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## Duplication of the Gall Bladder; A Report of Two Cases

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### 重複胆のう；2例報告

原爆傷害調査委員会放射線部 国立予防衛生研究所

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(昭和45年7月29日受付)

胆のう造影で発見された2例の重複胆のうを報告した。これらの診断には透視下撮影が最もよく、断層撮影も補助となつた。2例とも胆石を合併していた。

### Introduction

Duplication of the gallbladder is a rare anomaly. Review of the medical literature available to us revealed less than 200 cases. The two cases reported here are participants in the Adult Health Study (AHS) of the Atomic Bomb Casualty Commission (ABCC) and the Japanese National Institute of Health (JNIH), all of whom routinely have complete biennial physical examinations in the ABCC clinic. These subjects are members of selected samples of the Hiroshima and Nagasaki populations, originally numbering 20,000 people. Except for routine posteroanterior and lateral chest roentgenograms, roentgenologic studies are performed as clinically indicated. These two cases were detected among 1143 cholecystograms conducted on members of this sample.

CASE I (MF # 470787): A 52-year-old Japanese male received a complete physical examination at ABCC 15 years previously. His past history revealed an episode of jaundice of one month's duration 23 years ago. Since then he had intermittent dull pain and discomfort in the right upper quadrant. Physical examination was unremarkable, except for slight tenderness on palpation in the right upper quadrant. Hematological findings were: Hemoglobin, 16.5 g/100 ml;  $538 \times 10^4$  erythrocytes; 4,900 leukocytes; serum cholesterol, 250 mg/100 ml; serum albumin, 4.6 g/100 ml; and serum globulin, 2.24 g/100 ml.

Other tests including urinalysis were within normal limits. Oral cholecystography was performed at that time because of right upper quadrant pain, and a double gallbladder containing calculi was suspected. Intravenous cholangiography and cholecystography with 35 cc cholographin was then performed (Fig. 1).

Two gallbladders were identified. The duct of the lower gallbladder enters the common duct below that of the upper gallbladder.

The patient has been asymptomatic since that examination. Intravenous cholangiography and cholecystography with tomography following the injection of 30 cc Biligrafin was performed in December 1969 to delineate the structures more clearly. The patient was fluoroscoped 10 minutes post-injection, with spot films in the 45 degree left anterior oblique projection. Tomography in the same projection visualized the two cystic ducts better, though a moderate amount of feces in the colon interfered, as shown in Fig. 2.

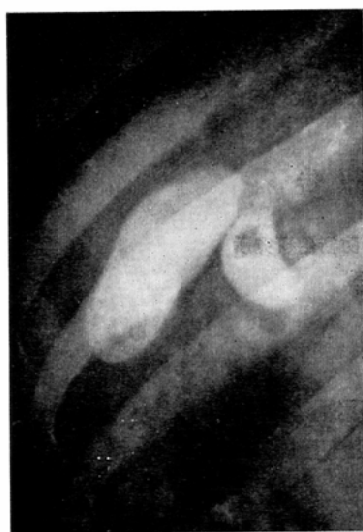


Fig. 1. Case I (MF #470787); intravenous cholangiogram showing two separate gallbladders each with calculi. The duct of lower gallbladder enters the common duct below that of the upper gallbladder.

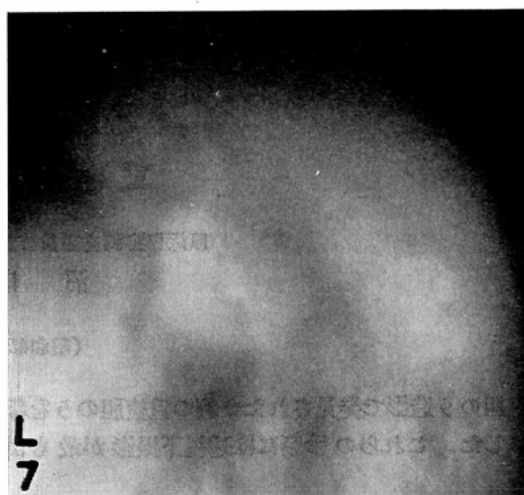


Fig. 2. The same case; intravenous cholangiogram with tomogram showing two gallbladders each with a cystic duct.

CASE II (MF #240835): A 64-year-old Japanese male received a complete physical examination at ABCC 8 years ago and had a history of severe jaundice 18 years prior to then. He had had mild hypertension and diabetes during the previous 5 years. Physical examination revealed slight tenderness on palpation in the right upper quadrant. His blood pressure was 148/98 mmHg. Hematological studies showed: Hemoglobin, 16.4 g/100 ml; erythrocytes,  $519 \times 10^4$ ; leukocytes, 7,450; serum cholesterol, 177 mg/100 ml; and blood glucose: 120 mg/100 ml; fasting and 180 mg/100 ml at 3 hours in a glucose tolerance test. Urinalysis revealed a trace of glucose. Other laboratory tests were normal.

Chest roentgenograms showed multiple calcific densities in the right upper quadrant suggestive of cholelithiasis. Oral cholecystography 3 months later demonstrated a double gallbladder, one with mul-



Fig. 3. Case II (MF # 240835); combined oral and intravenous cholangiogram [visualizing two gallbladder with granular calculi in the one gallbladder.

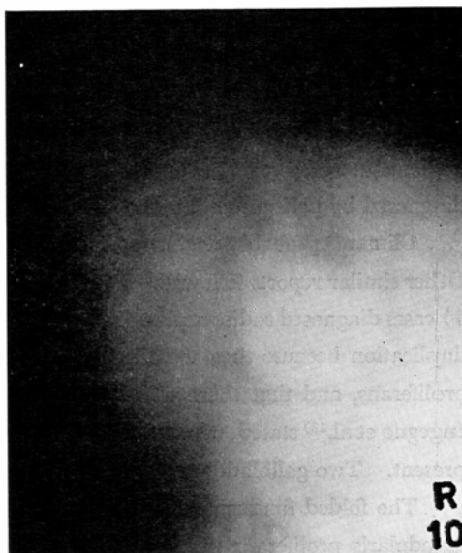


Fig. 4. The same case; intravenous cholangiogram with tomogram showing two cystic ducts entering the common bile duct.

tipple stones, but intravenous cholangiography 1 month later failed to visualize the cystic and common ducts clearly. Two days later, combined oral and intravenous cholangiography visualized them well, as shown in Figure 3.

In January 1970, combined oral and intravenous cholangiography with tomography demonstrated two gallbladders best in the right anterior oblique position at fluoroscopy. Tomography in this projection outlined two cystic ducts entering the common bile duct (Figure 4).

### Discussion

According to Guyer et al.,<sup>10)</sup> duplication of the gallbladder has been recognized in man since 31 B.C., and the first case was published by Gladius in 1674. According to Boyden<sup>2)</sup> and Wilson,<sup>30)</sup> its prevalence in the human is one in three to four thousand individuals. It is more common in domestic animals, such as cats.<sup>2)</sup> Several classifications of duplication have been proposed.<sup>2)</sup><sup>30)</sup> The types and subtypes of human double gallbladders, according to Ingegno et al.<sup>13)</sup> are shown in Figure 5.

In the ductular type, both cystic ducts usually enter the common bile duct, though one of them may open into a hepatic duct<sup>12)</sup> and may even be intrahepatic.<sup>28)</sup> There is insufficient data concerning the frequency of the divisa and duplex types. The ductular ("H"-shape) type appears to be the more common form of the duplex type, according to Stolkind<sup>27)</sup> and Moore et al.<sup>15)</sup>

Roentgenologically the septate and cleft types cannot be easily distinguished. Gross<sup>9)</sup> reviewed 148 cases of congenital anomaly of the gallbladder reported from 1901 to 1936 and found 28 cases of duplex, 9 of diverticular and 6 of the septate or cleft types among them. Flannery et al.<sup>7)</sup> reviewed 101 cases from 1936 to 1956, including 25 of duplex, 10 of diverticular and 5 of the septate or cleft type. In Japan, up to 1967, Naoe et al.<sup>16)</sup> found 33 cases of duplication including 11 cases of duplex, 13 of the diverticular and 9

of the septate or cleft type. An additional two of the duplex type,<sup>22)28)</sup> one diverticular type<sup>14)</sup> and five of the septate or cleft type<sup>21)25)</sup> were reported by 1969. Usually, duplication of the gallbladder is discovered at surgery or autopsy. Double gallbladder may be established radiographically only when the cystic ducts and their terminations can be visualized.<sup>18)</sup> In the two cases reported here they were successfully demonstrated, however they were of the ductular type. Shimada et al<sup>24)</sup> reported a septate or cleft type diagnosed by peritoneoscopy and roentgenography.

Climan<sup>4)</sup> probably documented the first case of double gallbladder detected roentgenologically alone. Other similar reports followed.<sup>8)9)11)</sup> Guyer<sup>10)</sup> reviewed 98 cases in the literature and commented on the 44 cases diagnosed radiographically. He stated that several of them could not be established as congenital duplication because they were indistinguishable from other conditions, such as cholecystitis glandularis proliferans, and that there was no definite delineation of the cystic ducts and their terminations. As Ingegno et al.<sup>18)</sup> stated, where there is appreciable separation of two gallbladders, the "H" form is probably present. Two gallbladders close together may be either of the "Y" or "H" type.<sup>18)</sup>

The folded fundus, Phrygian cap deformity, postural kink, hourglass gallbladder, and cholecystitis glandularis proliferans may sometimes be erroneously diagnosed as congenital duplication.<sup>1)5)6)18)</sup> It may be impossible to verify the divisa types radiographically because they have only one cystic duct, and

Fig. 5\* Classification and Characteristics of Accessory Gallbladder

| Type   | Subtype                            | Description  | Embryologic Origin  |
|--|------------------------------------|--|---|
| 1. Divided gallbladder<br>(Vesica fellea divisa) | a. Septate type                    | Septum divides cavity longitudinally more or less completely. Septum may be transverse. The loculi communicate. External appearance of gallbladder normal. One cystic duct   | Incomplete resolution of solid stage of development of the gallbladder  |
|  | b. Diverticular type               | Prevailing type in ungulates. Diverticulum, a sac-like protrusion, may arise from any portion of gallbladder, including fundus and neck. Diverticulum usually smaller but may be larger than main cavity and communicates with it. One cystic duct                           | Some, especially those near the neck, arise from persistence of cystic-hepatic ducts (ducts in embryo which pass from gallbladder bud or cystic duct into liver and normally regress). Others, especially near fundus, may be due to incomplete resolution of solid stage with pocketing off of a portion of fundus by a septum |
|  | c. Cleft type<br>(lobate or bifid) | Prevailing type in cats. There is division and separation of the fundic portion of gallbladder, extending to a variable degree down the body. Divisions may be unequal. Their cavities communicate with main cavity. Fundus has a lobed or bifid appearance. One cystic duct | The gallbladder primordium is partially split during the solid stage. When the viscus develops its cavity the fundic portions remain separate and may be unequal  |

| Type   | Subtype                                  | Description   | Embryologic Origin   |
|--|--|---|--|
| 2. Double gallbladder<br>(Vesica fellae duplex)      | a. "Y" type                              | Two gall bladders, usually close together or adherent and occupying the same fossa. Two cystic ducts which unite to form a common cystic duct. The latter then joins the hepatic ducts to form common bile duct. The gallbladders may be equal or unequal in size         | Probably as an accessory outpocketing of the cystic duct "subsequent to the formation of the definitive gallbladder" rather than as a "primary subdivision of the embryonic primordium" (Boyden) |
|  | b. "H" type<br>(ductular type of Boyden) | Two gallbladders, completely separate, and sometimes in different lobes of the liver. Two cystic ducts. The accessory cystic duct empties independently into an hepatic duct or the common duct. The accessory vesicle may be smaller or larger than the true gallbladder | As above, except that the accessory pouching occurs in embryo from the common duct or an hepatic duct  |
|  | c. Trabecular type                       | Two gallbladder in the gallbladder fossa. Two cystic ducts, one of which plunges directly into the adjacent liver substance. Very rare case reported by Croudace  | The accessory gallbladder arises as an outpocketing of liver cords or trabeculae bordering the gallbladder fossa and communicating with the smaller bile capillaries (Boyden)                    |
| 3. Multiple gallbladder<br>(Vesica fellae multiplex) |  | This has been reported in cats and ungulates. Its occurrence in humans is possible but not recorded. In this there may be three or more gallbladders, or two gallbladders with variations of the divisa type in one   |  |

\*Ref. 5, Ingegno et al

because of those conditions which may simulate it.

Alexander<sup>13)</sup> recommended lateral roentgenograms to differentiate duplication from the folded fundus or kinked gallbladder. We excluded several of our suspected cases of double gallbladder, disproven by this projection. However, in the two cases reported here, lateral roentgenograms failed to demonstrate the two gallbladders distinctly. Both cases were confirmed fluoroscopically with the subjects in various postures. Tomography was helpful in demonstrating the two cystic ducts in our second case.

A high percentage of duplication reportedly has associated calculi and cholecystitis.<sup>13)17)18)20)23)29)30)</sup> Other complications include cholesterosis and cholecystitis glandularis,<sup>23)</sup> papilloma,<sup>30)</sup> carcinoma,<sup>16)</sup> torsion,<sup>19)</sup> and limy bile.<sup>10)</sup>

Prevalence by age and sex is not clear. Moore et al.<sup>15)</sup> reviewed 36 cases reported up to 1954. The vast majority of cases had symptoms due to cholecystitis and cholelithiasis. Of those patients 75% were women. Age at onset of symptoms ranged from 19 to 69 years, with an average of 43 years. Each of our

patients was male and had jaundice at the age of 43 and 46 years, respectively. It is not clear whether the jaundice was truly due to cholecystitis or cholelithiasis. They had no other symptoms during the past 15 years or more.

### Summary

Two cases of duplication of the gallbladder were diagnosed by cholecystography. Both cases were of the duplex type. Fluoroscopy and, to a less extent, tomography were helpful in establishing the diagnosis in these two cases. Associated cholelithiasis was present in both cases.

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