Case Report

Journal Of Clinical & Medical Images Case Reports

Open Access, Volume - 2

An Unusual Presentation of Chylothorax Status Post Extensive Retroperitoneal Resection

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Received Date : June 13, 2022
Accepted Date : July 18, 2022
Published Date : Aug 10, 2022
Archived : www.jcmimagescasereports.org

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Abstract

Chylothorax may present as a postoperative complication most frequently after thoracic surgeries, although with an incidence rate of less than 1%. Chylothorax after retroperitoneal surgery is extremely rare and arises, subsequently, due to chylous ascites or chyloretroperitoneum. Diaphragmatic defects, however, have been shown to be responsible for the occurrence [16]. To the extent of our literature review, isolated chylothorax following a retroperitoneal surgery has never been reported.

We describe a case of this unusual complication, which became clinically apparent 13 days after extensive retroperitoneal surgery in a 65-year-old man. The chylothorax compromised the patient's respiratory and hemodynamic stability, requiring frequent thoracenteses, a low fat diet, octreotide, and total parental nutrition (TPN). After failing conservative management, interventional radiology performed a lymphangiography, which revealed a fistulous communication between the cisterna chyli and right pleural cavity. Iatrogenic trauma to the right hemidiaphragm and cisterna chyli was the suspected cause for this tract formation and subsequent development of an isolated chylothorax. Lymphatic duct/tract embolization was performed and the patient was continued on conservative management. Over the course of the next several days, the chylothorax progressively decreased and by post-operative day 5 imaging revealed only trace signs of an effusion. By post-op day 12 the patient continued to have persistent low-volume output from the pleural drain, and he was subsequently returned to Interventional Radiology (IR) for "touch up" percutaneous embolization of the coil mass using 5 mL of a (1:4) glue: lipiodol concoction. Output from the pleural drain ceased, and the catheter was ultimately removed on post-op day 15. The patient had an unremarkable subsequent hospital course and was discharged to home on post-op day 18.

Introduction

The cisterna chyli and thoracic duct are important structures of the lymphatic system that serve to transport lymph and chyle into systemic circulation [1]. Inadvertent injury to either structure is most commonly iatrogenic, particularly as a complication of thoracic or abdominal surgeries [2]. Postoperative chyle leaks are rare complications, occurring less than 1% [3]. They can vary in presentation, depending on the location of injury along the lymphatic tract. Chylothorax is due to the accumulation of chyle within the pleural space and is most commonly seen in the setting of thoracic duct injury during thoracic procedures, such as esophagectomy or during cardiac surgery [2, 3]. Damage to the cisterna chyli is a complication that can also occur in the setting of any retroperitoneal surgery. Cisterna chyli injury is likely to present as chyle accumulation in the retroperitoneum, known as chyloretroperitoneum [4]. Chylothorax after retroperitoneal surgery is extremely rare and there are only a few case reports in the literature [4-13]. In these reports, there has been simultaneous chyloretroperitoneum in addition to chylothorax. We present what we believe to be the first isolated chylothorax, following a retroperitoneal surgery. We review the literature of the pathophysiology and discuss treatment options, particularly the utilization of interventional radiology.

Case Report

A 65-year-old man with a past medical history of hypertension and psoriatic arthritis presented to our facilities for radical resection of a large right retroperitoneal mass. The mass was discovered on imaging during an evaluation for anemia at an outside facility. He had a colonoscopy and an esophagogastroduodenoscopy (EGD) performed that revealed benign polyps. During this evaluation, a computed tomography (CT) scan was performed revealing a large, heterogeneously enhancing

Citation: BK Grewal, Jeffrey Marteslo. An Unusual Presentation of Chylothorax Status Post Extensive Retroperitoneal Resection. J Clin Med Img Case Rep. 2022; 2(4): 1211.

right-sided retroperitoneal mass (**Figure 1**). On imaging, the mass displayed mass effect and displacement of the inferior vena cava, right kidney, right adrenal gland, and adhesion to the right diaphragm. The retroperitoneal mass was biopsied and pathology results revealed an undifferentiated spindle cell sarcoma. He was offered surgical resection by the outside facility but instead presented to our facilities for a second opinion, which ultimately also consisted of surgical intervention.

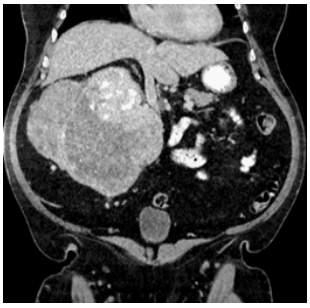

Figure 1: *A pre-surgical computed tomography scan of the abdomen and pelvis showing a large, heterogeneously enhancing right-sided retroperitoneal mass, with internal coarse calcifications. Mass effect and displacement of the inferior vena cava (IVC), right kidney, right adrenal gland is seen. Loss of fat planes with the IVC and the right diaphragm concerning for tumor infiltration/ adhesion.*

The patient agreed to surgical resection of the sarcoma and proceeded to undergo presurgical planning and workup. On the day of the operation, a large 40 x 18 cm sarcoma that encased the right kidney and densely adhered to the inferior vena cava was discovered. A radical resection of the sarcoma, right nephrectomy, right adrenalectomy, partial resection with repair of inferior vena cava, and partial omentectomy was performed. In addition, an umbilical hernia sac was resected and repaired. The patient required a nasogastric tube for the first 3 postoperative days and resumed a regular diet on postoperative day 6. He began having bowel movements, voiding, and ambulating at baseline by postoperative day 7. He was discharged on postoperative day 9 and was scheduled for an outpatient follow up 4 days later.

At the outpatient follow up, the patient was found to be profoundly hypotensive with systolic pressures in the 60-70s and diastolic pressures in the 30-40s. The patient was transported to our facility's emergency department for further evaluation and management. Upon a history and physical examination, the patient reported feeling weak, short of breath, and lightheaded for the past 48 hours. The patient also reported that he had not been able to maintain adequate oral intake, due to mild abdominal discomfort which he characterized as fullness. The patient denied fever, chills, vomiting, diarrhea, and constipation. Vitals at presentation included blood pressure of 55/30, temperature of 36.7 C (98 F), respiratory rate of 20, pulse of 72/minute, and an oxygen saturation of 96%. Physical exam revealed decreased air exchange of the right middle and lower lobes with normal respiratory effort, and a small surgical site infection with an area of fat necrosis and erythema around the lower incision. The abdomen was soft, nontender, and nondistended. Significant laboratory findings included a serum creatinine of 4.3 mg/dL, serum carbon dioxide of 32 mmol/L, white blood cell (WBC) count of 19.41 cells/mm3, and a lactate of 2.4 mmol/L. A chest radiograph was ordered that revealed a large right-sided pleural effusion (Figure 2). The patient was started on intravenous steroids, fluids, and antibiotics, per emergency department sepsis protocol. The patient's hypotension was fluid responsive, and he was admitted for further management.

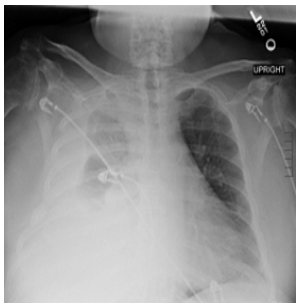

Figure 2: *An anterior-posterior chest radiograph showing a large right-sided pleural effusion.*

Pulmonology was consulted to perform a thoracentesis, which revealed 2.5 L of purulent outflow. During this procedure, pulmonology noted that a part of the right diaphragm did not appear to move in synchrony with the remainder of the diaphragm. The pleural effusion appearance along with the clinical findings were initially concerning for empyema. However, pleural fluid analysis revealed a triglyceride count of 651 mg/dL, raising concern for a chylothorax. The pleural fluid had a glucose of 117 mg/dL and a pH of 7.34, which were not suggestive of empyema. A right sided pigtail chest tube was placed to drain the high output chylothorax and to alleviate the patient's shortness of breath (**Figure 3**). The patient was initially placed on a nonfat diet and octreotide was initiated as conservative management for the suspected chyle leak. A CT of the abdomen and pelvis (CTAP) was ordered to assess for abdominal fluid collection (**Figure 4**).

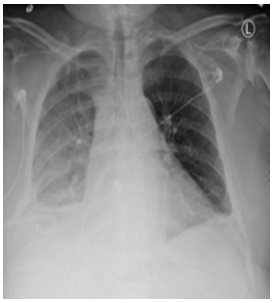

Figure 3: *An anterior-posterior chest radiograph showing a right-sided pigtail catheter placed in the right pleural space, in the collection of the effusion.*

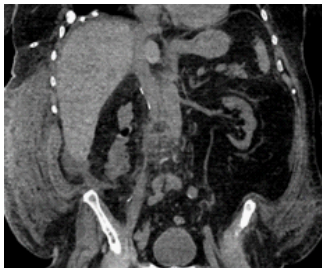

Figure 4: *A post-surgical computed tomography scan of the abdomen and pelvis revealing resection of the right-sided retroperitoneal sarcoma, right kidney, right adrenal gland. No fluid collection or residual tumor.*

The CT revealed trace ascites and mild flank edema. Pulmonology recommended draining 1-1.5 L/daily, then clamping the chest tube to avoid protein loss and fluid imbalance. The patient was also monitored with daily chest radiographs to assess the effusion size and to look for complications such as pneumothorax. By hospital day 3 the patient continued to have high chylous outflow (1-1.5 L/day) from the chest tube, despite conservative management. The decision was made to place a peripherally inserted catheter to start TPN and consult IR to evaluate with a lymphangiography. There was concern for trauma to the lymphatic tract due to recent surgery and persistent high outflow from the chylothorax. Therefore, IR scheduled the patient for lymphangiography with ad hoc intervention.

On hospital day 8, post-procedure day 0, the patient had no laboratory abnormalities: prothrombin time (PT) 12.5 seconds, internationalized normalized ration (INR) 1.0, platelet count of 224 cells/mm3. Subcutaneous heparin was held the morning of, and the patient was placed under general anesthesia for the entirety of the procedure. Using direct ultrasound guidance, multiple 25 gauge (G) spinal needles were used to access bilateral inguinal lymph nodes. Under intermittent fluoroscopic guidance, gradual injection of intranodal Lipiodol was performed to opacify the lymphatic channels. Contrast ascent was tracked through the lymphatic pathways until reaching the cisterna chyli (**Figure 5**). The cisterna chyli was adequately opacified at the level of L2-L3 and was accessed percutaneously with a 21 G Chiba needle. A microwire and microcatheter were advanced through this access and appeared to move laterally out of the cisterna chyli to the right. A lymphangiogram was subsequently performed through the catheter, with contrast, to allow for opacification and better visualization. Under fluoroscopy, a discrete foci of contrast extravasation arose laterally to the right of the cisterna chyli (**Figure 6**). Rather than dispersing throughout the retroperitoneal space, the contrast extravasation appeared to respect a well-formed boundary. The extravasated contrast ascended, cranially, towards the right hemidiaphragm and ultimately into the right pleural space, tracking towards the indwelling pleural catheter (**Figure 7**). Multiple micro coils were deployed through the microcatheter and into the nidus of the cisterna chyli injury. The deployed coils could also be seen coiling in the formed spaced previously outlined by the extravasated contrast. Coil deployment was halted after adequate contrast stasis was observed under fluoroscopy. Thereafter, a combination of 2:1 lipiodol: TRUFILL n-Butyl cyanoacrylate (n-BCA) embolization glue was slowly administered into the nidus of the cisterna chyli injury (Figure 8). The microcatheter was then removed, hemostasis was achieved, and sterile dressings were placed over the access sites.

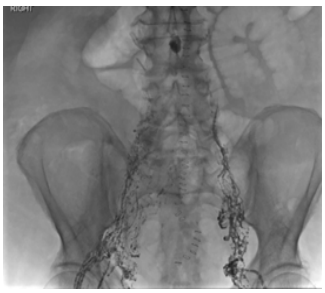

Figure 5: *A bilateral pelvic intranodal lymphangiography showing Lipiodol ascent tracking through lymphatic channels from bilateral inguinal lymph nodes to a dilated, saccular structure, known as the cisterna chyli.*

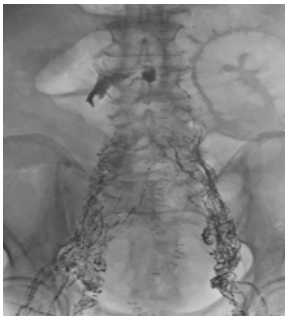

Figure 6: *Lymphangiography showing a discrete foci of contrast extravasation arising laterally to the right of the cisterna chyli.*

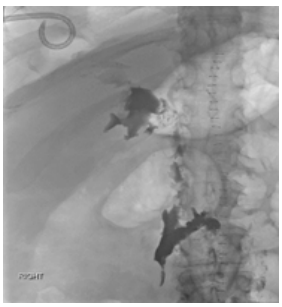

Figure 7: *Lymphangiography showing the extravasated contrast ascent, cranially, towards the right diaphragm, respecting a well-formed boundary rather than spilling in the retroperitoneal space. Ultimately, the extravasated contrasted reached the right pleural space, tracking towards the indwelling pigtail catheter.*

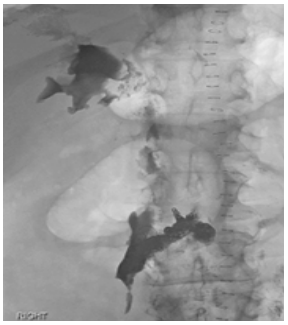

Figure 8: *Lymphangiography showing deployed coils coiled in the fistulous tract along with a combination of 2:1 lipiodol: TRUFILL n-Butyl cyanoacrylate embolization glue allowing for stasis of contrast.*

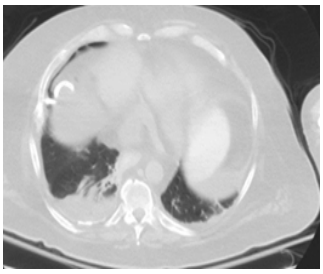

Figure 9: *Post-procedural day 5 computed tomography scan of the chest showing no loculated fluid. A trace effusion of stable size is present on the right.*

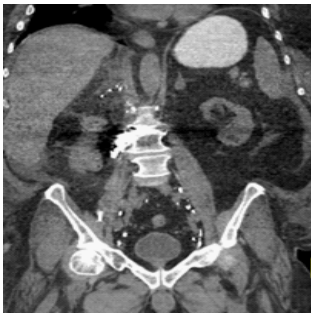

Figure 10: *Post-procedural day 5 computed tomography scan of the abdomen showing stable postsurgical changes of resection of the right kidney, right adrenal gland, and the large right retroperitoneal mass. No fluid collection or residual tumor. Mild ileus and trace ascites are present.*

Postoperatively, the right-sided pigtail catheter remained in place to allow for continued drainage of any remaining chylothorax. Daily chest x-rays were continued to monitor the size of the chylothorax and complications such as pneumothorax. A nonfat diet, TPN, and a daily octreotide regimen were also continued. Over the course of several days, the size of the effusion progressively decreased. On postprocedural day 5 a non-contrast CT of the chest and abdomen and pelvis were taken. CT of the chest showed a stable, trace pleural effusion on the right side (**Figure 9**). CTAP revealed trace ascites and no retroperitoneal fluid collections (**Figure 10**). The pleural catheter remained in place, as there continued to be persistent low-volume output. By postprocedural day 12, IR was again consulted for re-evaluation. Ultimately the decision was made to perform a "touch-up" percutaneous embolization of the coil mass using 5 mL of a 1:4 (glue: lipiodol) concoction (**Figure 11**). Subsequently, output from the pleural catheter ceased and was ultimately removed on post procedural day 15 (**Figure 12**). The patient had an unremarkable remaining hospital course and was discharged to home in stable condition on post procedural day 18.

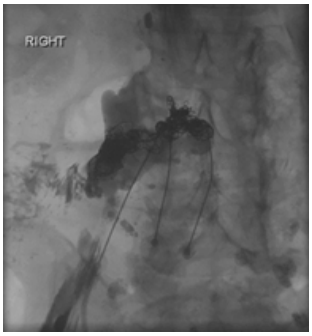

Figure 11: *Postprocedural day 12, a lymphangiography showing a percutaneous embolization of the previously embolized tract being embolized with additional coils and lipiodol/glue mix.*

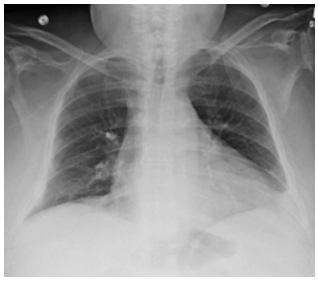

Figure 12: *Post-procedural day 15, an anterior-posterior chest radiograph showing the absence of a right-side pleural effusion and right pigtail catheter.*

Discussion

Chyle is a bodily fluid consisting of lymph, chylomicrons, proteins, lymphocytes, immunoglobulins, vitamins, and electrolytes. Chyle plays an important role in fluid balance, nutrition, and immunity. Two to four liters of chyle are transported through the cisterna chyli and thoracic duct each day [1]. The cisterna chyli is a dilated, saccular lymphatic structure arising in retroperitoneum, typically anterior to the first or second lumbar vertebrae. The thoracic duct arises from the cisterna chyli and ascends cephalad through the aortic hiatus to enter the posterior mediastinum, eventually terminating at the junction of the left subclavian and jugular veins [14]. Chyle leaks are a rare complication that manifest when either of these structures are damaged. Leakage may have several manifestations, such as chylothorax or chyloretroperitoneum.

The diagnosis of chylothorax is usually made clinically. Approximately 50% of chylous effusions are milky in appearance, the other half can present as bloody, serious, yellow, or green. Therefore, diagnosis cannot be made solely on appearance. Pleural fluid analysis is required and usually yields an exudate with a triglyceride level above 110 mg/dL. The presence of chylomicrons in the effusion can also confirm the diagnosis [3]. The etiology of chylothorax can be classified into three broad categories, spontaneous (non-traumatic), traumatic, and idiopathic [3]. Historically, non-traumatic chylothorax was the more common cause for chylothorax, accounting for two-thirds of all cases. Recently, traumatic chylothorax, particularly postoperative chylothorax, accounts for more than 50% of all cases described in the literature [15]. Postoperative chylothorax is almost exclusively in relation to thoracic surgeries: esophagostomy carries the highest risk of 5 to 10%, followed by lung resection with mediastinal lymph node dissection with 3 to 7% risk [3]. Other thoracic procedures carrying smaller risks include mediastinal tumor resection, thoracic aneurysm repair, sympathectomy, and other surgeries involving the base of the neck. However, chylothorax caused by injury to the cisterna chyli is extremely rare and it has only been reported in a few case reports [4-13]. In these reported cases the chylothorax subsequently developed from concurrent chylous ascites or chyloretroperitoneum. Our case is particularly unique due to the sole presentation of chylothorax due cisterna chyli injury following retroperitoneal surgery.

In this case, the patient had a large 40 x 18 cm retroperitoneal sarcoma that was directly adherent to the right diaphragm. The patient underwent extensive retroperitoneal surgery that radically removed the sarcoma, right kidney, and right adrenal gland. On postoperative day 13, a right-sided chylothorax was discovered. IR was consulted for a lymphangiography due to the high output chylothorax and concern for thoracic duct damage from recent surgery. During the lymphangiography, discrete contrast extravasation was found, ascending cranially from the cisterna chyli, through the right diaphragm and finally reaching the right pleural space. Injury to the thoracic duct below the 5th thoracic vertebrae results in pleural effusion on the right side, and damage above this level occurs as a left-sided pleural effusion [1]. Even though the injury was below the level of the 5th vertebrae, we believe the injury was suffered to the cisterna chyli rather than the thoracic duct. Contrast extravasation was adjacent to a dilated lymphatic structure in the regions of 2nd and 3rd lumbar vertebrae, which is the likely setting of the cisterna chyli. Cisterna chyli injury is likely to manifest as chyloretroperitoneum or chylous ascites [2]. Previous studies in the literature highlight the development of chylothorax after chylous ascites or chyloretroperitoneum due to diaphragmatic defects. One report states that congenital diaphragm weakness develops due insufficient deposition of tendon and muscle, which allow for the formation of blebs. These blebs form from the evagination of peritoneum and rupture secondary to increased intraabdominal pressure such as ascites [16]. An alternative explanation is porous diaphragm syndrome with the movement of fluid through diaphragmatic pores or defects. These porous defects allow subdiaphragmatic ascites to leak through into the thorax, similar to how a hepatic hydrothorax develops [17]. However, to the best of our knowledge no reports exist in the literature of an isolated chylothorax after a retroperitoneal surgery. Therefore, we speculate that the right diaphragm and cisterna chyli were damaged at the time of initial surgery. The injury

to these structures allowed for possible tract formation. This communication between the right diaphragm and cisterna chyli allowed for a chyle fistula to develop. Thus, chyle traveled this tract into the right pleural space instead of pooling in the retroperitoneal space.

The treatment approach for chylothorax can vary based on etiology (traumatic vs nontraumatic), effusion volume, and reaccumulation rate. In cases of chylous output less than 1 L/day, conservative management with dietary restriction is a reasonable initial approach [18]. This may include placing the patient on a diet plan that excludes long chain triglycerides such as a low to nonfat diet or medium chain triglyceride diet. The purpose of this is to slow lymph flow and allow spontaneous closure of the lymphatic duct [18]. TPN is considered an appropriate alternative or next step-up in treatment since it allows for better control over nutrient administration [19]. Although Octreotide and Somatostatin have not been confirmed for the indication of chylothorax, they are commonly used medications in addition to dietary management to slow chyle flow [20]. Therapeutic thoracentesis can be used in the setting of a large, symptomatic chylothorax. Drainage allows for alleviation of symptoms such as dyspnea, however, it does not reduce chyle flow. It is recommended to drain no more than 1-1.5 L/day due increased risk of immunosuppression and malnutrition [1].

When conservative treatment fails patients often require procedural interventions. General indications for surgery for postoperative chylothorax include chylous leak greater than 1 L/day for more than 5 days, persistent leak for more than 2 weeks despite conservative management, or development of insurmountable nutritional or metabolic complications [21]. Historically, the only remaining options were surgical. Surgical techniques reported in the literature included parietal pleurectomy, pleurodesis, pleuroperitoneal shunts, and thoracic duct ligation. In general, the main difficulty in surgical treatment is identifying the thoracic duct or leak [22]. Today, interventional radiology provides several image-guided procedures which offer a minimally invasive approach used to treat traumatic and non-traumatic chylothorax. Pelvic intranodal lymphangiography allows for visualization of the lymphatic system to assess for damage and leaks along the tract [23]. If a leak is identified, thoracic duct embolization has become a viable primary treatment for chylous leaks, particularly within the chest [24]. Embolization is performed when the thoracic duct is cannulated with a catheter proximally to the leak. First metallic coils are placed to provide a matrix for glue polymerization. Although it is well known that lymphangiography alone can result in cessation of chylothorax, having a success rate near 80% [23]. Studies suggest that the use of a combination of coils and glue is necessary to achieve optimal TDE. Embolization with coils alone result in lower success rates when compared with coils combined with glue (84% vs 91%) [24]. Although TDE is technically difficult, a 67% effective catherization is achievable, with successful management in over 90% of cases [25-27]. TDE is less invasive than surgery, associated with less complications (3% vs 38%), and has decreased mortality with no fatal outcome reported [28]. Hence, TDE is an important alternative and first line treatment to patients with traumatic and nontraumatic chylous leaks.

Our patient continued to accumulate a daily chylothorax, exceeding 1 L/day, for more than a week, despite being managed on a chyle leak diet, an octreotide regimen, chest tube drainage, and TPN. Consideration for high output in the setting of recent retroperitoneal surgery raised concern for thoracic duct injury. IR was present at our facility and well-experienced to access and treat lymphatic leaks. Therefore, IR services were called upon to perform a pelvic intranodal lymphangiography with possible embolization. The patient was discovered to have an injury at the level of the cisterna chyli, with a leak present on the right side. Contrast ascended along a fistulous tract, through the right hemidiaphragm and into the right pleural space, approaching the indwelling chest tube. This tract was suspected to be the cause of the chylothorax and was embolized with coils and glue.

In conclusion, the combined occurrence of chylothorax and chyloretroperitoneum is a very rare complication after retroperitoneal surgery and has been report in only a few cases. An isolated chylothorax after retroperitoneal surgery occurs even less and this may be the first reported case. Initial management with a low-fat diet, medium chain triglycerides, TPN, and therapeutic thoracentesis are simple and effective treatments. Surgical treatment should be reserved for patients who failed conservative measures. Interventional radiology provides alternative, first line treatments to surgical approaches that have high success rates and low morbidity and mortality.

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