

Venous pathology targeted surgical management in Hirayama disease: A comprehensive case series of nine cases exploring this potential etiology

ABSTRACT

Objective: Hirayama disease is a rare cause of cervical myelopathy predominantly affecting young individuals. The disease is classically characterized by muscle atrophy in the distal upper limbs. While various etiopathogenesis such as dural sac dysplasia, nerve root dysplasia, structural abnormalities of the spinal ligament, and venous dysplasia have been proposed, this study explores the potential role of venous pathology and surgical management on the basis of it.

Methodology: This is a prospective descriptive case series of nine cases. The diagnosis was made based on the Huashan diagnostic criteria which includes clinical manifestation, imaging, and electrophysiology. In cases where magnetic resonance imaging (MRI) failed to demonstrate engorged veins, a computed tomography (CT) venogram of the cervical spine was used as an imaging tool. All patients underwent cervical laminectomy and coagulation of the posterior epidural venous plexus with or without laminoplasty. All the patients were followed up regularly; clinical improvement and neck disability index were assessed.

Results: All nine patients were male and exhibited classical clinical features, electrophysiological abnormalities, and MRI findings except, in one patient where a CT venogram helped in establishing the diagnosis as the MRI was inconclusive. Postoperatively, all patients had neurological improvement and stabilization of the disease. All patients who underwent CT venogram and cervical spine X-ray in neutral and dynamic position demonstrated no recurrence of engorged venous plexus or significant instability except one patient developing kyphosis. One patient experiencing symptoms in the other limb underwent a second surgery.

Conclusion: This comprehensive case series strongly supports venous pathology as a potential etiology of Hirayama disease. Surgical management with laminectomy and venous coagulation with or without expansile laminoplasty has delivered consistent improvement in neurological outcomes and long-term disease stabilization without the restriction of movements and lesser complications. However, further research is warranted to elucidate the mechanism underlying cervical venous dilatation.

Keywords: Cervical myelopathy, cervical venous plexus engorgement, coagulation of venous plexus, Hirayama disease, laminectomy, venous pathology

INTRODUCTION

Hirayama disease was described in 1959 by a Japanese neurologist and is a well-known cause of lower cervical myelopathy in Asian countries.^[1] Hirayama disease is a very rare disease, and in a Japanese survey, the prevalence was estimated at 1/30,000 people.^[2] Several cases or small series of patients have been reported from various countries and regions, including India, North America, Europe, and Australia.^[3-6] Various etiopathogenesis have been proposed

DEEPAK NANKISHORE SHARMA, VAMSI KRISHNA YERRAMNENI, THIRUNAL YERRAGUNTA, GOVIND B. GAIKWAD, VASUNDHARA S. RANGAN, SASANK AKURATI
Department of Neurosurgery, Nizam's Institute of Medical Sciences, Hyderabad, Telangana, India

Address for correspondence: Dr. Vamsi Krishna Yerramneni, Department of Neurosurgery, Nizam's Institute of Medical Sciences, 5th Floor, Specialty Block, Hyderabad, Telangana, India. E-mail: vamsisky.ns@gmail.com

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Table of Contents Surgical Management Of Venous Disease

1. Understanding the eBook Surgical Management Of Venous Disease
 - The Rise of Digital Reading Surgical Management Of Venous Disease
 - Advantages of eBooks Over Traditional Books
2. Identifying Surgical Management Of Venous Disease
 - Exploring Different Genres
 - Considering Fiction vs. Non-Fiction
 - Determining Your Reading Goals
3. Choosing the Right eBook Platform
 - Popular eBook Platforms
 - Features to Look for in an Surgical Management Of Venous Disease
 - User-Friendly Interface
4. Exploring eBook Recommendations from Surgical Management Of Venous Disease
 - Personalized Recommendations
 - Surgical Management Of Venous Disease User Reviews and Ratings
 - Surgical Management Of Venous Disease and Bestseller Lists
5. Accessing Surgical Management Of Venous Disease Free and Paid eBooks
 - Surgical Management Of Venous Disease Public Domain eBooks
 - Surgical Management Of Venous Disease eBook Subscription Services
 - Surgical Management Of Venous Disease Budget-Friendly Options

6. Navigating Surgical Management Of Venous Disease eBook Formats
 - ePub, PDF, MOBI, and More
 - Surgical Management Of Venous Disease Compatibility with Devices
 - Surgical Management Of Venous Disease Enhanced eBook Features
7. Enhancing Your Reading Experience
 - Adjustable Fonts and Text Sizes of Surgical Management Of Venous Disease
 - Highlighting and Note-Taking Surgical Management Of Venous Disease
 - Interactive Elements Surgical Management Of Venous Disease
8. Staying Engaged with Surgical Management Of Venous Disease
 - Joining Online Reading Communities
 - Participating in Virtual Book Clubs
 - Following Authors and Publishers Surgical Management Of Venous Disease
9. Balancing eBooks and Physical Books Surgical Management Of Venous Disease
 - Benefits of a Digital Library
 - Creating a Diverse Reading Collection Surgical Management Of Venous Disease
10. Overcoming Reading Challenges
 - Dealing with Digital Eye Strain
 - Minimizing Distractions
 - Managing Screen Time
11. Cultivating a Reading Routine Surgical Management Of Venous Disease
 - Setting Reading Goals Surgical Management Of Venous Disease
 - Carving Out Dedicated Reading Time
12. Sourcing Reliable Information of Surgical Management Of Venous Disease
 - Fact-Checking eBook Content of Surgical Management Of Venous Disease
 - Distinguishing Credible Sources
13. Promoting Lifelong Learning
 - Utilizing eBooks for Skill Development
 - Exploring Educational eBooks
14. Embracing eBook Trends
 - Integration of Multimedia Elements

- Interactive and Gamified eBooks

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