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CASE REPORT

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Iatrogenic implantation of malignant meningioma to the abdominal wall

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Abstract We report a case of malignant meningioma that occurred in the abdominal operation scar of a 71-year-old woman. This tumor was a 13×8 cm gray–tan soft tumor, consisting of multiple nodules. Histologically, tumor cells proliferated in the subcutaneous tissue, displaying mostly a storiform pattern and a focal whorl formation with high mitotic figures. The immunohistochemical positivity for epithelial membrane antigen and negativity for CD34 enabled us to differentiate this tumor from a dermatofibroma protuberance or hemangiopericytoma. The patient had a history of operation for a recurrent orbital lesion of a malignant meningioma that initially developed in the frontal skull base. The present case probably resulted from iatrogenic transplantation of the orbital malignant meningioma to the lower abdominal wall, which had served as a donor site for adipose tissue used to pack the orbital defect.

Keywords Iatrogenic tumor · Meningioma · Malignant · Implantation

Introduction

Although a possibility of iatrogenic seeding of a neoplasm by means of surgical operation is well recognized by surgeons [4, 12], cases have unfortunately been repeatedly reported since 1907 [2, 9]. Pathologists may be involved in diagnosing iatrogenic tumors, but it is challenging to distinguish these tumors from those of other causes. Here, we report a case of malignant meningioma that might have been iatrogenically implanted in the lower abdominal wall, which was a donor site for adipose tissue used to pack the orbital defect due to resection of a recurrent malignant meningioma.

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Case history

In December 1992, a 71-year-old woman underwent a first operation for a tumor that occupied the frontal skull base and nasal cavity. The histological diagnosis was a transitional meningioma, but the pathologist stated that the tumor was potentially malignant because of the high mitotic count (5 per 10 HPFs) and increased cellularity (Fig. 1). The patient presented with right exophthalmos 5 months later due to an orbital tumor. The histological diagnosis was recurrence of meningioma, and she underwent a second operation for the recurrent tumor. During this operation, abdominal adipose tissue was used to pack the orbital defect due to the curettage of the tumor. A clinician noted a nodule 3 cm in diameter in the abdominal wound 5 months after the second operation. This nodule progressively enlarged. Since the biopsy specimen led to the suspicion of a dermatofibroma protuberance, the tumor was totally resected and submitted to West Japan Pathology Laboratory (Kurashiki, Japan). There was no evidence of additional tumor metastasis in any organs examined. However, the patient showed recurrence of the tumor in the frontal skull base and nasal cavity and died in April 1994.

Materials and methods

The specimen was fixed in formaldehyde and embedded in paraffin. Sections (4-μm thick) were stained with hematoxylin and eosin for histological diagnosis. In addition, the sections were immunostained using an avidin-biotin horseradish peroxidase complex method. For epithelial membrane antigen (EMA; 1:50; Dako, Kyoto, Japan), vimentin (1:40; Dako), cytokeratin (AE1/AE3; 1:400; Dako), α-smooth muscle actin (1:50; Immunon, Pittsburgh, Pa.), and CD34 (1:50; Immunotech, Marseille, France), a microwave antigen retrieval method was used. For MIB-1 (1:50; Immunotech), a pressure cooker antigen retrieval method was used. The sections were developed with diaminobenzidine and counterstained with hematoxylin. Negative controls were treated in the same manner but without the primary antibodies.

Pathologic findings

The specimen was a 13×8 cm gray–tan soft tumor comprising multiple nodules of 6×4.5, 2×2, and 3×2 cm. Mi-

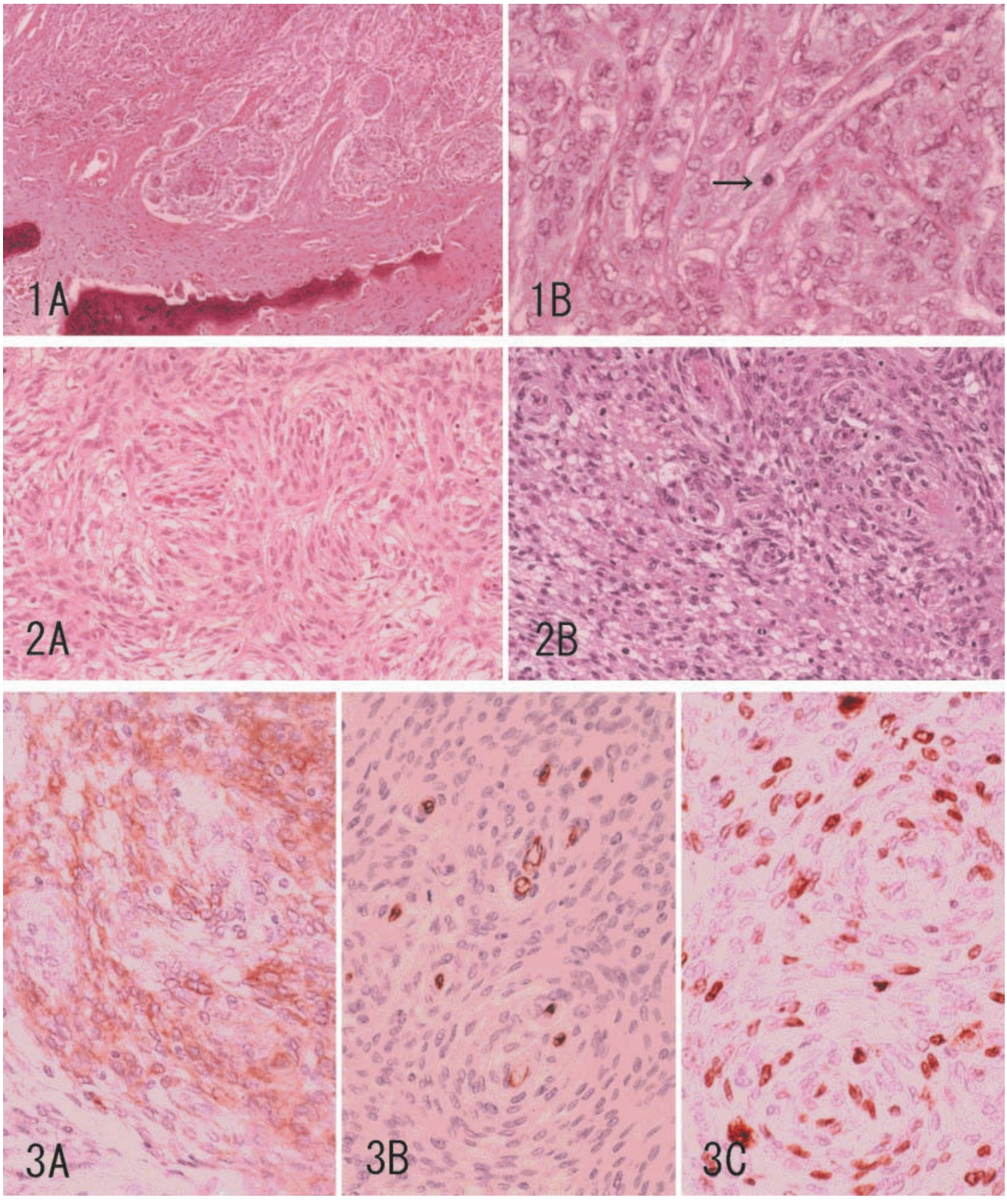

Fig. 1 **A** Histology of the primary meningial tumor. Tumor cells show cellular whorls and the invasive pattern at the periphery of the tumor. **B** Higher magnification of the tumor. Note the mitotic figure (*arrowhead*)

Fig. 2 Histology of the abdominal tumor. **A** Tumor cells proliferate in subcutaneous tissue displaying a storiform pattern, mimicking a dematofibrosarcoma protuberance. **B** They also show cellular whorls

Fig. 3 Immunohistochemical staining of the abdominal tumor. **A** Tumor cells show diffuse cytoplasmic and focal vacuolar staining for epithelial membrane antigen (EMA). **B** CD34 is not expressed in tumor cells but in endothelial cells of blood vessels. **C** Forty percent of the nuclei of the tumor cells are labeled with MIB-1

croscopically, spindle cells proliferated in subcutaneous tissue. The nodules displayed a storiform pattern in most parts (Fig. 2A) and focally formed cellular whorls (Fig. 2B). Neither necrosis nor calcification was found. The mitotic count was greater than 50 per 10 HPFs. A review of the orbital specimen from the second operation revealed that both abdominal and orbital tumors showed the same histology. Immunohistochemically, they were positive for EMA (Fig. 3A) and vimentin but negative for S-100 protein, cytokeratin, α-smooth muscle actin, and CD34 (Fig. 3B). Forty percent of the tumor cells were labeled with MIB-1 (Fig. 3C) [6]. Thus, the final diagnosis of the abdominal wall tumor was malignant meningioma implanted from the orbital lesion.

Discussion

Iatrogenic seeding of neoplasms may occur by various types of surgical interventions. Based on the patient's history and a histological review of the previous operation specimens, we concluded that the present case was due to an iatrogenic implantation of a malignant meningioma. It was suggested that the tumor cells were transplanted to the abdominal wall during the second surgical operation when the surgeon used an abdominal-free flap to pack the massive defects caused from resection of the orbital recurrent tumor. The present case was similar to the cases reported by Brandes et al. [1] and Cole et al. [2]. They stressed the clinical importance of avoiding possible tumor contamination of operative fields by means of meticulous instrument changes and by isolation of multiple surgical fields.

Implantation metastasis of a tumor of the central nervous system has been reported as a rare complication of surgical operations or needle biopsies [3, 5, 8, 11]. As for meningiomas, a case of a histologically benign meningioma with implantation metastasis in the temporalis muscle due to the surgical operation has been described [11]. The risk of tumor implantation may depend on several factors, including the degree of malignancy, the number of cells in the inoculum, and the nature of implantation site. In the present case, the malignant potential of the primary meningioma might have contributed to the rapid growth of the implanted tumor cells. In addition, the abdominal wall might be a suitable site for the survival and growth of tumor cells.

The present case indicates a need for caution and that surgical pathologists should pay attention to a patient's past history to avoid overlooking an iatrogenic tumor. To diagnose the abdominal tumor histologically, we must first differentiate the tumor from a dermatofibrosarcoma protuberance (DFSP), which often occurs in operation scars and shows a storiform pattern. Immunohistochemically, a DFSP shows a CD34+EMA– phenotype [13]. However, in the present case, the tumor cells showed a CD34–EMA+ phenotype [7], ruling out a DFSP. Similarly, this case could be differentiated from a hemangiopericytoma, which showed immunoreactivity for CD34 [7].

Since distant extracranial metastases of meningiomas have been reported to occur in less than 1 per 1000 meningioma cases [10], we should consider the possibility that the present case represents an extracranial metastasis of a malignant meningioma. However, this is unlikely because metastases of malignant meningiomas are usually multiple and often involve lungs, bones, liver, and lymph nodes [10]. Therefore, the absence of multiple metastasis and the unusual metastatic site suggest that the abdominal tumor was the result of surgical implantation. In conclusion, we reported a case of abdominal malignant meningioma, in which iatrogenic seeding was considered to be a possible cause.

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