G Chir Vol. 30 - n. 3 - pp. 93-95
Marzo 2009

casistica clinica

Ethmoidal adenocarcinoma with lung metastases: diagnosis and multimodal treatment

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SUMMARY: **Ethmoidal adenocarcinoma with lung metastases: diagnosis and multimodal treatment.**

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We report a case of 68-year-old patient underwent a magnetic resonance imaging (MRI) of the skull and a computed tomography (CT) of the thorax for rhinorrhea and dyspnea. The MRI showed an irregular ethmoidal lesion and the CT of the thorax underlined a solid nodular neoformation in the upper right pulmonary lobe. The patient underwent rhinoscopy with biopsies that showed an ethmoidal adenocarcinoma; excision of the tumour was carried out via trans-sphenoid. After one month the patient underwent wedge-resections in video-thoracoscopy (VATS). Perioperative histologic examination revealed a lung metastases due to an adenocarcinoma of the ethmoid.

The patient was treated with chemotherapy and did not show relapses after 12 months from VATS.

RIASSUNTO: **Adenocarcinoma dell'etmoide con metastasi polmonari: diagnosi e trattamento multimodale.**

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Paziente di 68 anni sottoposto ad una risonanza magnetica dell'encefalo e ad una tomografia computerizzata del torace in seguito a rinorrea e dispnea. La risonanza magnetica evidenzia una lesione irregolare a carico dell'etmoide mentre la tomografia computerizzata rileva la presenza di una neoformazione nodulare nel lobo superiore di destra. Una rinoscopia con multiple biopsie permette diagnosi di adenocarcinoma dell'etmoide; l'exeresi della neoplasia è stata effettuata per via trans-sfenoidea. A distanza di un mese, il paziente è stato sottoposto a resezioni atipiche polmonari in videotoracoscopia. L'esame istologico estemporaneo rilevava trattarsi di metastasi da adenocarcinoma dell'etmoide.

Il paziente è trattato con chemioterapia e non presenta fenomeni ripetitivi a 12 mesi dall'intervento toracoscopico.

KEY WORDS: Ethmoidal cancer - Lung metastasis - Surgery - Radiotherapy - Chemotherapy.
Cancro dell'etmoide - Metastasi polmonare - Chirurgia - Radioterapia - Chemioterapia.

Introduction

Ethmoidal tumour is a rare pathology. In most cases, it does not present symptoms but reveals itself later, involving the facial structures. Repeated epistaxis, late exophthalmos, periorbital space invasion, glabellar frontal and periorbital swelling are the clinical findings. Peripheral metastases are shown in skeleton, liver and lung. In 7-15% of cases lymph node diffusion is present, within 4 years from diagnosis (1), in relation with size and grading of the tumour. We analyzed the diagnosis and treatment of primary ethmoidal cancer and pulmonary secondary lesions in a 68 years old patient.

Case report

A 68-year-old patient showed persistent mucous haematic rhinorrhea, dyspnea, cough and evening fever. Magnetic resonance imaging (MRI) of the skull showed an ethmoidal lesion, with a diameter of 3 cm and irregular margins. The chest ray highlighted a right pulmonary opacity. Computed tomography (CT) of the thorax revealed the presence of a solid nodular formation with a 4 cm axial diameter, located in the ventral segment of the right upper pulmonary lobe and adherent to the mediastinal pleura and the superior cava vein. CT also showed a lymph node of 2 cm in diameter in the Barety ridge. The fiber-optic bronchoscopy was negative.

The histologic exam of the CT-guided needle biopsy revealed a

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carcinoma not well specified. The patient underwent rigid rhinoscopy with biopsies that showed an ethmoidal adenocarcinoma; trans-sphenoidal excision of tumour was carried out.

One month later the patient was submitted to a video-assisted thoracoscopy (VATS) of the right lung. The upper pulmonary lobe appeared partially adherent to the thoracic wall. After dissection of the parenchyma, we noticed an oval white neoformation in the ventral segment of the upper lobe, with a soft-elastic consistency. In the lower lobe we also found two neoformations with a hard-elastic consistency and a diameter of 0.8 cm. The frozen section examination revealed tumour with some pattern of the adenocarcinoma that could be assimilated to the previous neoplasm of the ethmoid. We performed wedge-resections of the masses with Endo-GIA 30 Blue (Ethicon Endosurgery) with support in Gore-Tex (Seamguard, W.L. Gore & Associates, USA); the mediastinal lymph node was removed.

The patient was discharged on the third day. He began 6 cycles of adjuvant chemotherapy treatment with Cisplatin (100 mg/m^2, day 1) and Gemcitabine (1000 mg/m^2, day 1 and 8) every 21 days. After 12 months the CT total-body did not show the presence of relapses.

Discussion

Our study showed that MRI and CT are essential to allow the diagnosis and the staying of ethmoidal carcinoma differentiating between opacities due to secretions and opacities due to masses. MRI represents the best technique to characterize the neoplasm from nearby tissues and to define the periosteal and connective limits. In T2-weighed sequences MRI allows us to distinguish tumour from inflammatory tissue through signal intensity based on the different protein concentration; T1-weighed sequences show anatomical structures of small size (2, 3). High resolution CT allows to evidence better bony erosions, because it displays the calcification component of the bone, and to find the distant metasthies.

In order to have a more accurate diagnosis it is also useful to perform a trans-nasal biopsy.

After radiotherapy these exams present variations in the intensity of the adipose tissue signal, explained both in the increase of the thickness of the periplast component and in the thickness of the muscles, due to the inflammation and edema, secondary to the treatment, and to alterations of the medullary component of the bone. These variations are much more visible with MRI, that shows hypo and hyperintense areas, rather than with CT. The initial edema will leave space to a progressive fibrosis and to a reduction of muscular thickness 6 months after radiotherapy.

Naso-sinusal cancers are uncommon (about 0.8% of all the cancer and about 3% of head and neck cancers). The prognosis of these malignant neoplasms is unfavorable notwithstanding the several strategies of treatment (surgery and/or chemotherapy and/or radiotherapy). It is conditioned by the extension of the pathology to near zones (orbit, basicranium, encephalon and cavernous sinus) and by the aggressive istological type (melanoma, neuroendocrinous tumor and indifferentiated cancer). At present the therapeutic gold standard is the external monolateral subtotal ethmoidectomy (EMST) and anterior craniofacial resection (RCFA). The medial ethmoidectomy and maxillectomy of Session and Larson (1977) is an external resection of all ethmoidal bone except the fovea, the cribrose lamina and the whole lateral wall of nasal fossa as far as the pavimentum. We use a lateral rhinotomy, than we remove a nasal bone and a part of maxillary bone process. When it is possible we preserve the nasolacrimal system. When the neoplasm does not allow EMST for its large diffusion, it is better to make a safety craniofacial resection (RCF). In 1941 Dandy proposed a transcranial and transfacial surgery approach. There are two different surgical approaches in the cancer of ethmoidal roof by Melecki and Ketcham: a) transcranial and transfacial approach with frontal craniotomy and nasal-maxillary bony flap; b) transfacial approach.

A RCFA begins with the access to the tumour by the detachment of bony flaps followed by the excision of the bony block and the fleshy parts containing the neoplasm and then the zone is rebuilt. The resection margins are defined by the diagnostic image. If the cancer invades the FCA, we remove the median part of its floor (the ethmoidal fovea and the cribrose lamina). The excision block is sometimes constituted of a part of FCA (dura mater, olfactory benderel and cerebral parenchyma), of both the ethmoidal blocks, of the medial bony wall of the orbit or of the papyraceous laminae of the two sides; it can also be part of the nasal septum, of the lateral walls of the nasal fossae as far as the pavement in the site where the tumour is more advanced. These surgical technique are very aggressive and they have often functional and aesthetic consequences.

Endoscopic rhinosinusal surgery was born in the Seventies. For fifteen years these technique was considered the best treatment of the phlogistic rhinosinusal pathology because of its microinvasiveness (4, 5). Later in the Eighties and Nineties the technique was also used for benign nasosinusal tumours (inverted papilloma, juvenile angiofibroma) and for the basicranium pathology (dural plasty for rhinoliquor fistulae and the treatment of the sellar pathology). In the second half of the Nineties a good technical development allowed their use for the nasosinusal malignant tumour pathology (6-8). In 1995 Jorrisen (6) applied this technique in 8 patients suffering from nasosinusal malignant tumour but he did not show the efficacy of the procedure. Goffart et al. (9) showed the efficacy of endoscopic surgical technique in 78 patients (66 of them were treated with the pure endoscopic approach). The study was

effected in two different centers; total survival at 2 and 5 years was respectively 63.4% and 52.3%. Ethmoidal adenocarcinoma had better results, with a survival of 89.8% and 63.8% respectively after 2 and 5 years. Jorrisen (6) has shown that tumours involving the fore and posterior ethmoide, the upper side and the medial part of the maxillary sinus or the sphenoidal sinus and the front recess can be treated by an exclusive endoscopic approach. This technique is not advised for those tumours involving the fore-inferior wall of the maxillary sinus and the frontal sinus. If the cancer included the maxillary sinus and frontal sinus the endoscopic surgery is not indicated. Therefore, the tumours extending beyond the limits of the paranasal sinus are not generally treated by intranasal surgery.

Radio and chemotherapy results are still debated. Preoperative radiotherapy stabilizes the disease. In the postoperative stage, it allows sterilization of the residual neoplasm; this combination guarantees a survival rate of 45-50% at 5 years. An interstial brachytherapy with filiform sources of Iridium^{192} may be used if the mass has limited dimensions. Radiotherapy makes use of external bundles of high energy photons of ray type (ERT) or Gy radiations of Cobalt. Doses of 60 Gy/20 fractions in 5 weeks are used in the first stages of the pathology (T_1-T_2, N_0) when the surgical margins are negative and in patients with risk factors who underwent chemotherapy. Greater doses of 70 Gy are used for patients with stage T_3-T_4, N_1-N_2-N_3 who underwent only radiotherapy and did not present risk factors (10). It is also possible to use a further dose of 16-20 Gy in 10 fractions in those cases in which the surgical resection margins are positive. Postoperative radiotherapy should begin 6 weeks after surgical intervention. The utilization of systemic chemotherapy aims at improving aggregate survival. High local doses of chemotherapy have been correlated to a lower incidence of collateral effects. The use of cisplatin supplied locally in high concentrations, improves the response and the control of the disease. A better control of the pathology, of the peripheral metastases and of survival is obtained with neoadjuvant chemotherapy, which reduces the tumour mass. On the contrary, adjuvant chemotherapy is less tolerated especially in the more advanced stages when radiotherapy is usually used.

Follow-up depends on factors such as treatment and the individual risk of relapse; it is generally performed every 1-3 months in the first year and every 6

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