THIONAMIDE – INDUCED AGRANULOCYTOSIS: CASE SERIES FROM A PERUVIAN HOSPITAL
Victor Hugo Noriega Ruiz1, Ruben Cruz Reyes1, Jaime Villena Chavez1, Miluska Huachin Soto1
1 Hospital Cayetano Heredia

Introduction: Agranulocytosis represents the rarest and rarest complication of treatment with thionamides (rate 0.11%-0.35%). Most events occur within 90 days of treatment. Associated risk factors are: age, re-exposure and dose ≥ 30 mg/dl. Objective: To describe general characteristics of in-patient thionamide-induced agranulocytosis (TIA) of a third level hospital. Methods: Descriptive-retrospective study, type series of cases. We reviewed medical records of patients diagnosed with TIA between 2009-2017. Demographic, management (dose, frequency, re-exposure) and hospitalization data were recorded, creating a database and subsequent analysis in SPSS v18. Central tendency and dispersion measures were determined. Results: We found 11 cases of TIA, all of them Grave’s disease (100%). The mean age was 32.6 ± 12.2 years and 64.6% being female. Disease duration was 7 ± 5.8 days. Most frequent presenting symptoms were fever (90.9%) and odynophagia (81.8%). At admission, SBP 110.2 ± 17.3 mm Hg, DBP 64.5 ± 12.1 mmHg, heart rate 110.3 ± 15.9 bpm and T° 37.9 ± 1.1 C° were recorded. On thyroid exam, all with diffuse goiter, 72.7% with WHO classification II and estimated weight of 74.6 ± 18.1 gr. In laboratory results, TSH 0.03 ± 0.4 mUI/ml, T4f 2.66 ± 1.6, leukocytes 1227.9 ± 2068/mm³ and granulocytes 241.6 ± 151.7/mm³. Methimazole mean dose was 28.2 ± 10.8 mg/d and all received 386.2 ± 716.7 days prior to the event. In the management, 81.8% was treated with lithium carbonate, 18.2% Lugol and all (100%) colony-stimulating factor (FSC) for 6.3 ± 2.7 days associated with beta-blockers, achieving resolution in 7.6 ± 2.7 days. Discussion: Grave’s disease (GD) is the most frequent cause of hyperthyroidism, with high rates in female patients, iodine-sufficiency areas and young adults (30-60y), explaining higher thionamides use and adverse effects in these groups. Dose ≥ 30 mg/dl carry a higher TIA risk, however, in recent studies risk increases from 25 mg/dl. 85% of TIA’s occur within 90 days of treatment, but may occur up to 12 months. Presenting symptoms were similar to previous reports. On exam, tachycardia could be secondary to fever or to decompensated hyperthyroidism, corroborated by finding in hormonal test. Lithium carbonate, beta-blockers and FSC were used in most cases, Lugol being the less used. Resolution time was lower than previous local report probably by standardization of FSC use.

CONSUMPTIVE HYPOTHYROIDISM: CASE REPORT OF HEPATIC HEMANGIOENDOTHELIOMAS SUCCESSFULLY TREATED WITH VINCRISTINE AND SYSTEMATIC REVIEW OF THE SYNDROME
Marina Weber Pasa1, Rafael Selbach Scheffel1, André B. Zanella1, Ana Luiza Maia1, José Miguel Dora1
1 Universidade Federal do Rio Grande do Sul (UFRGS)

Introduction: Consumptive hypothyroidism syndrome (CHS) is a severe form of hypothyroidism due to the high expression of thyroid hormone-inactivating enzyme type 3 deiodinase (D3) in tumoral tissues. Although the CHS has been initially noted in neonates and children with vascular tumors, it is not restricted to this age group nor this kind of tumors. To date, there is no summarized data of CHS on literature. Objectives: Describe one case of CHS and provide a summarized description of the cases reported to date. Methods: A case of CHS cured after treatment of hepatic hemangioendotheliomas is reported. A systematic review of the databases PubMed/Medline and Embase, using the term “Consumptive AND Hypothyroidism” was performed. From the 33 selected references, we extracted 42 case reports of CHS. A summarized description of the CHS clinical characteristics, treatments and outcomes was provided. Results: Here, we present the case of a seven-month-old female patient with a diagnosis of massive hepatic hemangioendotheliomas. After treatment with high doses of thyroid hormones, our patient started tumor-directed chemotherapy with vincristine. The tumor displayed excellent response, and euthyroid status was regained. Our systematic review summarizes 42 CHS cases reported: 36 children and 6 adults. The laboratory profile at diagnosis displayed high TSH and low T4 and T3 serum levels. The serum reverse T3 (rT3) and D3 activity levels were high in all patients tested. In children, 97% were vascular tumors, with only one case of a solid tumor – a fibrosarcoma; whereas in adults, 33% were vascular tumors, 33% fibrous tumors and 33% gastrointestinal stromal tumors. The conservative treatment was predominant in children, while in adults all cases were treated with surgery. Death occurred in 16% of children and 33% of adults. Conclusion: The CHS is a rare form of hypothyroidism, that occurs in children and adults, usually linked to hepatic vascular tumors. The condition is associated with high lethality. Prompt diagnosis and institution of appropriate high dose thyroid hormone replacement and tumor-directed therapy are the keys to optimize outcomes.