Superior Mesenteric Artery Syndrome Preceded by Diabetes Insipidus in Advanced Lung Cancer with Pituitary Metastasis – A Case Report

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The pituitary gland is an uncommon site of metastasis from most primary cancers of the breast in women and the lung in men. Because of the direct blood supply, typical pituitary metastases develop in the posterior lobe of the pituitary and present with symptoms of diabetes insipidus (DI). Superior mesenteric artery (SMA) syndrome is a rare disorder resulting from external compression of the third portion of the duodenum by the SMA anteriorly and the aorta posteriorly. The aortomesenteric angle is narrowed typically because of loss of the mesenteric fat pad due to rapid body weight loss or scoliosis surgery. We report the case of a 40-year-old male with advanced lung cancer presenting with progressive polyuria for months followed by postprandial vomiting. SMA syndrome due to severe dehydration and weight loss resulting from pituitary metastasis with DI was diagnosed. He then received desmopressin and nutritional support. The symptoms of DI and SMA syndrome resolved well within 2 weeks after restoring the body fluid status and increasing the mesenteric mass. ***(Thorac Med 2016; 31: 250-255)***

Key words: diabetes insipidus, superior mesenteric artery syndrome, lung cancer, pituitary metastasis

Introduction

The pituitary gland is an uncommon site of metastasis, with an incidence of 1~3.6% in patients with advanced malignant disease [1]. Most of the primary cancers are of the breast in women and the lung in men. Because of the direct blood supply, most pituitary metastases are to the posterior lobe of the pituitary and present with symptoms of diabetes insipidus (DI). Superior mesenteric artery (SMA) syndrome is a rare disorder resulting from external compression of the third portion of the duodenum by the SMA anteriorly and the aorta posteriorly. Typical symptoms of SMA syndrome include postprandial nausea and bilious vomiting, abdominal pain, and body weight loss. We report the case of a male with advanced lung cancer with SMA syndrome preceded by dehydration due to pituitary metastasis with DI.

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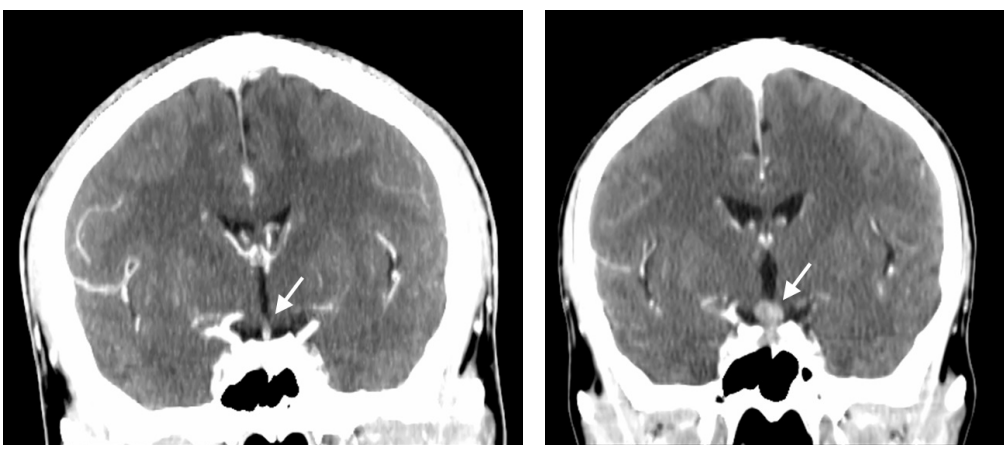
Case Report

A 40-year-old male construction worker with advanced lung cancer visited this hospital because of intermittent vomiting and frequent bowel movements for 1 month.

The patient had smoked cigarettes at 1 pack per day for 20 years. About 4 years prior to this entry, he presented with productive cough for months and was found to have diffuse micronodules in bilateral lungs. He was diagnosed as having adenocarcinoma of the lung, right lower lobe, cT4N0M1, cStage IV with bony metastasis (American Joint Committee on Cancer, 6th edition; AJCC 6). The patient subsequently received 4 cycles of first-line chemotherapy with gemcitabine plus cisplatin with a partial response (PR); and then, 6 cycles of docetaxel with a PR, and erlotinib for a total of 16 months, with a PR. Disease progression was noted 7 months prior to this entry, when multiple metastatic nodules in the brain parenchyma and 1 in the pituitary gland (Figure 1) were found. The patient received external beam radiation therapy to the whole brain and 8 cycles of pemetrexed, again, with a PR. He experienced grade 2 anorexia and vomiting after chemotherapy.

During the continuous pemetrexed treatment course, he became progressively thirsty for 6 months followed by postprandial vomiting for 1 month. Polyuria was noted with no evidence of hyperglycemia. His vomiting occurred mostly when in the supine position. Lateral decubitus would prevent this symptom. During this period, he also noticed body weight loss of 7 kg (from 59 kg to 52 kg). He was admitted for etiology survey. The urine output collection was reported to be more than 8000 milliliters a day. His urine sodium was 30 mmol/L, urine osmolality: 93 mOsm/kg, serum sodium: 145 mmol/L, serum osmolality: 296 mOsm/Kg, thyroid-stimulating hormone (TSH): 14.69 μIU/ml (0.35~5.5 μIU/ml), free thyroxine (free T4): 0.44 ng/dl (0.7~1.8 ng/dl), adrenocorticotropic hormone (ACTH): 38.9 pg/ml (5~77 pg/ml),



(A) (B)

Fig. 1. Computed tomography of the brain. (A) Computed tomography of the brain revealed no space-occupying lesion in the pituitary gland (arrow); image taken 6 months earlier. (B) A new contrast-enhanced pituitary tumor (arrow) protruding into the hypophyseal fossa while the patient developed polydipsia and polyuria. Effacement of the sulci indicated intracranial pressure might be increased in both figures.

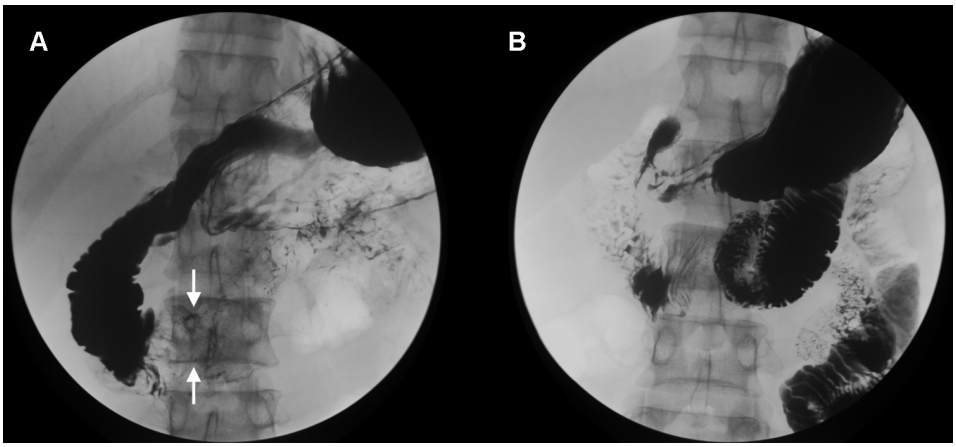


Fig. 2. Upper gastrointestinal series. (A) Upper gastrointestinal series with the patient in a supine position showed a vertical band-like indentation (arrows) at the third portion of the duodenum, implying external compression by a tubular or vascular structure. Note there was only minimal sluggish barium passage beyond this level. (B) With the patient in a prone position, the barium passage seemed normal. The dynamic change confirms the clinical diagnosis of SMA syndrome.

and cortisol (8AM): 3.5 μg/dl (4.3~22.4 μg/dl). A water deprivation test revealed a dramatic fall in urine output, from 360 to 35 ml, in the first hour; urine osmolality increased from 93 to 568 mOsm/kg after vasopressin therapy. The patient was diagnosed as having central type DI secondary to pituitary metastasis. Desmopressin (DDAVP) was prescribed.

An upper gastrointestinal endoscopy reported erosive esophagitis and superficial gastritis, which may have explained his poor appetite and postprandial vomiting. Upper gastrointestinal series with barium swallowing confirmed the diagnosis of SMA syndrome by revealing barium obstruction at the third portion of the duodenum with band-like indentation (Figure 2). His symptoms of SMA syndrome responded to DDAVP and nutritional support within 2 weeks after restoration of body fluid status and mesenteric fat content, although his weight did not show a significant gain for the rest of his life. The patient succumbed to his disease 7 months later.

Discussion

The pituitary gland is an uncommon site of metastasis. The incidence of pituitary metastasis was reported to be around 1~3.6% in patients with malignant tumors [1]. Most pituitary metastases occurred in the 6~7th decades of life, with breast tumors accounting for most cases in women (20~30%) and lung tumors in men (30~50%) [2]. The posterior lobe is the most common site of metastasis chiefly due to the direct blood supply, the large area of contact with the adjacent dura mater, and the lack of blood supply to the anterior lobe [3]. Some cancers, especially those of the breast, metastasize preferentially to the anterior lobe of the pituitary gland [1,3].

The majority of pituitary metastases are asymptomatic due to the advanced stage of the disease or the patients being too weak for the metastasis to become clinically evident [4-5]. In rare cases, symptomatic pituitary metastasis may be the first sign of a distant cancer. The

most common symptom is DI, which occurred in 45~70% of cases [6-7]. Up to 20% of patients diagnosed with DI actually had pituitary metastases [8]. Other manifestations include ophthalmoplegia, headaches, visual field abnormalities and anterior pituitary dysfunction. Differentiating between a benign pituitary adenoma and metastasis may be difficult. High-resolution computed tomography (HRCT) and magnetic resonance imaging are sensitive, but lack specificity [9]. The rapidly developing pituitary tumor in our case accompanied with multiple brain metastases rendered enlargement from a preexisting adenoma unlikely.

The management of pituitary metastasis is essentially palliative and depends on the extent of the disease and the symptoms. Radiation therapy and chemotherapy are usually the initial treatment modalities, in association with replacement hormones to relieve symptoms. Surgical decompression may be needed when suprasellar extension causes visual impairment or pain. The prognosis remains poor, not because of the metastasis per se, but because of the aggressiveness of the underlying end-stage disease. Median survival was 6~7 months in reported series [7-8].

SMA syndrome is a rare disorder resulting from external compression of the third portion of the duodenum due to a reduced angle and reduced distance between the SMA anteriorly and the abdominal aorta posteriorly [10-11]. The incidence of SMA syndrome is estimated to be 0.013~0.3% in the general population; it is more common in females and occurs in patients from 35~45 years of age [12]. The aortomesenteric angle is narrowed typically because of loss of the mesenteric fat pad with rapid body weight loss, scoliosis surgery, or a high insertion of the ligament of Treitz. Other etiologies include mechanical compression by retroperitoneal tumor and bariatric surgery. Patients with SMA syndrome typically present with postprandial nausea and bilious vomiting, and abdominal pain associated with weight loss [11]. Because of the rarity and nonspecific symptoms, SMA syndrome can be easily misdiagnosed. The diagnosis can be established by showing an abrupt cutoff in the third portion of the duodenum in an upper gastrointestinal series or by showing a reduced aortomesenteric angle and reduced aortomesenteric distance in a contrast-enhanced CT scan.

Conservative treatment of SMA syndrome usually starts with weight gain in an attempt to increase the mesenteric fat pad. Nasojejunal tube feeding or total parenteral nutrition may be needed in patients unable to tolerate oral intake. Surgical decompression by division of the ligament of Treitz may be necessary if medical treatment fails. Laparoscopic duodenojejunostomy recently has become the preferred treatment modality [10].

Our case is unique in that the patient first had a lung cancer with pituitary metastasis and resultant DI, which in turn caused dehydration and weight loss precipitating the development of SMA syndrome. The symptoms responded well to DDAVP and nutritional support. It is assumed that increasing the mesenteric mass corrected the narrowed aortomesenteric angle and increased the aortomesenteric distance.

SMA syndrome is usually related to a vicious cycle in terminal cancer status. Physicians that treat cancer patients should maintain a high index of clinical suspicion of this condition when treating patients with unexplained postprandial vomiting.

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晚期肺癌因腦下腺轉移引發的尿崩症，隨後發生上腸繫膜動脈症候群－病例報告

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轉移癌並不常發生在腦下腺，發生在腦下腺的轉移癌在女性以原發性乳癌最常見，而在男性則是肺癌。由於直接的血流供應，典型的腦下腺轉移癌好發於後葉，常伴隨著尿崩症狀。上腸繫膜動脈症候群這個罕見疾病，是由於十二指腸的第三部分，其外部前方受到來自上腸繫膜動脈以及後方受到來自主動脈的壓迫所致。典型的上腸繫膜動脈症候群致病機轉來自於腸繫膜的脂肪層變薄，導致上腸繫膜動脈及主動脈的夾角變小；而造成這樣的主要原因包括不同疾病導致的快速體重減輕或接受脊柱側彎手術。在此我們報告一位 40 歲男性晚期肺癌病例，表現出多尿症狀數個月後發生飯後嘔吐症狀。經診斷為上腸繫膜動脈症候群，而此症候群是由於腦下腺轉移造成尿崩症，進而引發嚴重脫水所致。他接受了去氨加壓素治療與營養支持，恢復其體液狀態並增加腸繫膜容積後，他的尿崩症狀以及上腸繫膜動脈症候群，在兩周內有顯著改善。*(胸腔醫學 **2016; 31: 250-255**)*

關鍵詞：尿崩症，上腸繫膜動脈症候群，肺癌，腦下腺轉移

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