The patient was referred for oncologic evaluation and treated with cisplatin/etoposide chemotherapy for four cycles. Her β-HCG levels gradually decreased after surgery to normal levels by 3 months after surgery. She was in good condition 12 months after surgery with no evidence of recurrent disease on computed tomographic imaging.

Comment

Epithelioid trophoblastic tumor is a very rare tumor, which is grouped into a spectrum of gestational trophoblastic disease, along with hydatidiform moles, invasive mole, and choriocarcinoma [2]. Epithelioid trophoblastic tumor was first described in 1989 and, to date, fewer than 60 cases have been described in the literature. Epithelioid trophoblastic tumor is similar to choriocarcinoma but typically displays a monomorphic pattern of atypical trophoblastic cells, as was seen in our case, versus the dimorphic pattern that is seen in choriocarcinoma [3]. Epithelioid trophoblastic tumor usually has elevated β-HCG levels, but the amount of elevation is typically lower than the typical choriocarcinoma, in which β-HCG levels are commonly well above 2,500 mIU/mL.

Epithelioid trophoblastic tumor most commonly appears in women of reproductive age, with a mean age at presentation of 38 years. Epithelioid trophoblastic tumor generally originates in the uterus, and abnormal vaginal bleeding is the characteristic presentation. Metastatic spread to areas such as the lung and spine has been described previously [4]. Our case is unusual in that our patient had no uterine mass present, either on pelvic ultrasound or positron emission tomography imaging.

Epithelioid trophoblastic tumor has varying response rates to chemotherapy. Agents such as cisplatin, methotrexate, etoposide, and 5-fluorouracil have been used in case reports with different degrees of success. Because the diagnosis of primary pulmonary ETT is exceedingly rare, neoadjuvant chemotherapy is not feasible. Although we chose to treat our patient with chemotherapy after surgery, there is no consensus management strategy present in the literature.

The typical pelvic-based ETT has metastatic disease present approximately 25% of the time. Ten-year survival is approximately 90% [5]. Surgical resection appears to be the best treatment for these lesions. Metastatic disease present at the time of surgery and high mitotic index may be negative prognostic indicators. In patients with elevated β-HCG levels, a lung mass, and a negative gynecologic examination, primary ETT of the lung should be part of the differential diagnosis.

References


An Unusual Case of Pleural Chordoma

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Chordoma is a rare slow-growing neoplasm arising from notochordal remnants. In the United States, the annual incidence of chordoma is 0.08 per 100,000 and is more common in men than in women. The most common locations of chordoma are the cranial (32%), spinal (32.8%), and sacral (29.2%) regions [1]. We report an unusual case of pleural chordoma in a 45-year-old man.

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evaluation, which revealed a right vocal cord lesion. The patient was scheduled for a biopsy procedure of the right cord lesion under laryngoscopy. However, chest roentgenograms preoperatively showed a mass-like opacity in the left lung field. Follow-up computed tomography (CT) of the chest without contrast showed a small pleural effusion with a lumpy contour, innumerable small pleural nodules, and lymphadenopathy seen in the anteroposterior window (Fig 1).

The patient was referred for thoracic surgical intervention evaluation, and a thoracoscopic pleural biopsy procedure was performed. On inspection, the patient had innumerable yellowish white pleural nodules along the chest wall, diaphragm, mediastinal pleura, and visceral pleura (Fig 2).

The tumor was composed of large epithelioid or polygonal cells with copious clear cytoplasm and mild atypical vesicular nuclei. Immunostaining failed to provide any clear evidence of a specific line of differentiation. An initial pathologic diagnosis of clear cell carcinoma of the left pleura was made. The patient had a negative metastatic workup including positron emission tomography/CT, CT of the abdomen and pelvis, and brain and spine magnetic resonance imaging (MRI). He had excellent results on pulmonary function testing and a V/Q scan showed 36.5% of perfusion to the left lung. Based on the patient's young age, extensive pleural disease, and diagnosis of clear cell carcinoma, a left radical extrapleural pneumonectomy and resection of the diaphragm and pericardium with reconstruction with Gore-Tex (W.L. Gore and Associates, Flagstaff, AZ) mesh was performed. Intraoperatively, it was noted that there was extrapleural extension of the tumor overlying the T6–8 vertebral bodies next to the aorta. This area was resected en bloc with the rest of the specimen. The surgical specimen consisted of the paraspinal tumor, the left lung with attached parietal pleura, and a portion of the diaphragm. The paraspinal tumor consisted of 3 × 3.5 × 0.5 cm reddish soft tissue. There were multiple firm tan nodules involving and forming adhesions between the parietal pleura, visceral pleura, chest wall, and diaphragm. The largest nodule measured 10 cm and involved the deep surface of the parietal pleura. On cut section, there were areas of intraparenchymal extension of the nodules into the left lung as well as the chest wall. The hematoxylin and eosin-stained slides showed round nuclei and abundant vacuolated cytoplasm representing physaliferous features, and immunohistochemical evaluation demonstrated epithelial membrane antigen, cytokeratin (CAM5.2), and S-100 positivity in tumor cells. Therefore, the final diagnosis of metastatic chordoma was made and the paraspinal tumor seemed to be the primary lesion. Interestingly, we reviewed the preoperative thoracic spine MRI scan after the operation and did detect some signal heterogeneity at the T6 level, but the radiologist read it as nonspecific and it can represent hemangioma. The patient recovered well from the procedure and completed adjuvant radiation therapy to the left hemithorax with a boost to the paravertebral spine region, for a total dose of 6660 cGy. The patient was disease free at 1 year follow-up.

Comment

Chordoma is a rare tumor arising from notochordal remnants. It is more common in men than in women and is rare among patients younger than 40 years. Median survival has been reported to be 6.29 years, with 5- and 10-year survival rates of 67.6% and 39.9%, respectively [1]. Chordoma has an indolent course and often appears radiographically as destructive bone lesions [2]. The distinct feature of chordoma is the local invasion of the intervertebral disc space, which is seldom seen in osteosarcoma and chondrosarcoma. However, in our patient, there was no intervertebral lesion detected on

Fig 1. Computed tomography (CT) of chest without contrast showed small pleural effusion with lumpy contour and innumerable small pleural nodules.

Fig 2. Multiple yellowish white pleural nodules along the chest wall, diaphragm, mediastinal pleura, and visceral pleura.
CT and MRI. Microscopically, chordoma manifests as 1 of 3 types: classic, chondroid, or dedifferentiated [3]. The mainstay of treatment of chordoma is en bloc excision with wide margins and postoperative external-beam radiation therapy. Radical resection of chordoma with negative tumor margins significantly decreases local recurrence rates compared with incomplete excision [4, 5]. The addition of radiation therapy can lead to 5-year local control of 10% to 40% and increases overall survival after incomplete resection [2]. However, the proximity of the spinal cord to the chordoma limits the maximal deliverable dose of conventional radiation therapy to 40 to 60 Gy. Newer techniques in radiation therapy, such as the 3-dimensional conformal technique combining photon and proton beams, can allow higher doses of radiation to be delivered to the target tumor site while minimizing injury to the surrounding tissue, especially the spinal cord [2, 6]. Overall, conventional chemotherapy has been proved ineffective in the treatment of chordoma.

In summary, chordoma is a very rare tumor arising from notochordal remnants. It has a high local recurrence rate despite aggressive debulking operations combined with radiation therapy.

References

Rare Case of Lung Abscess Caused by a Swallowed Denture

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Accidentally swallowed dentures can cause serious complications when they are not diagnosed and treated promptly. We report an extremely rare case of a lung abscess caused by a swallowed complete denture. Chest computed tomography and endoscopic examination revealed that a swallowed denture migrated to the right upper lobe through an esophageopulmonary fistula, and a lung abscess developed. A life-saving and curative operation was performed with no significant postoperative complications. To the best of our knowledge, such a clinical condition has not yet been described in the literature.

Swallowing dentures and dental plates is not so rare among elderly patients. Most patients are aware of the diagnosis before seeking treatment; exceptions are in the mentally handicapped, the demented, and those under the influence of alcohol [1]. Managing swallowed dentures is problematic in that they are sharp and have been associated with high morbidity and mortality because of abscess formation or perforation causing mediastinitis. Most swallowed dentures land in the gastrointestinal tract, but some persist in the esophagus. Once a foreign body is impacted in the esophagus, life-threatening complications, such as esophageal perforation, penetration to the great vessels, esophageotracheal fistula, and mediastinitis, become grave concerns. We present the case of an 85-year-old woman who accidentally swallowed her denture and received a radical operation, along with a brief review of the available literature.

An 85-year-old woman first presented to a medical practitioner with a 1-week history of anorexia and cough. Because she could not explain a clinical history and symptoms adequately on account of her dementia, her family believed she had a common cold. Chest roentgenograph showed a strange shadow similar to human teeth in her upper lung field (Fig 1). She was diagnosed with a pulmonary foreign body and sent to our hospital. At the initial visit, it was very difficult to communicate with her as a result of her dementia, but the lack of denture could be confirmed. Chest computed tomography revealed the existence of the swallowed denture in the right upper lobe with abscess formation. Computed tomography also showed an esophago-pulmonary fistula connecting to the lung abscess (Fig 2). Esophagoscopy was performed, which revealed the esophageotracheal fistula in the right wall at a level of 18 cm from the incisors; a small part of swallowed denture and necrotic lung tissue were observed in the abscess cavity (Fig 3). However, because catastrophic hemorrhage was a major concern, an attempt was not made to remove the denture endoscopically during esophagoscopy.

The emergency operation was performed through a standard right posterolateral thoracotomy. Although inflammatory adhesion of the lung parenchyma with parietal or mediastinal pleura was significant, there were no complications when they are not diagnosed and treated promptly. We report an extremely rare case of a lung abscess caused by a swallowed complete denture. Chest computed tomography and endoscopic examination revealed that a swallowed denture migrated to the right upper lobe through an esophago-pulmonary fistula, and a lung abscess developed. A life-saving and curative operation was performed with no significant postoperative complications. To the best of our knowledge, such a clinical condition has not yet been described in the literature.

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