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Muhammad Imran Butt, Muhammad Asif Shahzad

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Case Report: A 68-year-old female was admitted to hospital for investigation of delirium associated with polydipsia and polyuria. She developed hematemesis in hospital which required further investigations. Patient was found to have GIST, diagnosed histologically, associated with diabetes insipidus. She required ICU admission and developed nosocomial pneumonia. Despite appropriate treatment, our patient died.

Conclusion: Gastrointestinal stromal tumors usually present as subepithelial neoplasms of gastrointestinal tract (GIT). However, there has been no association described so far between GISTs and diabetes insipidus and needs further work up to evaluate the link between the two entities.

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CASE REPORT

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Keywords: Diabetes insipidus, Gastrointestinal, Stromal, Tumor

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INTRODUCTION

Gastrointestinal stromal tumors (GISTs) account for less than 1% of gastrointestinal tumors, however, these are the most common mesenchymal tumors of gastrointestinal tract [1]. Given the relatively low annual incidence of clinical GISTs, only a few microscopic tumors would grow into a clinically significant size with malignant potential. Further study is needed to confirm these observations and to clarify the genetic events responsible for the transformation of microscopic GIST lesions into clinically relevant GISTs.

CASE REPORT

A 68-year-old female was presented to the hospital with a two-week history of polydipsia, polyuria, severe lethargy, dehydration and fluctuating state of consciousness. There was no history of abdominal pain or gastrointestinal bleeding. She was admitted under endocrinology unit for further investigation. She had a background history of stable asthma and osteoporosis being treated with alendronate.

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On examination, she was hemodynamically stable except for mild tachycardia. Her Glasgow coma scale (GCS) was 15/15. However, a fluctuating sensorium was identified. Systemic examination was unremarkable in particular no abdominal mass was identified, and there was neither neurological deficit nor visual field defects.

Initial biochemistry of the patient revealed serum sodium levels of 150 mmol/L (range of serum sodium during admission: 141–155 mmol/L), serum osmolality 291 mmol/L (range of serum osmolality during admission: 291–311 mmol/L), urine sodium 16 mmol/L (range of urine sodium during admission: 16–84 mmol/L) and urine osmolality was 195 mosm/kg (range of urine osmolality during admission: 95–394 mosm/kg). Serum glucose, urea and creatinine were within normal range, however, she did have mild elevation of liver function tests which got further deteriorated later in the admission.

To hunt the cause of delirium; septic screen was performed but no infective cause was identified hence diabetes insipidus was thought to be the most likely cause of delirium. She was commenced on desmopressin and a strict fluid balance was maintained. She displayed minimal improvement even with higher dose of desmopressin. Subsequently, amiloride was also added with no significant effect. Due to her ongoing delirium, water deprivation test was difficult to perform. Computed tomography scan of brain was unremarkable, however, an MRI scan of brain was not possible without sedation hence delayed. Due to continued delirium, septic screen was repeated (including lumbar puncture) and on this occasion urinary tract infection (UTI) was identified (which was most likely hospital acquired) for which intravenous antibiotics were commenced.

On day-6 of admission she had a coffee ground vomitus requiring urgent upper gastrointestinal endoscopy which revealed gastric ulcer (most likely GIST related) but no mass and treatment with intravenous pantoprazole was commenced. After this procedure her conscious level deteriorated significantly to the extent to require intensive care and subsequent endotracheal intubation.

During her ICU stay she continued receiving desmopressin, however, Amiloride was stopped. Magnetic resonance imaging scan of brain was organized which did not show any abnormality particularly in hypothalamus or pituitary gland. Incidentally serum ammonia levels were found elevated (as high as 118 μ mol/L) and on further investigation she was diagnosed with urea cycle disorder for which she required a special diet through nasogastric tube. During ICU admission she also developed ventilator associated pneumonia with *Klebsiella pneumoniae* which was also treated with appropriate Intravenous antibiotics according to the culture results. Screening for autoimmune disorder was negative.

A hunt continued to find the cause of delirium and resistant diabetes insipidus for which CT chest, abdomen and pelvis was arranged. CT abdomen revealed Omental caking and normal liver and pancreas. Her CA-125 level

was 183ku/L whereas CA-19.9, CEA, AFP and hCG were within normal limits. Further to that a CT guided biopsy of omentum was arranged which confirmed the diagnosis of GIST by showing spindle cell morphology on histology (Figure 1). Additionally, immunophenotyping showed predominance of CD117 on stain for c-Kit (Figure 2). Unfortunately, the patient passed away before the histological diagnosis of GIST.

DISCUSSION

Gastrointestinal tract neoplasms originating from mesenchyma typically present as subepithelial tumors. They are divided into two large group amongst which GISTs are the most common. They originate mostly in the stomach and proximal small intestine, but can occur anywhere in gastrointestinal tract including omentum, mesentery, and peritoneum.

Figure 1: Gastrointestinal stromal tumors with spindle cell morphology (H&E stain, x40)

Figure 2: Stain for c-Kit (CD117).

The cellular morphology of GISTs ranges from predominantly spindle-shaped to epithelioid in character. Histologically, the appearance of these tumors usually falls into one of three relatively uniform categories including spindle cell type which is most common “70%” and our patient also had spindle cell morphology. Other two types include epithelioid type (20%) and mixed type (10%). GISTs frequently metastasize to liver and peritoneum and rarely to regional lymph nodes. They uncommonly metastasize to the lungs, the most common site of metastasis for most soft tissue sarcomas. There are no case reports of GIST being metastasized to pituitary gland or hypothalamus. The CD117 antigen is a known part of the KIT transmembrane receptor tyrosine kinase that is the product of the KIT (c-kit) proto-oncogene. More than 90% of GISTs are KIT positive where KIT gene leads to formation of a variant of KIT protein which is then abnormally activated enabling the oncogenic signaling in the cell.

Central diabetes insipidus results from diminished secretion of antidiuretic hormone (ADH). It is mostly idiopathic (likely due to immune mediated injury to ADH-secreting cells). It can also result from trauma, surgery of pituitary gland, or hypoxic brain injury. Some familial cases have been described in literature but their occurrence is rare [2].

Nephrogenic diabetes insipidus is characterized by pathologically varying degrees of renal resistance to ADH. It is usually mild and occurs more commonly in elderly or patients with renal impairment due to decreased concentrating ability.

The patients who have partial urea cycle enzyme deficiency may have chronic hyperammonemia or occur only during metabolic decompensation [3]. However, it is usually not severe. Plasma ammonia concentration should be measured at the time of decompensation because ammonia may be normal during healthy periods.

CONCLUSION

The most common mesenchymal neoplasms affecting the gastrointestinal tract are collectively referred to as gastrointestinal stromal tumors. To our knowledge no link has been reported between gastrointestinal stromal tumors (GIST) and diabetes insipidus but we think that GIST may cause micrometastasis to hypothalamus which may explain the presentation for our patient. Further study and literature are required in this regard.

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Author Contributions

Muhammad Imran Butt – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Muhammad Asif Shahzad – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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