IDIOPATHIC CHOLEDOCHUS CYST

WITH REPORT OF A CASE CURED BY CHOLEDOCHODUODENOSTOMY

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In consideration of the rare nature of the disease and hoping to cast light upon its interesting pathogenesis, I should like to give an account of a case of severe cystic dilatation of the ductus choledochus, or, as it is designated in literature, idiopathic choledochus cyst, which was operated on by me last year.

The case is that of a ten-year-old girl, C. T., who was received into the county hospital of Lidköping on May 15, 1916.

Remarks.—No case of malformation in the family, no case of gall-stone, but, on the other hand, several cases of tuberculosis. The patient had whooping-cough and measles when two years old, and scarlatina in 1915, since which she has been delicate and weakly.

According to the statement of the mother, the patient ever since the age of three has had about once each year an attack of abdominal pains, sometimes accompanied by vomiting, usually lasting one or two days, during which time she was obliged to keep to her bed. Jaundice never showed itself during these attacks.

For three weeks before admission to the hospital the patient has occasionally had pains in the right side, but not so severe as to prevent her going to school. Three days ago, on the evening of May 12, the patient became ill with rather severe pains in the right side of the abdomen, repeated attacks of vomiting, but no shivering. The pains went through to the back, and were especially severe during deep respiration, but were somewhat alleviated when the patient assumed a half-sitting position. The following day, May 13, the patient felt better, the pains having diminished considerably. On May 14 the pains again became more severe with vomiting several times during the course of the day. The physician, who was called in, found there was fever and prescribed medicine. As the pains continued the following day, the patient was brought to the hospital. During her illness the patient has had normal stools on May 13 and May 15.

Present Condition (May 15, 1916).—The patient is of slight build, rather thin, the general condition somewhat depressed, complexion pale without icterus, the scleræ show, however, a slight icteric tinge. Temperature 38.1, pulse 100. Urine clear, dark-green, containing gall-pigments and a trace of albumen.

Heart and lungs, nothing to be remarked upon.

The abdomen, as a whole, sunken. In the right upper part of the abdomen, about in the middle, a slight protuberance appears.

in the wall of the abdomen about as large as half-a-crown. To palpation the above-named part corresponded to a tender resistance of about the size of a fist, oblong in form, with the long axis vertical, reaching downwards to the region of the umbilicus, and upwards to a finger's breadth below the costal margin, with a breadth from side to side of about 7 cm., lying in its greater part behind the right muscularis rectus, but protruding somewhat outside its outer edge. The tumor has a smooth surface and appears to be of rather firm consistency, but this is difficult to determine on account of the extreme tenderness. The tumor shows no respiratory mobility, does not permit displacement either from side to side or from above downwards; it does not give the impression of engaging the abdominal wall, nor does it appear to have any connection upwards with the liver, which as well as the gall-bladder is not palpable. The percussion tones over the tumor are somewhat shortened.

The patient had been sent in for supposed appendicitis. The palpable tumor might, if it had been situated somewhat further laterally, possibly be supposed to be caused by a retrocæcal abscess from an appendix, either unusually long, or situated abnormally high up, but the existing high and medial position seemed to put appendicitis quite out of the question, nor could the tumor be localized to any special organ as its origin. The preceding attacks of pain, as well as the existing slight icterus, might point to a cholelithiasis, although this disease is very rare in childhood, and the resistance could be explained as a hydrops vesicæ fellæ. The definite limit of the tumor upwards, without any connection with the liver, was against the theory of dilatation of the gall-bladder, further, its low degree of mobility and, in some measure, its form also. Opposed to the theory of the kidney as point of origin was the elevated and medial position of the tumor, as well as the absence of swelling in the flank. A cyst of the pancreas could scarcely be supposed, as they are very seldom lying so decidedly to the right of the medial line. It was not possible, therefore, to make a positive diagnosis, but it seemed to me quite evident that operative treatment was necessary.

An operation was performed the same evening under ether anæsthetic. A 10 cm. long vertical incision was made through the middle of the right rectus muscle from the costal margin to the level of the umbilicus, somewhat curved upwards towards the middle line. In the cavity of the abdomen an inconsiderable quantity of clear fluid. Nothing of note as regards appendix. The gall-bladder free and mobile, of normal size, with somewhat hyperæmic serosa. No stones to be felt. Below the gall-bladder lies the tumor which had been perceptible to the touch, in a retroperitoneal position, covered on its outer part by the transverse colon and the hepatic flexure and medially by the duodenum. It is of tense elastic consistency, in position and form corresponding in some degree to an enlarged kidney, so that my first thought was that it was a case of hydronephrosis. When better space had been

gained by a 5 cm. long transverse incision outwards from the middle of the abdominal cut, the peritoneum was divided laterally of the ascending colon and flexure which were pushed towards the middle and the tumor, evidently of a cystic nature, was exposed, by blunt dissection, first on its front side, where, however, its connection especially medially with the peritoneum was very intimate; in addition to this the difficulty of detaching it was increased by a rich development of veins. Downwards it was easy to get round the rounded lower pole of the tumor, as well as to pass upwards on its posterior side. It is found then that no connection exists with the kidney, which lies quite normal and free behind the tumor. The opinion is now in favor of puncturing the tumor, and while waiting till the puncture syringe is boiled, the tumor is raised upwards in order to examine more closely from behind its medial connection, especially with the pancreas, which, however, shows nothing abnormal in palpation. During these manipulations, the remarkably thin walls of the cyst burst and discharge its contents—dark-colored clear bile. This is absorbed in gauze, and amounts to about 200 c.c. The opening is widened upwards with scissors, so that the inside can be examined. The inside is pale, smooth, and of an equal surface with no perceptible alteration in the walls. No concrement can be discovered. It is evident that the sac consists of the highly cystically dilated choledochus; on account of its considerable depth it is impossible to see from the inside its connection with the upper gall-passages. When all the bile has been dried out of the sac, however, one could plainly perceive how the contents of the gall-bladder, on compression, emptied itself into the upper part of the sac. On examination of the continuation of the cyst downwards no continuous passage from the inside of the cyst could be discovered. After some searching, however, a small lumen was found, immediately to the left of the lower pole of the cyst, and this was found to belong to the upper end of the ductus choledochus lying behind the duodenum. An ordinary probe, inserted into this passage, went direct into the duodenum, without meeting any hindrance. The passage is 3 to 4 cm. in length and possibly somewhat narrower than a normal choledochus. Evidently the rupture of the cyst had occurred just at the junction between the cyst and the lower part of the choledochus. With a few catgut sutures the upper end of the stump is implanted in the cyst in its original position. As the position of the incision on the posterior side of the cyst was not suitable for anastomosis, it was sewed together with 2 rows of interrupted catgut sutures, after which an anastomotic opening 1.5 cm. long was made between the forward part of the cyst and the front side of the duodenum in the upper part of the pars descendens. The sutures were made in two rows, the outer one interrupted, the inner one continuous round about through the entire wall, both of catgut. The part where the choledochoduodenostomy was made as well as the suture line on the posterior side of the cyst and the part where the lower choledochus stump was

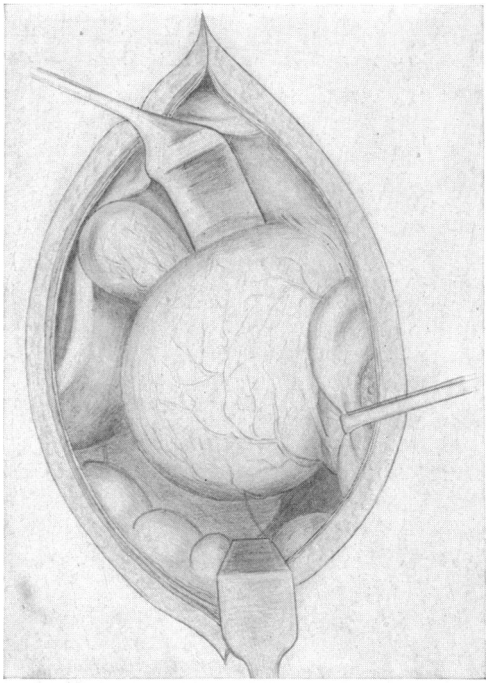

FIG. 1.—Cystic dilatation of the ductus choledochus.

implanted were wrapped round with a wisp of the omentum. A small rubber tube was inserted against the outer part of the cyst, at a little distance from the place of the sutures, and was led out through the lateral angle of the transverse incision. The abdominal wound was then sewed up.

The patient was considerably exhausted after the operation, the pulse was feeble and extremely rapid; she revived quickly, however, after stimulants. Two days after the operation nothing could any longer be discerned of the above-mentioned trace of icterus in the scleræ, or the gall-pigment in the urine. During the first week after the operation there was a rising in the patient's temperature; this, however, went down afterwards. On the seventh day the drain-tube was removed, on the eighth the stitches. Healing by first intention took place. On May 29 the patient was allowed to be up. Twice, on June 4 and June 7, occurred an evening rise of temperature to 40.3° and 39.2° respectively, with vomiting, but no other symptoms. On both these occasions the temperature was normal the following day. On June 10 the patient was discharged, healed and well.

It appears from the observations made at the operation: (1) That the cyst (see accompanying drawing, Fig. 1) was developed from the middle and upper part of the choledochus; (2) that the lowest portion of the choledochus, 3 to 4 cm. in length, was not engaged in the dilatation; (3) that the above-mentioned part of the gall-passage, representing the communication between the cyst and the duodenum, did not show obstruction, and (4) that none of the otherwise usual obstacles to the passage of bile into the choledochus, in the form of stone, tumor, stricture, or pancreatitis, were found to exist. These circumstances exclude the presumption that this was a retention cyst in the ordinary sense; the case must be explained as one of idiopathic dilatation, a so-called real choledochus cyst, corresponding in all its details with all others of analogous type.

Previous to the operation this disease was unknown to me, and I was thereby led to an examination of the literature on the subject, the result of which seems to cast light upon the disease in question.

When Langenbuch in 1897 published his great work on the surgery of the gall-passages, he only knew of one case, that of Konitzky in 1888. In 1909 Ebner gave a summary of 11 new cases, described by Douglas, Edgeworth, Seyffert, Heid, Russell, Nicolaysen, Rostowzew, Dreesmann, Broca and Bakes, as well as a case of his own, operated on by Lexer. Schloessmann in 1911 reports 4 more cases—Wettwer's, Arnold's, Weiss's and one operated on by Sprengel. From Japan, in the following year, Mayesima gave a new observation of the disease. Kehr, in his last great work, Neue Deutsche Chirurgie, 1913, mentions 2 more cases, Letulle's and Goldammer's, and finally, 1916, Seeliger in a summary of the preceding cases, reports still another new case which he observed in 1913. Besides these cases, 20 in all, I have been able to

discover no less than 14 others, reported by Arnison, Butters, Twain, Ebner, Sternberg, Clairmont, Brun-Hartmann, Ashby, Kolb, Lavenson, Hildebrand, Ipsen, Smit and Heiliger. The sum total therefore of known cases, my own included, would be thirty-five. Common to all these cases is the circumstance that the disease could not be diagnosed clinically, in most of the cases, not even at the operation. For this reason and on account of an irrational method of operation most cases have terminated fatally; only a few having been restored to health by the operation. Among the latter I find only one case, that of Bakes, in which, as in the case operated on by me, an anastomosis was made primarily between the cyst and the intestine.

The idiopathic choledochus cyst, says Konjetzny, who has closely investigated the disease from the pathological anatomical point of view, is dependent upon a congenital anomaly in the course of the common duct. It differs distinctly from the not uncommon dilatations of the choledochus, which appears as a secondary circumstance in previous diseases of the gall-passage or pancreas as a result of the purely mechanical obstacles these diseases produce. The dilatation of the gall-passage in question includes, as a rule, the gall-duct in its entirety, converting it into a uniform cylindrical tube, retaining its original form, and never—even when of year-long standing—exceeding a certain degree of dilatation, corresponding about to the thickness of the small intestine—while the circumstances in the case of a choledochus cyst are quite different. It is here a question of what one might call independent primary enlargement of the common duct, only engaging its upper and middle portion, with free passage of its lower undilated part, forming a spherical or ovoid sac-like enlargement, a sort of "biliary cloaca" (Letulle), generally of very considerable size. Thus, in most cases, the dimension of the cyst has been about that of a child's or a man's head, in some instances, the quantity of fluid it has been capable of containing was as much as 4 or 5 litres. In my case the cyst was remarkably small, in comparison to the other cases. I estimated it as about the size of a fist, a volume which seems to be rather exceptional. The position of the cyst is below the liver, usually lying closely pressed against its lower surface. Sometimes, as in my case, when it had not advanced so far in its size, it lies a little below the liver. It often extends over the middle line, and goes backwards towards the spinal column, more or less further downwards, in not a few cases as far as the pelvis. It generally adheres firmly to the surroundings. The duodenum is forced more or less medially, to a degree corresponding to the size of the cyst. In one case, Seeliger's, it was pressed quite flat against the front cyst wall; the stomach is pushed over to the left, and not seldom turned on edge with the great curvature forward. The right part of the transverse colon and the hepatic flexure are pushed to the left and downwards. The upper part of the gall system, including the gall-bladder, shows

either no dilatation or a very inconsiderable one in proportion to the choledochus. The liver, in the far advanced cases, has, when enlarged, exhibited the appearance of a biliary cirrhosis. As already mentioned, the lower, more fixed portion of the choledochus, lying behind the duodenum, is never affected by dilatation, and long retains its normal calibre. For the explanation of the manner of origin of the cysts, the minute examination of the condition of this part of the choledochus has been of great importance. It has been found several times that the end portion has undergone a development deviating from the normal, with a more or less oblique opening into the intestine in a direction from left to right, or from the front backwards, instead of the normal direction from right to left. A more or less marked kink has been thus produced, and this has been explained as an obstruction of the passage of congenital origin, causing the dilatation of the passage lying above. In my case, no such abnormal insertion in the duodenum could be found, and besides, it is difficult to think that this alone could be the real cause of the dilatation, when in all cases, signs of difficulty in evacuation have been existent only for a relatively short time, while the choledochus, to judge from the reports of the illnesses, has performed its function normally for years previously. Another circumstance which might be of greater significance is that in most of the operated cases a more or less sharp transition has been found to exist between the cyst itself and the upper part of the end portion. From the fact that the enlargement took place to the right and downwards, the developed diverticulum has been pressed with its left wall against the fixed end portion of the choledochus, compressing the latter, so that on cutting open the cyst, it actually looked as if this part of the choledochus lay like a fine canal in the medial cyst wall itself. Inside the sac, it had the appearance of a valviform duplication of the wall, and this valve has during life acted as a ventile with intermittent action. This circumstance would be, therefore, to a certain degree, analogous with hydronephrosis, produced by an obliquely inserting ureter or with a kink in the cystic duct, with transformed gall-bladder the so-called cholecystosis remittent. In the case operated on by me the existence of such a fold could unfortunately not be ascertained in consequence of the rupture of the tumor which occurred just at the point in question; but in view of the fact that the dilatation took place so far down that the lower pole of the cyst lay below the pars horizontalis inferior duodeni, consequently below the place of entry of the common duct into the intestine, I should consider it most probable that such was the case. The mechanism of the emptying of the cyst would then be as follows: With a certain fulness of the sac the fold is pressed close to the opposite wall, rendering emptying for the time impossible; by a continuous supply of bile from the liver, however, the pressure inside the sac is continuously increased, the cyst wall expands more and more, the effect of which will be that the spur-like fold of the mucous

membrane at the place of the kink is gradually pressed somewhat to the side, leaving a portion of the lumen free, and permitting the discharge of a quantity of bile into the duodenum. The pressure now relaxes again somewhat in the sac, and the wall contracts and forms a valve again; this hinders a further discharge and prevents the cyst from becoming completely empty. In many cases which have been sufficiently long under observation (Rostowzew's, Heid's, Wettwer's, Smit's) the working of this intermittent valve-mechanism has been clinically observed, showing itself in a peculiar and striking increase and diminution of the tumor with respectively increased and reduced tension or consistency and tenderness of the same, and it has in reality given the clinical picture of the disease a very characteristic feature. It may be mentioned in parenthesis that, in some cases, a similar valve or fold formation has been found, not only at the junction of the cyst with the end portion of the choledochus, but also farther up at its union with the cysticus and even with the hepaticus.

The above-described valve formation gives a sufficient explanation of the question, why the fully formed cyst cannot empty itself, and why it undergoes an ever-increasing enlargement of volume. But it is impossible to suppose that the valve can be developed until a part of the duct has been widened into a sac-like cavity and the existence of the valve is therefore not sufficient to explain what causes the first enlargement. Many hypotheses have been put forward for the explanation of this primary dilatation, which later leads to valve-formation and ventile obstruction. It has been supposed that there exists congenital weakness of the wall, with defective development of the elastic and muscular elements, a sort of atony, as predisposing circumstance, and, as immediate cause, a catarrhal mucous swelling, a stone, or spasmodic contraction of the choledochus sphincter. Further, Dreesmann has suggested the idea of a congenital primary dilatation of the passage. Kehr also mentions this, but says at the same time that proof of this possibility is, up to the present, absent from literature. It seems to me, however, that such proof really does exist, although not mentioned in any of the accounts published of operatively treated choledochus cysts, not even in Seeliger's of the last year. It is to be found related by Heiliger in 1910 in a dissertation from the Women's Hospital in Giessen. In postmortem examination of an almost mature male fœtus, he happened to find, besides a diaphragmatic hernia (containing stomach, spleen and small intestine) also an alteration in the choledochus in the form of a 3 cm. wide, 2½ cm. long, tense, cyst-like distention of the same. He says, "through this sac-like distention of the choledochus, exclusively confined to its distal portion, which, in relation to the normal, must be regarded as a monstrous enlargement, and which only in a slight degree passes into the orifice of the hepatic and cystic ducts, the proximal end of the choledochus, which had its course in the wall of the choledochus sac, was compressed to such a degree that it was

only after some trouble and by means of a fine bristle that it was possible to probe this part. It must, however, be supposed that sufficient bile had come out into the intestine, for the intestinal contents were normally bile-colored, and there was not a single sign of bile stagnation, nor any icteric coloring of the tissues."

This observation of Heiliger's seems to me to give indisputable proof that the choledochus cyst itself can exist already formed at birth, that it can be really congenital. But from the fact of its existing from birth it does not necessarily follow that it should show immediate symptoms. On the contrary, all observations seem to indicate that it is in the beginning absolutely latent to its owner. This period of latency then, as appears from the accounts of the disease, can evidently, in different cases, vary very considerably, and as the cyst evidently has a marked tendency to develop continuously in size, it seems to be the natural conclusion that what determines the point of time when the first symptoms of the disease begin is the size of the fetal choledochus sac. If this, at the time of birth, has already attained large dimensions, the troublesome symptoms it produces come earlier than if the individual is born with a smaller cyst. The youngest of the patients observed with serious symptoms of the disease was 2 years old, the oldest 25—between these two ages lie all stages. The average age when the patient began to suffer from the disease seemed to have been from 12 to 14.

The principal *symptoms* produced by choledochus cyst are icterus, tumor and pains. The icterus has been intermittent and, in a number of cases, has passed into a continuous form of varying severity; in nearly all cases it has been remarkably strongly marked, in a couple to a very high degree—icterus melas. Only in one case, Smit's, it has been completely absent; in my case it existed only in subicteric coloring in the scleræ and urine. The most important symptom is, without doubt, the tumor, lying in the right hypochondriac region and almost always attaining a very considerable size. In none of the published cases was it of so small dimensions as in mine, where it was of the size of a fist. In several cases it has been as large as a child's or a man's head, or even larger. As a rule, it is situated immediately close to the liver, in certain stages, as in my case, it is clearly separated from it by a region with tympanitic percussion tones. In 5 of the published cases (Dreesmann's, Mayesima's, Arnold's, Seeliger's and Russell's) could be observed over the great tumor another smaller one, varying from the size of a hen egg to that of a goose egg, lying under the edge of the ribs, and having its origin in the equally palpable, somewhat distended gall-bladder. In the cases which have been observed long enough, the great tumor, as I have already mentioned, exhibited a peculiar variation in size and consistency, and this variation has been ascertained to result from the taking of food into the stomach; the tumor swells and becomes harder some hours after meal times, evidently in connection with the fact that the secretion of bile is most active at this time. The pains have varied

from more or less severe flatulency of the abdomen to the most violent colic pains. Their violence is usually in proportion to the icterus and the size of the tumor. These symptoms have generally been accompanied by fever, and the general condition has been more or less affected. Three times (in Ashby's, Mayesima's and Wettwer's cases) ascites has occurred, caused by the compression of the portal vein by the tumor. Further, the disease has generally developed at intervals spread over a varying period of time, and as a rule, finally showing very evident symptoms; once (Seeliger's case) exhibiting a real intestinal obstruction. A peculiarity of this disease is that it appears par préférence in the female sex; only in 5 of the known cases (Russell's, Weiss's, Mayesima's, Clairmont's and Butters') has it attacked boys. No one has given a satisfactory explanation of this very remarkable fact.

As regards to the *diagnosis* of the disease, this, as has been already mentioned, has hitherto not once been possible previous to operation. It has usually been mistaken for an echinococcus, pancreas or liver cyst. It seems, however, that if one is only acquainted with the fact of the existence of such a disease it would not be impossible to recognize it. Its appearance during the first two decades, as a rule, usually among females, with repeated attacks in the form of a large, cystic, sometimes in size alternating swelling in the right hypochondriac region, more fixed than a distended gall-bladder, conjoined generally with icterus, fever, more or less violent pains; these features, says Dreesmann, are so characteristic that a diagnosis of probability of choledochus cyst would be authorized.

With regard to the *therapeutics* of this disease, an investigation of the 30 cases in which operative measures were employed shows the following: In the majority of cases, namely in 21, the operative treatment consisted only in suturing the cyst in the abdominal wound and the application of an external fistula, in some cases after a preceding puncture through the abdominal wall. All these cases ended fatally a longer or shorter time after the operation, either from exhaustion caused by the enormous loss of bile through the fistula or on account of subsequent icteric hemorrhage or infection. In 1 case, however (Clairmont's), the patient lived three years with this fistula, and died of phthisis. In Ebner's case death occurred in the remarkable form of purpura hæmorrhagica fulminans three months after operation. In 3 of the 21 choledochostomy cases (Ashby's, Dreesmann's and Seeliger's) an attempt was made by means of a second operation to establish communication between the cyst and the intestine; death, however, resulted in all cases. In 1 case, operated on by Bakes, 1911, as well as anastomosis between the cyst and duodenum, a fistula at the same time was made externally, either on the choledochus or the gall-bladder (which does not clearly appear from the account given by the pathologist, Sternberg, who made the postmortem examination); this patient also sank from the operation. In 3 instances (Goldammer's, Lavenson's, and Smit's) the operation

consisted in extirpation of the sac. Death ensued after the operation in these cases also. There remain 5 cases which were restored to health. In 3 of these cases an external fistula was made primarily, and communication between the cyst and the intestine was opened secondarily. In the first of these cases, operated on by Twain in 1894, the anastomosis was made with Murphy's button to a loop at the jejunum drawn up through an opening in the mesocolon. In the second, operated on by Brun and Hartmann in 1897, after a previous marsupialization of the cyst, choledocho-enterostomy was performed in a manner not fully described. The third, operated on by Hildebrand in 1912, was so far remarkable in that the anastomosis made between the cyst and the duodenum could not produce healing of the external fistula, evidently depending on the fact that it was of insufficient size; only when, in a third operation, a new larger inner fistula opening was made did the outward flow of bile cease and definite healing take place.

In the 2 other cases, that operated on by Bakes, 1907, the other my own case, has, as primary measure, a lateral choledochoduodenostomy been made, and both were restored to health.

Bakes' case was examined two years after operation, and the patient was then in perfect health. My case occurred only ten months ago, but from what I have learned the patient has been well ever since the operation.

It appears, therefore, from what has been said, that the opening of communication between the cyst and the intestine is the only rational therapeutic, and this ought to be done primarily.

Dreesmann feared that the application directly of connection between the intestine and the cyst would cause infection of the latter by intestinal bacteria and lead to progressive cholangitis and hepatic suppuration; he proposed, therefore, in order to reduce the risk of infection, which he feared, that the operation should be performed in successive stages; first by employing a gastro-enterostomy; two or three weeks later exclusion of the pylorus according to von Eiselberg, and then making use of choledochoduodenostomy. Well thought out theoretically, this is not, however, to be recommended; partly because the patient is, as a rule, in a reduced icteric condition, which does not permit several trying operative encroachments, partly because, judging from the experience gained from several quarters, the danger of infection from anastomoses between the biliary passages and the alimentary canal is practically not so great.

Finally is hereby given a survey of the cases of idiopathic choledochus cyst described in literature, with short extracts from the descriptions of the cases.

A. OPERATED CASES.

1. KONITZY, 1888. A woman of twenty-one. Ill six months. Rapidly increasing fluctuating tumor, extending from the liver to crista il. Icterus. Operation. Extreme cystic dilatation of choledochus sac, which was incised. Died eight days after operation.

2. SEYFFERT, 1888. Woman of twenty-three. Suffered from icterus for two

years, with intervals of four months, for one year, increasing swelling of the abdomen and attacks of colic. Beside the gall-bladder, which was normal, a tumor as large as a child's head, which proved to be a gall-filled cyst. Drinage. Patient died one month later of violent hemorrhage, which could not be stopped.

3. ARNISON, 1891. The tumor was supposed in operation to be a pancreas cyst and was drained. The patient, who was very much exhausted, lived only a few days. In the postmortem the operation was found to have been choledochostomy and the tumor the dilated common duct.

4. SWAIN, 1895. A girl of seventeen. Had been ill two years. Increasing icterus and swelling of the abdomen for ten months. An enormous abdominal tumor filling the whole of the right side, and extending even to the left half of the abdomen. Puncture, discharge of six pints of a gall-colored fluid. Tumor formed again after five days. Operation. The cystic tumor was emptied of seven pints of gall-colored fluid. The gall-bladder was normal, and lay above the tumor. Gall-stone, at first supposed to exist, could not be found. Anastomosis between the cyst and jejunum with Murphy's button through mesocolon transversum. Recovery.

5. EDGEWORTH, 1895. A girl of four. Ill one year. Repeated attacks of icterus. Large abdominal tumor, diagnosed as distended gall-bladder, which was drained. Death one week after operation. The sac was found to be the enormously distended choledochus, stenosed at its lower end. Cystic duct obliterated.

6. BRUN-HARTMANN, 1897. A girl of three and one-half years with congenital dilatation of the choledochus. The cyst was first marsupialized by Brun. At the next operation Hartmann performed choledocho-enterostomy with success.

7. RUSSELL, 1897. A boy of eight years. Acutely ill for five days with fever and icterus. Right flank filled by a large distended tender tumor, extending downwards to the crista il., forward to the middle line. Under the ribs another tumor, as large as a hen egg, insensitive, and not quite so distended. Diagnosis, echinococcus. Operation: Choledochostomy. Died four days after the operation of hemorrhage from the stitches, according to the account given. Postmortem showed that the cyst came from the choledochus and that the passage between this and the intestine was free. Russell regarded the formation as congenital and analogous with congenital hydronephrosis.

8. ASHBY, 1898. Girl of seven years. Ill two and one-half years with icterus, and increasing emaciation and finally swelling of the abdomen and œdema in the legs. After fifty ounces of ascites fluid had been tapped from the abdomen, a large cyst was observed in the right half of the abdomen; during a period of three months this was punctured eleven times and from eight to sixteen pints of gall-colored fluid drawn off each time. When the patient began to improve under this treatment a choledochostomy opening was made through which all the bile was drawn off. Two months later, an attempt was made to establish connection between the cyst and the intestine. Death from peritonitis ensued. "The cyst seemed to be formed by enormous distention of the common and cystic ducts. The hepatic duct opened into the cavity, but there was no connection between it and the duodenum."

9. ROSTOWZEN, 1898. A girl of thirteen years. For three years distention of the abdomen. Intermittent icterus for one year. Large tumor in the region of the liver, fluctuating in circumference and consistency. Temperature rising to 39°. The cyst which was firmly adherent to its surroundings was first emptied by puncture of two litres dark greenish brown fluid, then sewed fast to the wall of the abdomen and drained. Death the day after operation. Postmortem showed an enormous enlargement of the choledochus, the hepaticus also dilated. The cyst exhibited a kind of valve, and, like the gall-bladder, was empty. The choledochus was bent at an angle at the entrance into the duodenum, and passed obliquely through the front wall.

10. NICOLAYSEN, 1899. A girl of eight years. Icterus a year previously for three months. At her reception into Rig's Hospital in Christiania the patient showed a high degree of icterus, acholic stools, and a fluctuating resistance in the right side under the liver, seemingly as large as a man's head. A small incision was made in

the abdominal wall, with packing against the cyst in order to get cohesions forward. Six days later the cyst was opened and emptied of one and one-half litre of gall-colored fluid. Death ensued the following morning. The postmortem showed that the cyst had originated through enlargement of the hepatic duct and the greater part of the choledochus, the lower portion of which, to a length of two cm., which was fully permeable, was not engaged in the dilatation. The gall-bladder small, and communicating freely with the cyst. "As concrements were not found anywhere, nor disease in any of the adjacent organs, one cannot suppose compression or obstruction of the common duct or the hepatic duct to be the cause; the smallness of the cystic duct being also opposed to this idea. The solidity and thickness of the cyst are also in favor of the theory that enlargement may have been taking place for a long time, perhaps even from birth."

11. DREESMANN. Woman of twenty-four years. Even as early as at the age of six months the patient had been yellow for a short period. Since the age of three violent attacks, with vomiting. From the age of eighteen the abdomen had been very much distended. For two months severe icterus with fever. Below the liver, a swelling as large as a man's head, tender to the touch, and, distinct from this under the edge of the ribs, a fluctuating tumor as large as a fist. The distended gall-bladder, and the large cyst were both opened and drained. An abundant flow of bile up to 1800 cubic cm. daily. Four and one-half months later, an attempt was made to form a connection between the cyst and the duodenum. Sudden collapse of patient three days later. The postmortem showed the absence of peritonitis. The swelling was the enormously dilated choledochus, and the dilatation was continued up into the upper gall-passages. The lower part of the choledochus to a length of three cm. was not engaged in the dilatation and was easily probed, partly from the cyst, and partly from the papillæ vateri.

12. KOLB, 1905. A girl of ten years. For some months, distention of the abdomen and icterus. Below the enlarged liver a distended elastic fluctuating tumor extending downwards to the smaller pelvis, and to the left to the left mammillary line. Test puncture exhibited a gall-colored fluid, without echinococcus hooks. A fortnight later operation (6-Angerer). After tapping the enormous cyst of one and one-half litre, it was sewed to the abdominal wall and drained. Death ensued, not quite a month later. Postmortem showed a cystic tumor into which the upper gall-passages opened upwards. The connection of the cyst with the intestine was only found on microscopic examination and was situated somewhat to the side of the pancreatic duct.

13. ARNOLDS, 1906. A girl of thirteen years. Distention of the abdomen for two years. Signs of obstruction of the choledochus for seven months. During the last few weeks, strength greatly reduced. Below the enlarged, strongly lobed liver in the middle of the abdomen a distended elastic tumor as large as a man's head; to the right of this another smaller cyst. In laparotomy the right cyst proved to be the enlarged gall-bladder, the left the dilated choledochus, from which was discharged "half a bucket full" of gall-colored fluid. Death ensued after twenty-fours hours. In postmortem access could easily be obtained to two passages as wide as the forefinger from the upper pole of the cyst. These were the dilated hepatic ducts. Downwards, connection with the duodenum was found in a short passage with a double angle bend.

14. GOLDAMMER, 1906. A woman of twenty-one years. Had undergone parturition three months previous to her entering the hospital. During pregnancy no troublesome symptoms, but after confinement a swelling observed under the edge of the ribs on the right side, and increasing icterus made its appearance. In the upper part of the abdomen a fluctuating non-sensitive tumor about the size of a child's head and connected with the liver. Icterus in high degree. Operation (Rümmel). Extirpation of the cyst, which contained several litres of greenish-brown serous fluid. Death from collapse. Postmortem showed complete absence of the choledochus with the exception of its intraduodenal portion, and the cyst must therefore

have been the enormously dilated common duct. The gall-bladder was unaltered. The cut-off hepaticus branches at their entrance into the liver were about the size of the forefinger. The intrahepatic gall-passages were much enlarged. No cause of the choledochus dilatation could be established.

15. BAKES, 1906. A woman of twenty-two years. Had suffered for three months from icterus without any other symptoms, except increasing loss of strength. Beside an icterus melas, with acholic stools, only a certain tenderness below the right edge of the ribs could be observed clinically. At operation a rounded cystic tumor as large as a child's head was found between the ventricle and the liver. Above the lower pale of this growth lay the duodenum in a strongly forward displacement. Gall-bladder normal. Papilla duodeni felt normal, without tumor or stone. It was supposed to be pancreas-mesenterial or kidney cyst. Test puncture gave thin yellow bile. Incision of the cyst, probing from the same to the intestine failed. Choledocho-duodenostomy was performed and the patient recovered.

16. EBNER, 1909. A girl of eighteen years. At the age of nine temporary attack of gall colic. From the age of thirteen almost constant painful sensations in the region of the liver, with gradually increasing swelling there. Icterus for six months. Clinically a district of resistance of the size of a man's head in the right hypochondriac region, of distended elastic consistency. Operation (Leerer) showed a cyst lying spread out under the surface of the liver, and filled with thin fluid gall. The cyst had developed retroperitoneally in bursa omentalis. Was sewn to the abdominal wall and drained. Gall fistula. Lived three months in comparative health. At the end of this time an extremely violent purpura hæmorrhagica developed in a few days producing death. Postmortem showed an enlargement of the choledochus, of the size of a man's head, as well as dilatation of the duct hepaticus and orifice of duct cysticus. Remainder of cysticus as well as gall-bladder normal. The lowest undilated portion of the choledochus pierced the anterior wall in an oblique direction, and when probed showed a completely free passage.

17. LAVENSON, 1909. A girl of eight. Icterus one year. For three weeks a swelling in the upper part of the abdomen. Clinical examination showed a considerable cystic non-sensitive tumor in the upper part of the abdomen, attached to the liver, and extending somewhat below and to the right of the umbilicus, as well as a high degree of icterus, and discolored fæces. Operation (Frazier) a cyst as large as an ostrich egg, covered in its lower part by the duodenum, between the latter and the liver the somewhat distended gall-bladder. The cyst was loosened from its attachment to its surroundings and extirpated, and its pedicle ligatured. Death after three days. Postmortem showed that the tumor was a cyst from the choledochus 15x8x7 cm. in size, gall-bladder somewhat dilated. Ductus cysticus obliterated, duct hepatici dilated. The lower portion of the choledochus 3 cm. in length between the cyst and the papilla vateri was in the form of a chord, as thick as a goosequill, in its upper third provided with lumen, in its lower two-thirds without. As well as this, chronic perilobular pancreatitis. Obliteration of lower portion of choledochus was regarded as the result of cholangeitis.

18. WEISS, 1910. A boy of six years. Temporary attacks of pain in the right side, with icterus for more than a year. For half a year abdominal swelling. Clinical examination showed no icterus. Beneath the liver which was enlarged to a high degree a rounded tumor about as large as a fist, hard, mobile in respiration, diagnosed as echinococcus. After test puncture which yielded a clear yellowish-green fluid, symptoms of peritoneal irritation appeared, wherefore an operation was performed with sewing forward of the distended cyst in the abdominal incision, with three days later opening of the same. Death nine days later. Postmortem showed a flaccid cyst as large as a child's head proceeding from the choledochus, beginning at the junction of the cysticus and hepaticus. Sharply displaced upwards by a spur-like protuberance. Impossible to find duodenal opening. Hepaticus and extrahepatic gall-passages moderately dilated. Inner wall of cyst without epithelium. Weiss regards it as a congenital anomaly.

19. BUTTERS, 1910. A boy of five, extremely icteric and emaciated. Abdomen considerably distended, especially to the right, where a large tumor of smooth surface can be felt. In operation a tumor was found below the liver from which was drawn three litres of gall-colored fluid. Death after some days of exhaustion. Postmortem, choledochus cystically dilated to a high degree.

20. ERCNER, 1911. Woman of twenty-three. Icterus in a high degree for one month. Wrongly diagnosed for pancreas cyst and operated. Postmortem (Professor Weichselbaum) showed a case of cystic enlargement of the choledochus containing five litres (this case is probably identical with that demonstrated by Miloslavich at the conference of the Society of Army Doctors of Vienna (Verein der Garmsonsärtze) on the 18th of November, 1911).

21. CLAIRMONT, 1911. A man of twenty-two. The previous history was characteristic, and the clinical appearance that regularly met with in these cases: That, namely of a large area of resistance, not distinguishable from the liver, situated under the edge of the thorax, and showing indistinct fluctuation, also icterus. Operation (v. Eiselsberg), after puncture of the cyst, which was as large as a child's head and contained two litres of pure gall, it was sewed to the abdominal wall and drained. In the evening of the day of the operation, the patient's life was threatened by severe hemorrhage, which, however, was checked. Richly secreting gall fistula, which, however, interfered but little with the patient's comfort. Death three days later of phthisis, and hemorrhage from the lungs. In the postmortem was found a fold of the membrane between the extra- and intraduodenal parts. This had probably caused an obstruction.

22. STERNBERG, 1911. A woman of twenty-five years. Operation performed by Bakes consisted of cholecystostomy and choledochoduodenostomy resulting in death "after some time." At the postmortem performed by Sternberg the gall-passages were found to be greatly enlarged, the choledochus formed a very large cyst, fastened on one side to the wall of the abdomen, and on the other to the duodenum, and open in both directions. Neither in the papilla vateri nor in the lower narrow portion of the choledochus was any alteration to be found in the form of a scar or similar formation, and there was also an absence of concrements. It was, therefore, a case like Ebner's of so-called "idiopathic choledochus cyst." At the point where the narrow part of the choledochus penetrated into the cyst a half-moon-shaped fold was found.

23. SCHLOESSMANN, 1911. A girl of seven. Had suddenly become ill with symptoms of peritonitis and icterus, while at the same time a resistance appeared in the right hypochondriac region. The reaction from the peritoneum disappeared, but the tumor, which clearly was cystic, remained. A test puncture was made, giving clear greenish fluid. In connection with this arose violent abdominal symptoms which necessitated an immediate operation (Sprengel). Gall peritonitis with fatty necrosis in the abdomen. Below the liver a cyst of the size of a child's head, adherent to its surroundings; this was opened and drained. Death from sepsis six days later. The postmortem showed that the cyst was formed from the choledochus duct, which was enormously dilated from its beginning to the vicinity of the papilla vateri. The cysticus and hepaticus opened into the upper part of the cyst in the lower, the terminal part of the choledochus protruding at an acute angle; in all three places, a fold could be discerned from inside. Gall-bladder small. Under the microscope the cyst wall was found to be a fibrous membrane, without elastic elements, and containing very few muscular fibres.

24. MAYESIMA, 1912. A boy of two years and two months. For ten months the child had had a gradually increasing swelling of abdomen, for four months icterus. In the upper part of the abdomen a tumor about as large as a man's head, filling out the right side and lower thorax aperture, extending over to the left mammillary line, downwards a finger's breadth above the symphysis, backwards to the region of the loins. In the place of the gall-bladder another tumor, rounded, about as large as a goose egg. Also ascites, and slight icterus. Operation, puncture of the enormous

cystic tumor, and tapping of two litres of gall-colored fluid. In consideration of the child's precarious condition, further intervention was refrained from for the time. The puncture and the abdominal incision were sewed up. The patient got over the operation, but died two months later. Postmortem showed the cyst to be formed of the enormously dilated choledochus, its lower part only was of normal calibre, and had retained the lumen, but it was slightly kinked and passed in a valviform bend, obliquely downwards from the right, upwards to the left to the cyst.

25. HILDEBRAND, 1913. The patient, whose age and sex are not mentioned, had in the upper right hand portion of the abdomen a large cystic fluctuating tumor, also icterus in a high degree and acholic stools. In operation were found a distended gall-bladder, and a large cyst under the liver, which was pushed upwards by it. The cyst which contained four litres of bile, was opened and drained. There was a flow of bile outwards, but no bile in the intestine. At a new operation an anastomosis was made between the cyst and duodenum. In the beginning some of the bile went to the intestine, but not later on. The gall fistula would not close up. At a third operation, a new and larger anastomosis was made between the cyst and intestine. Sometimes the contents of the intestine entered the cyst and were discharged through the fistula. Hildebrand feared infection of the cyst, which, however, did not take place, and normal conditions were restored with the healing of the fistula. The patient was demonstrated in good health one year after the operation in Berlin's Gesellschaft für Chirurgie (Surgical Society of Berlin) July 14, 1913.

26. SEELIGER, 1913. A girl of thirteen years. The patient, who up to that had been healthy, had become ill some days previously with increasing ileus symptoms together with severe icterus. The abdomen, as a whole, much swollen, especially so in the right upper part, where there was considerable tenderness and rigidity. In the place of the gall-bladder, a smaller limited swelling. Operation showed a large cystic tumor on whose front side passed the duodenum, pressed quite flat, upwards to the liver the distended gall-bladder. Puncture, giving 750 cubic cm. of gall-colored fluid. The cyst was fixated to the abdominal wall and opened four days later. An abundant flow of bile and disappearance of icterus; fifteen days later at a second operation, an attempt was made to produce a connection between the cyst and duodenum, a very complicated operation. Death took place a week later. Postmortem showed limited peritonitis, the cyst had sunk together into a cavity as large as a child's head into which opened upwards duct cysticus and hepaticus; its lower connection with the duodenum could not, however, be ascertained with accuracy.

27. SCHILBE, 1913. The patient, a woman of twenty-five years, had never had any distrubance of the gall-passages previously; she came to the hospital with icterus of three months' standing, for some time high fever almost every day and a tumor twice the size of a fist, below the liver. Marsupialization of an enormous cyst between the liver and duodenum and ventricle, giving nearly four litres of gall-colored fluid. Fever continued, and icterus did not diminish, there was a strong flow of gall. Death took place eight days later from violent hemorrhage from the wall of the cyst. Postmortem showed that the cyst was the highly dilated common duct. The gall-bladder was small, the hepatic and the intrahepatic gall-ducts enlarged. Probing of the lower part of the choledochus from papilla vateri took place without any difficulty.

28. IPSEN, 1913. A girl of eighteen. Had suddenly become ill, six weeks before her reception into a county hospital, with pains under the right ribs. Three days later, icterus with dark urine and acholic stools; her condition remained unchanged. Was received into Rig's Hospital, Division C, June 6, 1912. Icterus in high degree. The right upper part of the abdomen occupied by a considerable swelling. Supposed to be caused by choledochus stone with gall stagnation in the liver. At operation, June 10, an enormous enlargement of the choledochus was found, and this contained about two litres of bile-like clear fluid. The hepatic duct and gall-bladder also much enlarged. No stone, no opening into the intestine to be

seen. Drainage of gall-bladder and choledochus cyst. Severe cholangeitic hemorrhage for a week, after which, however, the patient recovered somewhat, and the icterus diminished. The gall entered the intestine a month later, July 11, 1912, but death resulted from fresh hemorrhage. Postmortem showed the much shrunken choledochus cyst, which communicated with the intestine by a cord, fine as a hair upwards, but somewhat thicker downwards.

29. SMITH, 1915. A girl of seventeen. For five years increasing swelling of the abdomen, for four years periodic pains in the epigastric region, occurring regularly for three or four hours in the afternoon, and lasting on towards the evening. Never icterus. Protuberance of upper part of abdomen and lower part of thorax, proceeding from a considerable tumor which occupied the right hypochondriacal and the epigastric region. Diagnosis, liver cyst (possibly echinococcus) or pancreas cyst. Operation, January 28, 1915. A rounded cystic tumor of the size of a man's head, firmly adhering to the under surface of the liver. Puncture, giving gall-colored fluid. "As one could not be certain regarding the point of origin of the cyst, and an incision with drainage does not appear to be very satisfactory, extirpation of the same was performed." In the course of this it was necessary to ligate four string-like formations, three from the upper part of the cyst going into the liver, and one from its lower part, 3 cm. long and as thick as a match, going towards the pancreas, all provided with lumina. The cyst contained three and one-half litres gall; in its wall lay the gall-bladder like a blind sac of the thickness of a lead pencil opening into the cyst, its place of entrance as well as those of the upper and lower gall-passages covered by crescent-shaped folds. Death eight days after operation. Postmortem showed that the duodenal part of the choledochus, which had been ligated 2 cm. from the papilla vateri, did not exhibit any stricture.

B. NON-OPERATED CASES

30. DOUGLAS, 1852. A girl of seventeen. For an uncertain time pains in the liver, icterus in a moderate degree, and a swelling gradually increasing in size in the right hypochrondriac region. This was accompanied by fever, wasting, and œdema in the legs. Test puncture showed gall-colored fluid. Death a fortnight later. Excessive enlargement of the choledochus, containing half a gallon of very ill-smelling gall, hepatic branches as thick as a finger. Gall-bladder and cysticus not enlarged. In the lower part of the cyst a funnel-formed opening, provided with a valve, leading into the lower, not dilated portion of the choledochus.

31. HEID, 1893. A girl of fourteen. For about six weeks pains in the region of the umbilicus, increasing icterus, and swelling of the abdomen. A considerably enlarged liver had been observed, below which was found a rounded tumor of a size and consistency varying with different occasions. Postmortem showed a liver weighing 3035 grams, a cystic enlargement of the choledochus as large as a child's head, its lower part 2.1 cm. long was not engaged in the dilatation, and was kinked, but could be easily probed from the intestine.

32. BROCA, 1898. At the postmortem of a ten-year-old girl was found a cystic enlargement of the size of a child's head. The passage showed no obstruction or obliteration, but an angular curvature between the cyst and duodenum.

33. WETTWER, 1905. Girl of fifteen. From the age of five had been subject to attacks of cramp of the stomach with vomiting, from the age of seven occasional icterus. In the epigastric region a rounded, soft tumor, sensitive to the touch, the size and consistency of which were increased after meals. Besides this icterus, fever and signs of ascites. Postmortem showed tumor twice the size of a fist, originating from dilatation of the choledochus. The lower part of the passage bent at an angle, going downwards from the cyst upwards to the duodenum was easily probed.

34. HEILIGER, 1910. Described in text.

In all the cases quoted above, the diagnosis of idiopathic choledochus cyst seems to be without doubt. In the literature on the subject there are, however, several cases related, of a high degree of cystic dilatation of the choledochus, which, either on account of incomplete data or from the existence of a real hindrance in the form of stone, tumor formations in the lower part of the choledochus, or chronic pancreatitis, cannot be reckoned offhand as belonging to this category, despite the fact that the pathogenesis not improbably—at least for some of them—seems to be the same.

To these cases belong Todd's (1817), Fabre's (1831), Barlach's (1876), Legg's (1876), Raynaud and Sabourin's (1879), Oxley's (1883), Wilkes-Moxon's (1889), Brunner's (1889), Mayo Robson's (1893), Eve's (1906), and Milner's (1909).

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