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NEUROLOGICAL MANIFESTATIONS FOLLOWING PARTIAL EXCISION IN SPINAL MENINGIOMA: CASE REPORT
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NEUROLOGICAL MANIFESTATIONS FOLLOWING PARTIAL EXCISION IN SPINAL MENINGIOMA: CASE REPORT

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SUMMARY

Presented here is a 16-year-old girl who was referred on 30th January 1996 with diagnosis of cord compression with spastic paraplegia with sensory level at T7/T8. CT scan myelogam confirmed soft tissue density mass displacing cord to the left with no dye being seen beyond T3. Thoracic spine decompressive laminectomy was performed on 1st January 1996 at Nairobi West Hospital extending from T3 to T6 level, which revealed a fibrous haemorrhagic tumour. Histology showed meningioma (mixed fibrous type and meningoepitheliomatous type) with many psammoma bodies. She had a stormy post-operative period, with infection and wound dehiscence. This was treated with appropriate antibiotics and wound care. She was eventually rehabilitated and was able to walk with the aid of a walking frame because of persistent spasticity of right leg. She was seen once as an outpatient by author on 6th July 1996, she was able to use the walking frame, but the right leg was still held in flexion deformity at the knee. She was thus referred to an orthopaedic surgeon for possible tenotomy. She was able to resume her studies at the University ambulating using a wheel chair and walking frame. She presented with worsening of symptoms in 2001 (five years after her first surgery). MRI scan thoracic spine revealed a left anterolateral intradural lesion extending from T3 to T5 vertebral body level compressing and displacing the spinal cord. She had a repeat surgery on 6th March 2001 at Kenyatta National Hospital; spastic paraparesis and urinary incontinenece persisted. She also developed bed sores and recurrent urinary tract infections. She was followed up by the author and other medical personnel in Mwea Mission Hospital where she eventually succumbed in 2005, nine years after her first surgery. This case is presented as a case of incompletely excised spinal meningioma to highlight some of the problems of managing spinal meningiomas when operating microscope and embolisation of tumours are not readily available. Also the family experienced financial constraint in bringing the patient for regular follow-up, and getting access to appropriate antibiotics, catheters and urine bags.

INTRODUCTION

Spinal intradural extramedullary tumours account for two thirds of all intraspinal neoplasms annually represented by meningiomas and schwanomas, with the former accounting for 25% to 46% of all primary intraspinal tumours (1). Technically advanced imaging techniques, magnetic resonance imaging (MRI) and surgical procedures have brought significantly better clinical results in the last two decades (1,2). However follow-up on patients under 50 years of age is limited (3).

Spinal meningioma generally responds favourably to surgical excision and has a low recurrence rate (3) it constitutes 5.75% to 15% of all meningiomas (4-6). Meningiomas are second most common intradural tumours accounting for 25% to 32% of spinal neoplasms; secondly only to neurofibroma (4,7-9). About 75% to 80% are thoracic, 16% to 26% are cervical and 4% to 7% are lumbar (5,10,11). Sacral meningiomas are extremely rare. Extradural meningiomas are the most common in the spine where 15% incidence has been reported with as many as 14% of the reported tumours being in children (13,14).

Cushing and Eisenhardt (4) reported successful removal of spinal meningioma. All major series have reported good to excellent results on surgical excision and recurrence rates of 3% to 7% (1-4). However most patients included in these series were older than 50 years (3). Cohen – Gadol *et al* (15) reported recurrence rate of 22% (nine out of 40 patients) in patients younger than 50 years (control cohort) in who the resection of spinal meningioma was performed over the same period. The higher the recurrent rate among the younger patients was attributed to aggressive histological subtypes, longer follow up periods, greater prevalence of extradural extension and enplaque tumours.

Sloof *et al* (14) at Mayo clinic found that among 1,322 intraspinal tumours occurring in patients of all ages, 29% to be nerve sheath tumours and 22.5% to be meningiomas; a total of 85% of meningiomas and 62% of nerve sheath tumours were totally intradural. Nerve sheath tumours were more common in cervical and lumbar region and in males, whereas meningiomas were most common in the thoracic area approximately 80% of these tumours were in females. In contrast Fortuna and co-workers (23) found 136 nerve sheath tumours (10.9%) and 54 meningiomas (4.3%) among 1,242 paediatric tumours in collected report. A total of 63% of childhood nerve sheath tumours were in male and there was a similar preponderance with meningioma contrary to adult statistics. The duration of symptoms to discovery average between 9 to 12 months for these tumours in children, compared with several years in adults. Among children both this tumours were most likely to appear between the age of 10 and 15 years and both tumours were frequently associated with multiple neurofibromatosis (von Recklinghausen's) disease with children (24). Meningiomas in children presented first with pain then paresis dumbbell meningiomas have not been reported in children. Meningiomas often show calcification; nerve sheath tumours are not radiosensitive, there have been mixed experiences with radiation therapy in a few cases of spinal meningiomas as in children (25,26) as with their intracranial counter parts spinal meningiomas in this age group can show malignant degeneration.

CASE REPORT

The case presented is that of a 16-year-old girl who at the time of presentation was experiencing slight pain and numbness of both lower limbs associated with weakness of the legs, episodes of spasms in the legs and the right leg lagging behind while walking and being unable to know the position of the legs. The above symptoms started gradually but got worse in November 1995 (two months prior to referral to authors care). She was initially seen at a local district hospital from 10th January 1996 to 24th January 1996 where she was diagnosed to have cord compression with sensory motor loss at T4 with gonococcal infection. She was given Benzathine penicillin 2.4 mega units on 25th January 1996 and then was referred to a neurologist who in turn referred her to my care for urgent decompression.

She had spastic paraplegia with sensory level at T7/T8. The right lower limb was in persistent spastic flexion at the knee joint. CT myelogram of C6 to T12 showed blockage of contrast at T3 level and a soft tissue density mass displacing the spinal cord to the left. The mass was of relatively high density.

On 1st February 1996, an extensive decompressive laminectomy from C7 to T4 was done. An intradural extramedullary fibrous haemorrhagic lesion was found. Piecemeal excision was done. Attempts were made to find a plane of cleavage between the tumour and the grossly compressed spinal cord; but this was greatly hampered by the bleeding from the haemorrhagic tumour. There were also no facilities for embolisation of the tumour and no operating microscope was available. Thus it was not possible to completely excise the extradural tumour completely. Histology showed features of a meningioma (mixed fibrous type and meningoepitheliomatous type) with many psammoma bodies.

She had a stormy postoperative period, wound infection, wound dehiscence and urinary incontinence. She was managed with appropriate antibiotics, wound dressings, and subsequent secondary suturing of the wound. She was then rehabilitated by physiotherapy. She improved and was able to control her urine and ambulate using a walking frame. However spasticity of her right leg persisted. She was able to resume her studies at the University in September 1996 (14 months after presentation to a medical institution) using a wheel chair to facilitate her mobility.

She was followed up as an outpatient by the author who referred her to an orthopaedic surgeon because of persistent spasticity of the right lower limbs. He advised a course of physiotherapy and probably later calipers.

On 7th December 1998, she was referred back to the author because of severe spasticity of lower limbs and urinary incontinence. On 22nd January 2001 an MRI scan of thoracic spine showed a left anterolateral intradural extramedullary lesion extending from T3 to T5 vertebral body level causing cord compression. Thus she underwent a second decompressive laminectomy. There was extensive fibrosis from previous surgery making dissection and delineating the plane between the tumour and normal spinal cord difficult. However attempts were made to remove as much of the tumour as possible from the extradural space as well as intradurally. Post operatively she underwent a further course of physiotherapy but spastic paraparesis of lower limbs and stool incontinence persisted. She was discharged after about two months of in patient rehabilitation on a wheel chair, to continue with physiotherapy at home. However she developed bed sores and was admitted to the nearest district hospital where she eventually succumbed in 2005, nine years after the first surgery.

DISCUSSION

Spinal meningioma generally responds favourably to surgical excision and has a low recurrence rate (3). It constitutes 5.75% to 15% of all meningiomas (4-6). Meningiomas are the second most common intradural tumours accounting for 25% to 32% of spinal neoplasms second only to neurofibroma (4,7-9). About 75% to 80% are thoracic, 16% to 26% are cervical and 4% to 7% are lumbar (5,10,11). Sacral meningiomas are extremely rare.

Extradural meningiomas are the most common in the spine, where 15% incidence has been reported with as many as 14% of the reported tumours being in children (13,14).

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The patient presented was 16 years old at the time of first surgery. She presented numbness, spasms of the lower limbs and dragging of the right leg for a period of two months prior to presentation. The two months period of presentation of symptoms is rather short, for meningioma but this could be that the patient ignored the early symptoms and signs of cord compression. This could explain the delay in presentation. The symptoms never completely resolved because of incomplete excision of the tumour; but there was improved function and prevention of further deterioration of neurological function for five years after first surgery as she was able to resume her studies for two years. After the second surgery the paraplegia and urinary incontinence persisted and she was not able to resume her studies this time. She was in and out of hospital because of urinary tract infection and bed sores. She eventually succumbed to this complications four years after the second re-exploration.

Nadkarni *et al* (3) reported a case of a 45-year-old woman who was 27 years old at the time of her first surgery. She had a recurrence 18 years after initial surgery, a notably longer period than the other series (1,2,4).

Using statistical analysis, Mirimanoff *et al* (16) computed the progression free survival rate to be 63%, 45%, and 9% at 5, 10 and 15 years respectively after subtotal excision of meningioma. This was in contrast to recurrence free survival rates of 93%, 80% and 68% during respective periods following total excision.

Calcification of meningioma is another variable in tumour recurrence as it adds to incomplete removal of the tumour (3,7). In the patient presented here, the tumour was reported as mixed fibrous type and meningoepitheliomatous type with many psammoma bodies, which is the pathological variant most likely, associated with calcification.

Nadkarni *et al* (3) and Doita *et al* (18) reported similar cases of psammomatous spinal meningiomas, which recurred at 18 years and 33 years respectively after the initial surgery. In the second patient, the recurrent tumour was detected on a plain radiograph appearing as a calcified tumour at the same thoracic level as the site of the initial operation. In our patient

the tumour was reported to be of relatively high density probably indicating calcification.

Anterior dural attachment has also been suggested as an adverse variable (19). This appears to have been the case in our patient as the tumour was extending anteriorly at the first surgery. However, in the case reported by Nadkarni *et al* (3), this was not so as the tumour had lateral and posterior attachments at second surgery.

Extradural attachment of the meningioma has not yet been established as a cause of recurrence, but multiplicity of intraspinal meningioma is shown to increase the rate of recurrence (1,20,21). There was evidence of extradural extension in patients presented here.

Gezen *et al* (22) reported in a series of 36 patients, two recurrences over a mean follow-up period of nine years (range 2–15 years). Both patients were treated with radiotherapy. Radiotherapy is recommended when there is tumour recurrence early after removal of the tumour mass, when surgery is hazardous because of the tumour location or when there are risks involved in surgical management (22). Our patient did not undergo radiotherapy. The progressive deterioration of the patient's neurological status and subsequent complication of urinary tract infection and bed sores was basically due to incomplete excision of the meningioma; because of technical problems of the tumour being haemorrhagic at surgery. Also at the time of surgery, an operating microscope was not available as well as facilities for embolisation of tumour before surgery.

CONCLUSION

The case presented here was a 16-year-old girl who had subtotal excision of intradural extramedullary meningioma at T3 level. The tumour was mixed fibrous meningoepitheliomatous type with plenty of psammoma bodies. There was anterior extension as well as extradural attachment. She improved partially after the first surgery and resumed her studies. However, five years later she presented with deterioration of neurological manifestation and regrowth of the tumour as evidenced by subsequent imaging. She underwent a second decompressive laminectomy. However she did not improve after the second surgery. She had erratic follow up and developed complications of urinary tract infections and bed sores to which she succumbed to nine years after the first surgical intervention. The complications she succumbed to were preventable, but because of financial constraints the family experienced there was delay in seeking medical intervention.

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