

Forestier disease: A rare cause of an emergency tracheostomy

Ghalib Mohammed Al Sayed; Yahya Al Qahtani; Montasir Junaid*; Khalid Assiri

*Montasir Junaid

Department of Otorhinolaryngology, Armed Forces Hospital Southern Region, Khamis Mushayt, Saudi Arabia

Phone: 05 31 039 914; Email: montsj@gmail.com

Abstract

Diffuse idiopathic skeletal hyperostosis or Forestier disease is a relatively rare disease causing obstructive symptoms in elderly patients such as dysphagia which is the most common while symptoms of upper airway obstruction are rare finding and much rarer is the difficulty in intubation leading to tracheostomy. The disease is usually missed and treated symptomatically and being identified incidentally; the cause almost always is cervical osteophyte causing compressive symptoms.

We present one such case of an elderly male who ended up having tracheostomy after being identified with Forestier disease.

Keywords

Forestier disease; non-inflammatory disease; dysphagia

Introduction

Forestier disease or diffuse idiopathic skeletal hyperostosis (DISH) is a non-inflammatory disease leading to ossification and osteophyte formation in ligaments, tendons and fascia of the anterior spinal column [1]. First description of such pathology was by Mosher in 1926 who described a case of dysphagia secondary to cervical spine osteophyte [2], 1938 Iglauer reported excision of cervical osteophyte in a similar case [3] while Foriester coined the term ankylosing disease of spine or senile ankylosis hyperostosis (Forestier disease) for such cases [4]. Resnick and Niwayama in 1970 set a classification criteria requiring involvement of at least four continuous vertebral bodies, preserved intervertebral disc space, absence of apophyseal joints or sacroiliac inflammatory changes. They termed this condition DISH [5]. Other criteria by Julkunen and Ultsinger has also has been proposed for this condition [6,7]. DISH is usually seen in elderly while male to female ratio is 3:1 [8,9]. Clinical examination is usually unremarkable and patients are identified incidentally on routine neck X-rays performed for other reasons. Ossification and excessive calcific changes in longitudinal ligament at C-spine may result in dysphagia while dyspnea or dysphonia is a rare finding [9].

We report a similar case in which patient underwent emergency tracheostomy after trial of intubation failed. We believe there are only few case reports in literature with patient having difficulty intubation secondary to cervical osteophyte.

Case Presentation

A 73 years old diabetic, hypertensive male presented with history of back pain, on and off fever, inability to walk properly and progressive weakness of dorsiflexion of left foot. Patient was admitted to the hospital and diagnosed to have spondylodiscitis with brucellosis having subdural abscess at the level of L3 and L4 vertebrae on x-ray. So patient underwent decompression of the spinal canal and drainage of subdural abscess, and was intubated with some difficulty. In the post-operative period, ENT consult was generated to re-evaluate patient for mild difficulty in swallowing and snoring while patient denied any dysphonia, or difficulty breathing. Flexible fiber optic endoscope showed a significant bulge of posterior pharyngeal wall opposing the epiglottis but bilateral vocal cords were mobile. A CT scan of cervical spine showed marked compression effect of the larynx and the esophagus from the level of C2 to the level of C6 due to osteophytes formation 'double beak sign' (figures 1 and 2).

Plan was made to admit the patient in another occasion for the decompression of the C-spine osteophytes after the brucella infection had subsided. 8 weeks later, patient was re-admitted for cervical osteophytes decompression.

After routine pre-operative work up and informed consent from the patient, he was taken to the operating room for cervical osteophytes decompression by orthopedic spine surgeon. After trial of intubation which remained failed, ENT team was called upon for an emergency tracheostomy which was performed in usual fashion under local anesthesia. After securing the airway, spine surgeon proceeded for decompression of the osteophytes from C2 to C6 through right anterior neck approach. There were no intraoperative or post-operative complications apart from mild weakness of the right vocal cord which recovered completely in three weeks time. The patient was observed for two days in the ICU and after that he was shifted to the general surgical ward in which he received a regular tracheostomy care and feeding through the nasogastric tube. The tracheostomy tube was removed after one week of operation and the patient started on oral diet; progressing from liquid to soft and then regular diet. 1 week after, patient was discharged on 7th post-op day after achieving complete recovery. Patient was seen on first follow-up after four weeks and found to have complete recovery of airway obstruction and dysphagia. Now patient is on regular follow-up with spinal surgeon and no new ENT complains were identified.

Discussion

Forestier disease or DISH still remains a pathology of unknown origin but has been correlated with diabetes, hypercholesterolemia, gout and obesity. Though asymptomatic it may present with GIT, Respiratory, Neurological and spinal instability [9]. These symptoms are masked and treated only symptomatically till the time a radiological evidence is acquired for another reason [10].

Cervical and thoracic region are the most common affected sites while male being more affected presents in their 6th or 7th decade of life [8]. Differential diagnosis usually includes ankylosing Spondylitis and spondylosis deformans. Other conditions described in literature to be associated with DISH include gout, rheumatoid arthritis, pagets disease and chondrocalcinosis [11]. Due to nonspecific clinical symptoms radiological investigations are crucial to identify the disease such as plain cervical X-rays, Computed tomography (CT) and Magnetic resonance imaging (MRI). Speech, swallowing evaluation along with Upper GI and Respiratory tract endoscopies are routinely performed but with caution to rule

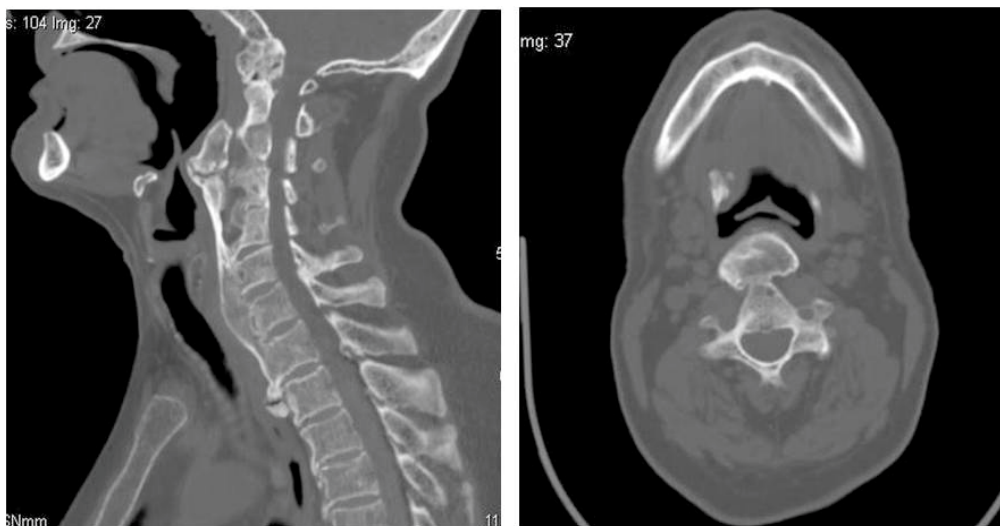
out lesions in the same [11]. No specific treatment is available till date to halt the progression of disease process [8]. The Treatment protocol can be broadly divided into conservative and surgical modalities, the former includes physical therapies (chiropractic, acupuncture) improving range of movements with spinal stiffness, pharmacological (NSAID, Muscle relaxant, Glucocorticoid bolus) and dietary modification have also been tried. Surgical intervention is reserved for failure cases; this usually includes procedures such as osteophyctomy and feeding tube insertions [1], while tracheostomy is reserved for cases in which intubation is difficult or not possible [12, 13, 14].

C1 and C7 osteophyctomy in recent literature is the only treatment which shows promising result in term of resolution of aero digestive symptoms when conservative treatment has failed; while complications may range from recurrent laryngeal nerve injury, Horner's syndrome, cervical instability to persistence of symptoms [14]. Osteophyctomy (cervical) approaches can be anterolateral, posterolateral, or transoral with later being aesthetically more pleasing but surgically more challenging in terms of exposure [15].

Conclusion

DISH/Foresteir Disease as a cause of dysphagia in elderly is well defined in the literature but is an unsuspected entity for difficult airways in operating room and could pose an airway threat to anesthesia team during intubation of elderly patients. We recommend that in such patients undergoing any procedure under G/A, back up tracheostomy plan should always be included while elderly patients with dysphagia should be evaluated for DISH and one must rule out DISH and cervical osteophytes in all patients with difficult airway in elderly patients.

Figures



Axial CT scan (Figure 1) & Sagittal CT scan (Figure 2) showing marked compression of larynx and the esophagus

References

1. Khairullah A, Shahrul H, Brito S. Diffuse idiopathic skeletal Hyperostosis: A rare cause of dysphagia. *Phillipp J Otolaryngol Head Neck Surg* 2014; 29(2):34-36.
2. Mosher HP. Exostoses of the cervical vertebrae as a cause for difficulty in swallowing. *Laryngoscope* 1926, 36:181-182.

3. Iglauer S. A case of dysphagia due to an osteochondroma of the cervical spine-oesteotomy-recovery. *Ann Otol* 1938;47:799-803.
4. Forestier J, Rotes-Querol J. Senile Ankylosing hyperostosis of the spine. *Ann rheum Dis* 1950, 9:321-330.
5. Resnick D, Niwayama G. Radiographic and pathological features of spinal involvement in diffuse idiopathic skeletal hyperostosis (DISH). *Radiology* 1976, 119:559-568.
6. Julkunen H, Heinonen OP, Knekt P, Maatela J. The epidemiology of hyperostosis of the spine together with its symptoms and related mortality in a general population. *Scand J Rheumatol* 1975;4:23-7.
7. Utsinger PD. Diffuse idiopathic skeletal hyperostosis. *Clin Rheum Dis* 1985;11:325-51.
8. Diore K, Facon D, Dupuch KM, Bobin S. An Underrecognized cause of dysphagia: Forestier's disease. *Fr ORL* -2005; 88:124-127.
9. Yurter A, Kaloostain E.P. Double cervical Bird Beak sign resulting in dysphagia. *Open journal of modern Neurosurgery* 2013,3, 63-65.
10. Constantoyannis C., Papadas T and Konstantinou D, diffuse idiopathic skeletal hyperostosis as a cause of progressive dysphagia: a case report. *Cases journal* 2008, 1:416.
11. Krishnarasa B, Vivekanandarajah A, Ripoll L, Chang E and Wetz R. Diffuse Idiopathic skeletal Hyperostosis (DISH) – A rare Etiology of Dysphagia. *Clinical Medicine Insights: Arthritis and Musculoskeletal Disorders* 2011;4 71-75.
12. Karkas AA, Schmerber SA, Gay EP, Chahine KN, Righini CA. Respiratory distress and vocal cord immobilization caused by Forestier's disease. *Otolaryngol Head Neck Surg.* 2008;139:327-8.
13. Papakostas K, Thakar A, Nandapalan V, O'Sullivan G. An unusual case of stridor due to osteophytes of the cervical spine: (Forestier's disease). *J Laryngol Otol.* 1999;113:65-7.
14. Carlson ML, Archibald DJ, Garner DE, Kasperbauer JL. Surgical management of dysphagia and airway obstruction in patients with prominent ventral cervical osteophytes. *Dysphagia.* 2011 Mar;26(1): 34-40.
15. Pulcherio Janaina Oliveira Bentivi, Velasco Cláudia Márcia Malafaia de Oliveira, Machado Rosane Siciliano, Souza Wallace Nascimento de, Menezes Daniella Rossi de. Forestier's disease and its implications in otolaryngology: literature review. *Braz. j. otorhinolaryngol.* [Internet]. 2014 Apr [cited 2017 May 08]; 80(2): 161-166.

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Authors Information: Ghalib Mohammed Al Sayed¹; Yahya Al Qahtani¹; Montasir Junaid^{1*}; Khalid Assiri²

¹Department of Otorhinolaryngology, Armed Forces Hospital Southern Region, Khamis Mushayt, Saudi Arabia

²Department of Orthopedics, Armed Forces Hospital Southern Region, Khamis Musayt, Saudi Arabia

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