# **ISSUES IN DERMATOLOGY**

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# The Rediscovery of Previously Described Dermatoses

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ho discovered America? By some accounts, it was Christopher Columbus, who left Palos, Spain, on August 3, 1492, and dropped anchor off the coast of Santo Domingo, the Dominican Republic, not far from Haiti, on October 12 of the same year. Other historians, paleographers, and polemicists offer different accounts based on literary and archeological evidence suggesting that first the Phoenicians and later the Vikings arrived on these coasts and discovered the New World well before Genoa's maritime genius. Even assuming these latter accounts are true, we cannot ignore the outstanding achievements of Columbus: he introduced Europe to the new continent and initiated a new adventure in human endeavor, what have come to be known as "modern times." Nevertheless, only a small part of the New World remains associated in name with this discoverer—Columbia. Place names have been unfair in that respect, and the name "America" was proposed to designate a quarter of the world in honor of Amerigo Vespucci, who made several expeditions to the lands discovered by Columbus and who succeeded him as the *piloto mayor*.<sup>1</sup> "But what," you are no doubt asking yourself, "does this have to do with skin diseases?"

> By analogy, this same process of discoveryrediscovery-renaming occurs in the field of medicine and, *a fortiori*, in dermatology. The eponyms of many diseases do not necessarily refer to their first discoverers. The variability of authorship, authors' deliberate ignorance of previous publications, and a lack of interest in articles written in other languages have all contributed to "pseudodiscoveries."

> This article is not a systematic attempt to completely devalue these "rediscoveries"; in some cases, they serve to rehabilitate neglected or forgotten entities and may even generate new interest and stimulate research. This was the case, for example, with Alan Lyell<sup>2</sup> in 1956, who wrote the most highly cited article ever to appear in *The British Journal of Dermatology*<sup>3</sup> and paved the way for the rediscovery of "dermatitis exfoliativa neonatorum syndrome" described by

Gottfried Ritter von Rittershain<sup>4</sup> in 1878 and "erythroderma with epidermolysis" described by Debré et al<sup>5</sup> in 1939. Its rediscovery generated a considerable amount of research, especially on staphylococcal exfoliations, immunologic mechanisms of severe bullous toxicoderma, and pharmacokinetics in dermatology. This research in turn contributed to the categorization of the "Lyell syndrome" into its 3 component entities also recognized by Lyell<sup>3</sup>: staphylococcal scalded skin syndrome (SSSS), exanthematous necrolysis (a major form of the Stevens-Johnson syndrome), and drug-induced toxic necrolysis.

## REDISCOVERY: THE NEGATIVE SIDE

However, a rediscovery is not always so useful or enlightening, nor are the achievements of the rediscoverer always so valuable. At times, the rediscovery is attributable not to the rediscovering authors

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themselves but to the fact that the large centers involved in collecting and identifying international scientific data use predominantly English as their research language. Despite their international calling, they do a less than thorough job researching data, and the selection process gives priority to English-language journals. The implications of such a scenario are not difficult to imagine. The French linguist Claude Hagège<sup>6</sup> denounced it with a polemical tone, noting

Either the articles in French are missing from the bibliographies because authors using the most widely available resources cannot find them; or, even worse, an English-speaking researcher does find the articles, but plagiarizes them. A researcher may plagiarize non-English articles with impunity because, without an archiving system, the scientific community is able to ignore their presence in the original language. And when the authors or their affiliates discover the larceny, it is too late to seek the rightful credit for an idea or discovery that an act of piracy has whisked away without due process.

Here is just 1 recent example of this phenomenon: the rediscovery by Hunt et al<sup>7</sup> of microvenular hemangioma that was described barely 2 years earlier under another name in a German-language journal of dermatology.<sup>8</sup>

## EPONYMOUS ATTRIBUTIONS: GOOD AND BAD

The eponymic route<sup>9</sup> is one road to immortality for the rediscoverer. Such eponymy is sometimes fortunate. For example, Lyme disease<sup>10,11</sup> was instrumental in the discovery of borreliosis caused by *Borrelia burgdorferi*,<sup>12</sup> and its discovery led to the etiological reclassification of entities such as erythema chronicum migrans and acrodermatitis chronica atrophicans. It is probably *Lyme disease* or the more accurate *Lyme borreliosis* that will enter into general usage and endure.

In some cases, the eponym is only partially deserved. For Netherton syndrome, several authors provided partial descriptions of a set of symptoms: Comèl<sup>13</sup> described ichthyosis linearis circumflexa in 1949; Bazex and Dupré<sup>14</sup> described variable circinate erythemas, periorificial erythemas, and hypotrichosis in 1956; Netherton<sup>15</sup> described the bamboolike hairs characteristic of this disease, which Wilkinson et al<sup>16</sup> then named Netherton disease in 1964. This entity has had numerous discoverers, all worthy researchers. But despite protests by Bazex and Dupré,<sup>17</sup> only the name of the person who described the hair shaft defects received an eponymous role.

# ACRONYM DISGUISES

Resorting to acronyms is another way of completely or partially stripping away historically significant events. Hence, the syndrome with the acronym CRST (calcinosis, Raynaud phenomenon, sclerodactyly, telangiectasia), which has become CREST syndrome (because of esophageal involvement) and even, pseudoeponymically, "the Crest syndrome," has expropriated the contributions of Thibierge and Weissenbach,18 who provided a remarkable description in 1911 of the 4 cardinal signs of what, in their estimation, was only a symptomatic variant of scleroderma. Surely CR(E)ST is more descriptive and less a mouthful than Thiebierge-Weissenbach disease, but it does not give the actual discoverers their due credit.

The REM syndrome (reticular erythematous mucinosis) described by Steigleder et al in 1974<sup>19</sup> turned out to be identical, according to the authors,<sup>20</sup> to plaquelike cutaneous mucinosis described in 1960 by Perry et al,<sup>21</sup> but the acronym REM seems to have stuck.

In some cases, the acronym is the best name: CHILD syndrome<sup>22</sup> (congenital hemidysplasia, unilateral ichtyosiform erythroderma, and limb defects) is infinitely more communicable than the titular descriptor proposed simultaneously by other authors<sup>23</sup> for the same disease.

# SHIFTING TERMINOLOGY

The choice of terminology is a fundamental issue in many cases. Thus, keratoacanthoma first described by Hutchinson<sup>24</sup> in 1889 (crateriform ulcer) has been rediscovered several times: by Dupont<sup>25</sup> in 1930 (sebaceous cyst with vegetative wall); by MacCormac and Scarff<sup>26</sup> in 1936 (molluscum sebaceum); and by Rook and Whimster<sup>27</sup> in 1950 (keratoacanthoma). But the term keratoacanthoma seemed better than all the others, even according to Dupont,<sup>28</sup> who luckily still retains credit for its discovery.

However, it does not always turn out that way; the choice of certain terms often precludes the citation of certain articles despite their earlier publication dates. The expression "sinus histiocytosis," proposed by Rosai and Dorfman,29 provides no link to original research by Destombes,<sup>30</sup> whose name rarely appears in publications using the eponym and never shows up in works using the anatomopathological term to designate the disease. The concept of multiple endocrine neoplasias that appeared in 1968 in the wake of research by Pearse<sup>31</sup> on the APUD (amine precursor uptake and decarboxylation) system has either eliminated or reduced the citation of previous works where such pathological relationships were already described. Hence, the seminal work by Bazex and Dupré<sup>32</sup> on "cutaneous myelinic neuromas" is never cited by articles on the MEN (multiple endocrine neoplasia) 2b syndrome.

Finally, there are more complicated situations where even a bookworm would feel disoriented. The syndrome associating multiple primitive carcinomas of the colon and cutaneous tumors (keratoacanthomas, adenomas, and sebaceous carcinomas, in particular) was described by Muir et al in 196733 and by Torre in 1968.34 There has never been a conflict concerning these authors and if the eponym Torre syndrome generally prevails in the citations, this would seem linked to the less than rigorous searches of later authors who duplicate each other's work without checking sources.

# SOME REDISCOVERIES ADVANCE OUR KNOWLEDGE

Successive rediscoveries do at times improve our understanding of the disease process. Examples abound. For instance, carcinoid papillomatosis,<sup>35</sup> considered to be pseudocancerous, first became florid papillomatosis,<sup>36</sup> a precancerous disease, and next verrucous carcinoma,<sup>37</sup> a low-grade cancer. Most, if not all, of the poikilodermas have been described and rediscovered, and rarely are the first authors credited eponymously.<sup>38</sup>

It is not always possible to explain the outcome of a discovery or the designation of a name. And this does not just involve the medical field. In the automotive industry, 2 models undergoing development with the utmost secrecy may wind up being identical when they hit the market, as their designers integrate the same features using the latest technology. But in fact, 1 of the 2 models will enjoy greater success commercially contrary to all logical predictions.

A medical example of this apparently random phenomenon is the association of scleroderma and primitive biliary cirrhosis, remarkably described by Murray-Lyon et al<sup>39</sup> and Reynolds et al<sup>40</sup> at the same time. However, only the term Reynolds syndrome prevailed.

### CULTURAL CHAUVINISM

Unfortunately, cultural chauvinism is present everywhere: Dubreuilh's melanosis is to the Frenchspeaking world what Hutchinson melanosis is to the Englishspeaking world; Darier disease in Europe becomes White disease in the United States, although the 2 names should be systematically combined because this disease was described simultaneously on both sides of the Atlantic in 1889.41,42 Sometimes discoveries occur simultaneously on both sides of a battle front and the division persists into posterity. The Fiessinger-Leroy-Reiter syndrome is better known in Germany as the Reiter syndrome.43

### CONCLUSIONS

So what does all this mean? First, it means that we should regard any apparently original ideas with some scientific skepticism and do a careful literature search. Access to data banks facilitate such work, but direct consultations with colleagues sometimes yield other information or cruel disappointment.

It also means that we should learn how to publicize our discoveries, using all existing means of communication. The choice of words, the timing of a written or an oral presentation, and the relevant forum are assets as valuable as the quality of the research itself.

Finally, we have to show respect for our discoveries by tailoring them to the scientific movements of their time and allowing them to gain recognition as such. Pautrier (in personal oral communication with E.G. on many occasions) said that in order to succeed, one has to have the knowledge, communicate the knowledge, and use the knowledge. With respect to medical discoveries, these 3 principles should be applied systematically if one does not wish to remain the anonymous Indian or Viking or Phoenician who discovered the New World, but, instead, desires to emulate Christopher Columbus, who not only discovered the New World, but also made his discovery known.

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#### REFERENCES

- Tomb R. De la redécouverte des maladies cutanées antérieurement décrites. Strasbourg, France: Université Louis-Pasteur, 1989:3-4.
- Lyell A. Toxic epidermal necrolysis: an eruption resembling scalding of the skin. Br J Dermatol. 1956;68:355-361.
- Lyell A. Smoke without fire. *Curr Contents Clin* Med. 1991;16:8.
- Ritter von Rittershain G. Die exfoliative Dermatitis jüngerer Säuglinge. *Centralzeit Kinderheilk*. 1878;2:3-23.
- Debré R, Lamy M, Lamotte M. Un cas d'érythrodermie avec épidermolyse chez un enfant de 12 ans. *Bull Soc Péd Paris*, 1939:37:231-238.
- Hagège C. Le Français et les siècles. Paris, France: Editions Odile Jacob, 1987:189.
- Hunt SJ, Santa-Cruz DJ, Barr RJ. Microvenular hemangioma. J Cutan Pathol. 1991;18:235-240.
- Bantel E, Grosshans E, Ortonne JP. Zur Kenntnis mikrokapillärer Angiome: Beobachtungen bei schwangeren bzw unter hormoneller Antikonzeption stehenden Frauen. Z Hautkr. 1989;64:1071-1074.
- Garfield E. What's in a name? the eponymic route to immortality. *Curr Contents Life Sci.* 1983;47: 5-16.
- Steere AC, Malawista SE, Snydman DR, et al. Lyme arthritis: an epidemic of oligoarticular arthritis in children and adults in three Connecticut communities. Arthritis Rheum. 1977;20:7-17.
- Steere AC, Malawista SE, Hardin JA, Ruddy S, Askenase PW, Andiman WA. Erythema chronicum migrans and Lyme arthritis: the enlarging clinical spectrum. *Ann Intern Med.* 1977;86:685-698.
- Burgdorfer W, Barbour AG, Hayes SF, Benach JL, Grunwaldt E, Davis JP. Lyme disease: a tickborne spirochetosis? *Science*. 1982;216:1317-1319.
- Comèl M. Ichthyosis linearis circumflexa. Dermatologica. 1949;98:133-136.
- Bazex A, Dupré A. Génodermatose à érythèmes circinés variables. Ann Dermatol Syph. 1956;83: 610-612.
- Netherton EW. A unique case of trichorrhexis nodosa: "bamboo hairs." Arch Dermatol. 1958;78: 483-487.
- Wilkinson RD, Curtis GH, Hawk WA. Netherton's disease. Arch Dermatol. 1964;89:46-54.
- Bazex A, Dupré A. Syndrome de Netherton: observation précédemment publiée sous le titre de "érythèmes circinés variables, érythèmes périorificiels, hypotrichose congénitaux. Bull Soc Fr Dermatol. 1971;78:641-644.
- Thibierge G, Weissenbach RJ. Concrétions calcaires sous-cutanées et sclérodermie. Ann Dermatol Syph. 1911;42:129-155.
- Steigleder GK, Gartmann H, Linker U. REMsyndrome: Retikuläre erythematöse Mucinosis (Rundzellerythematosis). *Z Hautkr.* 1974;49:235-238.
- Braddock SW, Davis CS, Davis RB. Reticular erythematous mucinosis and thrombocytopenic purpura: report of a case and review of the world literature, including plaque-like cutaneous mucinosis. J Am Acad Dermatol. 1988;19:859-868.
- Perry HO, Kierland RR, Montgomery H. Plaquelike form of cutaneous mucinosis. Arch Dermatol. 1960;82:980-985.
- 22. Happle R, Koch H, Lenz W. The CHILD syndrome: congenital hemidysplasia with ichtyosi-

form erythrodema and limb defects. *Eur J Pediatr.* 1980;134:27-33.

- Laplanche G, Grosshans E, Gabriel-Robez O, et al. Hyperplasie épidermique et hémidysplasie corporelle hypoplasique congénitales homolatérales (démembrement du syndrome de Solomon). Ann Dermatol Venereol. 1980;107: 729-739.
- 24. Hutchinson J. The crateriform ulcer. *Arch Surg.* 1889;1:241.
- Dupont A. Kyste sébacé atypique. Bull Soc Belge Dermatol. 1930:177.
- MacCormac H, Scarff RW. Molluscum sebaceum. Br J Dermatol. 1936;48:624-626.
- 27. Rook A, Whimster I. Le kérato-acanthome. *Arch Belg Dermatol.* 1950;6:137-146.
- Dupont A. Le kérato-acanthome (kyste sébacé végétant, molluscum sebaceum). *Ann Dermatol Syph.* 1854;81:621-633.
- Rosai J, Dorfman RF. Sinus histiocytosis with massive lymphadenopathy: a newly recognized benign clinicopathological entity. *Arch Pathol.* 1969; 87:63-70.
- Destombes P. Adénites avec surcharge lipidique de l'enfant ou de l'adulte jeune, observées aux Antilles et au Mali (quatre observations). *Bull* Soc Pathol Exot. 1965;58:1169-1175.
- 31. Pearse AG. Common cytochemical and ultra-

structural characteristics of cells producing polypeptide hormones (the APUD series) and their relevance to thyroid and ultimobranchial C cells and calcitonin. *Proc R Soc Lond B Biol Sci.* 1968; 170:71-80.

- Bazex A, Dupré A. Neuromes myéliniques muqueux à localisation centro-faciale et pharyngée: neurome des lèvres, de la langue, des paupières, des narines, du larynx (entité nouvelle?). Ann Dermatol Syph. 1958.85:613-641.
- Muir EG, Yates-Bell AJ, Barlow KA. Multiple primary carcinomata of the colon, duodenum and larynx associtaed with kerato-acanthoma of the face. *Br J Surg.* 1967;54:191-195.
- Torre D. Multiple sebaceous tumors. Arch Dermatol. 1968;98:549-552.
- Scheicher-Gottron E. Papillomatosis mucosae carcinoides der Mundschleimhaut bei gleichzeitigem Vorhandensein eines Lichen ruber der Haut. Z Hautkr. 1958;24:99-101.
- Rock JA, Fisher OR. Florid papillomatosis of the oral cavity and larynx. Arch Otolaryngol. 1960; 72:593-598.
- Grinspan D, Abulafia J. Oral florid papillomatosis (verrucous carcinoma). *Int J Dermatol.* 1979;18: 608-622.
- 38. Marghescu S. Kongenitale Poikilodermien. *Z Hautkr.* 1989;64:167-169.

- Murray-Lyon IM, Thompson RPH, Ansell JD, et al. Scleroderma and primary biliary cirrhosis. Br Med J. 1970;3:258-259.
- Reynolds TB, Denison EK, Frankl HD, et al. Primary biliary cirrhosis with scleroderma, Reynaud's phenomenon and telangiectasia: new syndrome. *Am J Med.* 1971;50:302-312.
- 41. Darier J. De la psorospermose folliculaire végétante. Ann Dermatol Syph. 1889;10:597-612.
- White JC. A case of keratosis (ichthyosis follicularis). J Cutan Genito-Urin Dis. 1889;7:201-209.
- Tomb R. De la redécouverte des maladies cutanées antérieurement décrites. Strasbourg, France: Université Louis-Pasteur; 1989:84.
- Grosshans E, Tomb R. 1492-1992: Il y a 500 ans Christophe Colomb découvrait l'Amérique. Ann Dermatol Venereol. 1992;119:7-9.

### Submissions

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