INTRASPINAL ENTEROGENOUS CYST ASSOCIATED WITH SPONDYLOLISTHESIS AND SPINA BIFIDA OCCULTA

Report of a Case

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A lorry driver aged thirty-five had complained of progressive backache in the lumbo-sacral region for seven years. The pain radiated into the buttock and lateral aspect of the thigh and into the right foot. It was relieved slightly by rest but never completely disappeared. It was not exaggerated by coughing. Just before his first attendance further pain radiated down the back of the left leg. Systematic enquiry revealed occasional dysuria.

On examination the patient appeared to be in good general health although rather obese.

There was some increase in the lumbar lordosis and a "step" could be felt at the lumbo-sacral level. Movements of the spine were full and painless, and there was no local tenderness or muscle spasm. Straight-leg raising was to 90 degrees on both sides, and Laségue's sciatic stretch test was positive on both sides. O'Connell's femoral stretch test was negative. All leg reflexes were normal apart from some reduction of the right ankle-jerk. No motor or sensory impairment could be detected. Examination of the other systems revealed no abnormality.

Investigations—The chest radiograph was normal. Antero-posterior, lateral and oblique views of the lumbar and sacral spine showed a first-degree spondylolisthesis of the fifth lumbar vertebra on the first sacral vertebra, with a defect of the lamina and pars articularis but not of the body of the fifth lumbar vertebra (Figs. 1 and 2). The following laboratory results were obtained: haemoglobin 15.1 grams/100 millilitres; white cell count 6,900/cubic millimetre, with normal differential; erythrocyte sedimentation rate (Westergren) 3 millimetres in first hour; electrocardiograph normal; cerebrospinal fluid-globulins negative, protein 30 milligrams,
1 lymphocyte/cubic millimetre, no red blood cells. Myelography showed a round filling defect at the level of the third lumbar vertebra, indicating an intrathecal space-occupying lesion (Fig. 3).

**Treatment**—Conservative treatment with a Goldthwait support was initiated but the patient returned after two months complaining that he had obtained no relief. After preliminary myelography operation was undertaken. An intrathecal tumour about 2 centimetres in diameter was found lying in the cauda equina opposite the third lumbar vertebra (Fig. 4). It was shelled out easily from the cauda equina but was found to be attached to one nerve root. After removal of the tumour and closure of the dura, posterolateral spinal arthrodesis was performed at the lumbo-sacral level between the transverse process of the fifth lumbar vertebra and the ala of the sacrum on both sides.

**Pathology**—Macroscopically the specimen measured 2·4 by 1·9 by 1·5 centimetres (Fig. 5) and appeared to consist mainly of yellow adipose tissue with a few fibrous trabeculae and a soft pinkish nodule approximately 0·5 centimetre in diameter. Sections revealed a cyst lined with epithelium and containing mucin, similar to that contained within small groups of acinar glands in its wall. The cyst appeared to be derived from distension of one of these acini. The tissue was considered to be ectopic aberrant intestinal epithelium. A small ganglion was found in the neighbouring tissue together with some degenerate nerve fibres.

**Progress**—After operation the patient complained of numbness over the toes of the left foot but this resolved after three weeks. At first there was some urgency of micturition but after four days micturition became normal. At follow-up after one month the patient complained of pain over the right iliac crest—the donor site for the bone graft. After two months the patient still remained free of back pain. He then discarded the Fisher support worn since the operation and resumed wearing a Goldthwait support.

**DISCUSSION**

Although enterogenous cysts located in the mediastinum have received considerable attention (Gross, Neuhauer and Longino 1950), it was stated by Harriman (1958) that enclosed intraspinal enterogenous cysts are rare. He pointed out, however, as did Rhaney and Barclay (1959), that some of these intraspinal lesions may formerly have been misdiagnosed as teratoid tumours.

Enterogenous cysts, regardless of their location, are commonly associated with spinal abnormalities (Fallon, Gordon and Lendrum 1954; McLetchie, Purves and Saunders 1954). The patient reported here showed two such anomalies, spina bifida occulta and spondylolisthesis of the fifth lumbar vertebrae due to infraction of the pars interarticularis.

The association of posterior spina bifida in the lumbo-sacral region with an intraspinal enterogenous cyst appears to have been previously reported in only one case, that of Rhaney and Barclay in 1959. They described an infant with multiple congenital abnormalities including...
hydrocephalus, a short neck and an Arnold Chiari malformation, together with an intraspinal enterogenous cyst of the cervical cord and posterior spina bifida of the lumbo-sacral spine. These deformities were incompatible with life and the infant died after twelve hours.

Sacral spina bifida combined with congenital spondylolisthesis has been documented by Laurent (1958) and by Newman (1963). In the case reported here the spondylolisthesis was probably congenital and due to a deficiency in the pars interarticularis, possibly resulting from spina bifida of the fifth lumbar vertebra, with some horizontal flattening of the facets of the first sacral and fifth lumbar vertebrae (Newman’s group I). In spite of these associations, a thorough search of western literature has failed to reveal a case of spina bifida occulta, congenital spondylolisthesis and intraspinal enterogenous cyst all in the same patient.

The position of intraspinal enterogenous cysts, often ventral to the spinal cord or cauda equina, has aroused much speculation into their embryological formation. It was postulated by Rhaney and Barclay that these lesions result from abnormal separation of germ layers in early embryonic development; splitting of the notochord, with traction between a connected endodermal tube and neurectoderm, permits primitive gut cells to be drawn into an ectopic intraspinal location. The features of the case reported here, together with a spectrum of congenital abnormalities that have been associated with enterogenous cysts, further substantiate this theory. Some mediastinal cysts derived from the foregut have been shown by Gross et al. to be connected to the vertebral column by a strand of tissue termed a neurenteric band. Saunders (1943) reported the coexistence of anterior and posterior spina bifida, and Feller and Sternberg (1929) have demonstrated anterior spina bifida with the abdominal contents actually in contact with the spinal cord. The association of spinal abnormalities of varying types with enterogenous cysts has been recorded by numerous authors including Fallon et al. (1954), McLetchie et al. (1954) and Knight, Griffiths and Williams (1955).
Guillery (1937) reported a case in which two small enterogenous cysts were found, one dorsal and one ventral to the same vertebral body, and each with a funnel-like projection as if to communicate through the vertebra. To complete the spectrum, Harriman (1958) has described an intraspinal enterogenous cyst in the absence of congenital spinal anomaly.

The radiographic changes produced by spinal tumours were well described by Elsberg and Dyke (1934). In the present case, however, the plain films of the spine, although showing spina bifida occulta and spondylolisthesis, did not suggest an intraspinal lesion. The cystic lesion was located at the level of the cauda equina, an area where there is considerable space for expansion, and probably for this reason the effects of local pressure on the vertebrae were not detectable. Myelography, indicated by progressive clinical findings, revealed the space-occupying lesion that otherwise would have remained undiagnosed until changes appeared on the plain radiographs.

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SUMMARY

1. An enterogenous cyst lying in the cauda equina opposite the third lumbar vertebra, and associated with spina bifida occulta of the fifth lumbar vertebra and spondylolisthesis of the fifth lumbar on the first sacral vertebra, is described in a man aged thirty-five suffering from chronic low back pain and sciatica.
2. Current embryological theories concerning the formation of intraspinal enterogenous cysts from primitive gut cells are further substantiated by the features of this case.

REFERENCES


