



## Case Report

# Solitary late spinal metastasis from apocrine salivary duct carcinoma: Case report

Bianca Maria Baldassarre<sup>1</sup>, Federica Penner<sup>1</sup>, Luca Bertero<sup>2</sup>, Giuseppe Di Perna<sup>1</sup>, Marco Ajello<sup>1</sup>, Nicola Marengo<sup>1</sup>, Francesco Zenga<sup>1</sup>, Diego Garbossa<sup>1</sup>

<sup>1</sup>Department of Neuroscience, Neurosurgery Unit, Città Della Scienza e Della Salute, <sup>2</sup>Department of Pathologic, Città Della Scienza e Della Salute, Turin, Italy.

E-mail: Bianca Maria Baldassarre - baldassarrebianca@gmail.com; Federica Penner - federica.penner@gmail.com; Luca Bertero - luca.bertero@gmail.com;

\*Giuseppe Di Perna - dr.giuseppediperna@gmail.com; Marco Ajello - m.ajello84@gmail.com; Nicola Marengo - nicolamarengo@icloud.com;

Francesco Zenga - zengafra@hotmail.com; Diego Garbossa - dgarbossa@gmail.com



### \*Corresponding author:

Giuseppe Di Perna,  
Neurosurgery Unit, Città  
Della Scienza e Della Salute,  
Turin, Giuseppe Di Perna, Via  
Cherasco 15, Turin - 10128,  
Italy.

dr.giuseppediperna@gmail.com

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## ABSTRACT

**Background:** The salivary duct carcinomas (SDCs) are rare, high-grade neoplasms involving major salivary glands. Parotid is the most frequently involved gland (85%). Apocrine phenotype (histological presence of decapitation secretions) and androgen receptor expression define SDC. The clinical course of these tumors is characterized by aggressive local behavior with extraglandular extension, high recurrence rates, early metastases, and poor prognoses. Despite aggressive surgical/radiation therapy management, the rates of locoregional and metastatic relapses are high, and the mortality rates over 48 months approach 65%. Notably, there is no treatment algorithm available for managing vertebral metastases from apocrine SDC.

**Case Description:** An elderly male presented with MR/CT findings of an isolated T11 vertebral metastasis attributed to a previously treated parotid SDC. On both CT/MR, it was an osteolytic lesion and demonstrated spinal canal infiltration. The patient underwent surgical biopsy/decompression/resection, following which the lesion histopathologically proved to be a SDC. The patient was subsequently treated with 30 Gy in 10 fractions within 2 weeks of discharge. One-month later, the MRI confirmed adequate epidural decompression without recurrence, and 9 months post-operatively, patient remained disease free.

**Conclusion:** Isolated metastasis attributed to parotid SDC followed by radiation therapy may result in tumor control.

**Keywords:** Apocrine salivary duct carcinoma, Carbon fiber, Salivary gland tumors, Separation surgery, Spinal metastases

## INTRODUCTION

Salivary duct carcinomas (SDCs) are rare and account for approximately 6% of all malignant salivary gland tumors. SDC are malignant, rapidly-growing aggressive tumors with a high local recurrence rate, and high frequency of early metastases.<sup>[7,10,14,16,18]</sup> In this report, we present a 77-year-old man male with an isolated T11 vertebral metastasis from a primary apocrine SDC of the parotid gland who was adequately managed with biopsy/surgical resection followed by radiation therapy.

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## CASE REPORT

### Initial presentation of SDC

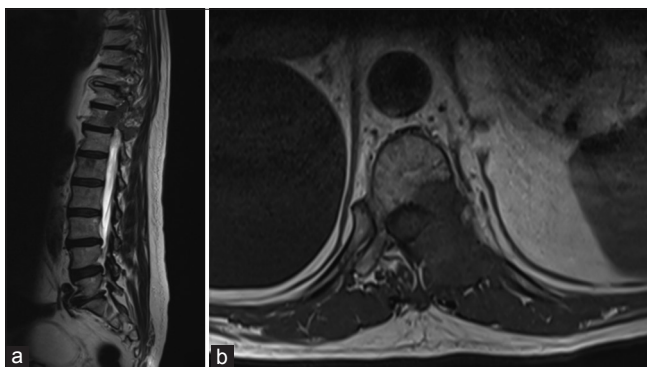
A 77-year-old male with a medical history of the left-sided parotid carcinoma presented with the MR/CT finding of an isolated metastatic T11 vertebral lesion diagnosed following a radical parotidectomy. The histological diagnosis was consistent with a SDC with apocrine differentiation. At that time, 3 lymph nodes were positive, and the patient, therefore, underwent subsequently adjuvant local radiotherapy.

### Presentation with T11 metastatic SDC disease

A chest-abdomen CT scan, performed 3 months later, showed an increase in size of the vertebral lesion and highlighted spinal instability. Further, the spinal MRI demonstrated infiltration of the spinal canal at the T11 level due to the SDC (i.e., Bilsky Grade 2 epidural compression) [Figures 1a and b].<sup>[2]</sup> The patient underwent biopsy/surgical decompression/resection of the tumor utilizing a left T11 laminectomy. At surgery, the left T11 pedicle was removed, followed by partial T11 corpectomy and the posterior application of two carbon rods from T10 to T12 [Figure 2a]. The postoperative spinal CT scan confirmed that the fixation system was accurately placed and that the spinal cord was sufficiently decompressed [Figures 2b and c].

### Histology

The initial histological diagnosis was consistent with a SDC with apocrine differentiation and 3 lymph nodes were positive. The T11 histopathology was the same. Immunohistochemical stainings for gross cystic disease fluid protein-15 (GCDFP15) and androgen receptor (e.g., for the T11 biopsy material) were diffusely positive; while cytokeratins 8/18 were focally positive [Figures 3a, b, c, d and e], all remaining consistent with the diagnosis of a SDC.



**Figure 1:** Sagittal (a) and axial (b) preoperative MRI showing epidural compression and circumferential vertebral involvement.

### Adjuvant radiation therapy

Subsequently, patient underwent adjuvant local radiotherapy and received 30 Gy in 10 fractions within 2 postoperative weeks. One-month post-operatively, the MRI confirmed adequate epidural decompression without tumor recurrence [Figure 2d]. Nine months later, the patient remained disease free [Table 1].

## DISCUSSION

SDC are rare, high-grade neoplasms involving major salivary glands, and represent approximately 6% of all salivary gland cancers.<sup>[12,15]</sup> The parotid gland is the most frequent involved (75–85%). The peak of incidence occurs in the 6<sup>th</sup>–7<sup>th</sup> decades of life, and there is a male predominance. Patients present with enlarging parotid masses, frequently associated with VII cranial nerve dysfunction, plus aggressive local behavior (i.e., with extraglandular extension), high recurrence rates, and early metastases correlating with poor prognoses.<sup>[10,14,16,18]</sup>

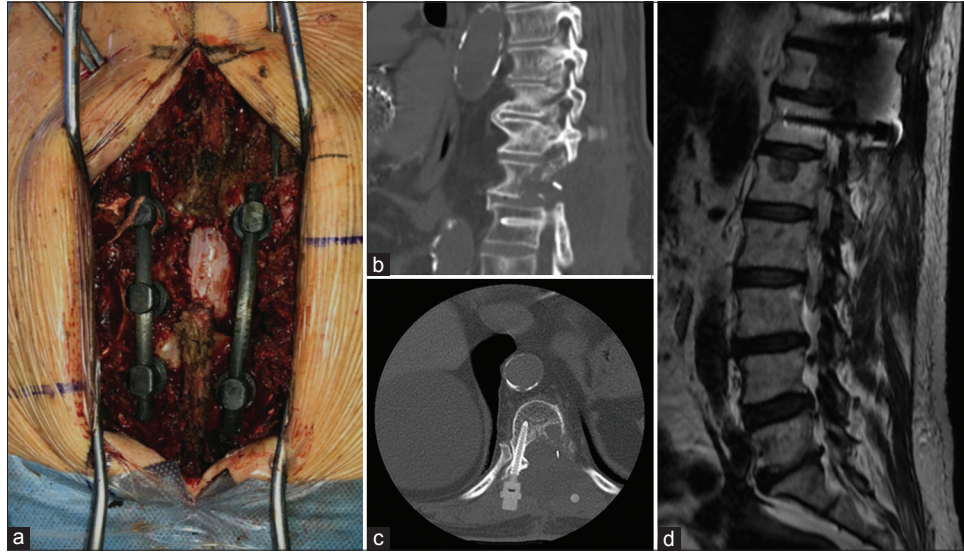
### Prognosis with surgery and radiation therapy

Although there is no specific management algorithm for the treatment of vertebral metastases from SDCs, those with single osteolytic lesions and high-grade epidural compression may be successfully treated with aggressive gross-total surgical resection followed by radiation therapy.<sup>[1,3-6,8,9,13]</sup> Due to the frequency of SCD's invasion and their aggressive

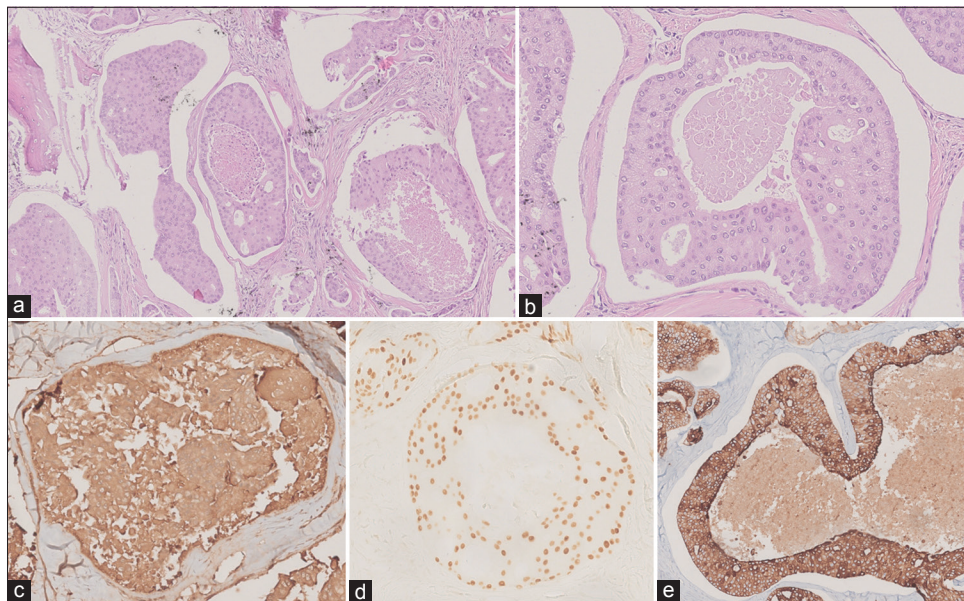
**Table 1:** Case report details.

Primary tumor	<ul style="list-style-type: none"> <li>• Left-sided salivary duct carcinoma with apocrine differentiation</li> </ul>
Primary tumor treatment	<ul style="list-style-type: none"> <li>• Radical parotidectomy+selective lymph nodes resection and neck dissection</li> <li>• Adjuvant radiation therapy</li> </ul>
Metastasis	<ul style="list-style-type: none"> <li>• Single osteolytic lesion involving T11 vertebral body</li> </ul>
Preoperative assessment	<ul style="list-style-type: none"> <li>• ESCC grade (Bilsky score): 2</li> </ul>
Treatment	<ul style="list-style-type: none"> <li>• Separation surgery with anterior-lateral decompression (bilateral) laminectomy, left pediclectomy, and ventral separation</li> <li>• Radiation therapy (30 Gy in 10 fractions)</li> </ul>
Immunohistochemistry	<ul style="list-style-type: none"> <li>• GCDFP15 + (diffusely)</li> <li>• AR + (diffusely)</li> <li>• Cytokeratins 8/18 + (focally)</li> <li>• NTRK/NTRK2/NTRK3 translocations</li> </ul>

SINS: Spinal instability neoplastic score, ESCC score: Epidural spinal cord compression, NSE score: Neurological stability epidural compression score



**Figure 2:** Intraoperative image (a) displaying the circumferential decompression and vertebral fixation with carbon fiber system. Postoperative sagittal (b) and axial (c) CT scan showing the accuracy of fixation system placement. Postoperative MRI image (d) demonstrating the epidural decompression and the absence of local disease recurrence.



**Figure 3:** Histological findings after vertebral lesion resection. H and E image (a: 100X, b: 200X) shows a bone infiltrating metastatic carcinoma with duct structures and comedonecrosis. Neoplastic cells were mildly pleomorphic with an eosinophilic cytoplasm and features consistent with apocrine differentiation. Immunohistochemical stainings showed a diffuse positivity for GCDFP15 (c) and androgen receptor (d), while cytokeratins 8/18 were focally positive (e).

nature, surgical treatment should include a radical/total parotidectomy with sacrifice of the facial nerve and ipsilateral lymph node dissection, followed by postoperative chemotherapy/radiotherapy. Even with such aggressive management, there is a nearly 65% locoregional rate of metastatic relapse, and patients typically succumb from recurrent and/or progressive disease within 48 months.<sup>[10]</sup>

### Histology

Apocrine phenotype (histological presence of decapitation secretions) and androgen receptor expression defines SDC that may metastasize to the spine (e.g., to the T11 level in this case) warranting surgical extirpation followed by adjuvant radiation therapy.<sup>[11,17]</sup>



## CONCLUSION

Three-year delayed metastatic parotid carcinoma of the T11 vertebral body was adequately managed with separation surgery achieved through an extracavitary transpedicular partial anterior corpectomy and posterior pedicle screw fusion, followed by radiation therapy.

### Ethical committee approval

All clinical and radiological data were collected and retrospective analyzed. This study does not require any variations in patient's treatment and no formal ethics committee approval was required.

### Authors' contributions

BMB writing and editing; FP conceptualization and revision; LB data collection; GD revision; MA data collection; writing and revision; FZ revision; DG conceptualization and supervision.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

## REFERENCES

- Barzilai O, Robin AM, O'Toole JE, Laufer I. Minimally invasive surgery strategies: Changing the treatment of spine tumors. *Neurosurg Clin N Am* 2020;31:201-9.
- Bilsky M, Smith M. Surgical approach to epidural spinal cord compression. *Hematol Oncol Clin North Am* 2006;20:1307-17.
- Cofano F, di Perna G, Alberti A, Baldassarre BM, Ajello M, Marengo N, *et al.* Neurological outcomes after surgery for spinal metastases in symptomatic patients: Does the type of decompression play a role? A comparison between different strategies in a 10-year experience. *J Bone Oncol* 2020;26:100340.
- Cofano F, di Perna G, Marengo N, Ajello M, Melcarne A, Zenga F, *et al.* Transpedicular 3D endoscope-assisted thoracic corpectomy for separation surgery in spinal metastases: Feasibility of the technique and preliminary results of a promising experience. *Neurosurg Rev* 2020;43:351-60.
- Cofano F, di Perna G, Monticelli M, Marengo N, Ajello M, Mammi M, *et al.* Carbon fiber reinforced vs titanium implants for fixation in spinal metastases: A comparative clinical study about safety and effectiveness of the new "carbon-strategy". *J Clin Neurosci* 2020;75:106-11.
- Cofano F, di Perna G, Zenga F, Ducati A, Baldassarre B, Ajello M, *et al.* The neurology-stability-epidural compression assessment: A new score to establish the need for surgery in spinal metastases. *Clin Neurol Neurosurg* 2020;195:105896.
- Cofano F, Monticelli M, Ajello M, Zenga F, Marengo N, di Perna G, *et al.* The targeted therapies era beyond the surgical point of view: What spine surgeons should know before approaching spinal metastases. *Cancer Control* 2019;26:1-13.
- di Perna G, Cofano F, Mantovani C, Badellino S, Marengo N, Ajello M, *et al.* Separation surgery for metastatic epidural spinal cord compression: A qualitative review. *J Bone Oncol* 2020;25:100320.
- Gasbarrini A, Cappuccio M, Mirabile L, Bandiera S, Terzi S, Bròdano GB, *et al.* Spinal metastases: Treatment evaluation algorithm. *Eur Rev Med Pharmacol Sci* 2004;8:265-74.
- Hosal AS, Fan C, Barnes L, Myers EN. Salivary duct carcinoma. *Otolaryngol Head Neck Surg* 2003;129:720-5.
- Jaspers HC, Verbist BM, Schoffelen R, Mattijssen V, Slootweg PJ, van der Graaf WT, *et al.* Androgen receptor-positive salivary duct carcinoma: A disease entity with promising new treatment options. *J Clin Oncol* 2011;29:e473-6.
- Jayaprakash V, Merzianu M, Warren GW, Arshad H, Hicks WL Jr., Rigual NR, *et al.* Survival rates and prognostic factors for infiltrating salivary duct carcinoma: Analysis of 228 cases from the surveillance, epidemiology, and end results database. *Head Neck* 2014;36:694-701.
- Laufer I, Rubin DG, Lis E, Cox BW, Stubblefield MD, Yamada Y, *et al.* The NOMS framework: Approach to the treatment of spinal metastatic tumors. *Oncologist* 2013;18:744-51.
- Roh JL, Lee JI, Choi SH, Nam SY, Kim SO, Cho KJ, *et al.* Prognostic factors and oncologic outcomes of 56 salivary duct carcinoma patients in a single institution: High rate of systemic failure warrants targeted therapy. *Oral Oncol* 2014;50:e64-6.
- Salovaara E, Hakala O, Bäck L, Koivunen P, Saarilahti K, Passador-Santos F, *et al.* Management and outcome of salivary duct carcinoma in major salivary glands. *Eur Arch Otorhinolaryngol* 2013;270:281-5.
- Wee DT, Thomas AA, Bradley PJ. Salivary duct carcinoma: What is already known, and can we improve survival? *J Laryngol Otol* 2012;126 Suppl 2:S2-7.
- Williams L, Thompson LD, Seethala RR, Weinreb I, Assaad AM, Tuluc M, *et al.* Salivary duct carcinoma: The predominance of apocrine morphology, prevalence of histologic variants, and androgen receptor expression. *Am J Surg Pathol* 2015;39:705-13.
- Williams MD, Roberts D, Blumenschein GR Jr., Temam S, Kies MS, Rosenthal DI, *et al.* Differential expression of hormonal and growth factor receptors in salivary duct carcinomas: Biologic significance and potential role in therapeutic stratification of patients. *Am J Surg Pathol* 2007;31:1645-52.

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