Case Report Recurrent Tolosa Hunt Syndrome, After Cervical Spine Surgery: Case Report

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Citation Daneshi A, Masoudi O, Darvishnia S, Mohajeri SMR, Ghazvini H. Recurrent Tolosa Hunt Syndrome, After Cervical Spine Surgery: Case Report. Iran J Neurosurg. 2025; 11:E6. http://dx.doi.org/10.32598/irjns.11.6

doi http://dx.doi.org/10.32598/irjns.11.6

Article info:

Received: 14 Jun 2024 Accepted: 25 Dec 2024 Available Online: 09 Mar 2025

Keywords:

Tolosa hunt syndrome (THS), Cranial nerve palsy, Laminectomy, Ophthalmoplegia, Surgical site infection (SSI)

ABSTRACT

Background and Importance: Tolosa hunt syndrome (THS) is a rare ophthalmoplegia that occurs in one person per million people. Clinical presentation, magnetic resonance imaging (MRI) findings, and a good response to steroids are used to diagnose THS.

Case Presentation: We present a 59-year-old man with right ophthalmodynia and ophthalmoplegia after cervical laminectomy and surgical site infection (SSI). After the first course of low-dose dexamethasone, the patient's symptoms relapsed within a short time. During the second course of treatment, high-dose dexamethasone was administered. The symptoms were treated without relapse within one year.

Conclusion: Infection, such as SSI, can trigger THS. Also, high-dose dexamethasone and longer treatment periods can prevent short-term recurrence of THS.

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Highlights

Tolosa Hunt syndrome (THS) can occur in inflammatory conditions, such as surgical site infection (SSI).

 Higher corticosteroid doses and longer treatment durations may prevent the short-term recurrence of Tolosa Hunt.

More research is necessary to be conducted on the dose of corticosteroids and the duration of treatment.

Plain Language Summary

THS is a rare eye movement disorder. We present the case of a 59-year-old man with right-eye paralysis after cervical surgery. His symptoms persisted 20 days after surgery. He was treated with low-dose dexamethasone, and his symptoms resolved within a few days; however, the patient's symptoms relapsed within a short time. During the second course of treatment, high-dose dexamethasone was administered. The symptoms were treated without relapsing within one year. We suggest that high-dose dexamethasone and a longer treatment period can prevent the recurrence of THS symptoms.

1. Background and Importance

olosa Hunt syndrome (THS) is a rare syndrome characterized by eye pain. It occurs in one to two people per million people [1]. Diagnostic criteria include ophthalmodynia, paresis of the cranial nerves for days to weeks, and spontaneous improvement. Ophthalmoplegic attacks can recur for months or years [1, 2]. Cavernous sinus involvement can occur as granuloma [3].

THS responds well to steroids, but there is no consensus on dosage and duration. A patient's cranial nerve defect can remain permanent or recover for a long time [2].

On magnetic resonance imaging (MRI), isointense lesions in the cavernous sinus are seen on T1-weighted images and isointense to hypointense lesions on T2weighted images, which show increased enhancement after injection. To diagnose, MRI findings and patient history and eye pain that responds to steroids will be helpful [1, 4].

2. Case Presentation

A 59-year-old man of Hispanic race with a history of diabetes was hospitalized for cervical laminectomy. Two weeks after surgery, surgical site infection (SSI) occurred, and the patient was treated with clindamycin 600 milligrams (mg) every 6 hours. Five days after hospitalization, he experienced pain in the right eye and the right side of the head without nausea and vomiting.

Forty-eight hours after the pain started, the patient had ptosis of the right eye. The other cranial nerves were normal. Also, the pupils in both eyes responded to light. Forty-eight hours after ptosis, he developed right eye movement paralysis (frozen eyes with cranial nerve III, IV, and VI involvement). No problems were observed in biochemical, thyroid, and liver functional tests. After brain MRI, FLAIR hyperintensity in the right cavernous sinus was observed (Figure 1).

Suspecting THS, the patient was treated with lowdose dexamethasone (4 mg every 8 hours) for 5 days. The patient's eye pain partially improved 24 hours after receiving the first dose of dexamethasone. Two weeks after stopping dexamethasone, the patient's eye pain recurred. Therefore, high-dose dexamethasone was started (8 mg every 6 hours) and continued for 10 days. At follow-up, the patient's symptoms improved, and no new episodes occurred after one year. Only paresis of the right third cranial nerve remained.

3. Discussion

THS is a rare painful ophthalmological syndrome involving cranial nerves III, IV, VI, and V1 [1, 5]. Hunt described six patients with recurrent unilateral painful ophthalmoplegia and suggested that non-specific cavernous sinus inflammation can cause THS [6].

Because our patient was hospitalized for SSI, the syndrome was diagnosed earlier. Also, because THS is an inflammatory condition and our patient was admitted with SSI, it suggests that THS can be triggered by SSI



Figure 1. Flair T2 MRI showing hyperintensities in right cavernous sinus



MRI: Magnetic resonance imaging.

inflammation. On MRI, FLAIR hyperintensity of the right cavernous sinus was observed. Other studies reported that the soft tissue of the supraorbital fissure area was markedly enlarged [1, 5].

In the first treatment course with low-dose dexamethasone, the patient responded well to treatment, but his symptoms relapsed after two weeks. In the second course of treatment with high-dose dexamethasone, no recurrence of the syndrome occurred during the one-year follow-up. Herrera et al. treated a 17-year-old woman with one gram per day of methylprednisolone for 3 days, followed by 40 mg of prednisolone for 6 weeks [5].

Recurrent THS over a short period may be due to the low dose of dexamethasone in the first course of treatment. In other studies, short-term recurrence has not been observed.

4. Conclusion

Comparing the findings of our study with those of other research studies revealed that, a higher corticosteroid dose and a longer treatment period can prevent shortterm recurrence of the syndrome. In addition, THS can be triggered by various types of infections (SSI after surgery).

Ethical Considerations

Compliance with ethical guidelines

Written consent was obtained from all patients for participation.

Funding

Daneshi A, et al. Tolosa Hunt Syndrome After Cervical Surgery. Iran J Neurosurg. 2025; 11:E6.

This research did not receive any grant from funding agencies in the public, commercial, or non-profit sectors.



Authors' contributions

Conception and design: Abdolhadi Daneshi, Omid Masoudi, and Seyed Mohammad Reza Mohajeri; Data collection: Omid Masoudi, Seyed Mohammad Reza Mohajeri, and Hossein Ghazvini; Data analysis and interpretation: Omid Masoudi, and Seyed Mohammad Reza Mohajeri; Drafting the article: Omid Masoudi, Saina Darvishnia, and Seyed Mohammad Reza Mohajeri; Critically revising the article: Abdolhadi Daneshi; Reviewing submitted version of manuscript, and approving the final version of the manuscript: All authors.

Conflict of interest

The authors declared no conflict of interest.

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