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TOCILIZUMAB FOR THE TREATMENT OF A CASTLEMAN'S DISEASE PAEDIATRIC PATIENT: A CASE REPORT

E. Nogué Pujadas¹, J.A. Esteban González¹, M. Bosch Peligero¹, M. Mendez Hernandez², X. Bonafont Pujol¹.

¹University Hospital Germans Trias i Pujol, Pharmacy Department, Badalona, Spain.

²University Hospital Germans Trias i Pujol, Paediatrics Department, Badalona, Spain.

Background

Castleman's disease (CD) is a heterogeneous group of lymphoproliferative disorders associated in a subset of cases with the human immunodeficiency virus (HIV) and human herpesvirus-8 (HHV-8). CD comprises at least two distinct diseases (unicentric and multicentric) with different approaches. There are currently no licensed drugs for the management of CD in paediatric patients.

Purpose

To describe a paediatric case of CD in which an antagonist of the interleukin (IL)-6 receptor (tocilizumab) was successfully used.

Material and Methods

Retrospective case report and literature search related to the treatment of CD. The information was obtained from electronic medical records, PubMed and Uptodate.

Results

An 11-year-old boy (weight 32.2kg, p3-10) was admitted to paediatric ward for study of asthenia, anorexia and body weight loss of 5-month evolution

Laboratory test values at the time of admission included:

- Hb 9.8mg/dL
- MCV 76.2fL
- CRP 65mg/L
- ESR 90mm
- Transferrin saturation 7%
- Renal and liver function tests: within normal limits

The screening of several | infections included:

- Epstein-Barr virus
- Cytomegalovirus
- Tuberculosis
- HHV-8
- HIV

All with negative results

It was detected an alteration of IL-6 (54pg/mL)

PET-SCAN showed multiple adenopathies suggesting a lymphoproliferative syndrome.

The adenopathies' biopsy

The adenopathies' biopsy was not conclusive

having excluded other pathologies, the case was oriented as a MULTICENTRIC CASTLEMAN'S DISEASE due to the symptoms and analytic results

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Given the age of the patient, treatment with TOCILIZUMAB 8mg/kg/month was initiated despite not being licensed for its use in CD After 5 doses of treatment, the patient presented clinical improvement
but acute phase reactants were still high and symptoms reappeared a few days before the next dose

As a consequence, the frequency of the treatment was reduced to 8mg/kg/21 days

After a year of treatment, the patient showed a great response with standardization of acute phase reactants

(CRP 0.2mg/L, ESR 6mm) and body weight gain of 22kg (current weight: 54kg)

Conclusions

In this case of a paediatric patient with CD, the offlabel use of tocilizumab showed to be safe and effective when reducing the intervals of administration to 21 days. Nevertheless, more studies are needed to demonstrate its efficacy and safety profile.



