

## Reidel's thyroiditis and Tolosa-Hunt syndrome, a rare association

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### Abstract

A previously undescribed association of Reidel's thyroiditis with Tolosa-Hunt syndrome is reported. A 33-year-old Caucasian female presented with a two-year history of painful progressive goitre, histologically a Reidel's thyroiditis. She underwent left partial thyroidectomy. Post-operatively she developed severe right retro-orbital pain, right temporal headache and a partial right oculomotor nerve palsy. A diagnosis of Tolosa-Hunt syndrome was made. Some important aspects of this case are discussed.

**Key words:** Thyroiditis; Tolosa-Hunt Syndrome

### Case report

A 33-year-old Caucasian female presented with a two-year history of progressive painful goitre and hypothyroidism. Incisional biopsy of the goitre showed characteristics of Reidel's thyroiditis. She underwent a left hemithyroidectomy and in the recovery room she developed transient severe right retro-orbital pain and a right temporal headache which settled gradually in six hours. Three weeks later the pain recurred with a more severe intensity associated with diplopia and drooping of the right upper eyelid diagnosed by an ophthalmologist as ophthalmoplegia affecting the right oculomotor nerve. A neurological diagnosis of Tolosa-Hunt syndrome was strengthened by the fact that the pain and the ophthalmoplegia resolved after treatment with prednisolone. The magnetic resonance image (MRI) showed evidence of inflammation and thickening of the lining of the right cavernous sinus and partial obliteration with intimal thickening of the intra-

cavernous portion of the internal carotid artery consistent with Tolosa-Hunt syndrome.

### Discussion

Tolosa<sup>1</sup> first described in 1954 a disease process affecting the intracavernous portion of the internal carotid artery and the contents of the cavernous sinus. His patient underwent a surgical exploration of the parasellar region, which was macroscopically unremarkable, but the patient died post-operatively on the third day. The autopsy report revealed granulomatous inflammation of the affected carotid artery and cavernous sinus.

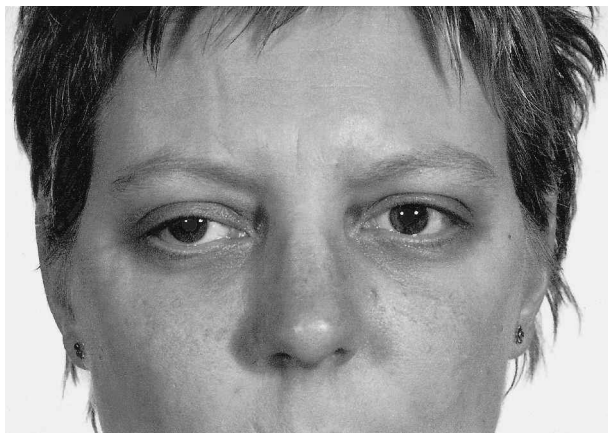


FIG. 1

Ptosis on the right side secondary to partial IIIrd nerve palsy.



FIG. 2

MRI scan showing the thickening of the wall of the cavernous sinus and narrowing of the internal carotid artery on the right side.

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TABLE I  
DIAGNOSTIC CRITERIA FOR TOLOSA-HUNT SYNDROME

Orbital pain	Episode or episodes of unilateral orbital pain for an average of 8 weeks if untreated
Cranial nerve palsy	Association with paralysis of one or more of the IIIrd, IVth and VIth cranial nerves which may coincide with the onset of the pain or follow it by a period of up to 2 weeks
Response to corticosteroid	Pain is relieved within 72 hours after the initiation of the corticosteroid therapy
Imaging	Exclusion of other causative lesions by neuroimaging and (not compulsory) carotid angiography.

In 1961 Hunt<sup>2</sup> further characterized this syndrome by reporting the therapeutic efficacy of systemic corticosteroids with prompt and dramatic improvement of the signs and symptoms.

- **Tolosa-Hunt syndrome is characterized by the formation of granulation tissue in the anterior cavernous sinus or superior orbital fissure, producing a painful ophthalmoplegia**
- **This paper presents a patient who developed this syndrome shortly following resection of the thyroid for Reidel's thyroiditis. It has been previously reported in patients with Hashimoto's syndrome and autoimmune polyglandular syndrome**
- **The authors conclude that Tolosa-Hunt syndrome may be associated with Reidel's thyroiditis but this conclusion is necessarily speculative and no objective evidence is available to support their claim**

In 1966, Smith and Taxdal<sup>3</sup> were the first to apply the eponym 'Tolosa-Hunt syndrome' and stressed that the dramatic response to steroids in 48 hours allowed the prompt differentiation of these cases from other conditions.

The International Headache Society<sup>4</sup> has hereby set the guidelines for this syndrome as follows:

- (1) *Description*: Episodic orbital pain associated with paralysis of one or more of the IIIrd, IVth, or VIth cranial nerves, which resolves spontaneously but may relapse and remit.
- (2) *Diagnostic criteria*: (Table I).

Other clinical manifestations reported in this syndrome include disturbances in sensory perception of the ophthalmic and maxillary divisions of the Vth nerve and in rare instances of some other sites such as the IIInd nerve and VIIth nerves.<sup>6-11</sup>

Our case satisfied the above criteria and the diagnosis of Tolosa-Hunt syndrome was made<sup>4,5</sup> with the MRI scan demonstrating a thickening of the wall of the involved cavernous sinus and a narrowing with intimal thickening of the wall of the intra-cavernous portion of the internal carotid artery<sup>12,13</sup> with an absence of any accountable structural lesion. There was also a dramatic response to a course of high dose of steroids with resolution of the pain instantly and a gradual resolution of her ophthalmoplegia.<sup>3</sup>

Reidel's thyroiditis is a rare condition of unknown aetiology characterized by a focus of dense fibrosis that completely replaces the affected part of the thyroid. Obliterative vasculitis affecting both veins and arteries is usually prominent in the fibrotic area and there are chronic non-specific inflammatory changes around the lesion. This disease is sometimes associated with multifocal fibrosis<sup>14</sup> and has been reported to involve the retroperitoneal area, mediastinum, superior vena cava and superior orbital fissure (orbital pseudotumour).<sup>15</sup>

The aetiology of Tolosa-Hunt syndrome remains unknown. No information is available as to what triggers the inflammatory process in the region of the cavernous sinus or superior orbital fissure. It seems that the syndrome falls within the range of idiopathic orbital inflammation (pseudotumour).<sup>16</sup> This non-specific inflammation typically causes an orbitopathy, if it is located more posteriorly it affects the cavernous sinus producing the Tolosa-Hunt syndrome.<sup>17</sup>

There has been one report of Tolosa-Hunt syndrome with Hashimoto's thyroiditis<sup>18</sup> and another report of Tolosa-Hunt syndrome with autoimmune polyglandular syndrome type 111 (diabetes type 1, Basedow disease, alopecia).<sup>19</sup> The histological features of the Reidel's thyroiditis and the close temporal association of the surgery for Reidel's and the Tolosa-Hunt syndrome together with the MRI evidence of the inflammation of the cavernous sinus in this patient lead us to conclude that there is an association between Tolosa-Hunt syndrome and Reidel's thyroiditis.

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