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Abstract

A case of intramural pure pigment gallstones, which were fortuitously found in post-mortem examination, is presented. The incidence, mechanism of formation of the stones and roentgenological diagnosis of the intramural gallstones, porcelain gall bladder, are mentioned.

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INTRAMURAL PURE PIGMENT GALLSTONES A CASE REPORT

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Gallstones usually form in the gall bladder and occasionally in the biliary ducts. Recently we experienced intramural pure pigment gallstones which were found fortuitously in an autopsy case. One of the forms of the intramural gallstones; "porcelain gall bladder", in which the wall of the gall bladder becomes impregnated with calcium, has been described in the European and American lituratures but only a few cases are reported in Japan. The following case illustrated here is not a typical porcelain gall bladder, but a gall bladder with pure pigment stones which are found scattered disseminatedly almost in the entire wall of the gall bladder throughout. A search of the literature fails to report of a similar case and brief report seems warranted.

CASE REPORT

Clinical Summary. The patient, 52-year-old male, whose pulmonary tuberculosis was first noticed in August 1939, had bedrest or hospitalization until April 1943. In October 1956 his pulmonary tuberculosis reactivated and he was given SM and PAS. In May 1958 he developed right exudative pleurisy, received SM, PAS and INH, and in April 1960 he was admitted to Kokuritsu Okayama Ryoyosho (Okayama National Sanatorium) under the diagnosis of pulmonary tuberculosis, far advanced, active, with right old pleurisy.

Physical examination on admission revealed a slightly reduced male in no distress. The heart was normal in size with regular rhythm without any murmur. The right chest showed a dullness and diminished breath sounds associated with moist rales. The abdomen revealed no pathological findings. Otherwise negative. Chest X-ray revealed two giant cavities with many other small cavities in the right upper lung area and right old pleurisy. Laboratory studies such as C. B. C., urinalysis were within normal limits. Red cell sedimentation rate 65 mm per hour. Vital capacity was 1450 ml. Per cent vital capacity was 61%. Sputum examination showed many acid fast bacilli on smear.

Hospital Course: In June and July 1960 he had right thoracoplasty, resected portion of I to VII ribs, followed by antituberculous chemotherapy. In

January 1962 he had a curetting of the remaining right VI rib and the anterior area of the right thoracic wall on the suspicion of the tuberculous caries associated with a cold abcess. In April 1962 his general and cardio-pulmonary conditions became poorer without any improvement of the pulmonary tuberculous lesion; the heart with grade III systolic murmur at the apex, P_2 stronger than A_2, vital capacity 1100 ml., per cent vital capacity 31%, antecubital venous pressure 17 cm H_20. He complained of shortness of breath, loss of appetite, belching distress, nausea, discomfort or pain in the epigastric region. Electrocardiograms from April 1962 to October 1962 revealed typical pulmonary P and two to one right bundle branch block which was followed by intermittent right bundle branch block that later changed to persistent complete right bundle branch block. Several liver function tests and other routine laboratory studies showed within normal limits in February 1963. On March 11, 1963 he suffered upper respiratory infection followed by severe generalized malaise and developed acute exacerbation of the chronic cor pulmonale. He was given cardiac drugs, bronchodilators and antibiotics. Because of persistent complaining of the loss of appetite and epigastric pain, upper G. I. series was done on March 20, 1963 and revealed a niche in the first portion of the duodenum. He was put on antacids, anticholinergic-antispasmodic drugs and ulcer diet. Laboratory studies on March 29, 1963 showed R. B. C. 3,500,000, Hb 12.5 gm dl. and positive occult blood of the stool. His general and cardio-pulmonary conditions grew still worse. On April 4, 1963 large amounts of soft tarry stool were noted and epigastric pain became more severe. He had a downhill course with continuous defecation of tarry or reddish stool and expired on April 12. 1963.

Autopsy : The body is that of an emaciated, elderly male, 164 cm tall and weighing approximately 38 kg. The principal gross anatomical findings are illustrated. The right thorax shows status, postthoracoplasty. The heart weight is 420 gm. Right heart is enlarged. The wall of the right ventricle measures 0.8 cm in thickness. The right upper lobe of the lung contains nodular tuberculous lesions and some small cavities including yellowish-whitish tenacious purulent material. A cavity measuring $10 \times 9 \times 8$ cm and containing moderate amounts of yellowish-white tenacious purulent material is present in the anterior and lateral portion of the right supradiaphragmatic area. The peripheral area of the left upper lobe contains bullous changes of the pulmonary tissue measuring up to 3 cm in diameter. The stomach is filled with large amounts of blood. The gastric mucosa shows no pathological findings. The duodenum has a round mucosal defect measuring 1.5×1.0 cm in diameter, in the center of which a blood vessel measuring 0.7 mm in diameter is explored, in the upper wall 1.5 cm from the pylorus ring. The liver weighs 1000 gm and is pale in color. The gall bladder measures 7 cm in length and adheres in the midportion of the

body to the liver. It contains yellow and slightly turbid bile. The wall contains black and exactly fragil solid materials measuring up to 3 mm in diameter, about thirty in number. They scatter disseminately almost in the entire wall throughout. The mucosa has a velvety lining and the bile tree is patent throughout.

Microscopically (Figs. 1 and 2), almost all of the epithelium of the gall bladder is lost. The mucosa is mounted on coarse folds. Gland-like spaces (the Rokitansky-Aschoff sinuses) extend from the lining to the moderately hypertrophied muscular coat. There is an increase of connective tissue in the lamina propria and between the muscle bundles. All the connective tissue is infiltrated with lymphocytes, plasma cells, fibrocytes and mononuclear cells. Some of the Rokitansky-Aschoff sinuses contain darkish-brown homogenous material.

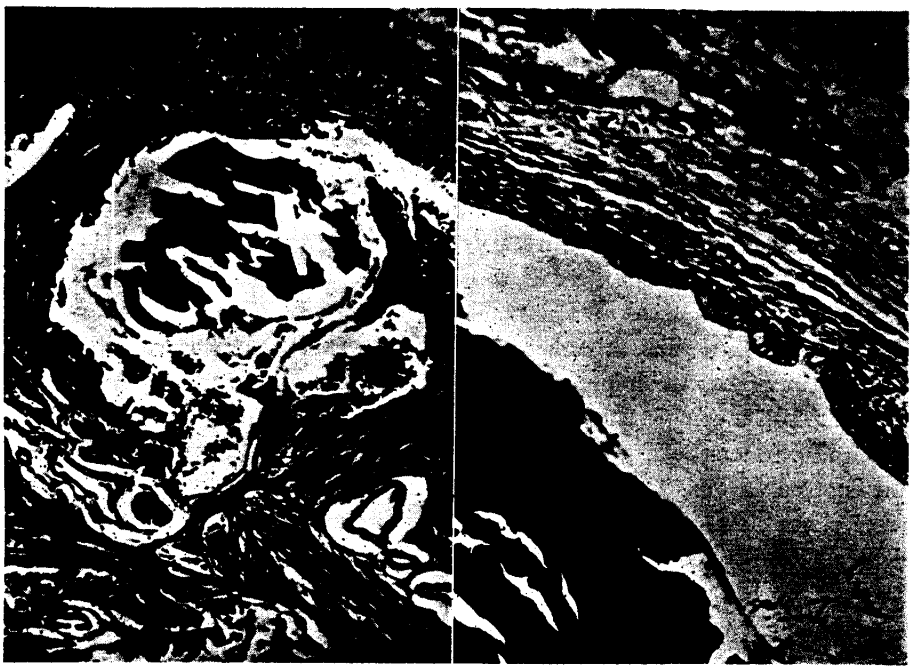

Fig. 1 Wall of the gall bladder. Proliferated lamina propria and muscle layer. The Rokitansky-Aschoff sinus extends to the perimuscular layer and contains pure pigment stone.

Fig. 2 Photomicrograph of a part of the Rokitansky-Aschoff sinus and pure pigment stone. There are a tall columnar epitherial lining and many infiltrated cells in the lamina propria.

COMMENT

European and American liturratures describe that gallstones usually form in the gall bladder and only occasionally in the biliary ducts, while many observa-

tions[1] in Japan reveal that gallstones are found in frequency of 50 to 80 per cent in the gall bladder, 20 to 50 per cent in the biliary ducts and 1 to 5 per cent in the liver. But intramural gallstones are very rare. In 1929 FLOERCHEN[2] first described porcelain gall bladder, in which the wall of the gall bladder becomes impregnated with calcium so that a cracking sound may be elicited on touch. CAPUA[3], TASCHENDORF[4] and SCHINZ[5] described it in the roentgenological studies. In Japan, YAMAMOTO *et al.*[6] reported two cases suggestive of porbelain gall bladder in the roentgenological study in 1958. Recently FUJII *et al.*[7] experienced a surgical removal of a gall bladder with intramural calcium bilirubinate stones. The failing report of the intramural pure pigment gallstones in clinical experience is presumed responsible for radiolucency of the bilirubinate stone and rarity of this stone in the Western countries.

Chronic cholecystitis may be induced by various factors, such as the sequel of an acute inflammation of the gall bladder, the presence in the viscus of pure gallstones or of the mulberry-shaped mixed gallstones and intermittent or continuous abnormal composition of the bile. Attenuated or usually nonpathologic enteric organisms may also produce chronic cholecystitis. The most important of these various conditions is the almost invariable presence of gallstones in the gall bladder. In some instances the gallstones are the cause of and in others are the sequel to the chronic inflammatory state of the gall bladder.

Grossly, the wall of the gall bladder with chronic cholecystitis is increased to several times its usual thickness. The folds are coarse and in advanced stages obliterated or absent with trabeculations of the muscular surface. Occasionally the wall may become impregnated with calcium so that a cracking sound may be elicited on touch. In this condition it is called "porcelain gall bladder". Microscopically the epithelium dips down to line outpouchings of the mucosa toward the external layers. These outpouchings, the Rokitansky-Aschoff sinuses, usually form along blood vessels and penetrate the thickened muscular coat to varying depths. Their fundi frequently reach the perimuscular layer. In the porcelain gall bladder the Rokitansky-Aschoff sinuses contain calcium. It is assumed that the same phenomena may occur in our case illustrated above, in which the stones contained in the sinuses are pure pigment but not calcium.

TASCHENDORF[4] states the following possibilities of the roentgenological diagnosis of the porcelain gall bladder: 1) Calcified gall bladder in normal size; 2) Calcified gall bladder with atrophy. In this condition long standing and repeating G. B. trouble persists; 3) Calcified gall bladder with simultanous presence of gallstones. In this condition troubles are elicited by the stones in the bile ducts; 4) Atrophied gall bladder with stones and faint but certain presence of calcified wall, which is not completely deformed yet and still noticeable tenderness; and 5) Partial calification of the wall.

However, the roentgenological diagnosis of the intramural pure pigment gallstones is presumed to be very difficult because of radiolucency of these stones and it is thought that there is only a possibility to be fortuitously found in surgery under the diagnosis of chronic cholecystitis. In view of this, in the treatment of gall bladder disorders one should keep in mind a possible presence of this kind of gallstones.

SUMMARY

A case of intramural pure pigment gallstones, which were fortuitously found in post-mortem examination, is presented. The incidence, mechanism of formation of the stones and roentgenological diagnosis of the intramural gallstones, porcelain gall bladder, are mentioned.

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