ISSN: 0975-833X

Available online at http://www.journalcra.com

International Journal of Current Research
Vol. 8, Issue, 10, pp.40671-40675, October, 2016

INTERNATIONAL JOURNAL OF CURRENT RESEARCH

RESEARCH ARTICLE

EPIDURAL DORSOLUMBAR SPINAL METASTASIS OF CHORIOCARCINOMA A RARE CASE REPORT

***Dr. Milan K Senjaliya**

Department of Neurosurgery, B.J. Medical College and Civil Hospital, Ahmedabad

ARTICLE INFO

Article History:
Received 23rd July, 2016
Received in revised form
05th August, 2016
Accepted 12th September, 2016
Published online 30th October, 2016

Key words:

Choriocarcinoma,
Epidural metastasis,
Chemotherapy.

ABSTRACT

Aims: To present and review a rare case of metastatic choriocarcinoma in the dorsolumbar spine.
Methods: A 21-year-old woman presented with complains of acute sudden onset paraplegia since 15 days with past history of uterine dilatation and curettage before 1 month for abnormal products in uterus and vaginal bleeding and laparotomy for uterine perforation before 20 days. No report of biopsy was available. Dorsolumbar magnetic resonance examinations revealed epidural lesion at the level from D5 to L1 on posterior and right lateral side which was hypointense on T1 weighted image, hyperintense on T2 weighted image and shows post contrast enhancement with compression of spinal cord. Patient was operated for D5 to L1 laminotomy and biopsy from that lesion which was very vascular.
Results: Biopsy report revealed infiltration of invasive hydatidiform mole. Serum level of βHCG was 242958 miu/ml. Patient was further investigated for contrast enhanced computer tomography of thorax and abdomen showed nodular metastasis of lung with pleural effusion and ascites. Despite of chemotherapy and radiotherapy patient has paraplegia at present.
Conclusion: We have reported a rare case of dorsolumbar epidural metastasis and pulmonary metastasis of choriocarcinoma. Choriocarcinoma is a highly anaplastic malignancy derived from trophoblastic cells characterized by the secretion of human chorionic gonadotropin (βHCG) and early hematogenous metastasis. However, metastatic choriocarcinoma in the spine is extremely rare. Very few cases of metastasis in lumbar and/or sacral vertebra have been reported. Chemotherapy is treatment of choice for metastatic choriocarcinoma.

Citation: Dr. Milan K Senjaliya, 2016. "Epidural dorsolumbar spinal metastasis of choriocarcinoma a rare case report", *International Journal of Current Research*, 8, (10), 40671-40675.

INTRODUCTION

Choriocarcinoma is a malignant, trophoblastic cancer, usually of the placenta. It is characterized by early hematogenous spread. It belongs to the malignant end of the spectrum in gestational trophoblastic disease (GTD) (Rosenberg *et al*., 2008). Characteristic feature is the identification of intimately related syncytiotrophoblasts and cytotrophoblasts without formation of definite placental type villi. The prognosis is considered to be very poor, and metastases often develop early (Beşkonakli *et al*., 1998). The most common sites for metastasis are the vulvo-vaginal region, the lungs, the liver and the brain (Beşkonakli *et al*., 1998; Lurain, 1998). The spinal metastasis of choriocarcinoma is extremely rare (Naito, 2009). Four cases of metastasis to the lumbar vertebral column and 2 cases to the epidural space have been reported (Beşkonakli *et al*., 1998; Kuten *et al*., 1978; Lee *et al*., 2010; Menegaz *et al*., 2004; Naito *et al*., 2009 and Vani, 1993).

Corresponding author: Dr. Milan K Senjaliya
Department of Neurosurgery, B.J. Medical College and Civil Hospital, Ahmedabad

In this study, we report the case of a patient with multiple metastases to the lungs, the dorsolumbar spinal column, in epidural space.

Case Report

A 21-year-old woman was presented with complain of acute sudden onset weakness in both lower limbs for 15 days. Patient was nulliparous and had past history of uterine dilatation and curettage before1 month for abnormal products in uterus and vaginal bleeding at other hospital. Patient had complication of uterine perforation during curettage and operated for midline laparotomy and suturing of that perforation before 20 days at another hospital. After laparotomy, on 5th post operative day, patient developed sudden, acute onset bilateral lower limbs weakness. Weakness increased and patient developed paraplegia within one day. No report of biopsy was available. On neurological examination, we found, patient was conscious, alert and well oriented. Higher functions and cranial nerves examination was normal. Tone, power, sensation, reflexes and joint position sensations in bilateral upper limbs were normal.

Patient had decreased tone, grade 0 power, hypo reflexes, altered joint position sensation in bilateral lower limbs. Sensations for touch, pain, temperature and vibration were absent below spinal cord level D7 on examination. Patient was investigated for routine investigations in the form of blood investigations, x- rays, ECG, etc., which were normal. Dorsolumbar magnetic resonance examinations revealed long segment epidural lesion at the level from D5 to L1 on posterior and right lateral side which was hypo intense on T1 weighted image (image 1), hyper intense on T2 weighted image (image 2) and shows post contrast heterogeneous enhancement (image 3). There was mass effect in form of compression and displacement of lower dorsal spinal cord and conus towards left side with resultant significant cord edema secondary to the epidural lesion with clumping and aggregation of nerve roots.

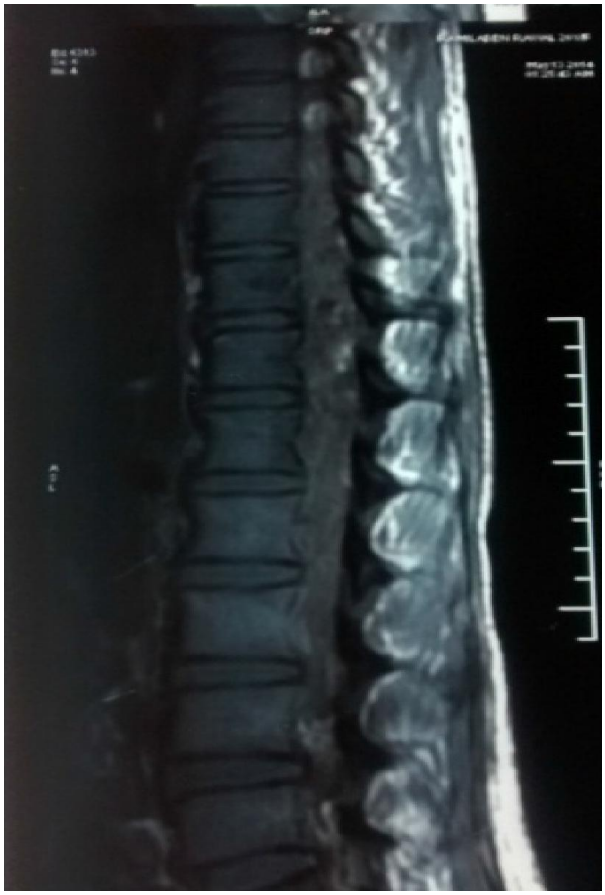


Figure 1. T1W sagittal view of MRI spine

Patient was operated for the spinal lesion. D5 to L1 laminotomy was done. Long segment epidural very vascular lesion was found on posterior and right lateral side of the cord. Partial excision of the lesion was done and biopsy was sent for frozen section report which was suggestive of invasive hydatidiform mole.

RESULTS

During post operative period patient remained paraplegic. Biopsy report was suggestive of infiltration of invasive hydatidiform mole. Histopathological picture revealed syncytiotrophoblasts, large multi-nucleated cells with eosinophilic cytoplasm, often surrounded the cytotrophoblasts, reminiscent of their normal anatomical relationship in chorionic villi. Cytotrophoblasts are polyhedral, mononuclear cells with hyper chromatic nuclei and a clear or pale cytoplasm. Then patient was further investigated for other metastasis in form of contrast enhanced computer tomography of thorax and abdomen (Image 4 and 5), which were suggestive of multiple subserosal lesions in uterus with multiple metastatic nodes in lungs, pleural effusion and ascites.

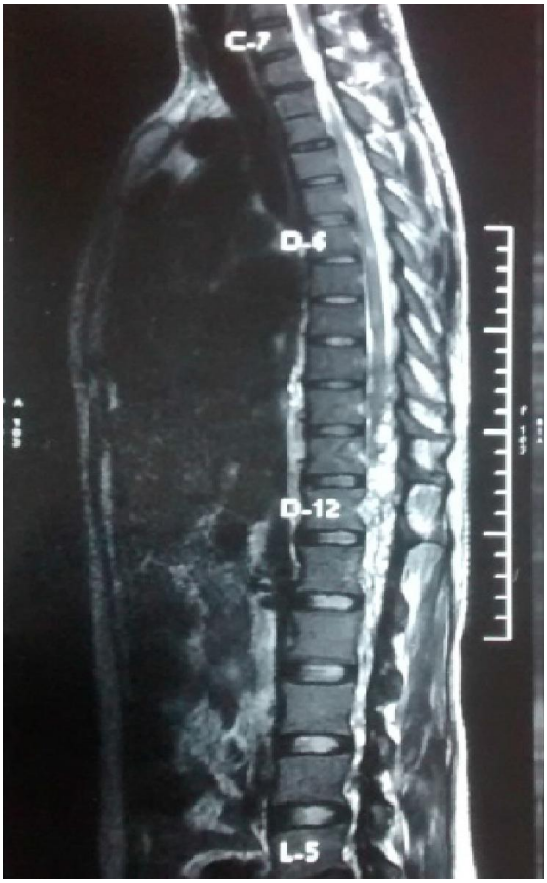


Figure 2. T2W sagittal view of MRI spine

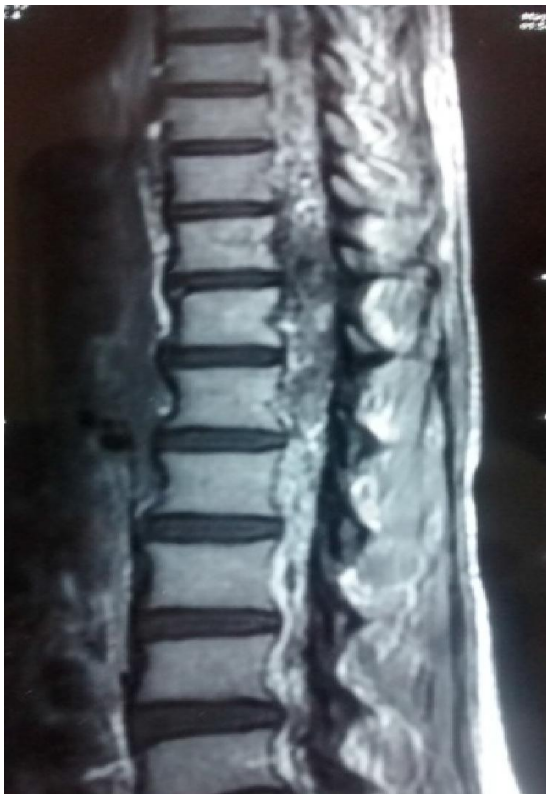
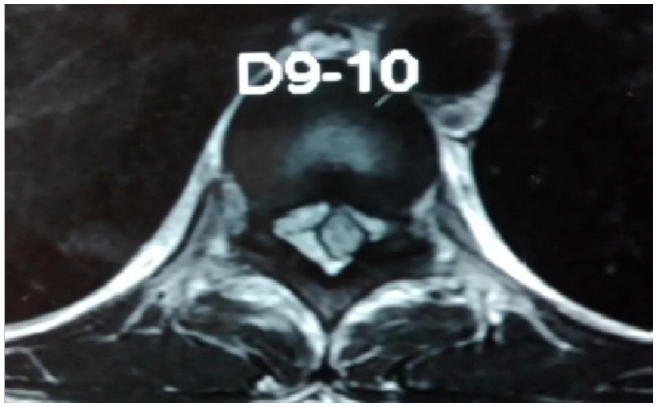
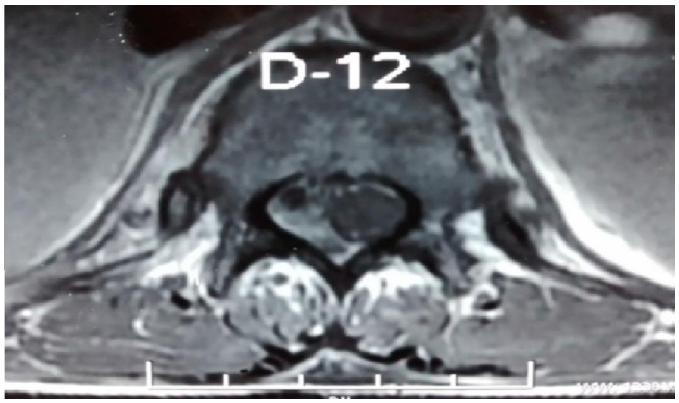


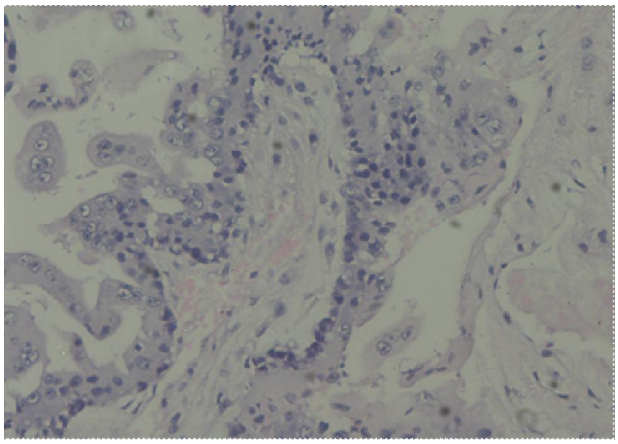
Figure 3. Post contrast sagittal view of MRI spine



Contrast T1 image



Axial T2 image



Histopathology picture of Choriocarcinoma

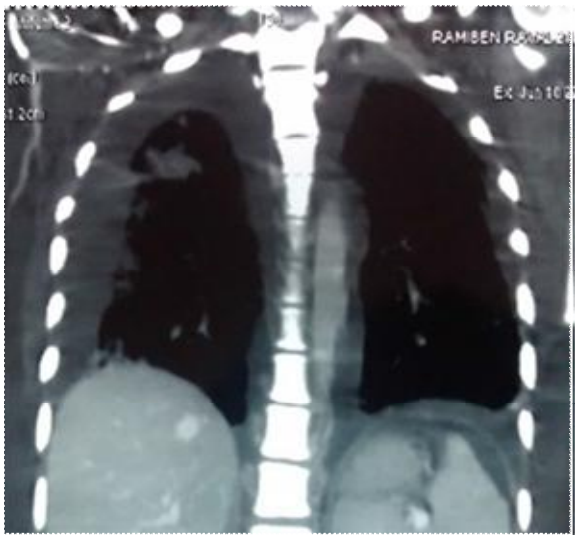


Figure 4. CECT Thorax

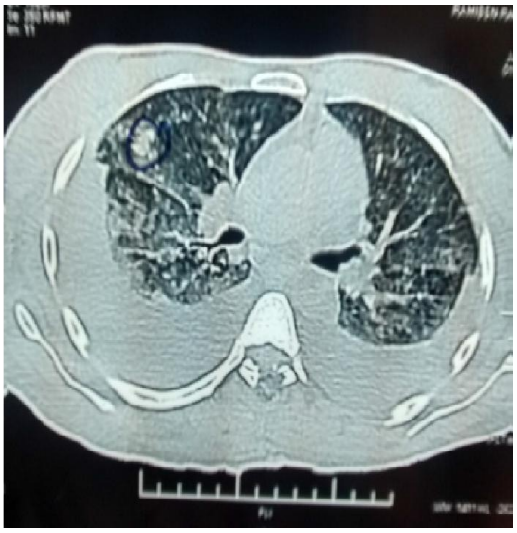


Figure 5. CECT Abdomen

Table 1. Comparison of various studies

Authors & year	Age (yrs), sex	Location of metastasis	Treatment	Prognosis
Beşkonakli et al., 1998	44, F	Thoracic epidural space	Surgery (decompression) with chemotherapy	Died during chemotherapy
Kuten et al., 1978	20, F	Lumbar epidural space	Surgery (decompression) with radiotherapy and chemotherapy	Tumor remission
Lee et al., 2010	33, F	L3 vertebral body, lumbar epidural space, lung, brain	Surgery (decompression with fusion) with chemotherapy	Chemotherapy continues
Menegaz et al., 2004	45, F	Lumbar vertebrae, sacrum, lung	Chemotherapy and radiotherapy	Died during chemotherapy
Naito et al., 2009	38, F	L2 vertebral body, lumbar epidural space, lung	Surgery (enbloc resection with fusion) with chemotherapy	Died during chemotherapy
Vani et al., 1993	27, F	Fifth sacrum, lung	Chemotherapy and radiotherapy	Worsen (follow-up loss)

Magnetic resonance examination of brain was normal. Serum level of human chorionic gonadotropin (βHCG) was 242958 miu/ml also support the diagnosis of metastatic invasive hydatidiform mole. Patient was treated with chemotherapy, EMA (Etoposide, Methotrexate, and Dactinomycin). Patient is paraplegic at present even after completion of chemotherapy.

DISCUSSION

Choriocarcinoma most often originates in the trophoblastic tissue of a hydatidiform mole but may also originate from the germinal epithelium of the testes, ovaries, or a normal placenta (Beşkonakli *et al*., 1998). Hydatidiform moles are thought to be benign, but choriocarcinomas develop in 1% of these patients (Beşkonakli *et al*., 1998 and Chandra *et al*., 1990). Early, hematogenous, and widespread metastasis is well documented (Beşkonakli *et al*., 1998; Lee *et al*., 2010 and Naito *et al*., 2009). Approximately 30% of patients with choriocarcinoma show metastases at the time of diagnosis. The favored sites of involvement are the lungs (94% of all metastatic choriocarcinoma), vagina (44%), liver (28%) and brain (28%), followed by the skin, gastrointestinal tract, kidney, breast, and bones (Beşkonakli *et al*., 1998; Lee *et al*., 2010). While metastasizing within the central nervous system, these tumors are known for their tendency to produce hematoma and intracranial hemorrhages, causing morbidity and mortality in these patients, but metastasis in spinal canal is very rare (Rosenberg *et al*., 2008). Although choriocarcinoma can follow any type of pregnancy, approximately 50% of the cases of choriocarcinoma are preceded by a hydatiform mole.

The remaining 50% are equally distributed between normal antecedent term gestational and abortion or ectopic pregnancy (Lurain *et al*., 1998; Naito *et al*., 2009; Scott *et al*., 1999; Seckl *et al*., 2010). The spinal metastasis of choriocarcinoma is very rare. Only 6 cases have been reported (Table 1). Despite improvements in treatment modality, the prognosis for these cases of spinal metastasis is unfavorable. Because these patients generally have a poor outcome within a few weeks after initiating presentation, early recognition and treatment of choriocarcinoma might enable a reduction the mortality rate. Surgery for a case of spinal metastasis of choriocarcinoma was firstly reported by Naito *et al*. in 2009 (Naito *et al*., 2009). Surgery is indicated when a massive mass effect is due to tumor mass or hematoma. In this situation, spine surgery such as spondylectomy or laminectomy is required to provide acute decompression or to control bleeding (Lee *et al*., 2010; Naito, 2009). Because choriocarcinoma is extremely hemorrhagic, it is quite possible the surgical procedures may have exacerbated the invasive and metastatic potential of this tumor. Thus, preoperative angiographic embolization, or at least angiography of the tumor should be used to reducing perioperative hemorrhages and evaluate vascularity of the tumor site, respectively (Lee *et al*., 2010; Naito, 2009). When central nervous system metastases are present, radiotherapy (whole brain irradiation) is usually given simultaneously with the initiation of systemic chemotherapy (Seckl, 2010; Lurain *et al*., 2011). Radiotherapy was performed as adjuvant treatment in four previous cases of spine metastasis of choriocarcinoma (Menegaz *et al*., 2004; Naito *et al*., 2009 and Vani *et al*., 1993). This therapy can also be utilized when spine metastasis are present (Menegaz *et al*., 2004; Naito *et al*., 2009). In our case, the patient was successfully treated by 6 courses of EMA-CO after laminectomy without radiotherapy. Choriocarcinoma is one of the malignant tumors most sensitive to chemotherapy (Bagshawe, 1984 and Lurain *et al*., 2011). Patients with a low risk have been treated with single agent methotrexate or dactinomycin, but EMA/CO (Etoposide, Methotrexate, Dactinomycin, Cyclosphosphamide, and Oncovin) therapy has been considered the most effective treatment regimen for patients with a high-risk[(2)]. Remission rates in the nonmetastatic stage of choriocarcinoma are 98% to 100% and more than 75% even in cases of metastatic choriocarcinoma (Rosenberg *et al*., 2008; Lurain *et al*., 1998).

Conclusion

We have reported a rare case of dorsolumbar epidural metastasis and pulmonary metastasis of Choriocarcinoma. Choriocarcinoma is a highly anaplastic malignancy derived from trophoblastic cells characterized by the secretion of human chorionic gonadotropin (βHCG) and early hematogenous metastasis. However, metastatic Choriocarcinoma in the spine is extremely rare. Very few cases of metastasis in lumbar and/or sacral vertebra have been reported. Any patient presented with long segment vascular epidural spinal lesion with uterine pathology, diagnosis of metastatic Choriocarcinoma should be kept in mind. Patients with a low risk can be treated with single agent Methotrexate or Dactinomycin, but EMA/CO (Etoposide, Methotrexate, Dactinomycin, Cyclosphosphamide, and Oncovin) therapy has been considered the most effective treatment regimen for patients with a high-risk. Despite improvements in treatment modality and the use of combined modality treatment with chemotherapy, surgery and radiation, the prognosis for these choriocarcinoma cases with spinal metastasis is unfavorable. Earlier diagnosis and multimodality treatment is crucial for significant reduction in mortality.

REFERENCES

Bagshawe, K.D. 1984. Treatment of high-risk choriocarcinoma. *J Reprod Med.*, 29:813–820.

Beşkonakli, E, Cayliğlu, S., Kulaço, S. 1998. Metastatic choriocarcinoma in the thoracic extradural space : case report. *Spinal Cord.,* 36:366–367.

Chandra, S.A., Gilbert, E.F., Viseskul, C., Strother, C.M., Haning, R.V., Javid, M.J. 1990. Neonatal intracranial choriocarcinoma. *Arch Pathol Lab Med.,* 114:1079–1082.

Kuten, A., Cohen, Y., Tatcher, M., Kobrin, I., Robinson, E. 1978. Pregnancy and delivery after successful treatment of epidural metastatic choriocarcinoma. *Gynecol Oncol.* 6:464–466.

Lee, J.H., Park, C.W., Chung, D.H., Kim, W.K. 2010. A case of lumbar metastasis of choriocarcinoma masquerading as an extraosseous extension of vertebral hemangioma. *J Korean Neurosurg Soc*., 47:143–147.

Lurain, J.R. 2011. Gestational trophoblastic disease II: classification and management of gestational trophoblastic neoplasia. *Am J Obstet Gynecol*., 204: 11-18.

Lurain, J.R. 1998. Management of high-risk gestational trophoblastic disease. *J Reprod Med.*, 43:44–52.

Menegaz, R.A., Resende, A.D., da Silva, C.S., Barcelos, A.C., Murta, E.F. 2004. Metastasis of choriocarcinoma to lumbar and sacral column. *Eur J Obstet Gynecol Reprod Biol*., 113:110–

Naito, Y., Akeda, K., Kasai, Y., Matsumine, A., Tabata, T., Nagao, K., *et al*. 1976. Lumbar metastasis of choriocarcinoma. Spine (Phila Pa) 2009; 34:E538–E543.

Rosenberg, S., DePinho, R.A. Weinberg RE, DeVita VT, Lawrence TS. 2008. *DeVita, Hellman, and Rosenberg's Cancer: Principles & Practice of Oncology*. Hagerstwon, MD: Lippincott Williams & Wilkins. ISBN 0-7817-7207-9.OCLC

Scott, J.R., Di Saia, P.J., Hammond, C.B. 1999. Gestational trophoblastic neoplasms. 8th edn. In: Scott JR, DiSaia PJ, Hammond, editors. Danforth's Obstetrics and Gynecology. Philadelphia, PA: Lippincott Williams & Wilkins. 927-937.

Seckl, M.J., Sebire, N.J., Berkowitz, R.S. 2010. Gestational trophoblastic disease. Lancet., 376: 717-729.

Vani, R., Kuntal, R., Koteshwar, R.K. 1993. Choriocarcinoma following term pregnancy with bone metastasis. *Int J Gynaecol Obstet.*, 40:252–253.

****