Respiratory Medicine Case Reports 25 (2018) 204–206

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

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

Case report

Metastatic pulmonary melanoma complicated with erythroderma and recurrent sepsis

Lining Wang[a,b], Ning Li[a], Ling Zhou[a], Xiaoyan Chen[c], Wei Tang[a,*]

[a] Department of Pulmonary and Critical Care Medicine, Ruijin Hospital, Shanghai Jiaotong University School of Medicine, Shanghai, China
[b] Department of Hematology, Ruijin Hospital, Shanghai Jiaotong University School of Medicine, Shanghai, China
[c] Department of Pathology, Ruijin Hospital, Shanghai Jiaotong University School of Medicine, Shanghai, China

ARTICLE INFO

Keywords:
Melanoma
Erythroderma
Sepsis
Lung cancer

ABSTRACT

Erythroderma is occasionally associated with lung cancer. Here we report a 69-year-old male who has history of melanoma from 8 years ago presented with erythroderma associated with skin, soft tissue and pulmonary infection. CT chest showed a single enlarging right upper lobe lung nodule. Biopsy showed evidence of adenocarcinoma, and eventually proven to be metastatic melanoma. The patient improved after antibiotics and antifungal treatment. To our knowledge, this is the first case of melanoma metastasis presented as erythroderma as paraneoplastic syndrome.

1. Case presentation

A 69-year-old male presented to our emergency department with febrile erythroderma, chills and dyspnea, worsen during steroid taper. He was recently admitted to another hospital for febrile erythroderma with desquamation. He also has an enlarging right upper lobe lung nodule first found 18 months prior, but was never biopsied. He was treated with high dose intravenous steroid and IVIG for his erythroderma, His symptom significantly improved and almost completely resolved when he was discharged on steroid. Unfortunately, he lost follow up but started self tapering steroid when symptom recurred.

The patient has a history of nasal malignant melanoma 8 years ago. He was initially treated with two cycles of chemotherapy (dacarbazine and cisplatin) and radiation (200cGy per doses for 33 doses). He then underwent surgery, which followed by 5 more cycles of adjuvant chemotherapy (temozolomide). After that, he had local relapse which required three more intranasal operations, again followed by chemotherapy with temozolomide and local radiation (200cGy per dose for 25 more doses). Most recently in 2015, a regional lymph node was found to be metastasis and was resected. He was further treated with one year of interferon alfa-2b and interleukin-2. He has no known history of dermatosis or autoimmune disease.

At admission, he was febrile at 40 °C, tachycardia at 120 bpm, blood pressure at 110/60 mmHg, and hypoxia at 78 mmHg at room air. Oxygen saturation improved to 99% with 3L/min oxygen. He was generally weak but remained awake and alert. He has diffuse edematous erythroderma at scalp, bilateral upper eyelids, trunk, scrotum and extremities with vesicular rash and desquamation. (Fig. 1). Lung exam showed decreased breathing sound throughout, and fine crackles at bilateral lung bases. There was no palpable adenopathy or hepatosplenomegaly.

CBC at admission showed a leukocytosis at 15.68 K/μL with left shift. Comprehensive metabolic panel showed very low albumin at 19 g/L, but normal creatinine, and normal liver function tests. Blood culture were positive for staphylococcus aureus (MRSA).

CT chest confirmed the right upper lobe nodule and bilateral pleural effusion and mediastinal adenopathy (Fig. 2).

Empirical antibiotics with Vancomycin and Imipenemn were immediately started. Urgent dermatology consult was obtained, and methylprednisolone was added to the treatment regimen. Central line had to be placed for access due to the significant edema.

The patient made quick improvement with the above regiment. A CT guided transthoracic biopsy was then performed. Cytopathogical analysis initially showed trace of adenocarcinoma. It was decided to taper the steroid to prepare for surgical intervention.

The patient then had relapsed of fevers, chills and desaturation one week after the start of tapering steroid and de-escalation of antibiotics. Blood cultures were repeated and this time it was positive for Candida parasilosis and MDR *Klebsiella pneumoniae*, the latter is only sensitive to tigecycline. The patient was then started on fluconazole and tigecycline. Meanwhile, the final pathology report confirmed its positive for S-100, indicating the lesion is metastatic melanoma (Fig. 3).

* Corresponding author.
E-mail address: tw10986@rjh.com.cn (W. Tang).

https://doi.org/10.1016/j.rmcr.2018.09.004
Received 3 December 2017; Received in revised form 5 September 2018; Accepted 5 September 2018

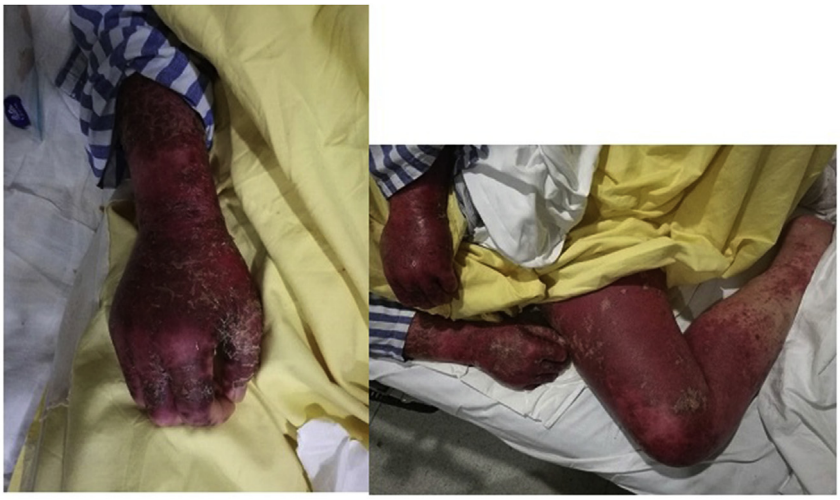

Fig. 1. Patient presented with systemic erythroderma with vesiculation and desquamation at admission.

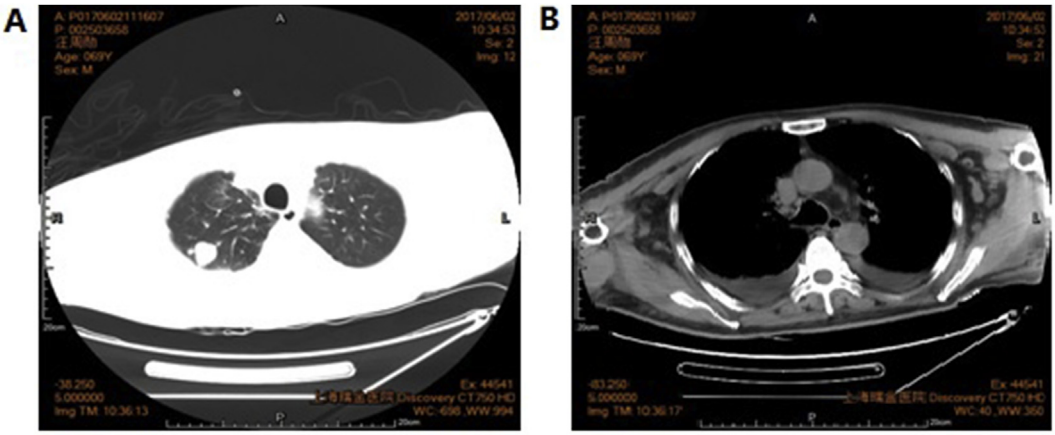

Fig. 2. Pulmonary nodule (A) and mediastinal adenopathy (B) demonstrated in the thoracic CT scan during admission.

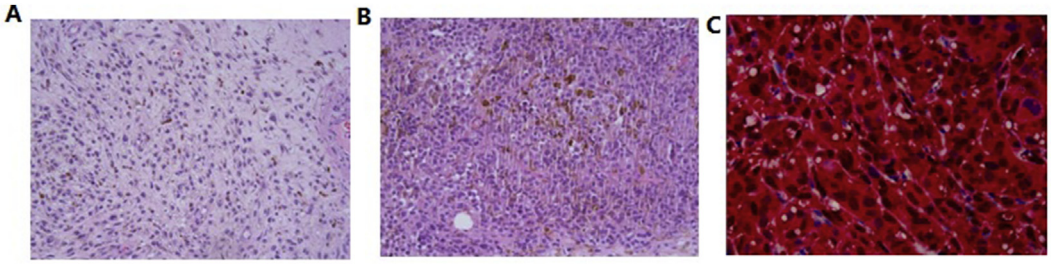

Fig. 3. A: Pathology of the nasal surgery sample which identified melanoma 8 years ago. Tissues observed under microscope (400×). B: Pathology of transthoracic needle aspiration tissue which identified pulmonary metastatic melanoma. Biopsy tissues observed under microscope (400×). C: Immunohistochemistry analysis of transthoracic needle aspiration tissue positive for S-100.

The patient eventually improved after the course of antibiotics and antifugul treatment and was discharged. Surgical resection of the pulmonary metastasis was offered, the patient is to consider but yet to decide.

2. Discussion

One-third of melanoma patients develop metastatic lesions and the most common distant organ affected is the lung [1]. Approximately 80% of these patients initially have one single site of metastasis just like our patient. The overall survival rate for patients with pulmonary metastatic melanoma is poor, 34%, 14% and 6% at 1, 2 and 5 years respectively despite of all therapeutic options available. The absence of other metastatic sites and the practice of metastasectomy are two factors associated with longer disease free survival [1,2]. Earlier discovery and treatment of the metastatic site is associated with better prognosis. Hence, routine systemic follow-up should be emphasized to all melanoma patients. As in our patient, he had a first local metastasis at the neck 8 years ago. He responded quite well to radiotherapy, which is rare for melanoma. He was found to have the right upper lobe lung nodule 18 months ago, which was enlarging at follow up, but unfortunately was never biopsied. One can only imagine if a biopsy then resection were done sooner, he might not have developed erythroderma and sepsis.

Erythroderma is a skin condition with erythema involves more than 90% of the total body surface. It may be secondary to various etiologies including autoimmune diseases, infections, drug reaction, psoriasis and malignant tumors [3]. If drugs and underlying dermatosis are excluded, the proportion of malignancy-associated erythroderma could be as high as 20% [4]. The malignancy-associated erythroderma is thought to be part of paraneoplastic syndrome. As for this patient, the erythroderma appeared much later than the lung metastasis, so we believed it's indeed a manifestation of the paraneoplastic syndrome. Melanoma, just like other cancers, gives rise to paraneoplastic syndromes which often affect retina or central nervous system. Cutaneous manifestations linked to melanoma are scarce and of unknown underlying mechanism. The diagnosis is of exclusion and should meet the criteria of Curth's postulates [5]. The best documented dermatoses in relation are dermatomyositis and pemphigus vulgaris [6]. Additionally, gyrate erythroderma [7], grover's disease [8] and Sweet's syndrome [9] have also been reported. We believe this is the first time that erythroderma is reported as part of cutaneous paraneoplastic manifestations of melanoma, although development of invasive melanoma or non-melanoma skin cancers in patients with congenital erythroderma has been reported previously [10,11]. We believe there is an unknown connection between these two diseases. Also, melanoma has been shown to be associated with autoimmune diseases such as SLE, RA and Sjogren [12,13] these autoimmune disorders may appear before the symptoms of melanoma or during its treatment [14,15]. Thus, melanoma patients suffering from erythroderma should be screened for autoimmune antibodies and ideally should have the skin lesion biopsied to rule out other coexisting condition(s).

Erythroderma is often associated with recurrent fever [3]. The cutaneous disorder itself and its usual treatment of systemic steroid, both impair the integrity of the skin which could lead to cutaneous infection or even sepsis. In the case of paraneoplastic erythroderma, as it is mentioned in Curth's postulates, improvement of the symptoms mostly happen after treating the original malignancy. Long term large dose systemic steroid thus is not desired due to its limited benefit, plus the increased risk of infections which could be fatal for late stage cancer patients, or at least could delay the more definitively treatment of surgical intervention or immunosuppressive therapy. As for our patient who was critically ill and hospitalized for two months, the best window for surgery may have passed.

3. Conclusion

This is a case of pulmonary metastatic melanoma who presented with erythroderma as paraneoplastic syndrome. Secondary infection associated with systemic large dose steroid should be promptly treated.

Authors' contributions

WT contributed to the conception and design of the study. LNW, NL, LZ and WT contributed to the patient care. XYC interpreted the histological examination and IHC stains. LNW and WT drafted and revised the manuscript. All authors read and approved the final manuscript.

Conflicts of interest

No potential conflict of interest was reported by the authors.

Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.rmcr.2018.09.004.

References

[1] R.P. Petersen, et al., Improved survival with pulmonary metastasectomy: an analysis of 1720 patients with pulmonary metastatic melanoma, J. Thorac. Cardiovasc. Surg. 133 (1) (2007) 104–110.
[2] H.B. Neuman, et al., Stage-IV melanoma and pulmonary metastases: factors predictive of survival, Ann. Surg Oncol. 14 (10) (2007) 2847–2853.
[3] N. Mistry, et al., A review of the diagnosis and management of erythroderma (generalized red skin), Adv. Skin Wound Care 28 (5) (2015) 228–236 quiz 237-8.
[4] G.F. Tan, et al., Causes and features of erythroderma, Ann. Acad. Med. Singapore 43 (8) (2014) 391–394.
[5] T. Tagami, et al., Paraneoplastic dermatosis in a patient with anaplastic large-cell lymphoma: case report and literature review, Dermatopathology (Basel) 3 (2) (2016) 39–43.
[6] R. Vyas, J. Selph, M.R. Gerstenblith, Cutaneous manifestations associated with melanoma, Semin. Oncol. 43 (3) (2016) 384–389.
[7] Z. Ahmad, W. Jaffe, N. Craven, Gyrate erythema and melanoma-a new clinical association, Ann. Plast. Surg. 71 (6) (2013) 631–633.
[8] L. Tognetti, et al., Grover's disease and cutaneous melanoma: a fortuitous association or a paraneoplastic case? G. Ital. Dermatol. Venereol. 150 (6) (2015) 756–758.
[9] S. Pintova, et al., Sweet's syndrome in a patient with metastatic melanoma after ipilimumab therapy, Melanoma Res. 23 (6) (2013) 498–501.
[10] P. Jaju, et al., Invasive melanoma in a patient with congenital ichthyosiform erythroderma, Pediatr. Dermatol. 34 (1) (2017) e35–e36.
[11] S. Yasar, et al., Development of multiple non-melanoma skin carcinomas in a patient with non-bullous congenital ichthyosiform erythroderma, J. Dermatol. 38 (8) (2011) 813–816.
[12] M.G. Bouwhuis, et al., Autoimmunity and treatment outcome in melanoma, Curr. Opin. Oncol. 23 (2) (2011) 170–176.
[13] U. Bottoni, et al., Association between autoimmune disease and cutaneous melanoma with regard to melanoma prognosis, Clin. Exp. Dermatol. 40 (3) (2015) 254–259.
[14] H.S. Sandhu, A.M. Kolomeyer, M.K. Lau, C.L. Shields, L.M. Schuchter, C.W. Nichols, T.S. Aleman, Acute exudative paraneoplastic polymorphous vitelliform maculopathy during vemurafenib and pembrolizumab treatment for metastatic melanoma, Retin Cases Brief Rep. (2017 Jun 13), https://doi.org/10.1097/ICB.0000000000000604 [Epub ahead of print].
[15] F. Brunet-Possenti, et al., Antiepidermis autoantibodies induced by anti-PD-1 therapy in metastatic melanoma, Melanoma Res. 26 (5) (2016) 540–543.