

## TWO CASES OF INTRALOBAR SEQUESTRATION OF THE LUNG

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Since Pryce's accounts of intralobar sequestration of the lung in 1946 and Pryce, Holmes Sellors, and Blair's in 1947, the pathology and clinical recognition of this condition have been placed on a firm basis. With the advance of thoracic surgery more cases have come to light and the dangers inherent in this condition have been recognized. Harris and Lewis in 1940 first pointed out the hazards of lobectomy when the presence of an anomalous artery is not recognized, their case ending fatally due to uncontrolled haemorrhage. Two other fatal cases have since been reported (Butler, 1947; Douglass, 1948).

The abnormality consists in the disconnexion from the main bronchial tree of a portion of lung tissue, and in the supply to it of a large artery arising from the lower thoracic or upper abdominal aorta. This bronchopulmonary mass or cyst lies within or in close proximity to the lower lobe, more commonly on the left side. Sometimes as a result of infection in the mass a communication develops between its bronchial spaces and the normal tree.

The anomalous vessel has the outward appearance and histological characters of a pulmonary artery. It is subject to atheromatous changes, perhaps due to associated infection in the mass, perhaps to the incongruity between the aortic pressure and its somewhat thin elastic walls. The abnormal vessel supplies either normally connected lung (Type 1), or the sequestered mass and adjacent normal lung (Type 2), or mass only (Type 3).

Bruwer, Claggett, and McDonald (1950) suggested that the anomalous vessel anastomosed with the main pulmonary artery, but Kergin (1952) showed by vinylite casts of the anomalous vessel and pulmonary artery and bronchial tree that no communication existed between the anomalous vessel and the pulmonary artery.

In the present cases, as well as in the earlier one from Green Lane Hospital (McDowell, Robb, Hinds, and Nicks, 1951), iodized oil injected into the anomalous vessel drained out of the pulmonary

vein, and radiographs taken of the specimen after injection of the oil into the anomalous vessels showed free communication between it and the inferior pulmonary vein. Most authors agree that the venous return is through the pulmonary vein.

The present cases are reported as a contribution to the subject, and one case is interesting in having two large anomalous vessels coming off the aorta. Douglass's case (1948) is the only other on record with two anomalous arteries. That case in addition had an associated vein draining into the azygos.

### CASE REPORTS

CASE 1.—J. T. S., a well developed boy, aged 10 years, was referred to the thoracic consultation committee on February 23, 1953, with a tentative diagnosis of left lower lobe bronchiectasis. At the age of 3 months he had an attack of pneumonia and since then four further attacks. The sides concerned were not specified. For years he had had a chronic cough with pale green, mucopurulent, inoffensive sputum. He often had colds.

The boy's complexion was "muddy" suggesting chronic toxæmia. There was slight flattening of the left lower chest anteriorly, the percussion note being impaired at the left paravertebral base from the ninth space downwards. Breath sounds were clearly audible over this area and did not differ much from those over the rest of the left chest or the right base. Coarse rhonchi were heard over both sides of the chest. There was a reduction in vocal resonance over this area and a dubious increase in vocal fremitus. There was clubbing of the toes with similar changes in the finger tips, but no drum stick broadening. The temperature on admission was 100° F.

The tentative diagnosis of intralobar sequestration was based on a critical consideration of serial radiographs. The earliest available films of January 8, 1952 (Fig. 1), showed a fairly dense opacity at the base of the left hemithorax partly relieved by relatively radiolucent areas and without a clear-cut upper margin. In association the heart was markedly displaced to the right in the normal inspiratory film. This displacement was increased on expiration without any suggestion of obstructive emphysema of the lung field

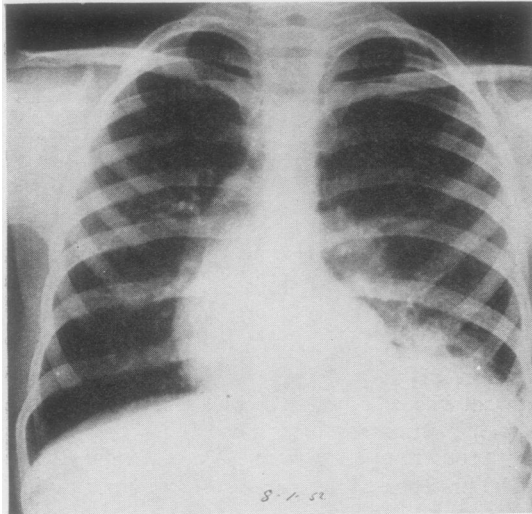


FIG. 1.—Case 1. Radiograph of the chest showing fairly dense opacity at the left base and cystic spaces. The heart is displaced to the right.

above the basal opacity. A film of January 13, 1953 (Fig. 2), shows the left basal opacity reduced in extent. The fluctuation in extent of the basal opacity is clearly seen in the left lateral films. There is a marked reduction from January 8, 1952 (Fig. 3), to March 4, 1953 (Fig. 4), with a substantial increase again two days later (Fig. 5).

Bronchography on March 11, 1953, demonstrated a left bronchial tree of standard pattern with the exception of an abrupt termination of the posterior basal bronchus 3 cm. from its origin after giving off one posterior and one anterior sub-segmental branch. There was no suggestion of bronchial dilatation and the outlined bronchi are displaced upwards, forwards, or laterally by the basal opacity.

This composite picture conforms in every respect with the case previously recorded from Green Lane Hospital (McDowell *et al.*, 1951), and may perhaps be taken as a pattern for the recognition of the condition of sequestration following the establishment of a communication with the normal bronchial tree. The picture is that of a semi-cystic mass situated typically in the postero-medial aspect of the hemithorax, susceptible to sudden changes in bulk related to partial evacuation of its contents or the trapping of air. In the latter event it may be capable of displacing neighbouring structures—diaphragm or mediastinum—in a manner comparable to that of obstructive emphysema of the lung. The additional features are displacements of otherwise normal branches of the bronchial tree and the presence of one (or possibly more) abruptly terminated bronchial fillings which represent the site of fistulous communication between the bronchial cysts of the sequestered mass and the normal bronchial tree.

**Operation.**—The left chest was entered on March 18, 1953, through the bed of the resected seventh

rib. The upper lobe was normal with a deep fissure separating the lingula from it like a middle lobe. There was partial separation of the apical segment of the lower lobe from the remainder by a fissure. This apical segment was large, and, together with the anterior basal region, constituted the only normal parts of the lower lobe. The lower posterior and medial parts felt indurated and functionless. Dense adhesions were present between them and the pericardium, diaphragm, and vertebral gutter, and over the aorta. After considerable dissection two large branches were defined coming off the descending aorta like twin funnels, each almost 1 cm. in diameter,  $1\frac{1}{2}$  cm. long, and lying about 1 cm. apart. There was  $2\frac{1}{2}$  cm. of normal aorta between the lower of these two vessels and the diaphragm. The branches of the pulmonary artery to the lower lobe were smaller than normal, as was also the branch to the lingula. The lower lobe was removed *in toto*—normal tissue plus indurated mass. The only unusual step was the clearing of the adhesions and the division of the two anomalous vessels. The chest was closed with two tubes to underwater seals.

The boy made an uneventful recovery, his pink complexion making a marked contrast to his former muddy appearance.

**Specimen.**—The specimen was inflated and the main bronchus was tied, thus maintaining the inflated state. A deep groove separated the lobe into an upper portion which was aerated and spongy and a lower consolidated and cystic portion. Low on the postero-medial aspect two arteries were seen entering the mass. (Fig. 6 shows the anterior view and Fig. 7 the lateral.)

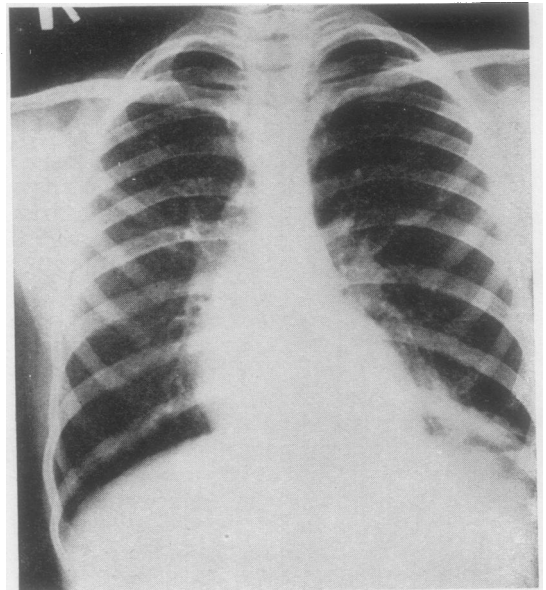


FIG. 2.—Case 1. Radiograph of the chest five days later, showing reduction of size of the mass in the left base. The cystic spaces are evident.

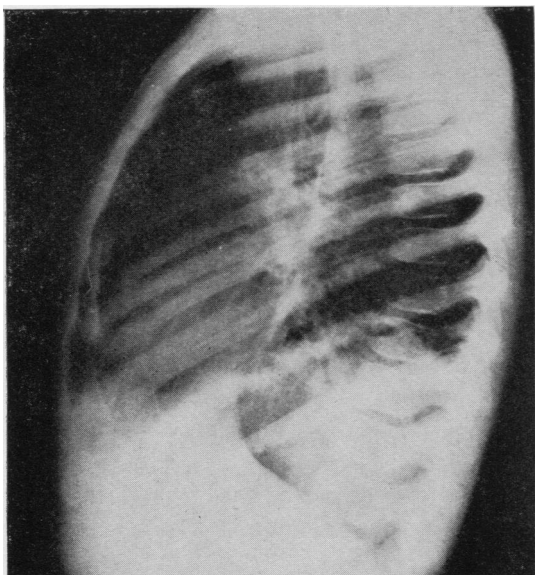


FIG. 3.—Case 1. Left lateral radiograph (January 8, 1952) showing sequestered mass and cystic spaces.

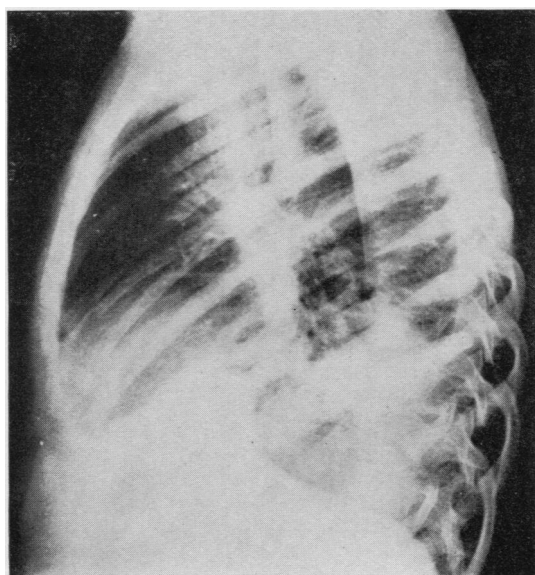


FIG. 5.—Case 1. Left lateral radiograph (March 6, 1953) showing increase in size of the mass.

Lipiodol was injected into these arteries and was found to run out of the inferior pulmonary vein which had been divided at the hilum. Radiographs were taken of the specimen after injection of the "lipiodol,"

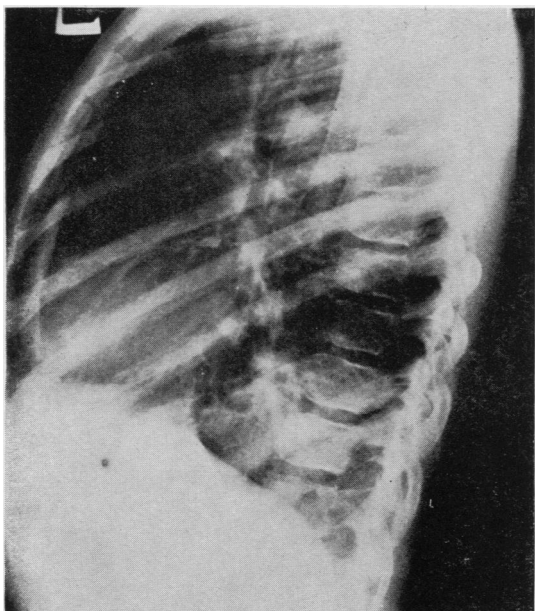


FIG. 4.—Case 1. Left lateral radiograph (March 4, 1953) showing reduction in size of the mass. The cystic spaces are very evident in this film.

and it was seen that the arteries supplied the sequestered mass only (Fig. 8) and that they communicated freely with the pulmonary vein (Fig. 9). The case thus belonged to Pryce's Type 3.

On section (Fig. 10) the upper part of the specimen consisted of pale lung tissue of normal appearance. The lower portion was composed of irregularly arranged cystic spaces. Most of these spaces were filled with inspissated mucopurulent secretion. Irregular bronchial channels connected some of the large cysts. The anomalous arteries could be seen branching through the lower portion and showed occasional small areas of atheroma.

*Microscopy.*—Microscopically the lower portion of the specimen was composed of distorted and inflamed pulmonary elements. Except for occasional strands of tissue no functioning lung parenchyma was found, the alveoli being represented by small, epithelial-lined spaces with thick fibrotic walls. The large cystic spaces were lined by normal bronchial epithelium and were filled with mucus and inflammatory debris. The abnormal arteries were thick-walled and composed predominantly of elastic tissue comparable in structure with the pulmonary artery entering the normal lung hilum. Occasional atheromatous plaques were seen in the intima. The parenchyma of the upper portion of the specimen was normal, although some of the bronchioles contained purulent secretion.

The specimen thus represented an intralobar sequestration in the lower lobe of the left lung, with two anomalous arteries from the descending aorta supplying the disconnected mass.

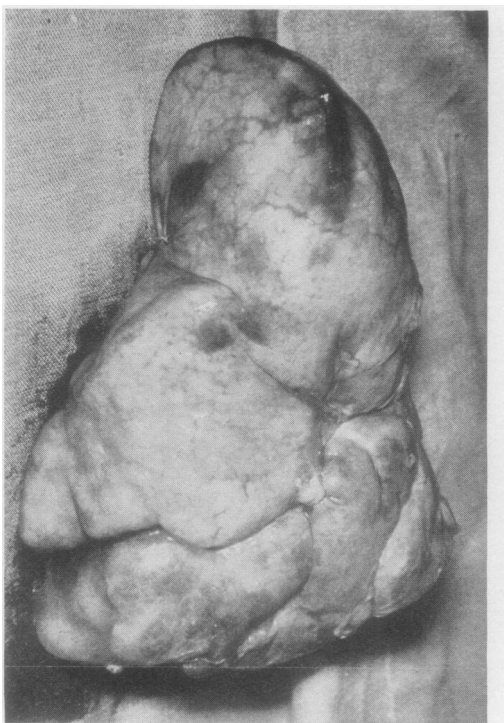


FIG. 6.—Case 1. Inflated specimen of the left lower lobe showing a marked groove which separates aerated normal lung above and thick sequestered mass below.

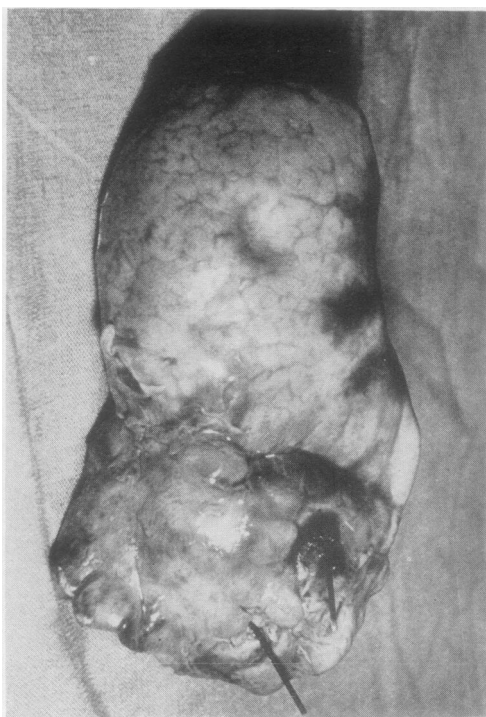


FIG. 7.—Case 1. The lateral view shows two pointers in the two anomalous arteries supplying the mass below.

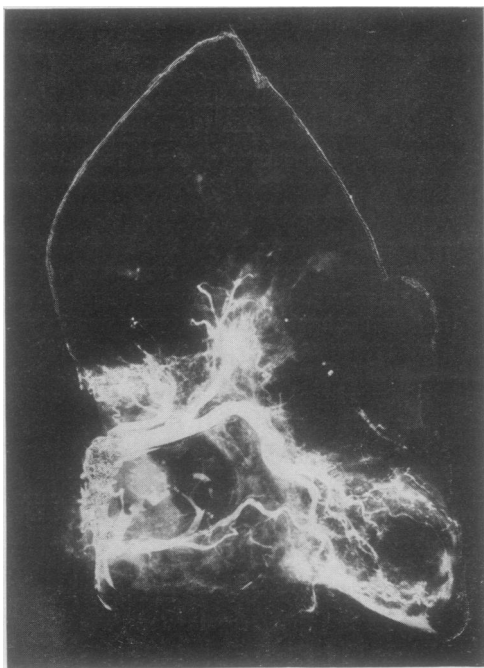


FIG. 8.—Case 1. Radiograph of specimen injected with "lipiodol." This shows that the anomalous arteries supply only the sequestered mass.

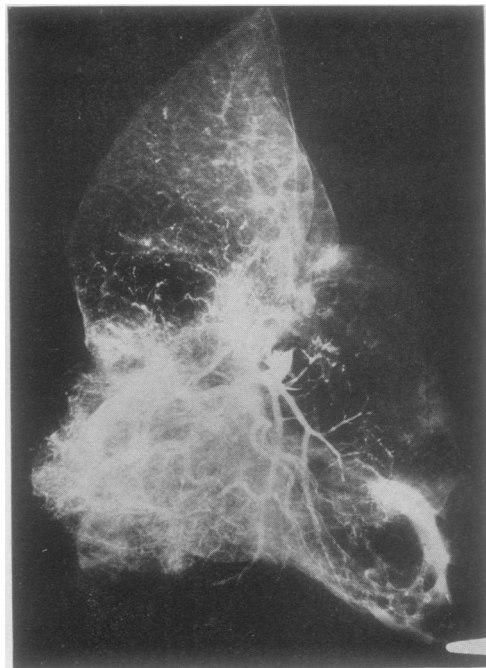


FIG. 9.—Case 1. Radiograph of the specimen injected with "lipiodol" through the accessory artery (lower right corner). The medium has collected and is draining out by the stump of the inferior pulmonary vein, seen clearly near the centre of the specimen.

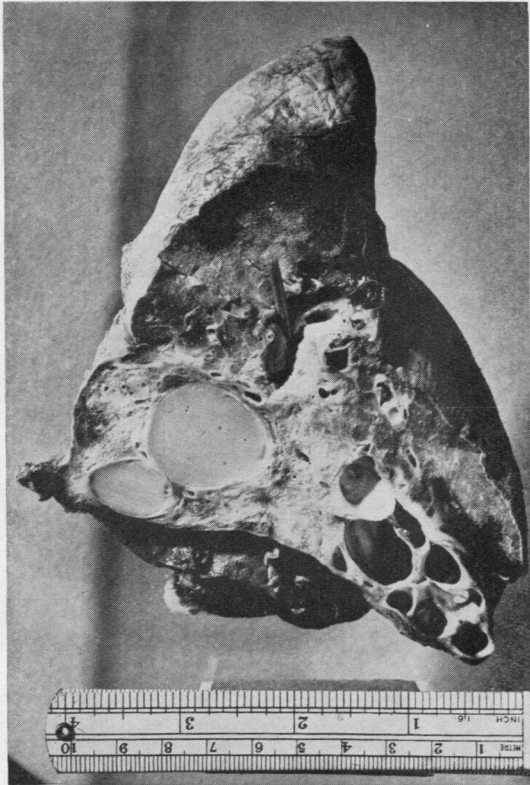


FIG. 10.—Case 1. Cut section showing normal lung tissue above and cystic spaces in the mass below.

CASE 2.—H. L., a Chinaman, 23 years of age, was in good general condition. He had had an occasional mild productive cough for 10 years. For six months he had had increased cough and yellow sputum, latterly blood-tinged, amounting to 1–2 oz. daily. Air entry over the left lower zone was diminished and the percussion note impaired. He had a thoracic scoliosis with concavity to the lower left thoracic region. Radiologically, a patchy consolidation in the left basal region, with a small amount of fluid, was noted. Minor changes were seen in both antra, but these soon cleared. No specific organisms were found in sputum or washings. Bronchoscopy was difficult, but creamy pus was seen coming from the apical segment of the left lower lobe. Over six months there was little change in the size of the affected area, but its quality changed—for example, an area of translucency in it appeared two months before the operation. On bronchography the dense area failed to fill. Otherwise the tree was normal except for being displaced forwards.

The diagnosis of sequestration was made, and a left lower lobe resection carried out on July 22, 1953. There was a cap of normal lung tissue on the upper and lateral aspect over a heavy, carneous, partly cystic mass 3 in. in diameter. A clear line marked the

boundary between normal and abnormal tissues. The mass was adherent to the diaphragm and mediastinal tissues. A thick-walled artery 1.5 cm. in diameter—almost as large as the aorta—arose from the aorta 3 in. above the hiatus, and entered the mass in the neighbourhood of the ligamentum latum. The

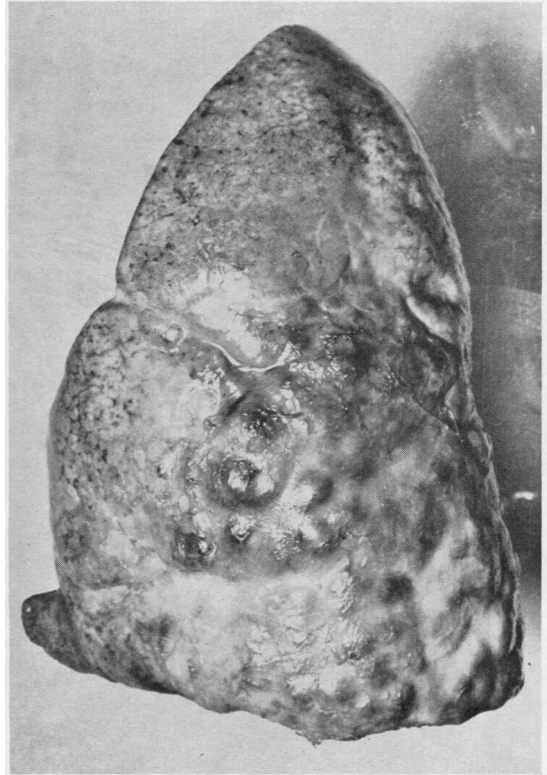


FIG. 11.—Case 2. Lateral view of the specimen—normal lung above, cystic mass below.

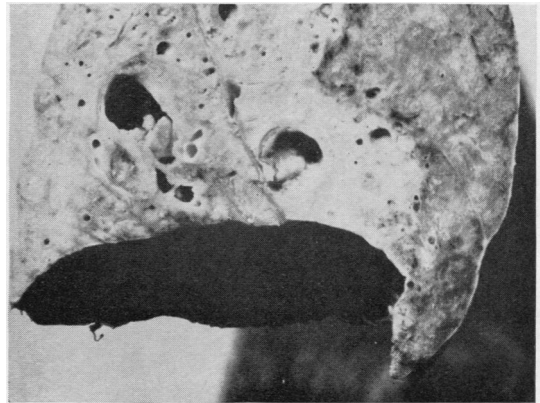


FIG. 12.—Case 2. Cut section of the specimen—normal lung above, cystic mass below.

branches of pulmonary artery to the lower lobe—two to the apical segment and two to the basal—were smaller than normal. The pulmonary vein was of normal size. Very large glands clustered round the fissure and the bronchus. The whole lower lobe was taken because of the difficulty in separating off the sequestered mass. The abnormal artery was carefully secured.

The post-operative course and progress were without incident and a recovery followed.

The specimen was similar to that in Case 1, and is shown in Figs. 11 and 12. It was not examined by "lipiodol" injection.

This case thus exemplifies a sequestration, Pryce Type 3, affecting the left lower lobe, having one large anomalous artery arising from the aorta. It did not show variation in size in the mass radiologically, as in Case 1, but its texture altered, translucent areas appearing while under observation.

#### SUMMARY

Two cases (making three in all from the Green Lane Hospital Unit) of intralobar sequestration of Pryce's Type 3 are described. Both affected the

left lower lobe and one exhibited two anomalous branches from the aorta.

The clinical presentation of the condition is discussed and a pattern for recognition is suggested.

The possibility of serious haemorrhage from the anomalous vessels, should they be unsuspected during resection, is emphasized again.

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