Rare disease

Ossification of the posterior longitudinal ligament in a Nigerian woman

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Ossification of the posterior longitudinal ligament (OPLL), though commonly reported among the Japanese population, has however been infrequently reported among other racial groups. It has not been reported among black Africans. A case of OPLL of the cervical spine seen in a 62-year-old Nigerian woman is hereby reported.

BACKGROUND

The posterior longitudinal ligament is located within the vertebral canal, extending from the axis and membrana tectoria at the upper cervical spine down to the sacrum.¹ It may, however, ossify affecting the cervical spine, especially at the levels C3–C5.

Ossification of the posterior longitudinal ligament (OPLL) has been extensively reported among the Japanese population and is sometimes called the ‘Japanese disease’.² It has also been commonly reported among non-Japanese Asians³ as well as in Caucasian Americans.^{4–5} It has also been rarely reported among American Blacks.^{2–6–7} There is, however, no documented report among black Africans.

A case of OPLL in a black African is hereby presented.

CASE PRESENTATION

A 62-year-old, non-obese housewife, initially presented to the rheumatology clinic in our teaching hospital in 2005. She was neither hypertensive nor diabetic. She presented then with a 6-month history of neck pain, neck stiffness and headache. Physical examination showed an elderly woman with tenderness over the cervical spine but no neurological deficit. Radiograph of the cervical spine showed straightening of cervical spine from muscle spasm without any bone changes. She was started on diclofenac, muscle relaxant (tizanidine) and referred for physiotherapy.

She did improve initially but then defaulted for follow-up. She, however, presented again after 2 years with worsening of her complaints and associated paraesthesiae in both hands. A repeat cervical spine radiograph showed the same changes as in her first visit but with additional findings of osteophytes at C4–C6 vertebrae, diagnostic of cervical spondylosis. She was treated as before.

She, however, defaulted again but came back after another period of 1½ years. Repeat cervical spine radiography showed OPLL of the cervical spine as well as presence of osteophytes at C3–C5 (figure 1). She was started on tramadol, naproxen and amitriptyline. She was also referred for physiotherapy and neurosurgical consultation. Surgery was, however, not contemplated as the patient declined.

INVESTIGATIONS

- ▶ Radiograph of the cervical spine
- ▶ fasting blood sugar 86 mg%.

DIFFERENTIAL DIAGNOSIS

- ▶ Ankylosing spondylitis
- ▶ diffuse idiopathic skeletal hyperostosis (DISH)
- ▶ cervical spondylosis.

TREATMENT

- ▶ Diclofenac
- ▶ tramadol
- ▶ tizanidine
- ▶ physiotherapy.

OUTCOME AND FOLLOW-UP

Pain was reduced with little change in neck mobility. Patient was eventually lost to follow-up.

DISCUSSION

OPLL is a multifactorial disease in which complex genetic and environmental factors interact.¹ Gene analysis has shown specific polymorphism that may be associated with OPLL in several collagen genes. Polymorphism in the nucleotide pyrophosphate gene involved in regulation of chondrocyte calcification may also be associated with OPLL.

Clinical presentation includes male preponderance and peak age at onset of the sixth decade. Manifestations may vary between the asymptomatic and neurological features such as paraesthesiae and even quadriplegia.⁸ OPLL has been associated with other medical conditions such as diabetes and obesity.⁹ It has also been associated with spondylosis, DISH and ankylosing spondylitis.¹⁰ Management is dependent on the severity of radiculopathy and especially myelopathy. Severity of myelopathy has been graded by two main scales – Nurick Scale which is a 6-point grading system¹¹ and a 17-point Japanese Orthopaedic Association Scale.¹² Spontaneous resolution of the OPLL and recovery

Figure 1 Ossification of the cervical spine and osteophytes at C3–C5.

of normal neurologic function can be expected with conservative treatment and education.¹³ Apparent neurological deficit may be an indication for surgery. The two major surgical strategies are anterior decompression with fusion and posterior decompression via techniques such as laminoplasty and laminectomy. The indications for anterior surgery include ossification of a small length of the posterior ligament. However, when there is multisegmental pathology or developmental canal stenosis, posterior decompression is indicated.¹⁴

The case presented is a female in her early 60s and has neurological features of paraesthesiae though without features of myelopathy. This rare condition is presented to raise awareness of OPLL in black Africans.

Learning points

- Rare disease unreported in black Africans.

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Competing interests None.

Patient consent Obtained.

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