

Cullen's Sign

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THE best description of Cullen's sign that has yet appeared in the literature is that of Schumann,¹ namely, peri-umbilical ecchymoses. The sign is due to the presence of blood pigments in the abdominal wall, usually in the region of the umbilicus or near operation scars, which look exactly like bruises. This sign appears to be of great rarity, for as far as I can ascertain it has only been described in the literature on 45 occasions, referred to on two other occasions, a similar sign mentioned twice, and in this paper I record 3 more cases; making 52 in all. Of the 45 cases 7 were not true Cullen's sign, but cases of what I call pseudo-Cullen sign, or Hofstatter's sign. His name has never been given to this sign, but, as I shall show, he was the first to describe this "Modification of Cullen's sign."

As a student I heard nothing of the sign, and first read of it in Zachary Cope's book "Early Diagnosis of the Acute Abdomen" in which he says that it is "an inconstant sign of ruptured ectopic gestation." The sign was first described by Professor T. S. Cullen² in 1918, and published in that year in the *American Journal of Obstetrics*. It was an account of the proceedings of the 43rd annual general meeting of the American Gynaecological Society, held at Philadelphia, May 16th to 18th, 1918. It was a very short clinical memorandum entitled "A New Sign in Ruptured Extra-uterine Pregnancy." The patient was a woman aged 38 who suddenly developed abdominal pain and distension. She was seen by Cullen 3 weeks later, and the umbilical region was bluish-black. History of injury was absent. Cullen diagnosed the presence of haemoperitoneum by a process of analogy from a case reported by Ransohoff in 1905. He opened the abdomen and found a right ruptured tubal gestation and 1½ quarts of free blood in the peritoneal cavity. The case of Ransohoff^{3, 4} referred to by Cullen was first recorded in 1905. The patient was a man aged 53, who showed vague abdominal signs and after some time the umbili-

cus and the surrounding region was stained a saffron-yellow colour. Laparotomy revealed a ruptured common bile duct, and the abdominal cavity contained a large quantity of free bile. Two popular beliefs have arisen from these descriptions; firstly, that the sign is seen only in connexion with ruptured ectopic gestation and, secondly, that a large quantity of free blood in the peritoneal cavity is essential for its appearance. Neither of these statements is correct.

Pseudo-Cullen Sign.

Before proceeding further it is necessary to consider the pseudo-Cullen sign, or Hofstatter's sign. This consists of a blueing of the umbilicus due to the colour of blood shining through an umbilical hernia. It has also been described in connexion with incisional and femoral herniae. The proof that this differs from Cullen's sign is given in a case recorded by Esau⁵ in 1934. In his case both the colour shining through an umbilical hernia and the peri-umbilical staining were present. At the operation a piece of omentum surrounded by blood was found adherent to the hernia. When this was released and the blood mopped away, the blue colour at the umbilicus disappeared, but the staining around persisted. This is the only case in the literature in which both signs appeared together. There are seven references to the appearance of the pseudo-Cullen sign in the literature^{6, 7, 8, 9, 10, 11, 12}. The sign was first described by Hofstatter in 1909.⁶ It is interesting to note that many continental writers refer to true Cullen's sign as the Hofstatter-Cullen-Hellendall sign. A perusal of the literature shows that these two authors have only described the pseudo-Cullen sign, and there is not any evidence to show that they have ever seen the true Cullen sign and, therefore, have no claim to associate their names with the American in the description of the sign.

Grey Turner's Sign.

Cullen's sign has been described in connexion with acute pancreatitis and further reference will be made to this later; a sign bearing a resemblance to Cullen's sign has been described by Grey Turner¹³ and referred to by Hamilton Bailey¹⁴ and others. Grey Turner described 2 cases showing the sign. The areas of discoloration were the size of the palm of the hand and dirty brown in colour. In both cases a moderate amount of dirty brown fluid was found in the abdomen. The areas were

raised and oedematous. In the first case the staining was around the umbilicus, but in the second it was situated in both loins. At the post-mortem examination on the second case the pancreas was found to be represented by a slough, and dissection showed a track leading directly to the stained areas of skin. Grey Turner considers that it is due to the direct action of the pancreatic ferments on the superficial tissues, and that in his first case the umbilicus was involved via the ligamentum teres hepatis.

Diseases in which Cullen's Sign Appears.

As I have previously pointed out an error arose from Cullen's original description, and it was thought that the sign was pathognomonic of ruptured ectopic gestation. One has only to think of Ransohoff's original case to see that it can occur with any cause of haemoperitoneum. By chance it happened in his case that the bile was escaping from a ruptured common duct, but the sign would probably have occurred just the same if the escape had been from the gall bladder or any other part of the biliary tract. Cullen saw a blue instead of a saffron-yellow discoloration, and argued rightly that its cause was blood. It so happened that the bleeding was coming from a ruptured Fallopian tube, but it is reasonable to suppose that the sign would have appeared from any other cause of haemoperitoneum. Beckwith Whitehouse¹⁵ says that it is not pathognomonic of extra-uterine gestation, but may occur with any intra-peritoneal haemorrhage. This statement is partly correct, but it will be seen that the sign can occur without intra-peritoneal haemorrhage. The accompanying table shows the number and distribution of the cases recorded. For the sake of completeness the cases of Grey Turner's sign and Hofstatter's sign are included.

Disease	Cullen's sign	Hofstatter's sign	Grey Turner's sign
Ectopic gestation	19 (5)	7	—
Acute pancreatitis	8	—	2
Ruptured uterus	3 (2)	—	—
Haematoma of rectus sheath	1	—	—
Ovarian cyst	1	—	—
Pyosalpinx	1	—	—
Hepatic haemorrhage	1	—	—
	34 (7)	7	2

(5) Cases described, the literature of which I have been unable to obtain.

(2) Cases apparently not recorded, but mentioned at a scientific meeting.

Ectopic gestation contributes by far the largest percentage in the above table, which is not surprising when one considers that this must be the commonest cause of haemoperitoneum. It seems rather strange that acute pancreatitis should come second on the list and not ruptured uterus but in most of the cases described there was a considerable amount of blood-stained fluid in the peritoneal cavity. It was not until 1937 that the sign was recorded in connexion with ruptured uterus. This was by Gerrard¹⁶ of Manchester. Two others recorded here by myself and two others mentioned in connexion with Gerrard's paper complete the list. There remain 4 miscellaneous disorders. It is of interest to note that the sign has been recorded by Petrivalsky,¹⁷ Fallis,¹⁸ and myself on 3 occasions each (the largest number recorded by a single observer), yet neither of us has seen it in connexion with ectopic gestation.

It will be seen from the table that 5 other cases in connexion with ectopic gestation have been recorded. I have had no opportunity of obtaining the journals concerned and am, therefore, not in a position to include them in the following analysis ^{19, 20, 21, 22, 23.}

Ectopic Gestation.

The first case described was Cullen's, mention of which has previously been made. Since 1918 the following cases have been described:

Novak²⁴ gives a short description of 2 cases which he has observed. In both there was a greenish-yellow discoloration around the umbilicus, and a considerable quantity of free blood in the peritoneal cavity. He reproduces an excellent colour plate of Cullen's original case. As regards the aetiology he believes that blood tracks through the lymphatic anastomosis around the umbilicus.

Jackson²⁵ observed the sign in an obese woman. It was not seen at first, but when the room was darkened and a strong light shone on the umbilicus it became quite obvious as a greenish-yellow stain. At the operation there was an unruptured tubal pregnancy, an ovarian cyst and gross haemoperitoneum.

Strube.²⁶ In this case the sign appeared 17 days before

evidence of tubal abortion occurred. By the time laparotomy was performed the sign was gone. Much free blood was found in the peritoneal cavity.

Pfeiffer. This case was recorded by Frankel.²⁷ The sign appeared here 7 weeks before laparotomy was performed. An unruptured ectopic gestation without haemoperitoneum was found.

Gold.²⁸ A blue stain appeared in a midline laparotomy scar which did not show any evidence of hernia. A ruptured ectopic was found at the operation. There was, in this case, a history of injury to the abdomen with a stick 2 weeks previously.

Solojew²⁹ remarks that he has seen the sign once only in over 100 cases. The diagnosis was in doubt until 3 days after admission, when the sign appeared. Its distribution was around the umbilicus, suprapubically, and near the anterior superior spine of the ilium. Laparotomy was performed and revealed a ruptured ectopic.

Forgue.³⁰ The only feature of interest in this case is that the omentum was adherent to the umbilicus. There was no hernia.

Lavell,³¹ in a review of his experience of ectopic gestation, states that in many cases he has only seen the sign twice, and that this occurred in the same patient in successive ectopic pregnancies.

Behney³² states in a similar review that it has appeared once in his experience of 167 cases. He describes it as 'a blue disc around the umbilicus, which is an inconstant sign in ectopic gestation.'

Tanner.³³ This case occurred in 1931. On October 17th the patient had a sudden attack of abdominal pain, and 3 days later observed some bruising on her abdominal wall. On the 30th she again had severe pain and was admitted to a nursing home. When seen by Tanner a day later there was bruising just below the skin, shaped like a circle the centre of which was free from staining. It was situated to the right and below the umbilicus which was stained red. Laparotomy revealed a ruptured ectopic and gross haemoperitoneum. The author holds the view that blood permeates the lymphatic anastomosis. This publication drew forth a letter from Blair-Bell³⁴ in which he said that he knew of the sign although a recent search of his writings fails to find any mention of it. Also arising out of Tanner's case Gordon,³⁵ of Newark, N.J., stated that he had seen the sign once in many cases of tubal pregnancy.

Schmitz.³⁶ In his case a blood red stain appeared in close proximity to an old appendicectomy scar to which omentum was adherent, and in which there was no hernia.

Esau.⁵ This case has been referred to previously as the only one in which Cullen's sign and Hofstatter's sign appeared together. Esau claims that this was a case of ovarian pregnancy.

Stabler.^{37, 38} In this case the patient had an attack of abdominal pain and noticed some bruises on her abdomen. She consulted her doctor, who at once recognized the sign and sent her to hospital with a diagnosis of ectopic gestation. One inch below the umbilicus and to the left was a black mark shaped like a comma, $\frac{3}{4}$ inch by $\frac{1}{4}$ inch. A little lower was a bruise, blue in colour, and one inch in diameter, while supra-pubically and to the left of the mid-line was a fresh bruise, reddish-purple in colour and $2\frac{1}{2}$ inches in diameter. Stabler diagnosed an intraligamentary rupture with blood tracking along the course of the round ligament. Operation, however, revealed an unruptured tubal pregnancy. Stabler is now of the opinion that an abnormal aperture, or break in the continuity of the peritoneum, is essential for the appearance of the sign. He has not seen it in any other case in his large experience of ectopic gestation.

Smith and Wright.³⁹ These two writers quote 2 cases. The first was in a woman aged 47, and the sign appeared near an appendicectomy scar in which there was no evidence of hernia. Pain had been present for 5 days before the appearance of the sign. The second case was in a woman aged 40. This sign was very faint and was seen better in the strong light of the operating theatre. It faded 2 weeks after operation.

Brady.⁴⁰ The patient was aged 35 and had had an appendicectomy in 1923. She complained of pain and irregular vaginal bleeding. She noticed that the appendicectomy scar had turned a dark blue colour. This happened 2 weeks after the onset of the irregular bleeding, and when admitted to hospital was gradually getting less. On examination the centre of the scar was bluish black and the surrounding area various hues. The umbilicus was normal. There was no hernia but the scar was thin. At the operation a left ruptured ectopic was discovered with 200 c.cs. of free blood in the abdomen. The discoloration did not disappear entirely until 2 weeks after the patient returned home from hospital.

Hofstatter's Sign.

For the sake of completeness short descriptions of these cases are included.

Hofstatter^{6, 7} described 3 cases of blood shining through hernial sacs. Two were umbilical and one ventral.

Schmid.⁸ Umbilical hernia, in 1913.

Hanak.⁹ Incisional hernia, in 1915.

Hellendall.^{10, 11} Umbilical hernia.

Souligoux and Laquiere.¹² Femoral hernia. The diagnosis made was one of strangulated hernia. A ruptured ectopic was found at the operation and the hernial sac was full of blood.

Acute Pancreatitis.

Reference in this connexion has already been made to Grey Turner's sign, which will not here be considered further. The earliest mention of Cullen's sign in this disease was made by Johnson⁴¹ in 1930. This patient showed a deep circle $1\frac{1}{2}$ inches in diameter around the umbilicus stained a deep yellow colour. Laparotomy revealed acute haemorrhagic pancreatitis. Drainage and cholecystostomy were performed and the patient recovered.

Petrivalsky¹⁷ recorded 3 cases of his own and one of Stutzer's.⁴² He subscribed to the lymphatic theory of origin.

Fallis¹⁸ described 3 cases of his own. In the first case, a woman aged 68, the sign appeared after 3 days of severe abdominal pain. The diagnosis was made on the appearance of the sign. Death followed laparotomy. In the second case the sign was seen only in the strong light of the operating theatre. In the third case (a male) there was a haemorrhage in the left half of the transverse meso-colon. These last 2 cases recovered. Fallis is the only person in the literature who recognizes the difference between Cullen's sign and that of Hofstatter. He also quotes 2 other cases of the same disease in which there was severe intra-peritoneal bleeding, and in which the sign did not appear.

Ruptured Uterus.

Gerrard¹⁶ was the first to record the sign in a case of rupture of the uterus. His patient was pregnant for the second time. Her first labour had been terminated by an upper segment Caesarean section after a long trial labour. There was some morbidity in the puerperium. At the time Gerrard saw her she was 37 weeks pregnant. Two days before he saw her she had an attack of abdominal pain. Some time later she

noticed a red stain on her umbilicus. At the time of examination she had no complaint, but around the umbilicus was a reddish stain the size of a shilling. The diagnosis was made on the appearance of the sign and laparotomy was performed. There was no haemoperitoneum, but a clot 2 inches by 1 inch was found attached to the old scar. When this was removed a rent in the old uterine scar 1 inch in length was found. Caesarean section was performed. Gerrard said that he could not find any reference in the literature to a similar case. In a discussion on this case at a meeting of the North of England Obstetrical Society, W. Gough⁴³ and W. D. Galloway⁴⁴ each said that they had seen a similar case. Gerrard⁴⁵ is quite definite that he would not have made the diagnosis if the sign had been absent. This brings me to my first personal case, the story of which is as follows:

The patient was pregnant for the fourth time and was admitted to hospital on May 26th, 1939. She had had two normal deliveries at term, the last one in 1929. She was admitted to hospital in 1934 in her third labour, which was obstructed by the presence of a large cervical fibroid. Upper segment Caesarean section was performed. Six months later the fibroid was removed by myomectomy. She was admitted at 5.30 p.m. on account of an attack of abdominal pain 12 hours previously. This had lasted 2 hours and there had been a slight show. On admission she had no complaints, her pulse-rate was 90 and her temperature normal. She was 36 weeks pregnant and the foetus was lying transversely. There were 2 abdominal scars, one a mid-line infra-umbilical incision through which the myomectomy had been performed and the other a paramedian, partly infra-umbilical and partly supra-umbilical through which the Caesarean section had been performed. This latter scar was very thin, but was not the site of the hernia. In the centre was a reddish-purple dot about a $\frac{1}{4}$ inch in diameter. The os was closed, the pelvis normal, and fibroids were not present. I was suspicious that this might be Cullen's sign, but the patient's condition was so good that I saw no reason to interfere. The next day her pulse was normal and she did not complain. The findings were the same. The following day, 28th, I was demonstrating abdominal palpation to the students and chose this patient as a subject. What I saw when the bed clothes were removed certainly surprised me. I found not only one small purple dot, but four, and three greenish-yellow patches looking like bruises and arranged around the

umbilicus. The largest was oblong and measured 2 inches by 1 inch. It was situated to the right and just above the umbilicus. The two others were circular in shape and about 1 inch in diameter. One was 1 inch to the left and level with the umbilicus, the second was situated over the linea alba and 2 inches below the umbilicus. There were no signs of internal haemorrhage, and no change in the previous findings except that the foetus was more easily palpable and was now lying as a breech. Kehr's sign (shoulder pain) was absent. I performed a laparotomy 2 hours later. The foetus, placenta, and membranes were lying loose and complete in the abdominal cavity, which contained a large number of clots and a considerable amount of free blood. The uterus was well retracted and empty, and not larger than it would have been at 4 months. The old scar had given way in its entire length. The myomectomy scar was visible and intact. The omentum was not adherent to either abdominal scar. The scar was resutured, the patient sterilized, and the abdomen closed. There was wound sepsis in the puerperium. Cullen's sign disappeared by the 9th day. The patient has since remained well.

My second case was admitted to hospital on January 16th, 1940. She was a booked case, but came in as an emergency at the 27th week of pregnancy. She had a contracted pelvis and had had 5 previous upper segment Caesarean sections. At 2 a.m. on the day of admission she was seized with violent abdominal pain and had a slight show. She was admitted at 8.30 a.m. complaining of severe abdominal tenderness. There was some rigidity, and the foetus was palpated with extreme ease. There was dullness in both flanks and Kehr's sign was present. A diagnosis of ruptured Caesarean scar was made. All the operations had been performed through the same abdominal incision, which, however, did not show any sign of hernia. In addition the patient was suffering from a severe attack of influenza, her temperature was 103°F., pulse-rate 116. Cullen's sign was absent. Operation was performed under local anaesthesia 1½ hours later. While the skin preparation (pure Dettol) was being applied a student drew my attention to the umbilicus. It was now coloured reddish-purple. When opening the abdomen I spent a considerable time in dissecting the abdominal wall to see if I could find out how the staining came about. The previous skin incisions had not involved the umbilicus. Cutting into this structure revealed the presence of fluid blood in the subcutaneous tissues. This blood appeared

to be coming from the rectus sheath. This structure was intact anteriorly, and when opened it was found to contain fluid blood. On retracting the muscle the posterior layer was found to be inefficiently repaired in some places. When the peritoneum was opened a small piece of omentum was found incorporated in the old scar and sticking through into the rectus sheath. Along this piece of omentum blood was running and gaining entrance to the rectus sheath. The foetus, placenta, and membranes were found lying free amidst a considerable quantity of fluid and clotted blood. The old uterine scar had given way completely. It was resutured, the patient sterilized and the abdomen closed. The next day the colour of the umbilicus had changed to a greenish-yellow, but the area had not increased. The sign had gone by the 8th day. The patient made a good recovery.

Haematoma of the Rectus Sheath.

This is my third personal case and as far as I can discover the only one of its type recorded. This patient was a woman aged 46, admitted to hospital, under the care of a physician, in November 1938, with a diagnosis of cardiac failure and auricular fibrillation. She complained of severe breathlessness, palpitations, swelling of the legs and a bad cough. The sputum contained blood. There was some vomiting. For a week prior to admission she had had a severe pain in the left side of the upper abdomen which had appeared suddenly after a very bad bout of coughing. On examination she was found to be suffering from cardiac failure associated with mitral stenosis. There was a large hard tender mass on the left side of the abdomen extending from the costal margin to three fingers' breadth below the umbilicus. When seen on December 12th, 1938, she had a very well marked Cullen sign, consisting of 5 "bruises" each about 1 inch in diameter. They were all situated on the left side over the lump. Two were above the level of the umbilicus, 2 below, and one level with it. The umbilicus was not discoloured. On January 5th, 1939, laparotomy was performed. By this time the sign had disappeared. At operation it was found that there was nothing abnormal in the peritoneal cavity, and that the swelling was in the abdominal wall. The abdomen was closed and a fresh incision was made over the swelling. It proved to be a large haematoma of the rectus sheath originating from a ruptured superior epigastric artery,

presumably brought about by a violent fit of coughing. Recovery was uneventful.

Other Diseases Showing the Sign.

There are 3 cases in this group, 2 of them described by Schumann.¹ The first case was diagnosed as an ectopic gestation on the strength of the presence of the sign. It proved to be an intra-uterine pregnancy complicated by a bilateral pyosalpinx. The second was a case of fatal intra-peritoneal bleeding from the liver due to post-operative acute yellow atrophy of the liver. The last case is recorded by Busch⁴⁶ and occurred in a woman aged 60.

Laparotomy revealed the presence of an ovarian cyst. There was no blood in the peritoneal cavity. I do not know how to explain this case and wonder if it really was a case of Cullen's sign. Miles Phillips⁴⁷ has mentioned a case in which the sign appeared without any haemorrhage. He does not say that it was a case of his, and it may well be Busch's case that he was quoting.

Although it is impossible to give any figures, or to get any idea of the incidence of the appearance of Cullen's sign, it will be seen from this paper, which is based on an almost complete review of the literature since 1900, that it is very rare. There are references in the literature emanating from men of great experience who state specifically that they have never seen the sign, although in many years of work they must have handled many cases of ectopic gestation. Luker,⁴⁸ in a short review of the signs and symptoms presented by cases of ectopic gestation admitted to the London Hospital over the course of a long period, does not make any mention of the sign.

Mode of Origin.

Most authors have been content to describe cases in which the sign has appeared and only a few have attempted to explain how it arises. Three theories have been advanced, viz.:

1. The simple soaking through of blood pigments from the peritoneum to the skin;
2. Blood pigments reaching the abdominal wall via the lymphatics;
3. Spread by means of an abnormal aperture in the peritoneum other than a hernia.

1. *The soaking through theory.* This theory was put forward by Ransohoff, who assumed that the stain was due to the substance in the peritoneum (in his case bile) soaking through the tissues of the abdominal wall. Why this should not

take place in every case of haemoperitoneum, if the theory is correct, is difficult to see. On the other hand why it should occur with only a few ounces of blood in the pouch of Douglas is equally incomprehensible. No other writer supports this theory.

2. *Lymphatic spread.* It is said that there is a very free anastomosis between the lymphatics of the peritoneum and the skin lymphatics of the abdominal wall, and that this anastomosis takes place around the umbilicus. It is assumed, therefore, by some authors that it is through this anastomosis that the blood pigments reach the abdominal wall. There must be thousands of cases of haemoperitoneum occurring annually in the world. This lymphatic anastomosis is normal and is not a freak of nature and, therefore, is present in every individual. Why, then, do we not encounter thousands of cases of Cullen's sign? Petrivalsky subscribes to this theory and overcomes the question that I have just raised by an explanation that Americans would rightly call phoney. He claims that if the omentum is normal and reaches to below the umbilicus then the blood lies behind it and is shut off from the umbilicus and the sign does not appear. If, however, the omentum is short then the blood does come in contact with the umbilicus, and the sign appears. I have many times seen the omentum reaching to below the umbilicus with blood lying *in front* of it. Also in some of the cases recorded there were only a few ounces of blood lying in the bottom of the pelvis and never coming anywhere near the umbilicus. Tanner and Novak also believe in this theory of lymphatic permeation.

3. *Spread by means of an abnormal aperture.* This is the theory which I support. Other supporters in the literature are Stabler and Gerrard. The belief is that blood escapes into the subcutaneous tissues from the confines of the peritoneal cavity through an abnormal aperture which may be either congenital or acquired, such as an operation scar. Abnormal apertures in the peritoneum are very rare; the anatomy and surgery books do not make any reference to them. Cullen's sign is also very rare. The visceral and parietal peritoneum have a different embryological derivation and the junction of the two might form the site of such an aperture. Several cases have occurred in operation scars where there might be breaks in continuity. In one of my cases I found such a break through which blood was gaining entrance to the rectus sheath. Once blood has entered this structure there are numerous apertures in the anterior

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layer through which vessels and nerves are transmitted, and through which blood could enter the subcutaneous tissues. In this case also a piece of omentum was forming a track along which the blood was running, and in several other cases recorded the omentum was adherent either to the umbilicus or to operation scars.

Lavell³¹ in many cases of ectopic gestation has seen the sign only twice, and this occurred in the same patient who had successive ectopic pregnancies. If this theory is correct it is to be assumed that if the sign appears once it should appear on every subsequent occasion in which the patient gets a haemoperitoneum. Conversely Hodgson⁴⁹ handled two successive ectopic gestations in the same patient and on neither occasion did the sign appear. I have also met a similar case.

How to explain the presence of the sign where no haemorrhage occurs anywhere as in Busch's case and one of Schumann's I do not know.

Summary.

1. Cullen's sign is described.
2. The difference between Cullen's and Hofstatter's signs are pointed out.
3. An analysis of the conditions giving rise to the sign is given.
4. A detailed review of the literature is given.
5. Its rarity is stressed.
6. A description of three personal cases is given.
7. Theories of its mode of origin are put forward.
8. It is assumed that the sign indicates the presence of blood in the peritoneal cavity or rectus sheath.

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