The Improvement of Stewart-Treves Angiosarcoma

through IL-23p19 inhibition

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Introduction

Stewart-Treves syndrome (STS) is a rare cutaneous angiosarcoma with very poor prognosis. Only early surgery with wide margins, which can include amputation or disarticulation, provide higher chances of survival. Median survival is estimated at 7 months.

Figure 1 - Clinical presentation of Stewart-Treves syndrome when patient before the first guselkumab injection.

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Erythematous plaque with coalescent purple bullous easy-to-bleed lesions on her left forearm surrounded by satellite hematic blebs (arrow).



Case report

We report the case of an 87-year-old white woman presenting with a large angiosarcoma confirmed by intralesional biopsy (Figure 1).

As the currently available therapeutic options (amputation, radiotherapy and chemotherapy treatments) were not feasible due to the patient's general condition, we sought out other options. Based on recent literature describing high levels of IL-23 within this type of tumour, an off-label compassionate use of guselkumab, an anti-IL-23 cytokine antibody, was considered in order to reduce tumour volume. After authorisation from the ethics committee to authorize the compassionate off-label use of guselkumab, Tremfya[®] (Janssen, Belgium) was administered by subcutaneous injection (100mg) once a month for three months. Pronounced clinical improvement was observed (Figure 2 after 2 and Figure 3 after 3 months). The only side effects of the treatment were headache and fatigue lasting a few days following the injections.

Figure 2 - Clinical presentation of Stewart-Treves syndrome after 2 months of guselkumab treatment.

Lesion was dryer and the bleeding tends to disappear.



Discussion

To date, the pathogenesis of Stewart-Treves syndrome remains unknown. Yoshida et al. recently published immunohistochemical analyses of two Stewart-Treves angiosarcoma. They detected a significant quantity of IL-23-producing cells and IL-17-producing cells at the edge of the tumour.

Immunohistochemical research in skin cancer has shown that IL-23 could play one

of the most important roles in the pathogenesis of tumours.

Therefore, we proposed guselkumab, a monoclonal antibody targeting IL-23p19,

as a treatment option to reduce tumor volume of our patient. Guselkumab is a well-established treatment for psoriasis. Adverse effects are limited and well reported.

Unfortunately, our patient died three months following her diagnosis, due to general physical deterioration in this geriatric and oncologic context. However, guselkumab treatment led to a better quality of life during her last months of life.

Figure 3 – Clinical presentation of Stewart-Treves syndrome after 3 months of guselkumab treatment.

The lesions had substantially decreased in size and the drip bleeding had disappeared. The satellite nodule was dryer and smaller (arrow).



Conclusion

We report the innovative use of guselkumab injections for the

treatment of Stewart-Treves syndrome. Recent data on IL-23 implication

in STS led us to propose this unconventional treatment to our patient.

Within three months, pronounced clinical improvement was observed.

This case is unusual and further investigations are necessary to prove the

efficacy of this IL-23p19 inhibitor in STS. Our case reveals a potential

novel therapeutic target for the treatment of Stewart-Treves angiosarcoma.