Anaplastic Oligodendroglioma with Cervical Lymphnode Metastasis

Report of a Case

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Abstract
A case of a patient operated on twice for a temporal parietal anaplastic oligodendroglioma, followed by radiotherapy, is reported. Although no intra cranial recurrence has happened, a biopsy-proven extracranial cervical lymphnode metastasis was diagnosed 15 months later. Surgical manipulation of the primary lesion by lymphatic and/or blood vessel invasion seem to be the main suspected factors in the development of CNS tumors metastasis in the head and neck region.

Key Words
Brain neoplasm; extracranial metastasis; immunohistochemistry.

Sinopse
Metástase em Linfonodo Cervical de Oligodendroglioma Anaplásico - Apresentação de Caso e Revisão de Literatura.
Os autores apresentam um caso de um paciente adulto operado em duas ocasiões para um oligodendroglioma anaplásico temporal-parietal direito, seguido de radioterapia complementar. Embora não haja ocorrido recidiva da lesão intracraniana, 15 meses após, foi diagnosticada e confirmada histologicamente uma metástase em linfonodo cervical. A manipulação cirúrgica da lesão primária com possível disseminação vascular e/ou linfática parece ser o fator primordial no desenvolvimento das lesões metastáticas cervicais nos tumores primários do SNC.

Palavras-chave
Tumores do SNC, metástases extracranianas, imuno-histocitomografia.

Introduction
Primary intracranial tumours rarely spread out of the skull[1, 3, 7, 8, 9, 13, 15, 19, 23, 24, 26], albeit the high frequency of the malignant intracranial neoplasms and, principally, the grade of malignancy of medulloblastomas and the majority of gliomas [18, 25, 26]. The occurrence of extracranial metastasis of intracranial tumours runs about 0.1 to 0.5% [13, 18, 19], meanwhile the dissemination within the Central Nervous System (CNS) is 1.5% [3]. Most of the cases do happen after craniotomies, the probable factor of its seeding, being surgical manipulation [1, 6, 9, 19, 20]. Intracranial tumours that most frequently produce extracranial secondary are, in order of frequency: meningiomas, undifferentiated gliomas, pituitary tumours, blood-veins tumours and rarely melanomas [12, 19]. As regards to glial tumours, the oligodendroglioma is the one that less frequently metastasizes out of the CNS [14, 21, 23], probably because of its relative low prevalence. The most frequent sites of extracranial metastasis are lung, liver, lymphnodes (mediastinal, cervical and abdominal) vertebrae and other bones, pleura and kidneys. [2, 6, 7, 9, 15, 19, 21]. Giannini and Yuan [3] noted that 33% of extracranial metastasis happened in the lungs and pleura and 23% in the regional lymphnodes.

The present report adds to the very few published cases in the literature [14, 21, 23], a case of a patient with an anaplastic oligodendroglioma, operated on and further irradiated, who presented with a big cervical lymphnode metastasis, with no evidence of intracranial recurrence.

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A 36-years-old white male presented with a history of headaches and vomiting for the last 4 months, aggravated in the last 30 days. Neurological examination showed bilateral papilloedema. Carotid angiogram and CT disclosed a right temporo-parietal space occupying lesion (Fig. 1) that was partially removed because of its infiltrative appearance. Histology showed a glial neoplasm constituted by cells with vacuolated cytoplasm, round or oval centrally located nuclei with “honeycomb” appearance; infiltration of cerebral parenchyma, with proliferation of vascular endothelium, numerous calcifications and mitosis. The Avidin-Biotin-Peroxidase immunohistochemical technique, utilizing glial fibrillary acid protein antibodies (GFAP-DAKO, 1:500 dilution) disclosed mild positivity for tumour cells. Conclusion: Anaplastic oligodendroglioma (Fig. 2).

Three months later, the patient returned with intracranial hypertension and recurrence of the lesion. Now, complete resection was performed, followed by radiotherapy. The patient got well, with no neurological symptoms but fifteen months later he noticed a “lump” in his right cervical region. On examination, a 10x5x5 cm hard mass, adherent to the deep muscles, extending from 2 cm below the mastoid process to the inferior insertion of the sternocleidomastoid was found. Neurological examination was normal. A biopsy of this lesion disclosed a lymphnode metastasis of anaplastic oligodendroglioma, with immunohistochemical confirmation (Fig. 2D). Neither operation nor radiotherapy were authorized and the patient was sent back home. His family doctor noticed progressive respiratory difficulty with no neurological symptoms and death ensued nine months later. Permission for autopsy was not allowed.

Discussion

The first case report of an extracranial metastasis of a primary CNS tumour dates to 1886 \(^{(21)}\), although only in 1928 Davis \(^{(4)}\) first detailed a case of a glioma with metastasis. The rarity of this situation is demonstrated by the few reported cases in the literature \(^{(1,2,6,8,9,11,20,21,23,25,26)}\). Although 17% of all malignant tumours are localized in the CNS \(^{(26)}\), being 50% of them represented by those of glaef origin - mainly glioblastomas with very agressive biological behaviour - the metastatic dissemination outside the CNS is a rarity, especially when one deals with oligodendrogliomas. These tumours are not frequent in the glial tumour series \(^{(13)}\) and metastasis are scarcely found.

In 1955, Weiss \(^{(25)}\) defined what he thought being the four criteria to be followed for the diagnosis of an extraneural metastasis of a brain tumour:

1. SNC tumour histological diagnosis;
2. First clinical presentation due to the primary lesion;
3. Identical pathological findings of both primary and secondary tumours;
4. Autopsy excluding the possibility of a primary tumour not being intracranial.

In 1974, Dolman \(^{(5)}\) after reporting a case of a lymphnode glioblastoma metastasis, suggested the second criteria should be excluded.

Every intracranial neoplasm can produce secondary lesions but medulloblastomas, meningiomas, ependymomas and undifferentiated gliomas most frequently do it \(^{(4,11,19)}\). Regarding to gliomas, one can reason (them being not only due to their frequency and grade of malignancy but also to their ability to invade dural sinuses \(^{(6,18,22)}\)). Meningiomas, especially those of malignant nature, have as a characteristic infiltrating the duramater and frequently the diploe, both richly vascularized tissues that could permit tumour cells reaching the sistemic circulation \(^{(6,10,30)}\).

The reason for the small metastasizing capacity of primary intracranial tumours is supposed to be due to \(^{(4,5,12,31)}\):

1. Absence of lymphatics in the CNS
2. CNS venous sinuses structure, externally involved by a dense dural layer, that holds off the penetration or invasion of neoplastic cells.
3. Small cerebral veins wall, that tend to collapse with the growing of an intracranial tumour and/or with the development of localized intracranial hypertension.
4. Neuronal tissue specificity, that makes difficult the development of metastatic glial cells in other sites.
5. Rapid evolution of malignant brain tumours that precludes the development of secondaries before intracranial hypertension or death happens.

It is largely accepted that the way neoplastic cells find to metastasize is through blood or lymphatic vessels (5,6,7,8, 16,19,21,22). The lymphatic dissemination although there being no encephalic lymphatic drainage, is considered in those cases in which there is head soft tissue invasion by tumour cells; blood dissemination is considered either through invasion of small intracranial vessels (rare) or through direct compromise of dural sinuses, when it may happen without vessel collapse (18). Seeding through CSF explains dissemination along CNS or, eventually, outside the CNS, in patients who have ventriculoperitoneal shunts (19).

Spontaneous metastasis of brain tumours have been reported (1,14,20,21) but those who happened without surgical manipulation are exceedingly rare (15,6). In almost every published cases (14,15,21,22) there was a previous craniotomy, so that is believed that surgical treatment proportionates malignant cell seeding (6,15,20). In this case, the patient had been operated on twice, plus radiotherapy, and that could be the reason for finding a homolateral cervical metastasis, one of the sites, along with lungs and pleura, most commonly affected (7,9,16,17,20).

References