and inhibiting presynaptic stimulation for the formation of aqueous humor, thereby reducing intraocular pressure. It is also used in the treatment of Horner syndrome because it increases the tone of Müller's muscle, causing rapid elevation of the upper eyelid by between 1 and 3 mm. It is not associated with many side effects, but cases of allergic blepharoconjunctivitis,²⁻⁴ burning sensation, eyelid retraction, and mydriasis have been attributed to it.

To our knowledge, only 3 cases of allergy to apraclonidine ophthalmic solution have been published^{2,5}; 2 of these were confirmed by means of skin patch tests using a 5% aqueous solution of apraclonidine and the other by means of a patch test using Iopimax, 0.5%, and a 10% aqueous solution of apraclonidine. In our patient, the allergy was confirmed by patch testing exclusively with Iopimax, 0.5%. We have performed patch tests with Iopimax on control patients who had undergone patch tests (standard battery and others) and have not detected any positive allergic or irritative reactions to date.

The explosion of new drugs in recent decades has led to substantial advances in the treatment of many diseases, including glaucoma. As a result, their use has increased sharply, occasionally causing medical problems, including contact allergic dermatitis. We conclude that this type of dermatitis should be considered as a rare but possible adverse effect of treatment with apraclonidine (Iopimax) ophthalmic solution. An alternative for these patients is the use of other α_1 -adrenergic drugs, such as brimonidine or clonidine.^{2,6}

Correspondence:
M.^a Teresa Bordel Gómez.
C/ Martínez Villergas, 6, 1^o B.
49003 Zamora, Spain.
maitebordel@aadv.es
matebordel@yahoo.es

Conflicts of Interest

The authors declare no conflicts of interest.

References

1. Holdiness MR. Contact dermatitis to topical drugs for glaucoma. *Am J Contact Dermatitis*. 2001;12:217-9.
2. Silvestre JF, Carnero L, Ramón R, Albares MP, Botella R. Allergic contact dermatitis from apraclonidine in eyedrops. *Contact Dermatitis*. 2001;45:251.
3. Gross RL, Pinyero A, Orengo-Nania S. Clinical experience with apraclonidine 0.5 %. *J Glaucoma*. 1997;6:298-302.
4. Butler P, Mannschreck M, Lin S, Hwang I, Alvarado J. Clinical experience with the long-term use of 1% apraclonidine. Incidence of allergic reactions. *Arch Ophthalmol*. 1995;113:293-6.
5. Armisen M, Vidal C, Quitans R, Suárez A, Castroviejo M. Allergic contact dermatitis from apraclonidine. *Contact Dermatitis*. 1997;39:193.
6. Shin DH, Glover BK, Cha SC, Kim YY, Kim C, Nguyen KD. Long-term Brimonidine therapy in glaucoma patients with apraclonidine allergy. *Am J Ophthalmol*. 1999;127:511-5.

Perineal Nodular Induration in Cyclists

R. González-Pérez,^a L. Carnero,^a N. Arbide,^b and R. Soloeta^a

^aServicio de Dermatología. ^bServicio de Anatomía Patológica, Hospital Santiago Apóstol, Vitoria, Álava, Spain

To the Editor:

Perineal nodular induration in cyclists, also known as ischial hygroma, third testicle, accessory testicles of cyclists, or cyclists nodule, is an entity that is relatively well known to sports medicine specialists and to professional and amateur cyclists, but practically unknown to other doctors.^{1,2}

A 49-year-old man with no relevant history visited our department with a tender lesion in the right ischial region that had appeared a year earlier. Physical examination revealed a 4-cm nodule of rubbery consistency, covered by normal skin. Ultrasound and fine-needle aspiration studies were performed but the results were inconclusive. Magnetic resonance imaging (MRI) (Figure 1) revealed a nodular lesion with a fibrotic appearance with a maximum diameter of 3 cm, in intimate contact with the lower surface of the right ischial ramus, and changes in trabeculation and infiltration of the surrounding fat, compatible with perineal induration of the cyclist. The contralateral region revealed similar, though less marked, changes. The patient was questioned again and confirmed that he was a cyclist.

The lesion was excised and histologic study revealed a tumor lesion with poorly defined borders, consisting of a multilocular cystic formation with a fibrinous content and not surrounded by epithelial or synovial cells; there were adjacent areas of aseptic necrosis (Figure 2). The rest of the lesion showed a fibrous proliferation with fusiform cells without atypia; in addition there were areas of hemorrhage, foci of myxoid degeneration, and, occasionally, groups of mature adipocytes trapped in the fibrous proliferation. Immune staining of the fusiform cells was positive for vimentin and factor XIIIa and negative for estrogen receptors, progesterone receptors, S-100, CD34, actin, and desmin. The Ki-67 proliferation index was 0%.

Clinically, perineal induration in cyclists usually manifests as 2 nodules (1 on either side of the perineal raphe), although it occasionally presents as a single nodule (third testicle), located immediately behind the scrotum, close to the ischial tuberosity. The skin covering the nodule presents a normal appearance and the nodule usually measures between 2 and 3 cm. Palpation reveals an fibroelastic consistency and adherence to the adjacent connective tissue may be appreciated.^{1,2} Our patient consulted with a nodule located in the right ischial region, although NMRI also revealed an incipient lesion of similar characteristics on the contralateral side.

Histologically, the nodule is characterized by a central pseudocystic formation, which develops in an area of

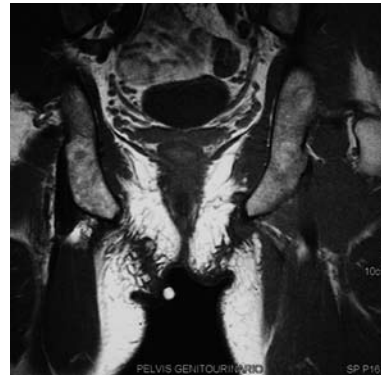


Figure 1. Magnetic resonance imaging (cutaneous marker on the studied lesion): nodule in the right perineal region, hypointense in T1 sequences.

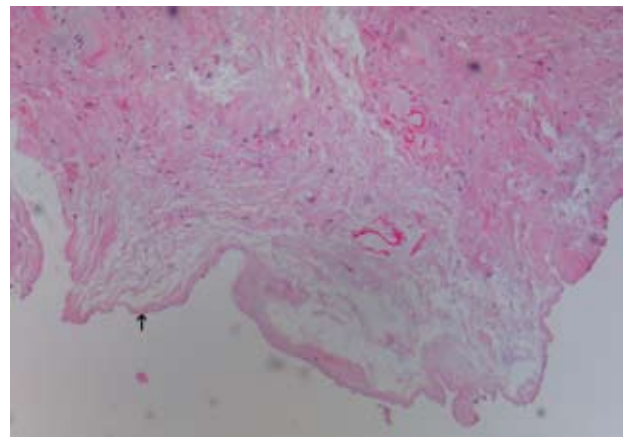


Figure 2. Cystic cavity lined by a fibrinoid material (↑) with no surrounding epithelial membrane. Below, lax conjunctive tissue with few cells and myxoid changes (hematoxylin-eosin, ×20).

aseptic necrosis that develops in the connective tissue of the surface perineal fascia. This tissue becomes homogeneous and loses its fibrous structure. Erythrocyte diapedesis and a mild reactive inflammatory infiltrate may also be observed.¹⁻³ Some of these findings are reminiscent of those of ischemic fasciitis found over bony prominences in elderly patients; this provides a clue to the etiology of this disorder.^{2,4,5} Although not universally accepted, it seems probable that the development of these lesions is linked to the compression of the soft tissue between the saddle and the ischial tuberosity, and to friction between the perineal fascia and the bony structures due to movement of the saddle; this could lead to collagen degeneration, myxoid changes, and formation of the pseudocyst.¹⁻³

Diagnosis of these nodules may be particularly difficult if a history of cycling is not detected. The differential diagnosis should be established with common minor disorders (cysts and lipomas), and with other, rarer and more important conditions such as aggressive angiomyxoma. This rare variety of myxoid tumor can infiltrate locally and is associated with a high risk of local recurrence after resection, but it has no metastatic potential. It generally affects young women and arises in the soft tissue of the pelvis and perineum. Rare cases have also been reported in men, where it develops in the scrotum and groin, presenting as masses or nodules in these areas.^{6,7}

Treatment of these perineal indurations in cyclists is generally surgical, although, if this is not possible because the patient is a professional cyclist, conservative treatment with rest and infiltrations of steroids or hyaluronidase may be tried.¹⁻³

These cyclist's nodules are a genuine handicap for professional cyclists and contraindicate cycling for amateur cyclists.¹

Correspondence:
Ricardo González-Pérez.
Servicio de Dermatología.
Hospital Santiago Apóstol.
C/ Olaguibel, 29.
01004 Vitoria, Álava, Spain.
RICARDO.GONZALEZPEREZ@osakidetza.net

Conflicts of Interest

The authors declare no conflicts of interest.

References

1. Vuong PN, Camuzard CT, Schoonaert MF. Perineal nodular indurations («accessory testicles») in cyclists. Fine needle aspiration cytologic and pathologic findings in two cases. *Acta Cytol.* 1988;32:86-90.
2. De Saint Aubain Somerhausen N, Geurde B, Couvreur Y. Perineal nodular induration: the «third testicle of the cyclist», an under-recognized pseudotumour. *Histopathology.* 2003;42:615-6.
3. Kohler P, Uterman S, Kahle B, Hartschuh W. [Biker's nodule-perineal nodular induration of the cyclist]. *Hautarzt.* 2000;51:763-5.
4. Perosio PM, Weiss SW. Ischemic fasciitis: a juxta-skeletal fibroblastic proliferation with a predilection for elderly patients. *Mod Pathol.* 1993;6:69-72.
5. Liegl B, Fletcher CD. Ischemic fasciitis: analysis of 44 cases indicating an inconsistent association with immobility or debilitation. *Am J Surg Pathol.* 2008;32:1546-52.
6. Chihara Y, Fujimoto K, Takada S, Hirayama A, Cho M, Yoshida K, et al. Aggressive angiomyxoma in the scrotum expressing androgen and progesterone receptors. *Int J Urol.* 2003;10:672-5.
7. Idrees MT, Hoch BL, Wang BY, Unger PD. Aggressive angiomyxoma of male genital region. Report of 4 cases with immunohistochemical evaluation including hormone receptor status. *Ann Diagn Pathol.* 2006;10:197-204

Perianal Verrucous Papules in a Patient With Bannayan-Riley-Ruvalcaba Syndrome

B. Casado,^a C. Gómez-Fernández,^a M. Feito,^a and M.J. González-Beato^b

^aServicio de Dermatología. ^bServicio de Anatomía Patológica, Hospital Universitario La Paz, Madrid, Spain

To the Editor:

Bannayan-Riley-Ruvalcaba syndrome (BRRS) is a rare autosomal dominant genetic disorder caused in approximately 60% of cases by mutations on the *PTEN* gene, a tumor suppressor gene on chromosome 10.¹ Typically, patients with this syndrome present a triad of macrocephaly, genital melanotic macules, and hamartomatous polyps in the intestine. Identical mutations of the *PTEN* gene have been reported in 80% of patients with Cowden syndrome, also known as multiple hamartoma syndrome.² Both syndromes have been reported in some families and some members may present phenotypes corresponding to both entities.³ In view of the clinical and genetic overlap of BRRS and Cowden syndrome, they are currently considered as different

phenotypic expressions of the same condition known as PTEN syndrome.^{1,3} This spectrum also includes Proteus and Proteus-like syndrome, in which *PTEN* mutations are found in a smaller proportion of cases.¹

There are numerous skin manifestations common to BRRS and Cowden syndromes, including oral, facial, and acral warts or verrucous papules, lipomas, trichilemmomas, and inverted follicular keratosis.¹⁻³ The presence of multiple trichilemmomas in the facial region is diagnostic of Cowden syndrome.⁴

We report the case of a 33-year-old man with no family history of interest who was diagnosed with BRRS on the basis of a range of abnormalities, the most prominent being marked macrocephaly with hypertelorism, slight mental retardation, genital lentiginosities, intestinal polyps,

gynecomasty, and pectus excavatum. Clinical suspicion was confirmed by genetic study, in which a deletion was detected on the *PTEN* gene.

In February 2007, he was diagnosed with colon adenocarcinoma, which was resected without any coadjuvant therapy. Fecal incontinence occurred as a sequela of the operation. Later, during a follow-up colonoscopy, a large number of perianal skin lesions were detected. Clinically these were suggestive of condylomata acuminata, and so the patient was referred to our clinic. The patient and his family denied that he had had sexual contact.

On physical examination, a verrucous erythematous pinkish plaque was observed on which multiple papillary and lobulated lesions of differing sizes were present. These were mostly sessile with a highly vascular appearance and bled on minimal contact (Figure 1). No other similar lesions were found on the rest of the body surface.

We excised one of the lesions for histopathological study. A polypoid lesion was observed with a connective tissue core, epidermal hyperplasia with hypergranulosis, and dermal papillae with numerous dilated vessels suggestive of a common wart (Figures 2 and 3). Typing was positive for human papilloma virus (HPV) 11, considered a mucosal subtype with low oncogenic risk.

In both BRRS and Cowen syndrome, loss of *PTEN* gene functionality leads to disorganized cell proliferation and failure of apoptosis. The 2 syndromes share a range of pseudoverrucous mucocutaneous manifestations, although these were first described in Cowen syndrome.¹ They might arise from an overgrowth of the epithelium of the follicular infundibulum due to the mutation.⁴ Clinically, all these lesions may resemble one another, and so histological analysis is necessary to correctly characterize them.^{2,5}

Different studies performed exclusively in patients with Cowen syndrome have attempted to clarify the role of HPV in pseudoverrucous lesions, but the results have been contradictory.^{5,6} Initially, in 1991, using immunohistochemical techniques, Hori demonstrated the presence of the HPV common antigen in an inverted follicular keratosis.⁴

In 2002, Schaller et al⁵ undertook typing of HPV in predominantly facial lesions. They managed to isolate DNA from the virus in most cases and concluded, therefore, that HPV could be the causative agent in most verrucous lesions associated with Cowden syndrome. However, in another study, Ruhoy et al⁶ attempted to isolate HPV DNA in inverted follicular keratoses excised from a patient with Cowden syndrome and obtained negative results.

Nevertheless, there are currently several studies that claim that HPV has a reservoir located in hair follicles and that it can remain latent for long periods without causing any clinically detectable skin lesions,⁷⁻⁹ and so an etiologic connection is difficult to confirm.⁵



Figure 1. Perianal verrucous erythematous pinkish plaque, with multiple polypoid lesions with a highly vascular appearance at the surface.

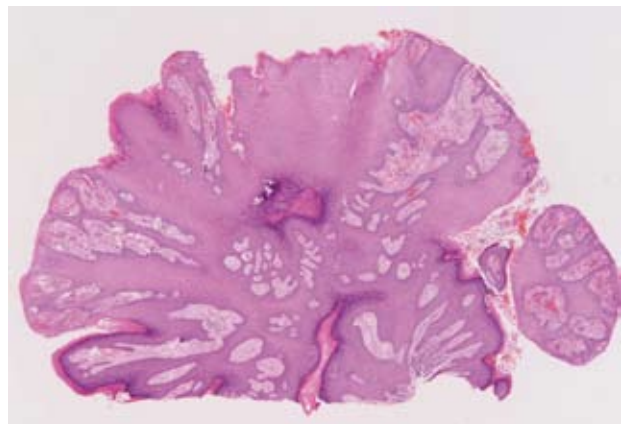


Figure 2. Hematoxylin-eosin, x10.

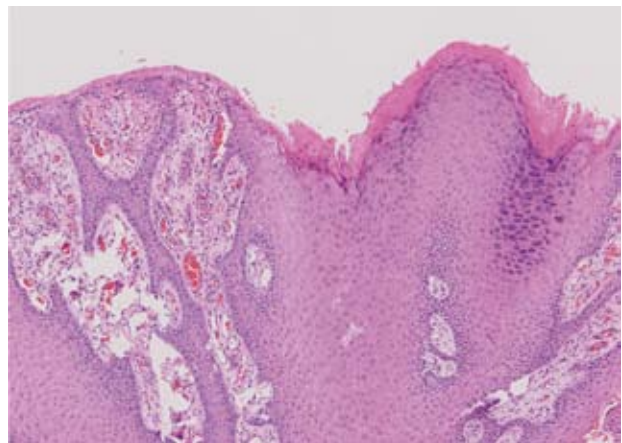


Figure 3. Hematoxylin-eosin, x40.

The patient is now receiving treatment with carbon dioxide laser light with good results. New lesions will continue to appear, however, while the patient remains anally incontinent.

We wish to highlight the importance of the numerous characteristic lesions associated with these PTEN syndromes. The pathogenesis has yet to be clarified, but often these lesions are the first manifestation of a set of genetic diseases in which there is a high risk of developing malignant tumors at different sites.

Correspondence:
Beatriz Casado Verrier.
C/ Paseo de la Castellana, 261.
Madrid. Spain.
bezcv@hotmail.com

Conflicts of Interest

The authors declare no conflicts of interest.

References

1. Eng C. Constipation, Polyps, or Cancer? Let PTEN predict your future. *Am J Med Genet A*. 2003;122A:315-22.
2. Eng C. PTEN: one gene, many syndromes. *Hum Mutat*. 2003;22:183-98.
3. Erkek E, Hizel S, Sanly C, Erkek AB, Tombakoglu M, Bozdogan O, et al. Clinical and histopatological findings in Bannayan-Riley-Ruvalcaba syndrome. *J Am Acad Dermatol*. 2005;53:639-43.
4. Larumbe A, Iglesias ME, Illarramendi JJ, Córdoba A, Gállego M. Queratosis acras y queratosis folicular invertida como manifestación de la enfermedad de Cowden. *Actas Dermosifiliogr*. 2007;98:425-9.
5. Schaller J, Rohwedder A, Burgdorf WH, Itin PH, Lautenschlager S. Identification of human papillomavirus DNA in cutaneous lesions of Cowden syndrome. *Dermatology*. 2003;207:134-40.
6. Ruhoy SM, Thomas G, Nuovo G. Multiple inverted follicular keratoses as a presenting sign of Cowden's syndrome: case report with human papillomavirus studies. *J Am Acad Dermatol*. 2004;51:411-5.
7. Boxman IL, Hogewoning A, Mulder LH, Bouwe Bavinck JN, Schegget J. Detection of human papillomavirus types 6 and 11 in pubic and perianal hair from patients with genital warts. *J Clin Microbiol*. 1999;37:2270-3.
8. Adachi A, Suzuki T, Tomita Y. Detection of human papillomavirus type 56 DNA, belonging to a mucous high-risk group, in hair follicles in the genital area of a woman no longer suffering from viral warts. *Br J Dermatol*. 2004;151:212-5.
9. Yell JA, Sinclair R, Mann S, Fleming K, Ryan TJ. Human papillomavirus type 6-induced condylomata: an unusual complication of intertrigo. *Br J Dermatol*. 1993;128:575-7.