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Pancreatic metastasis of renal cell carcinoma filling into the duct of Santorini.

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Abstract

A 78-year-old man who underwent right nephrectomy for renal cell carcinoma (RCC) 18 years ago visited our hospital complaining of abdominal pain. Imaging revealed that the pancreatic head tumor obstructed the Santorini duct. We suspected a pancreatic intraductal tumor, such as an intraductal tubulopapillary neoplasm or intraductal papillary mucinous neoplasm. Thus, the patient underwent subtotal stomach-preserving pancreaticoduodenectomy. Pathological findings confirmed the diagnosis of metastatic RCC. Herein, we report a case of pancreatic metastasis of an RCC that presented with a tumor in the pancreatic duct.

Key words: Pancreatic intraductal tumor, Pancreatic metastasis, Renal cell carcinoma

Introduction

The pancreas is a rare site for renal cell carcinoma (RCC) metastasis, accounting for 2% of all pancreatic neoplasms [1]. In terms of manifestation of the pancreatic metastasis following a diagnosis of RCC, most studies disclose mean time intervals of >10 years during which the patient may be disease-free [2]. On contrast-enhanced computed tomography (CT) images, pancreatic metastases of RCCs are typical well-demarcated and hypervascular tumors [3-6]. Additionally, pancreatic metastases from RCCs rarely affect the pancreatic duct [7-10]. We herein report a rare case of pancreatic metastasis from an RCC with a tumor in the Santorini duct.

Case report

A 78-year-old man with an 18-year history of right nephrectomy for RCC visited our hospital complaining of abdominal pain. Hematological investigations revealed the following: pancreatic amylase 99 U/I (16–52), lipase 236 U/I (16–60), carcinoembryonic antigen 3.1 ng/mL (≤ 5), and carbohydrate antigen 19-9 88.0 U/mL (≤ 37). CT revealed dilation of the main pancreatic duct (MPD) in the pancreatic body and tail (Figure 1a). Endoscopic ultrasound (EUS) revealed a hypoechoic tumor measuring 30 mm inside the duct of Santorini extending to the confluence of the MPD (Figure 1b, 1c). Color Doppler revealed blood flow into

the mass (Figure 1d). Contrast-enhanced harmonic imaging of EUS (CH-EUS) of the tumor revealed a hyper-enhancement pattern (Figure 1e). Magnetic resonance cholangiopancreatography and endoscopic retrograde pancreatography (ERP) revealed MPD disruption at the head of the pancreas and distal MPD dilation (Figure 2a, b). Intraductal ultrasonography revealed a tumor filling the duct of Santorini, extending to the confluence of the MPD (Figure 2c). Brush cytology and transpapillary biopsy findings were not conclusive. Based on the above findings, we diagnosed a pancreatic intraductal tumor, intraductal tubulopapillary neoplasm (ITPN) or intraductal papillary mucinous neoplasm (IPMN). The patient underwent subtotal stomach-preserving pancreaticoduodenectomy. The resected specimen revealed a tumor in the pancreatic head (Figure 3a). Pathologically, loupe findings demonstrated that most of the tumor was localized in the duct of Santorini; however, a few tumor cells were also observed in the pancreatic parenchyma (Figure 3b). Microscopic findings revealed that the tumor had filled the duct of Santorini. Tumor cells in the duct of Santorini and pancreatic parenchyma had a clear cytoplasm (Figure 3c). Immunohistochemical analysis revealed positive expression of CA9, CD10, and PAX8, which led to the diagnosis of metastatic RCC (Figure 3d, 3e). The postoperative course was uneventful, and the follow-up revealed the patient's survival for 6 months without further treatment or any evidence of recurrence or metastasis.

Discussion

RCC primarily metastasizes to the lung, liver, bone, and adrenal tissue. In only 2.8% of RCC patients, the tumor has been reported to metastasize to the pancreas [11,12]. Pancreatic metastasis of recurrent RCC has been reported in more than 85% of patients within 3 years after nephrectomy [13,14]. However, cases of recurrence after 3 years or more have often been reported [15].

There are two mechanisms of RCC metastasis: lymphatic metastasis and hematogenous metastasis. Hematogenous metastasis has been shown to be more common in cases of pancreatic metastasis [16,17]. Pancreatic metastases from RCC could be found in any part of the pancreatic parenchyma [18-20].

Pancreatic metastases from RCC exhibit abundant blood flow. Therefore, contrast-enhanced CT shows a characteristic intense homogeneous arterial phase enhancement in pancreatic metastases, higher than that observed in the normal pancreatic parenchyma [3-6]. In the present case, contrast-enhanced CT was not performed because of renal failure. Although CH-EUS showed a hyper-enhancement pattern in the early vascular phase, it was difficult to diagnose pancreatic metastasis from RCC because CH-EUS findings of pancreatic intraductal tumors are not well known.

In the present case, ERP and EUS revealed that the tumor filled the duct of Santorini. In general, pancreatic

metastases from RCC have a high affinity for the pancreatic parenchyma and causes non-destructive changes in the pancreatic duct [21]. The image findings of this case were not typical of pancreatic metastases of RCC. The present case was difficult to diagnose preoperatively owing to similarities with a pancreatic intraductal tumor. Although rare, a few cases of pancreatic metastases of RCCs affect the pancreatic duct have been reported (table 1) [7-10]. Compared to other reported cases, the unique feature of our case was the tumor morphology filling into the pancreatic duct, and the image resembled pancreatic intraductal tumor, such as an intraductal tubulopapillary neoplasm or intraductal papillary mucinous neoplasm. In addition, there were no reports of the pancreatic metastatic tumor of RCCs filled in the duct of Santorini.

Pathologically, most of the tumor was located in the duct of Santorini; however, most of the pancreatic duct epithelium was normal and was only partially invaded by the metastasis from RCC. Additionally, RCC metastasis was also observed in the pancreatic parenchyma. Based on the above findings, the RCC was assumed to have metastasized to the pancreatic parenchyma, partially invade the pancreatic duct, and then extend into the pancreatic duct.

In conclusion, we encountered a rare case of pancreatic metastasis of RCC, where the tumor filled the pancreatic duct.

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The authors have no conflicts of interest to disclose concerning this article.

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Legends to Figures

Figure 1

Computed tomography (CT) and endoscopic ultrasound (EUS) imaging.

(a) Abdominal CT. The main pancreatic duct (MPD) is dilated from the pancreatic body to the tail (yellow arrows). (b) EUS image and (c) schema. A hypoechoic mass measuring 30 mm is detected in the duct of Santorini. The mass extends to the confluence of the pancreatic ducts and dilates the downstream MPD. (d) Color Doppler EUS showing several blood flows in the mass. (e) Contrast-enhanced EUS: The mass showing a hyper-enhancement pattern.

Figure 2

Magnetic resonance imaging and endoscopic retrograde pancreatography (ERP).

(a) Magnetic resonance cholangiopancreatography image. The main pancreatic duct (MPD) showing disruption in the pancreatic head (yellow arrow) with distal pancreatic duct dilation. (b) ERP showing disruption of the MPD in the pancreatic head (yellow arrow). The MPD is dilated from the pancreatic body to the tail. (c) An intraductal ultrasonography image at the **【A】** line of the (b) image. A hypoechoic mass is located in the duct of Santorini (yellow arrow), extending to the confluence of the MPD (white arrow).

Figure 3

Pathological findings.

- (a) The resected specimen, a solid tumor, is found in the duct of Santorini (white arrow).
- (b) The loupe image of the area surrounded by the blue line in image (a). A tumor is observed inside the duct of Santorini (white arrow). Furthermore, another tumor was detected in the pancreatic parenchyma (yellow arrow).
- (c) Clear cells were observed in both the ducts of the Santorini tumor and the pancreatic parenchyma tumor.
- (d) Immunohistochemically, tumor cells showing CA9 expression.
- (e) Immunohistochemically, tumor cells showing PAX8 expression.

Figure 1

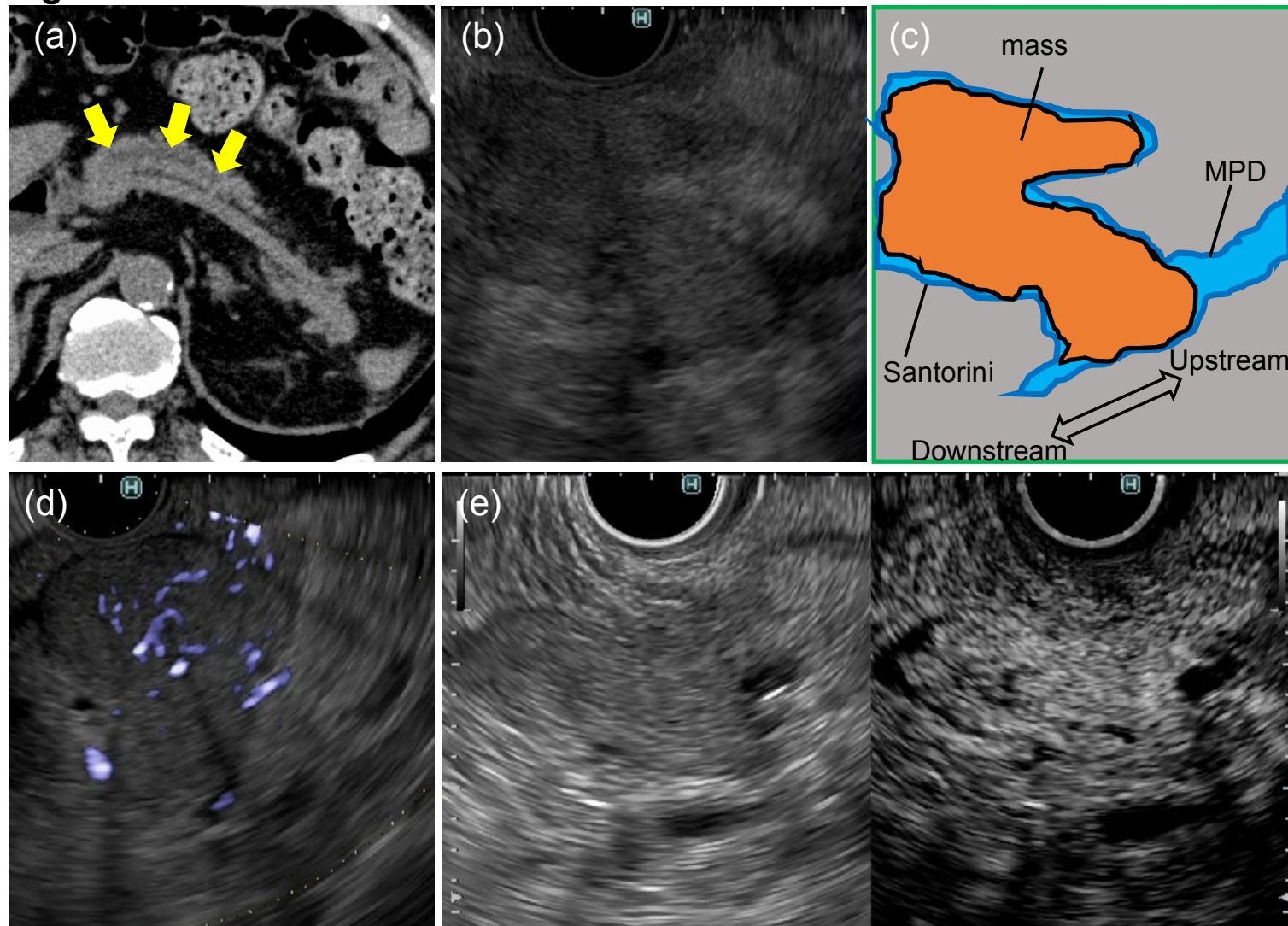


Figure 2

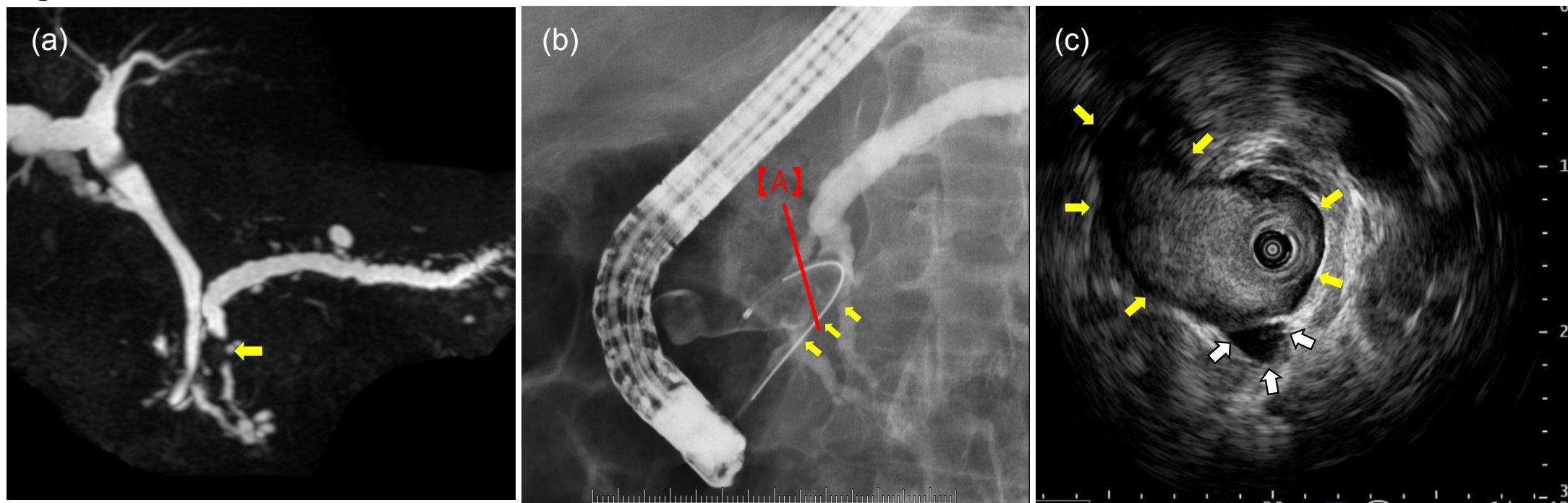
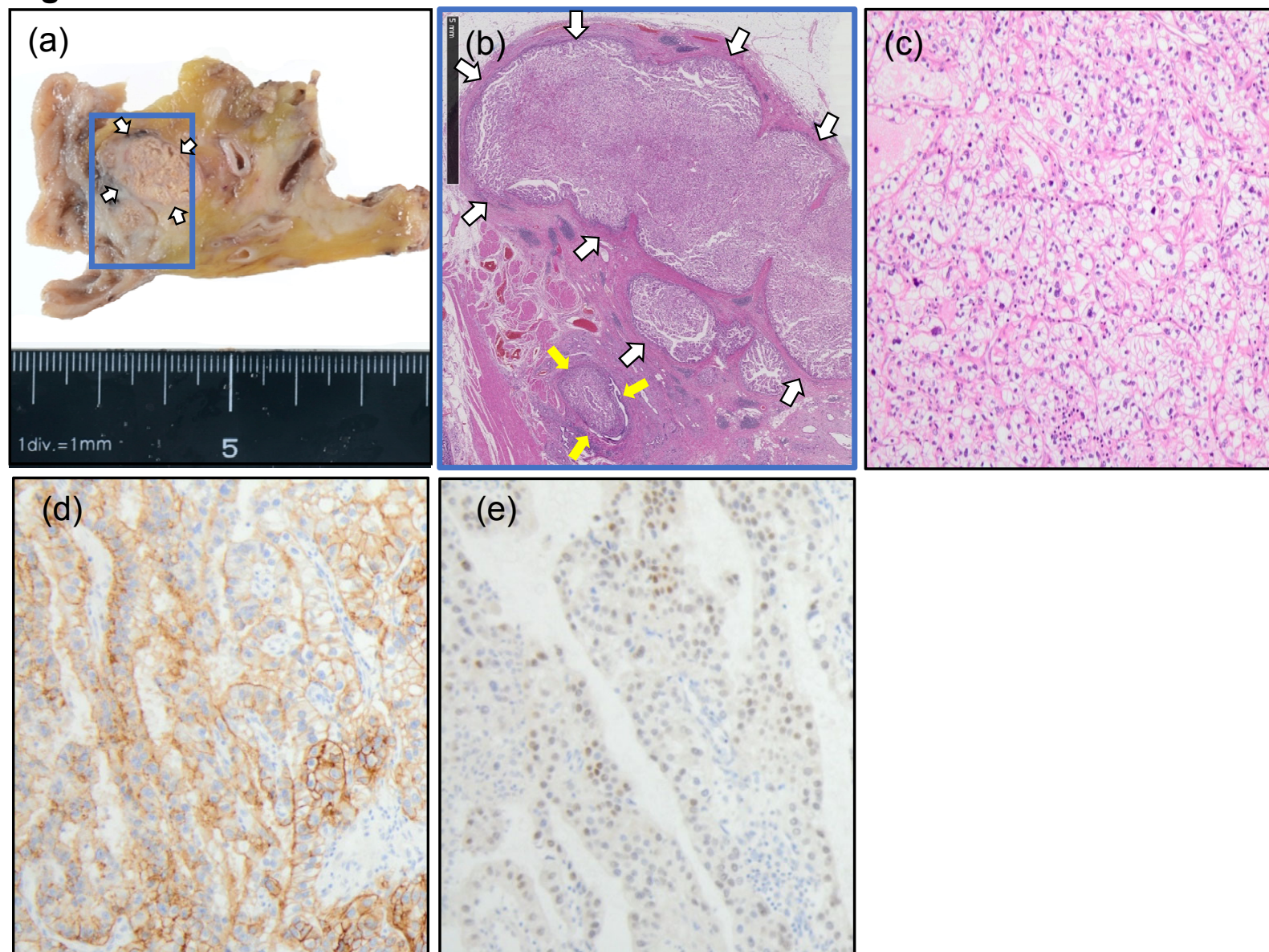


Figure 3



Case	Age (years)	Gender	Opportunity of diagnosis	Primary site	Interval from nephrectomy (years)	Local lymph node metastasis	Tumor location
1	66	Male	Diabetes mellitus	Right kidney	11	Absent	Main pancreatic duct
2	66	Male	Diabetes mellitus	Right kidney	17	N/A	Main pancreatic duct
3	60	Male	Pancreatitis	Right kidney	3	N/A	Main pancreatic duct
4	58	Male	Diabetes mellites	Right kidney	12	N/A	Main pancreatic duct
5	59	Male	Jaundice	Left kidney	14	N/A	Main pancreatic duct
6	65	Male	Jaundice	Left kidney	4	N/A	Main pancreatic duct
7	70	Male	Evaluated for renal transplantation	Bilateral kidney	4	N/A	Main pancreatic duct
This case	78	Male	Abdominal pain	Right kidney	18	Absent	Duct of Santorini

N/A: Data not available