

An Unusual Case of Diffuse Idiopathic Skeletal Hyperostosis without History of Any Metabolic Disease

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Received May 31, 2019; Accepted June 07, 2019; Published January 13, 2020

ABSTRACT

We present a 40 year old woman who suffering from chronic neck pain and dysphagia and shortness of breath. Lateral cervical x-ray and Cervical CT scan showed a large calcified anterior ligamentous mass in anterior part of body of C4, C5 and C6 vertebrae causing partial obstruction of esophagus. This patient was underwent surgery and the anterior neck calcification was removed and after a few days symptoms of dysphagia and dyspnea were resolved.

INTRODUCTION

Diffuse Idiopathic Skeletal Hyperostosis (DISH) is a rare systemic disorder that occurs in less than 10% of people over age of 50 and is very rare under age of 50 years [1,2] and male to female ratio is 2 to 1 [2,3]. It is characterized by continuous ossification of ligaments and entheses [2]. And thoracic spine is more involved than the cervical and lumbar spine [3]. The classification most commonly used for DISH today was proposed by Resnick and Niwayama [4-6]. DISH disease may be asymptomatic or with symptoms such as pain, limited range of spinal movements, dysphagia and increased susceptibility to unstable spinal fractures [7]. DISH tends to occur more in men and in those with diabetes, obesity, metabolic syndrome, gout, hypertension and advanced age [4,8-10]. Various causes have been described for DISH, including genetic causes such as Col6A1 gene defect [11] or non-genetic causes such as hypervitaminosis A can be causes of this condition [4,8-10]. DISH is a metabolic bone disease [11]. And it seems that IGF plays a role in the expansion of the DISH, Diabetes mellitus can also be an independent risk factor for DISH development [12-14]. Correlation between DISH and cardiovascular disease has also been reported [13]. DISH is a disease of obese and diabetic old people [13].

Cervical spine involvement and proliferative bone changes can lead to obstruction of respiratory tract and dysphagia [2].

CASE REPORT

The patient was a 40 year old woman suffering from chronic neck pain and dysphagia and shortness of breath. She was under investigation and imaging to diagnose digestive and respiratory diseases and pharyngeal disease that was healthy. But in the lateral cervical x-ray, a bone mass adhered to the anterior cervical vertebra was observed (**Figure 1**). This

patient was not obese (BMI=18.5) had no history of any rheumatoid, cardiovascular and metabolic disease and there was no evidence of disease in the studies. She did not have any neurological and motor disorder and sphincteric disorder. In the lateral cervical x-ray, a large calcified anterior ligamentous mass was observed in anterior part of body of C4, C5 and C6 vertebrae (**Figure 1**), causing partial obstruction of esophagus. In cervical MRI, in T1 view, isointense to hyperintense mass were observed on the anterior C4, C5, C6 and in T2 view, heterogeneous and isointense to hypointense mass was observed in the anterior cervical vertebrae (**Figure 2**). In thoracic spine and lumbosacral spine x-ray, bone mass was seen in anterior lumbar vertebrae (L2 and L3) (**Figure 3**), which had not caused any symptoms in the patient. This patient was underwent surgery and the anterior neck calcification was removed (**Figure 4**) and after a few days symptoms of dysphagia and dyspnea were resolved.

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Citation: Reihanian Z, Golsefid HN & Sigaroudi SNO. (2020) An Unusual Case of Diffuse Idiopathic Skeletal Hyperostosis without History of Any Metabolic Disease. J Neurosurg Imaging Techniques, 5(1): 245-247.

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Figure 1. In the lateral cervical x ray, a bone mass adhered to the anterior cervical vertebrae was observed.



Figure 2. In cervical MRI, in T2 view, heterogeneous and isointense to hypointense mass was observed in the anterior cervical vertebrae.



Figure 3. In and lumbosacral spine x-ray, bone mass was seen in anterior lumbar vertebrae (L2 and L3).



Figure 4. Patient was underwent surgery and the anterior neck calcification was removed.

DISCUSSION

DISH is a rare disease and it is more common in men [5] and is more than 50 years old [2]. Patients with DISH suffer from obesity and one of causes of this is impaired glucose tolerance [3,6,7]. The presence of cardiovascular disease is a feature of DISH [12,13], other diseases that can be seen with DISH include gout and hypertension [5]. All researches have argued that DISH is a metabolic disease. The patient surveyed in this report was a 40 year old, lean woman who had no evidence of metabolic and cardiovascular disease that here biochemical tests were normal and there was no metabolic, rheumatologic and cardiac disease in the examinations and studies. Depending on the symptoms and clinical features of DISH disease, this patient is different from all patients with DISH previously described. DISH Disease is common in over 50 year old men with obesity and metabolic diseases, while this patient was a 40 year old woman with no history and symptoms of a metabolic disease, with a large calcified anterior ligamentous mass at the C4-C5-C6 level with a compressive effect On the esophagus and the upper respiratory tract.

CONCLUSION

In patients with chronic dysphagia and chronic dyspnea, after examining digestive and respiratory systems, DISH, although rare, is a very important differential diagnosis for these symptoms and should be considered even if they do not have systemic DISH symptoms.

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