



Pediced jejunal interposition as esophageal substitute in pediatric patients. Technical considerations and long-term results

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ABSTRACT

The current study reports the surgical technique and the clinical long term results of pediatric esophageal substitution with a pediced jejunal loop in two patients with long gap esophageal atresia and one patient with severe esophageal stenosis due to a caustic lesions. Postoperative course was uneventful in all patients. At a long term follow-up of 29 and 43 years for the patients with esophageal atresia and 21 for the one with caustic stenosis, no feeding troubles were reported due to malfunction of the esophageal replacement nor to loss of its original functions and the patients referred a highly satisfying quality of life. Pediced jejunal interposition confirms as a good substitute of the esophagus in pediatric age with the advantage to maintain good performances for very long time. Although these patients do not share the same risks of illness as other patients still harboring their native pathologic esophagus, a far sighted surveillance is recommended due to long term potential morbidity risk.

1. Introduction

When the esophagus is congenitally defected as in the case of long-gap esophageal atresia or severely damaged because of caustic agents or other reasons, esophageal replacement becomes necessary.

It has been controversial what should be used as a substitute of the esophagus in infants and children, from the whole stomach, to a gastric tube, a segment of colon, or a jejunum tract [1–3].

Main characteristics of the esophagus substitute should ensure: i) a good peristalsis; ii) it should not dilate after positioned in the thorax or mediastinum; iii) no severe symptoms caused by the deficit of the organ itself.

Jejunum should be selected as shows a motility more similar to the esophagus, even if the number of comparative studies is limited and they are disomogeneous, being based only on short and medium follow up period.

Very long term results of our clinical experience in the reconstruction of the esophagus using the interposed pediced jejunum technique in two cases of esophageal atresia and in one case of caustic esophageal stenosis are here reported.

2. Case reports

Data from three patients, 2 with long gap esophageal atresia and 1 with severe esophageal stenosis due to caustic lesion were collected by the following sources:

- 1 hospitals records at the moment of esophageal reconstruction.
- 2 reports referring to check-ups performed at the outpatient clinic during the long term follow-ups, based on documents collected by patients.
- 3 recent telephone interview: specific questions concerning especially aspects related to social life, weight, height, swallowing habit, the presence of bad breath, diarrhea and other medical problems.

Long term follow-up period ranged between 21, 29 and 43 years. None of the three patients accepted to go through more diagnostic investigations.

All clinical records and the most recent phone interview were reviewed for medical and surgical history, operative technique, complications, and esophageal function.

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2.1. A) Esophageal atresia (case 1 and 2)

In two patients, esophageal atresia long gap, more than 6 vertebrae without fistula, was diagnosed. Weight at birth was Kg 2750 for the case 1, and Kg 2030 for the case 2.

No associated malformations were reported, but the second patient was diagnosed with Down syndrome. At birth both patients underwent gastrostomy surgery with permanent suction of upper esophageal stump by Replogle tube. Prior to esophageal reconstruction surgery, the gap between the two esophagus stumps were examined through investigations based on contrast agents: it was located over the sixth vertebra.

Both of them underwent esophageal substitution at the age of 11 months.

Esophageal reconstruction technique by pedicled jejunal interposition:

1) Laparotomy. With patient in supine position, a midline incision was made from the xiphoid process to and around the left side of the umbilicus. The gastrostomy was detached and closed with an interrupted suture. The pedunculated jejunum segment creation required an accurate study of jejunal mesenteric vessels. In case 1, the 2nd and 3rd mesenteric vessels were ligated in order to obtain a long vascular pedunculus: the jejunum was transected close to the Treitz ligament, leaving enough proximal jejunal length for the later restoration of continuity (Fig. 1). In case 2, the vascular arcade showed a complete interruption corresponding to the 2nd mesenteric vessel (Fig. 1-(A)): for this reason

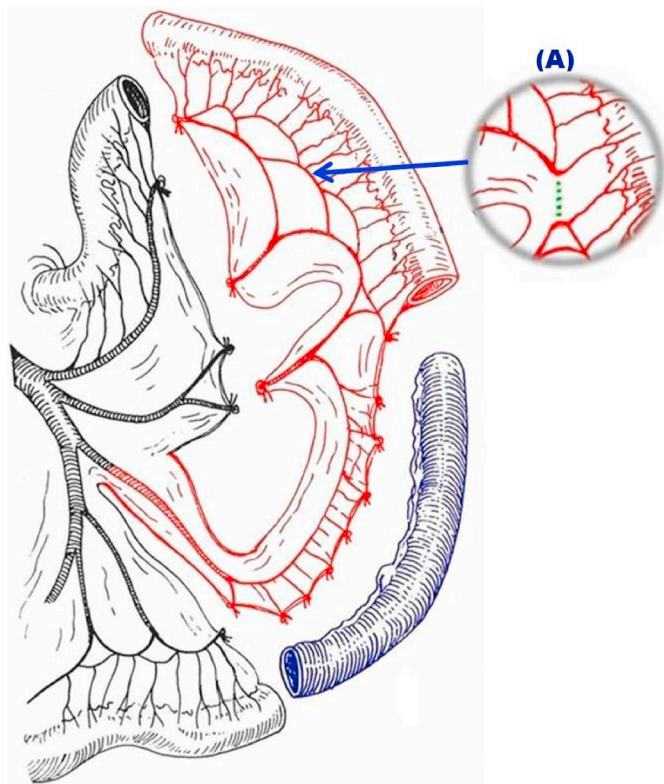


Fig. 1. Preparation of jejunal loop of 5–7 cm. to be transposed. In case 1 the first jejunal mesenteric vessel is preserved, the 2nd and 3rd mesenteric vessels are cut to ensure an adequate length to the vascular pedicle. The 4th jejunal mesenteric vessel maintained the vascularization of transposed jejunum. The jejunum was transected close to the Treitz ligament. (A): in case 2, the jejunum vascular arcade presented with a complete interruption at the 2nd mesenteric vessel level; for this reason, only the 3rd mesenteric vessel was ligated and the jejunum was at the 2nd mesenteric vessel level, for later restoration of continuity. Redundant part of the jejunum was resected.

son only the 3rd mesenteric vessel was ligated and the jejunum was transected at the level of the 2nd mesenteric vessel. Ligating just the 3rd mesenteric vessel was, fortunately, enough to obtain an adequate vascular pedunculus due to the abnormal length and the satisfactory calibre of the 4th mesenteric vessel.

In both cases the part closest to the jejunal loop was prepared to reconstruct the esophageal gap with a length of 5 and 7 cm, respectively; the distal part of the upper jejunum has been removed from the distal end, for a total length of 40 cm. As a result to the removal of this jejunal tract it was observed an additional elongation of the vascular pedunculus and a more satisfactory vascularization of the proximal jejunal loop.

The jejunal loop, previously prepared together with its long vascular pedunculus, was temporarily wrapped with transparent PVC foil to facilitate sliding and passed through retrocolic and retro pyloric position to be passed then into the thorax through a hole on the diaphragm opened beside the right hiatal crus.

Finally, the bowel continuity was restored with an end-to-end anastomosis between the two residual jejunal stumps.

2) Right thoracotomy: longitudinal thoracic skin incision on the axillary line, and thoracotomy at 4th and 7th intercostal spaces. Through thoracotomy at the 7th space the jejunal loop was transposed into the right hemithorax, by using a combined maneuver. From the 4th intercostal space incision end-to-side anastomosis between the upper esophageal stump and the jejunal loop, and from the 7th intercostal space incision end-to-end anastomosis between the inferior jejunum loop and the lower esophageal stumps (Fig. 2).

All anastomoses were performed using a single layer technique with interrupted synthetic resorbable sutures. Para-esophageal drain and a chest drain were placed. A nasogastric tube was passed through the neo esophagus into the stomach.

3) The thoracic and abdominal incision were closed.

2.2. B) Esophageal stenosis due to caustic lesions (case 3)

The patients, a child of 4 years of age, was subjected in vain to repeated esophageal dilations for more than 6 months. Due to an extensive esophageal stenosis caused by caustic agents starting from above one third of the thoracic esophagus until the gastro-esophageal junction (Fig. 3), the esophageal stenosis had been removed including the cardiac tract.

Esophageal reconstruction technique by pedicled jejunal interposition.

Due to the need to replace a long esophageal tract affected by stenosis, technical specificities compared to the other two cases were related to the esophagectomy and the length of isolated jejunum.

1) Laparotomy: esophagectomy started with a trans-hiatal approach, trying to preserve the vagus nerve branches. Through this trans-hiatal approach, esophageal isolation was performed at the highest possible location, proceeding then to esophagus transection from the stomach and subsequent suture of the stumps.

2) Right thoracotomy: the thoracic portion of esophagus affected by stenosis was isolated and removed through a thoracotomy incisions of the 4th and 7th intercostal space, as in the esophageal atresia patients. The jejunum to be transposed into the thorax had a final length of approximately 14 cm.

After end-to-side anastomosis between the upper esophageal stump and the jejunal tract from the 4th intercostal space incision, the jejunum distal portion was anastomized side to side to the anterior stomach wall (Fig. 4).

A nasogastric tube was placed to drain the stomach cavity.

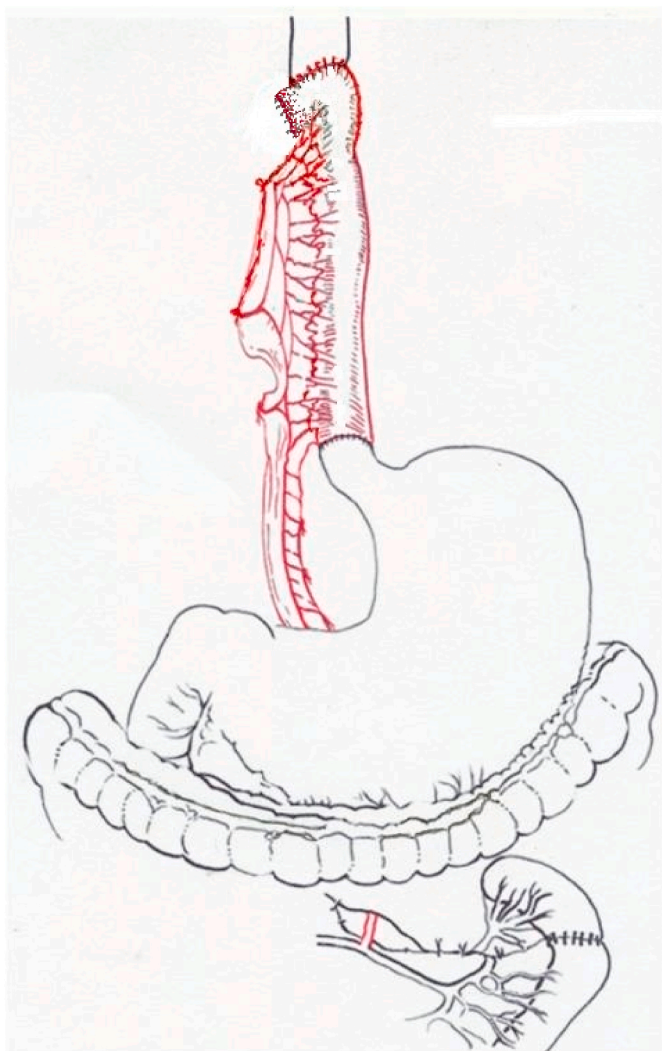


Fig. 2. Case 1 and 2. The jejunal loop is transposed in the right thorax cavity and anastomosed with upper and lower esophageal stump, respectively by end-to-side and end-to-end anastomosis.

2.3 Postoperative care. The post-operative course was uneventful in the three cases. Oral feeding started at days 8–11 following the surgery, after the contrast study that showed no leaks.

2.4 Long term follow-up. None of the three patients were readmitted to hospital after the esophageal reconstruction surgery: they were just subjected to routine check-ups at the outpatient clinic.

All clinical data were analyzed, as well as results from hematocemical, x-ray and instrumental investigations conducted along the years to observe the clinical evolution.

- 1) Esophageal atresia – case 1 (follow up 43 years): 29 years after the surgery an examination based on a barium meal test (Fig. 5) was performed, showing a good esophageal transit, with satisfactory peristalsis and no dilation of the jejunal neo-esophagus.

Currently, at year 43 after the surgery the patient is perfectly fine and, more specifically, has never complained with esophageal transit disturbances or drooling, halitosis, burping or any other sign of acid reflux.

This patient, having a weight of 57 kg and 159 cm tall, is now mother of two children of 13 and 9 years old: she went through a normal pregnancy and gave birth naturally.



Fig. 3. Case 3. An extensive esophageal stenosis caused by caustic agents starting from above one third of the thoracic esophagus until the gastroesophageal junction.

- 2) Esophageal atresia - case 2 (follow up 29 years): 10 years after the surgery a barium meal test examination (Fig. 6) was performed, showing a good esophageal transit, satisfactory peristalsis and no significant dilations of the jejunal tract.

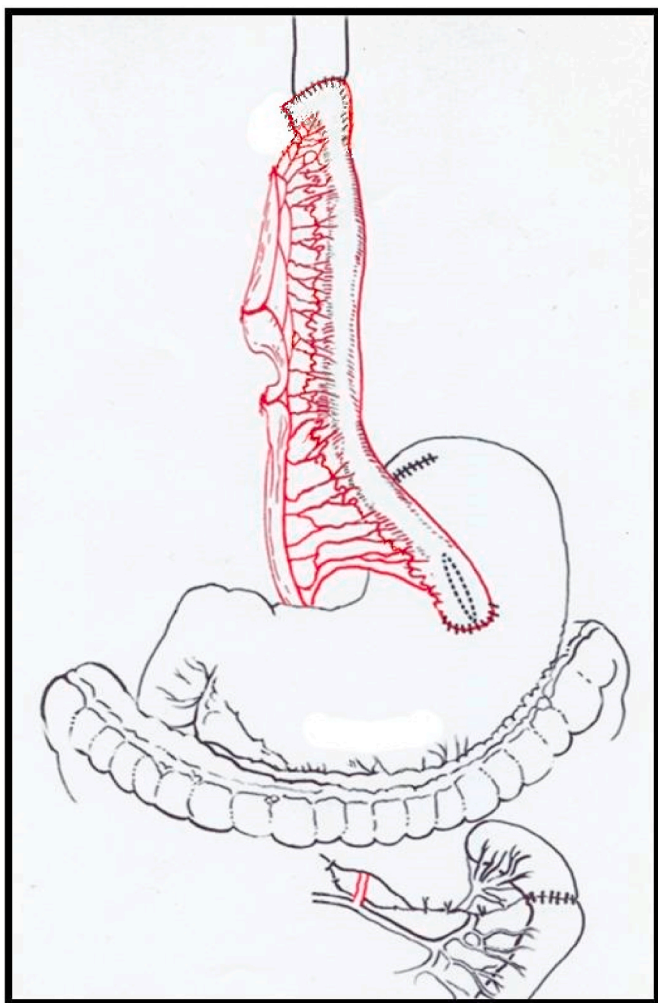


Fig. 4. Case 3. Esophageal stenosis: end-to-side anastomosis between the upper esophageal stump and the jejunal tract, and side-to-side anastomosis between distal jejunal loop with the anterior stomach wall.

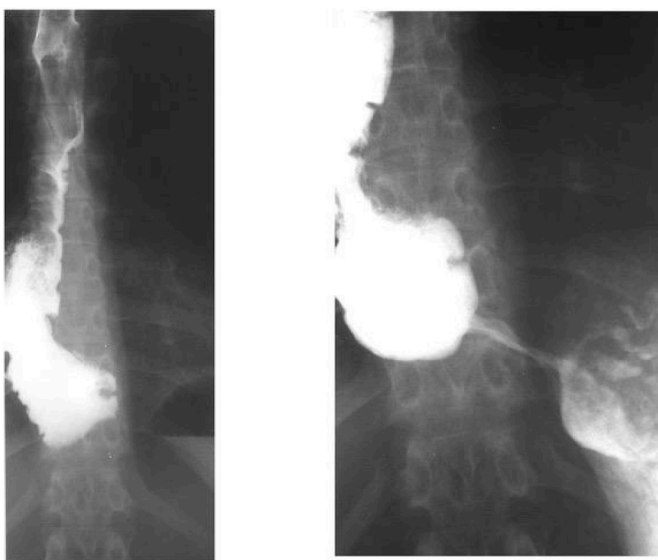


Fig. 5. Case 1. Esophageal atresia. Rx barium meal 29 years after the surgery: good esophageal transit and peristalsis, no dilation involving the neo-esophagus.

Currently, 29 years after the reconstruction, the patient is fine. The patient, 145 cm tall for 40 kg of weight, Down syndrome, was managed to achieve the secondary school diploma at Art School. She never complained with disturbances regarding the esophageal tract and no other disruptions related to the digestive system were observed.

3) Esophageal caustic stenosis – case 3 (follow up 21 years): 8 years after the surgery the patient was examined through a barium meal test: a good esophageal transit was shown (Fig. 7).

Nine years after the surgery a gastroscopy showed on the jejunal neo-esophagus a normal presentation of the mucous membrane from macroscopic and microscopic points of view both at thoracic and abdominal levels. No signs of peptic inflammation could be noticed involving the esophagus-stomach junction (Fig. 8).

Currently, 21 years after the esophageal reconstruction, the patient is still fine: no complaints of esophageal transit disturbances are reported. The patient is 177 cm, and weighs 88 Kg.

3. Discussion

Surgical management in newborns with long gap esophageal atresia is still controversial. Several different techniques are available, which can be divided roughly into 2 groups: surgical procedures utilizing only the esophagus and those that reconstruct the esophagus using another part of the digestive canal.

The first group of techniques are undoubtedly the best in theory, because it is acknowledged that there is no better esophagus than the patient's own also in long gap cases [4]. Recently, in long gap cases the technique of lengthening the esophageal upper pouch seems to lead to satisfactory results [5–7].

Regarding the esophageal stenosis secondary caustic lesions, the isolated short stenosis can be treated by dilations with good results. Longer and/or multiple stenosis, are in general hardly solved by dilations and require esophageal replacement [1].

The esophageal reconstruction technique based on an interposed and pedunculated jejunal loop has been extensively described by the Tohoku University Surgical School, Sendai, Japan, to treat esophageal tumors in adult patients [8,9] and in children with esophageal caustic stenosis [10]. Jejunum seemed to be a useful alternative method to restore enteric continuity as it has a diameter similar to the esophagus and an intrinsic peristaltic motility that facilitates esophageal transit lowering the risk of complications related to postoperative reflux.

Clinical reports on esophageal reconstruction using a pedunculated jejunal loop have been published since then. In adults surgical indications have been related to esophageal cancer with oncological limitations, or benign diseases [11–13], while in children indications were esophageal atresia and esophageal caustic stenosis [1,14–18].

Esophageal replacement can be extremely complicated when lesions involve the cervical esophagus as it occurs in adults due to larynx-tracheal tumors and in pediatric age in the case of esophageal atresia long gap in patients with cervical esophagostomy.

In some of these cases the esophagus was replaced as a free jejunal graft or through a “supercharged jejunal interposition technique” in which the jejunal loop was vascularized with an additional microsurgical vascular anastomosis to neck and thorax vessels [19–23].

Replacing the esophagus by using a pedunculated jejunal interposition technique is relatively more difficult and demanding than applying an esophagus-gastric or esophagus colonplasty.

However, these technical disadvantages are mitigated by the more efficient motility performance of the graft, as it has also been observed on our cases, based on a considerably long follow up.

The satisfactory jejunal peristalsis seems also to be the reason of the lack of peptic phlogosis in our case 3 (esophageal stenosis), being the one most at risk due to the exposure to gastric secretions.

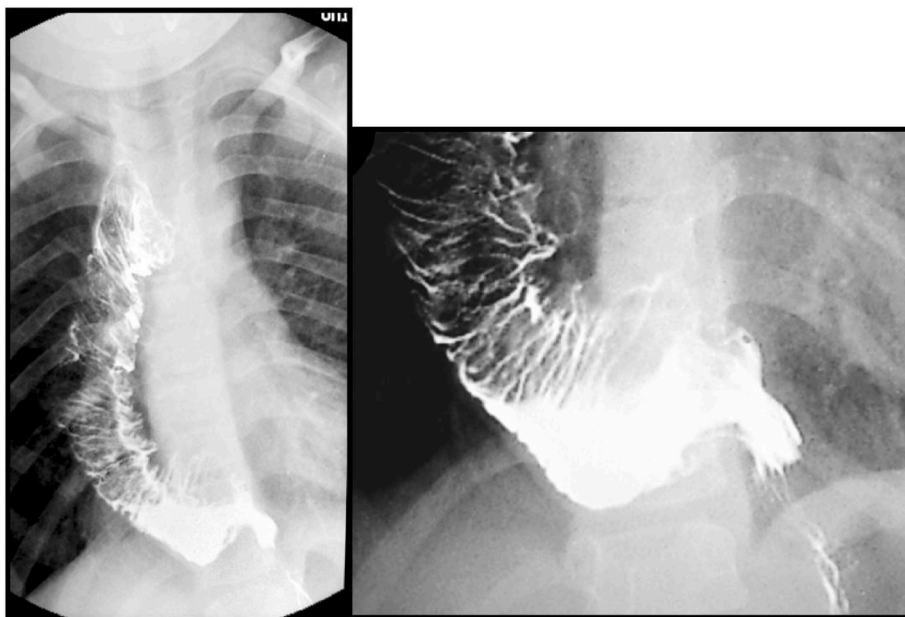


Fig. 6. Case 2. Esophageal atresia. Rx barium meal 10 years after the operation: good transit and peristalsis, no significant dilations involving the neo-esophagus.

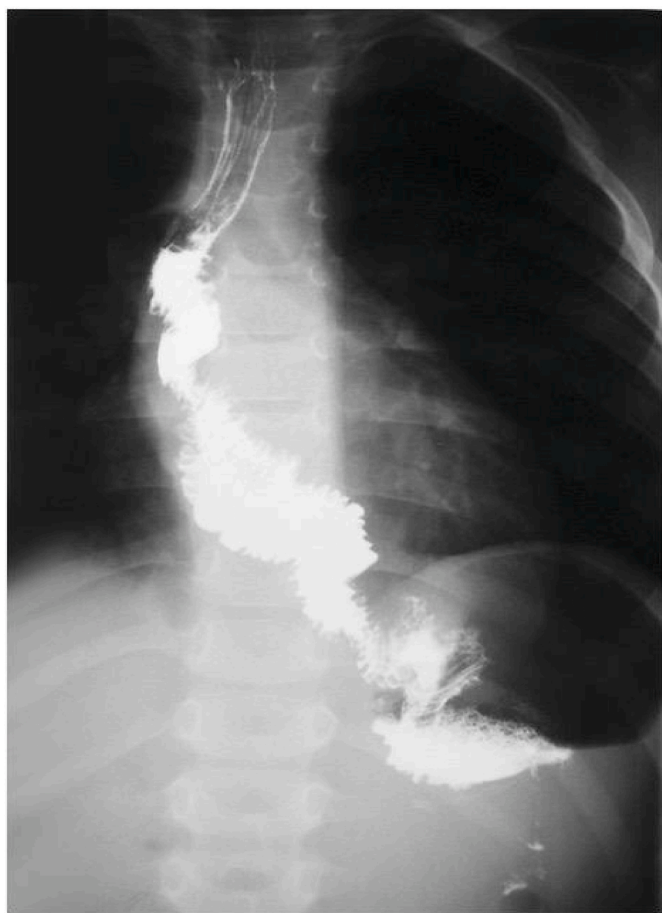


Fig. 7. Case 3. Esophageal stenosis. At a distance of 8 years following the surgery, the X-ray barium meal showed a good esophageal transit.

A lot of care should be taken at the moment of the esophageal stump isolations in the esophageal atresia, and during esophagectomy in esophageal stenosis patients, in order to avoid lesions to the posterior vagus nerve: its preservation is crucial to prevent gastric emptying disturbances.

For what concerns the technical level, problems can arise from the preparation of an adequate pedunculated jejunum with the right length in the case of anomalies involving the jejunum vascular arcade. In 20% of cases an interruption of the vascular arcade can be observed at the 2nd jejunal mesenteric vessel [24]. These are cases where it can be difficult to get a long vascular pedunculus, well efficient as it depends on the length of the next arcade.

To obtain a longer pedunculated jejunum, the anastomosis between the superior esophageal stump and the proximal jejunum loop should be end-to-side: this artifact makes it possible getting an extra length corresponding to the diameter of the transposed jejunum.

4. Conclusions

The use of the jejunum as a substitute for the esophagus is not very popular, due to the fact that the operation is believed to be cumbersome and time-consuming, and because of the necessity of discarding some jejunum length to create a sufficiently long vascular pedicle. However, the quality of motility function of this substitute can adequately match the very long life expectancy of children.

Though apparently healthy, our patients with congenital or acquired illness affecting the esophagus might be afflicted in the long term by significant morbidity. Chronic esophagitis and its consequences, namely gastric or intestinal metaplasia and cancer could be in some measure prevented by a jejunal substitute ensuring a good motility, however, such group of patients should be subjected to far sighted follow-up to collect information that are still warranted on the evolution in the long term.

In this limited experience, esophageal replacement by using the pedunculated jejunum showed to be extremely satisfactory as through a very long term follow-up (up to 43 years after surgery) no graft loss, neither mortality nor morbidity were observed.

These patients' quality of life has been absolutely satisfactory based, first of all, on subjective observation.

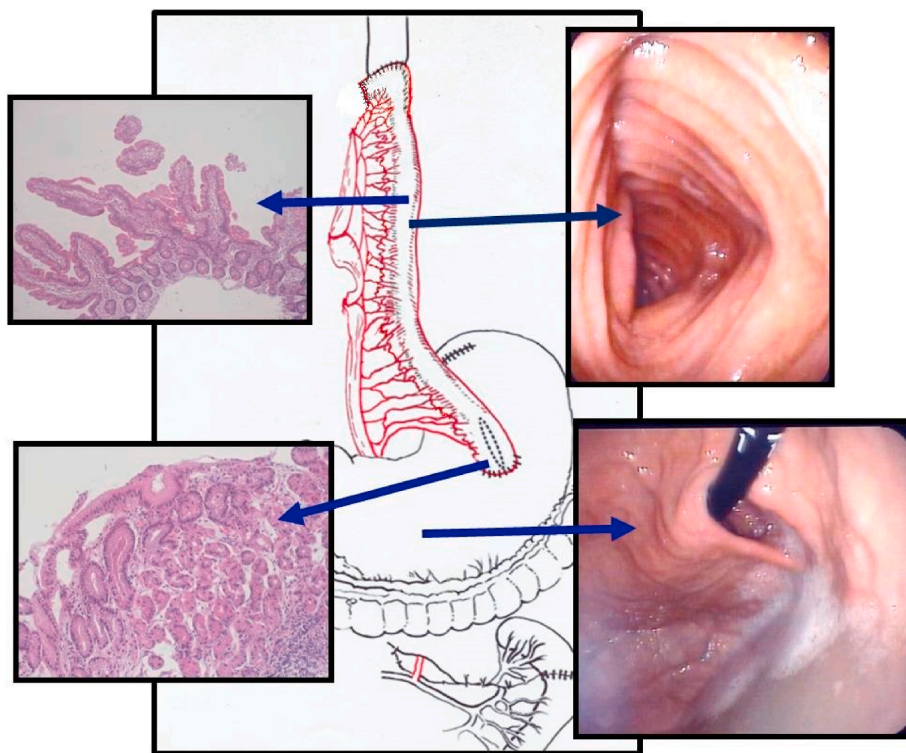


Fig. 8. Case 3. Esophageal stenosis. Nine years after the surgery, the patient was analyzed through esophageal gastroscopy that showed a jejunum neo-esophagus with a normal mucous membrane both in the thoracic and the esophagus-stomach junction. Mucosal biopsies conducted on two different tracts of the neo-esophagus did not show any inflammation.

On one hand, the lack of symptoms linked to esophageal reflux, slow gastric emptying and generic dyspepsia prevented us from being authorized to conduct instrumental investigations, on the other hand this has confirmed the safety and efficacy of such an intervention from the clinical point of view.

We believe that the pedunculated jejunum technique might be adopted as an appropriate method for esophageal substitution in infants and children as it can satisfactorily support esophageal functions free from complications for an extremely long time.

Patients consent

Consent to publish this case reports was not obtained. This report does not contain any personal information that could lead to the identification of the patients.case-report.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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