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RESEARCH ARTICLE

DIAGNOSTIC BRONCHOSCOPY FOR EVALUATION OF CONGENITAL STRIDOR:A CASE SERIES

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Abstract

Introduction: Stridor is a noise mechanically produced through partially occluded airway. Airway obstruction may be extrathoracic or intrathoracic. Stridor may be congenital or acquired. Timing in respiratory cycle determines anatomic location of lesion – inspiratory, biphasic, or expiratory. Gold standard for diagnosis is bronchoscopy which requires general anaesthesia in infants and small children. Major anaesthetic concerns are – possible difficult airway, sharing of an already compromised airway, airway oedema.

Case Description: 40 infants, 0 - 6 months age, with history of noisy breathing suggestive of congenital stridor, planned for diagnostic rigid bronchoscopy with or without therapeutic procedure, over one year period. Preoperative treatment – humidified oxygen, nebulization, dexamethasone, antibiotics, anti-reflux medication. Not premedicated, standard monitors applied. Induction of anaesthesia with inhalational oxygen and sevoflurane or intravenous propofol, fentanyl 1 mcg/kg, dexamethasone 0.5 mg/kg. Topical lidocaine 2% sprayed at vocal cords. 100% oxygen with propofol infusion for maintenance with spontaneous ventilation via nasopharyngeal airway. Patients requiring surgical intervention intubated using microcuffed endotracheal tube. Patients observed post-operatively. If ventilation was inadequate, intubated to control airway during recovery, extubated on restoration of spontaneous ventilation. After surgical intervention, babies shifted to ICU for elective ventilation for 48 hours.

Discussion: On bronchoscopy, laryngomalacia was the finding in majority of cases. Others had subglottic stenosis, tracheomalacia, vocal-cord paresis, laryngeal cyst. Out of 40 patients, 9 underwent therapeutic procedure and were electively ventilated, 26 resumed spontaneous breathing, 2 patients had delayed recovery and 2 had severe chest retractions and desaturations and they were managed accordingly. One baby aged 6 months diagnosed with grade III subglottic stenosis desaturatedand tracheostomy had to be done.

Conclusion: Anaesthesia for rigid diagnostic bronchoscopy is a significant challenge. Rigid bronchoscopy under general anaesthesia requires multidisciplinary approach and close cooperation between all team members.

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Introduction:-

Stridor is a noise mechanically produced through a partially occluded airway due to turbulent flow through narrowed lumen. Stridor is not a diagnosis, but a symptom of an underlying pathology[1]. Airway obstruction may be extrathoracic (nose, pharynx, larynx, trachea), or intrathoracic (tracheobronchial tree). Stridor may be congenital or acquired.Timing in respiratory cycle determines the anatomic location of airway lesion–Inspiratory: above the glottis, Biphasic: at the level of glottis or subglottis, Expiratory: lower trachea, tracheobronchial tree[2].

Causes of congenital stridor can be – congenital laryngomalacia, laryngotracheal stenosis(subglottic stenosis), vocal cord palsy, laryngeal cysts and webs, subglottic hemangiomas and papillomas, vascular compression of the airways[3]. Among these, congenital laryngomalacia is the most common cause (45% to 60%)[4]. One out of 10 infants have lesions in more than one anatomical site[2].

The gold standard for diagnosis of stridor is upper and lower airway endoscopy(diagnostic bronchoscopy) which requires general anaesthesia in infants and small children. Indications for diagnostic bronchoscopy are - severe stridor, progressive stridor, stridor with unusual features like; cyanotic attacks, apneic attacks, dysphagia, aspiration, failure to thrive, radiological abnormality, undue parental anxiety[2].

The anaesthetic management in these patients is particularly challenging. The major anaesthetic concerns during bronchoscopy for diagnosis and management of congenital stridor are – possibility of a difficult airway, sharing of an already compromised airway with the surgeon, airway instrumentation leading to airway oedema[5].

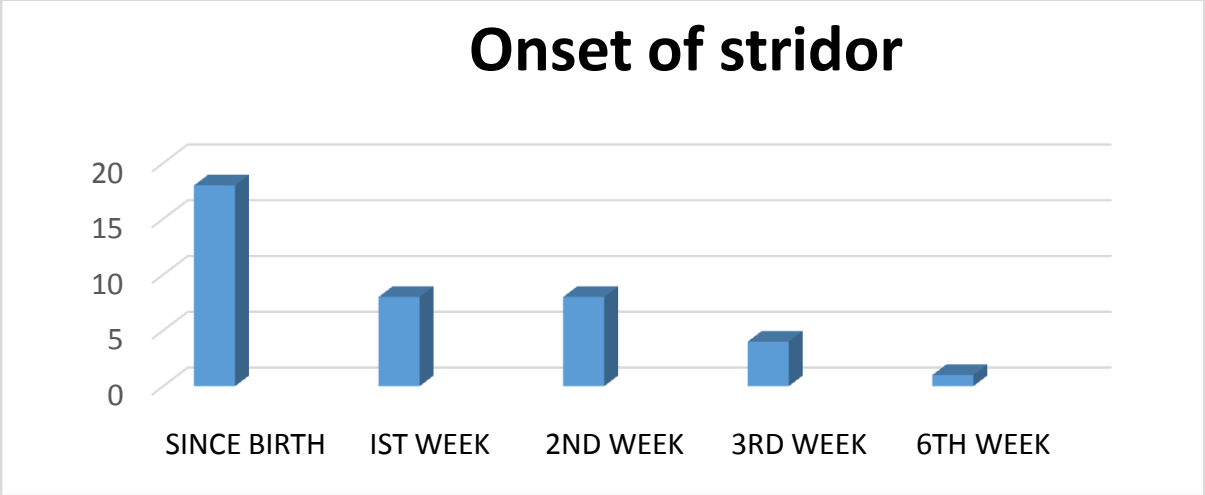
We are presenting a case series of the anaesthetic management of diagnostic bronchoscopy for evaluation of congenital stridor in infants from 0 to 6 months of age in our hospital during a period of one year.

Case Presentation:

We have taken 40 infants from 0 to 6 months of age at Indira Gandhi Institute of Child Health, Bangalore who presented with history of noisy breathing and other associated symptoms suggestive of congenital stridor, planned for diagnostic rigid bronchoscopy with or without a therapeutic procedure, for a period of one year, from January to December 2017.

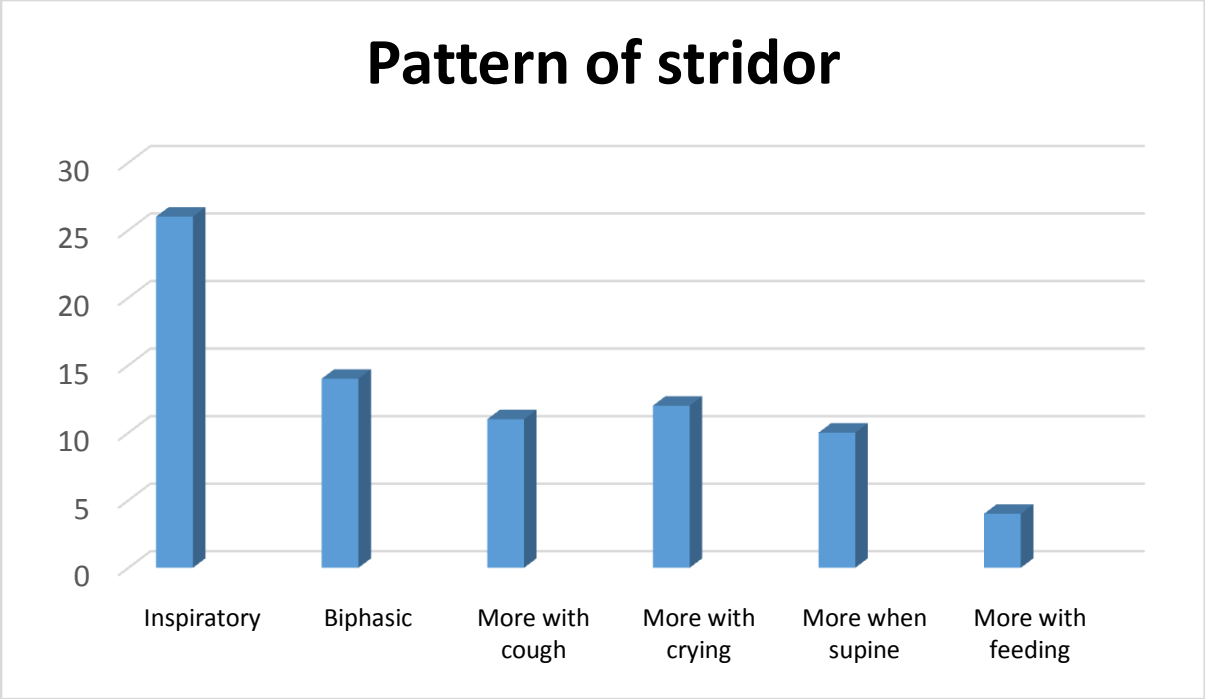
Demographic profile: Our study includes 40 infants, 0 to 6 months old, out of which 22 were male babies and 18 were female babies. 33 babies were born as full term, and the remaining 7 babies were preterm births.

Presenting signs and symptoms: All patients presented with stridor, most of them presented since birth (Figure 1).



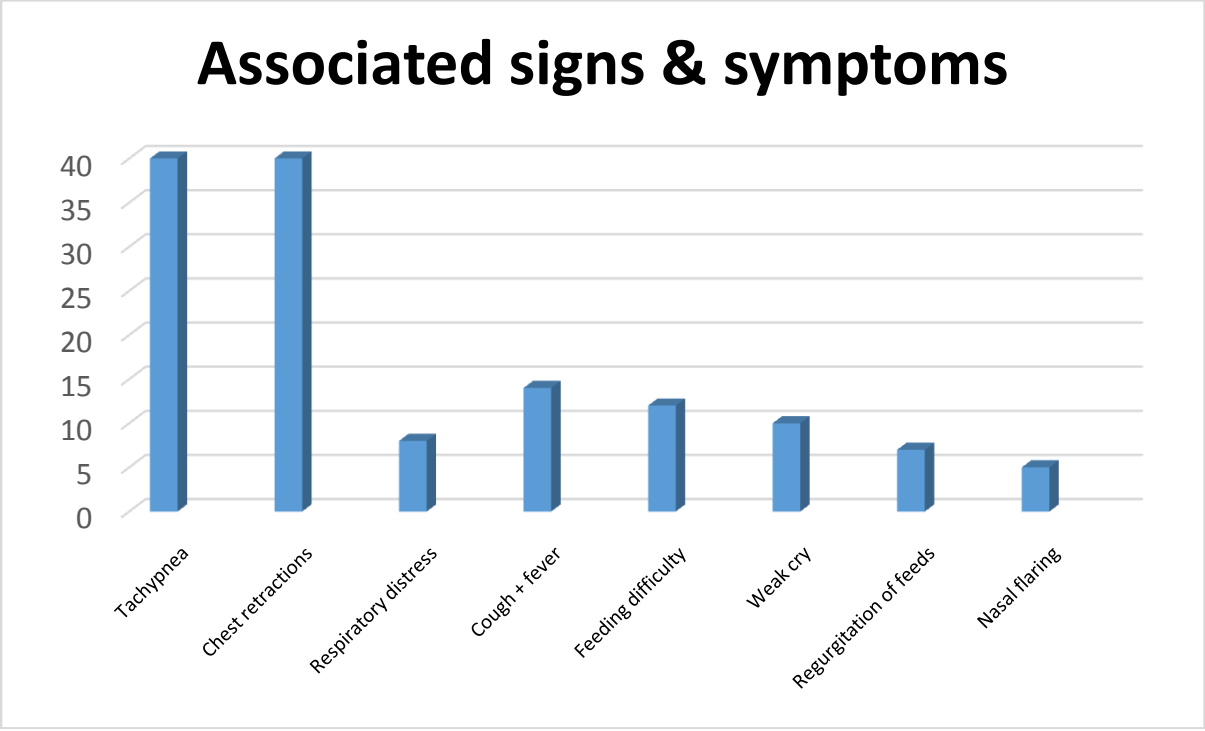
(Figure 1)

Pattern of stridor was inspiratory in majority of infants (Figure 2).



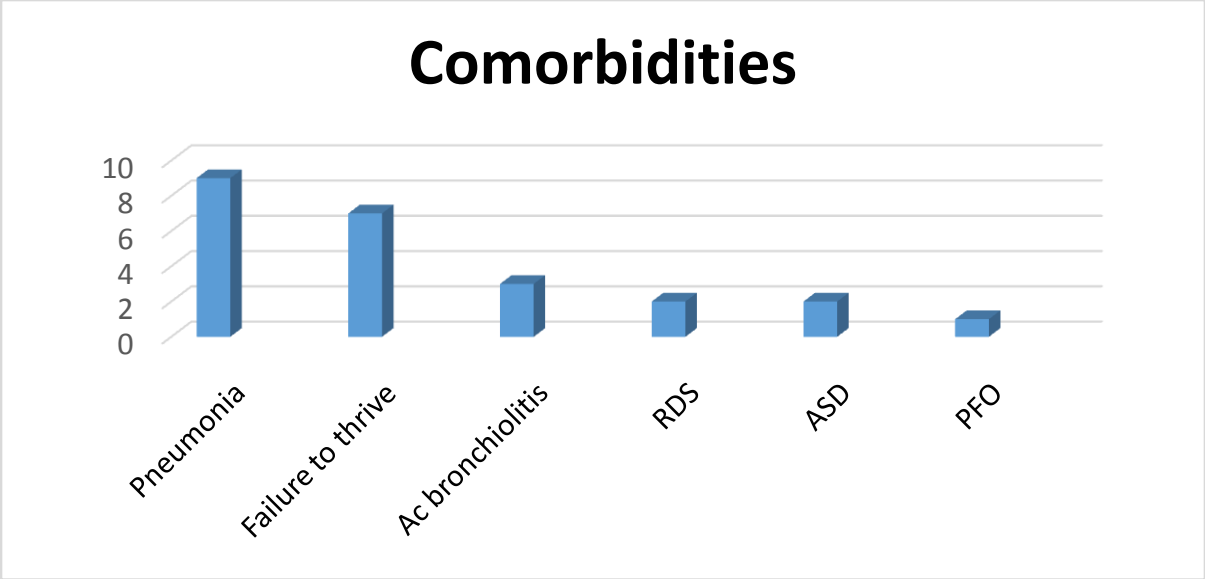
(Figure 2)

Associated signs and symptoms were tachypnea and chest retractions in all cases. Other associated signs and symptoms were respiratory distress, fever, cough, difficulty in feeding, weak cry, regurgitation of feeds, nasal flaring (Figure 3).



(Figure 3)

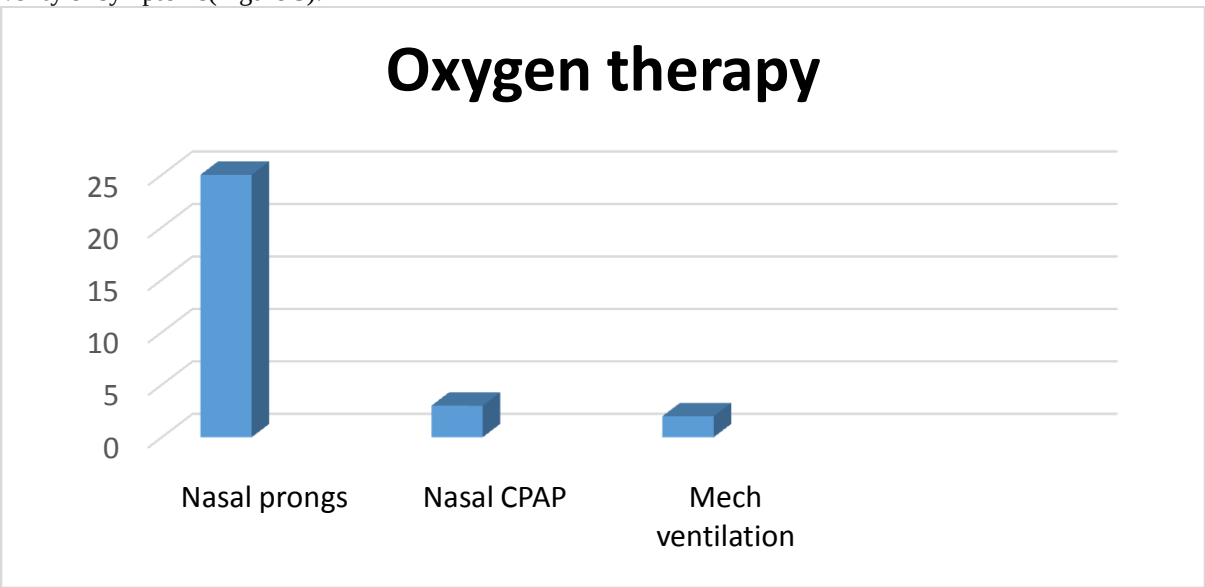
There were also associated comorbidities in many infants like; pneumonia, failure to thrive, acute bronchiolitis, respiratory distress syndrome, atrial septal defect, patent foramen ovale(Figure 4).



(Figure 4)

Management And Outcome:

Preoperative treatment received in all cases included – humidified air/oxygen therapy, nebulization with adrenaline(1:1000 0.5 ml/kg), systemic dexamethasone(0.1 mg/kg 8 hrly), intravenous antibiotics, anti-reflux medication. Oxygen therapy was given via nasal prongs, nasal CPAP or mechanical ventilation depending on the severity of symptoms(Figure 5).



(Figure 5)

A complete preoperative assessment was done in all the patients – a thorough history, complete physical examination, including SpO2 and FiO2, and imaging (chest X-ray and CT scan neck and thorax) was reviewed. All infants were taken as per the international fasting guidelines as 6 hours for milk and formula feeds, 4 hours for breast milk, and 2 hours for clear fluids.

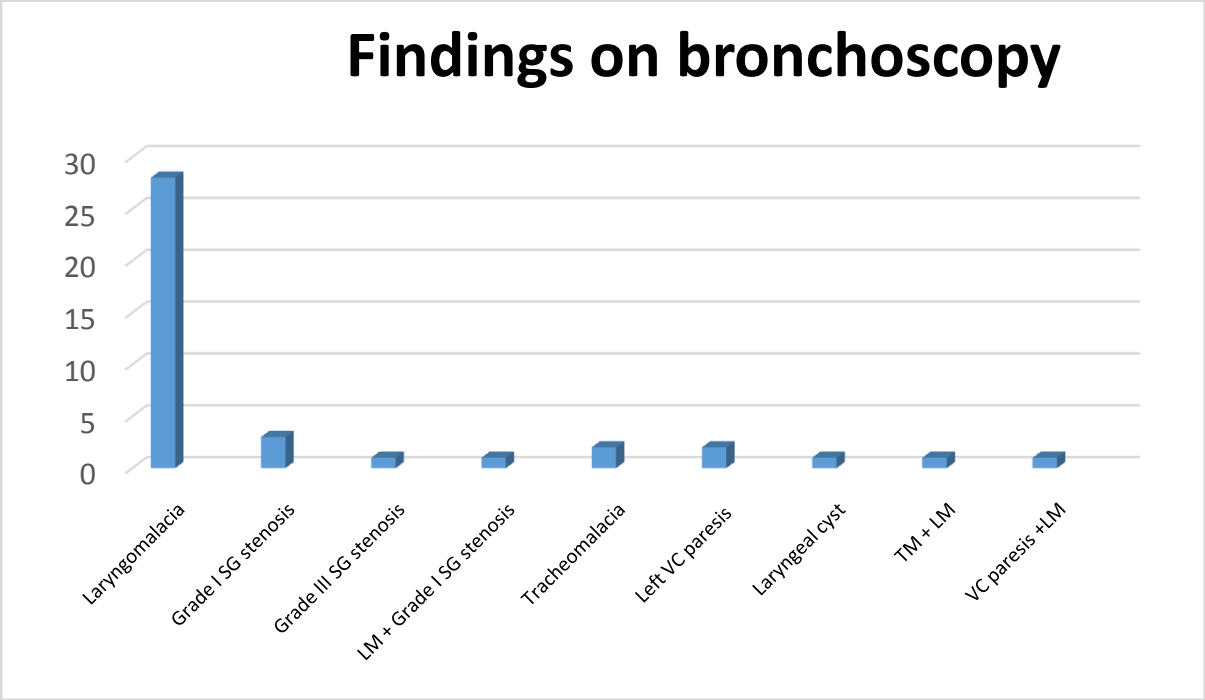
In the operating room, difficult airway cart was kept prepared as per the institutional protocol. No premedication was given to any of the patients, all standard monitors were applied - Pulse oximeter, Electrocardiogram, Non-invasive blood pressure, Temperature, Capnography.

Induction of anaesthesia was done with I.V. propofol 2 – 3 mg/kg b.w. in infants in whom I.V. line was already present, and with inhalational O2 with sevoflurane in those in whom I.V. line was not secured preoperatively. Fentanyl 1 mcg/kg was given for analgesia. Medications were slowly titrated for adequate anaesthesia, minimizing chances for laryngospasm, maintaining spontaneous ventilation.Dexamethasone 0.5 mg/kg I.V. was given to minimize airway edema. Topical lidocaine 2 % was sprayed at the vocal cords under direct laryngoscopic vision. 100% O2 with propofol infusion 50-100 mcg/kg/minute was used for maintenance of anaesthesia.

Diagnostic bronchoscopy was done using Hopkins optical telescope, which does not have an oxygenation port. All patients were maintained on spontaneous ventilation via nasopharyngeal airway of appropriate size connected to the breathing circuit.During bronchoscopy, saturation dropped to 90% in two infants and the surgeon was asked to stop and remove the endoscope, then mask ventilation with CPAP was done till saturation improved and the procedure was continued. Patients who required therapeutic/surgical intervention were intubated with smallest size 3.0 mm I.D.microcuffed endotracheal tube, and ventilation was switched over to inhalational – isoflurane for more easy titration. Neuromuscular blocking agent, atracurium was also used for maintenance.

After diagnostic evaluation, all babies were observed in the post operative period. If ventilation was adequate, babywas allowed to emerge breathing 100% oxygen via face mask.In case of inadequate ventilation, baby was intubated to control the airway during recovery and extubated only when regular spontaneous respiration was restored. All babies were shifted to ICU for observation.After surgical intervention, babies were shifted with ETT in situ to ICU for elective ventilation for 48 hrs.

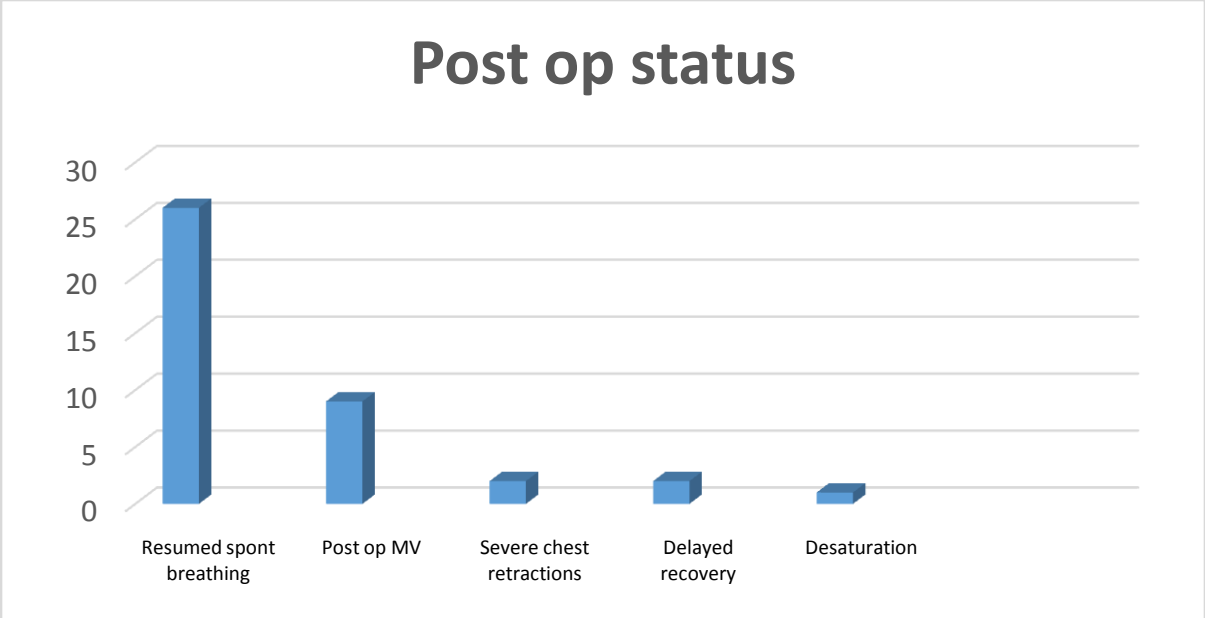
Findings on bronchoscopy were as – laryngomalacia in majority of cases i.e; 28 patients, 3 patients had grade I subglottic stenosis, 1 had grade III subglottic stenosis, 1 had laryngomalacia and grade I subglottic stenosis, 2 had left vocal cord paresis, 2 had tracheomalacia, 1 had laryngomalacia and tracheomalacia, 1 had vocal cord paresis and laryngomalacia, and 1 baby had laryngeal cyst(Figure 6).



(Figure 6)

In the immediate postoperative period (Figure 7) 26 patients resumed spontaneous breathing and were shifted to ICU for observation. 9 babies who were diagnosed as having severe laryngomalacia underwent surgical repair (aryepiglottoplasty and epiglottopexy) were shifted to ICU with endotracheal tube in situ and mechanically ventilated for 48 hours and then extubated in ICU uneventfully. 2 babies had severe chest retractions immediately in post operative period that was managed with CPAP ventilation via face mask and adrenaline nebulization (1:1000, 0.5 ml/kg), after which regular spontaneous respiration was restored. Both babies were monitored in the operating

room for some time and then shifted to ICU with oxygen inhalation. 2 babies had delayed recovery, and had to be intubated with 3 mm internal diameter uncuffed endotracheal tube and ventilation assisted till they recovered completely followed by extubation when complete recovery and regular spontaneous breathing started. One baby aged 6 months was diagnosed with grade III subglottic stenosis, and the decision for tracheostomy was taken. After diagnostic bronchoscopy, the baby immediately desaturatedupto SPO2 70%, positive pressure ventilation via facemask was immediately given till SPO2 improved, following which intubation was done with 2.5 mm ID tube as 3.0 mm ID tube could not be negotiated due to airway oedema. Then tracheostomy was performed by the ENT surgeon with 3.0 mm ID tracheostomy tube and the child was shifted to ICU after resuming spontaneous breathing.



(Figure 7)

One baby aged 26 days hadepiglottic cyst, partially covering the glottis. After confirmation of the finding it was decided to do a cystectomy. Intubation was done in left lateral position with 3.0 mm ID microcuffed tube taking care not to rupture the cyst. Post procedure the baby was extubatedand resumed spontaneous breathing.

Discussion:-

Rigid bronchoscopy under general anaesthesia requires a multidisciplinary approach and close cooperation between all the team members involved in the management, including anaesthetist, paediatrician, ENT surgeon and nursing staff. Pre-procedure assessment including history, physical examination and radiological examination is only a guide to the type and degree of pathology found during bronchoscopy. The type of anaesthesia and ventilation will be influenced by several factors including age, comorbidities, anaesthesiologist's comfort with an anaesthetic technique, purpose and goals for bronchoscopy. The approach to anaesthesia should be individualized to the patient and the preference of endocopist and anaesthesiologist[6].The anaesthetic plans and goals for surgery should be discussed with the ENT surgeon before-hand, emphasizing patient comorbidities including pulmonary function and potential for having a difficult airway.

Inhaled racemic epinephrine, heliox and steroids have been utilized to treat stridor preoperatively. Oxygen saturation should be monitored during treatment and arterial blood gas should be obtained to ensure that ventilation is adequate. This is important particularly if increased work of breathing is observed[4]. In our hospital we use adrenaline 1:1000 0.5 ml/kg for nebulization and systemic steroid dexamethasone I.V 0.1 to 0.5 mg/kg 8 hourly. Heliox is not available so we use humidified oxygen.

All children should be monitored with standard monitors. Continuous capnography is not always possible (particularly with Hopkin's telescope), so clinical observation of chest wall movement is important[5].Induction of anesthesia can be performed using an inhalational agent followed by I.V. placement or intravenous induction when

an I.V. line is already secured. Medications should be titrated slowly to adequately anaesthetize the patient while minimizing the chance for laryngospasm, while maintaining spontaneous ventilation. Bolus doses of additional medications or continuous infusions may be used to further blunt laryngotracheal reflexes and to ensure tolerance for the procedure[6]. Inhalational induction is the most commonly used technique and a volatile agent also has useful bronchodilatory effects. The induction may be prolonged due to the compromised airway, but with time and patience and modest amounts of CPAP, the airway can be maintained and the anaesthetic level deepened[7].

In our study most of the babies had I.V. line already secured and I.V. induction was done using slowly titrated doses of propofol. Only 5 babies came without an I.V. line and were induced with oxygen and sevoflurane and then I.V. line was secured.

Maintenance of spontaneous respiration throughout the technique maintains a degree of muscle tone and this helps enormously to maintain gas exchange allowing the anaesthetist time. This enables the surgeon to perform a diagnostic bronchoscopy assessing both pathology and any dynamic obstruction before any required operative surgery[7].Topically treating the vocal cords and subglottic airway with local anaesthetic (3 – 5 mg/kg lidocaine[7]) decreases the incidence of coughing or bucking during instrumentation, and allows the child to tolerate lighter levels of anaesthesia[5]. Although this helps the child tolerate the procedure, it may interfere with assessment of normal vocal cord movement. For that reason, a local anaesthetic is often topically applied after initial evaluation of the upper airway and before insertion of bronchoscope for evaluation of the distal airway[8].

In our study, after induction initially direct laryngoscopy was performed when the baby was spontaneously breathing to assess the movement of vocal cords, and then vocal cords and subglottis were sprayed with 2% lidocaine taking care not to exceed the dose of 3-5 mg/kg.

Possible ventilation strategies can be - spontaneous without endotracheal intubation (nasopharyngeal airway connected to breathing circuit/ETT placed in hypopharynx), insufflation, intermittent apnoeic ventilation, jet ventilation[6].Of the above choices, the use of a nasopharyngeal airway appears to be the most suitable[9]. In our study, since the babies were only upto 6 months of age the smallest size Hopkin's optical telescope was used for diagnostic bronchoscopy, which does not have an oxygenation port. So spontaneous ventilation was maintained via nasopharyngeal airway of appropriate size connected to the breathing circuit. The size of nasopharyngeal airway was measured from tip of nose to angle of mandible.

Spontaneous ventilation without an ETT allows an unobstructed view of the larynx[9]. Children must be spontaneously breathing so that vocal cords move freely. After movement of the vocal cords is observed and recorded, the anaesthetic level can be deepened as appropriate, a rigid bronchoscope (or just the telescope in small infants) is inserted through the vocal cords, and the subglottic area, the lower trachea, and the bronchi are evaluated. Goals of anaesthesia for bronchoscopy include – appropriate analgesia, an unconscious child, a quite surgical field. Coughing, bucking or straining during instrumentation during a rigid bronchoscopy may cause difficulty for the surgeon and damage the child's airway[5]. We took an utmost care to maintain appropriate depth of anaesthesia while maintaining spontaneous breathing by titrating the propofol infusion intraoperatively.

Although more than normal CO2 tensions are inevitable during the procedure, they are generally well tolerated. However, hypoxia is not well tolerated and the procedure should be stopped while the child is oxygenated. Because ventilation is intermittent and at times suboptimal, it is recommended to use 100% oxygen and high fresh gas flow rate during bronchoscopic examination, keeping in mind that the anaesthetist does not have complete control of the airway. Care must be taken to select a bronchoscope of proper external diameter to avoid damage to the laryngeal structures[5]. In our study, the smallest size endoscope was used and for maintenance 100% oxygen was used. In two babies when during the procedure oxygen saturation dropped to 90%, we asked the surgeon to stop and remove the endoscope and took control of the airway with CPAP through facemask till SPO2 picked up and the procedure was then continued. No other babies had any desaturation during the procedure.

After surgery to the airway in a small infant, close observation is essential to pick up worsening airway obstruction due to either swelling or retained secretions. Nebulized epinephrine (1:1000, 0.5 ml/kg) can be used after operation to improve the airway swelling. If the airway is not adequate, the baby should be intubated and ventilated for a short period and extubation attempted later. The tracheal tube size should be carefully chosen to avoid additional swelling of the airway. Extubation may take place in the operating theatre after the baby starts breathing adequately[7].If

ventilation is adequate and the anaesthetic depth is not excessive, the child can be allowed to emerge breathing oxygen via a face mask[5].

In our study, majority of patients resumed regular spontaneous breathing. Surgical repair for laryngomalacia was done in 9 patients after intubation, and were shifted to ICU with endotracheal tube in situ for elective ventilation for 48 hours and were extubated uneventfully in ICU. 2 of the babies who had delayed recovery were intubated post procedure and ventilation assisted till complete recovery, followed by extubation and shifting to ICU. 2 patients had severe chest retractions and were managed with CPAP via face mask and adrenaline nebulization. A baby diagnosed as having grade III subglottic stenosis on bronchoscopy had to undergo tracheostomy post bronchoscopy, desaturated after removing the bronchoscope and was managed with mask ventilation, then intubated with 2.5 mm ID endotracheal tube. Then tracheostomy was performed with 3.0 mm I.D. tracheostomy tube and shifted to ICU afer resuming spontaneous breathing. The reason for desaturation was severe airway edema that had further narrowed the subglottic stenosis.

Congenital laryngeal cysts are rare, with an incidence of 1.82 per 100,000 live births. They arise from the glottic area (58.2%), ventricular fold (18.3%), vallecula (10.5%), epiglottis (10.1%), and the aryepiglottic fold, as an order of frequency. A congenital laryngeal cyst may easily obstruct the smaller airway of a neonate. During anesthesia for patients with laryngeal cysts, anesthesiolosists can face the risk of obscured views of the larynx, loss of the airway, risk of rupturing the cyst, and potential aspiration of cyst contents[10]. In our study we had one case of epiglottic cyst partially covering the glottis. Once the diagnosis was confirmed and decision to do cystectomy was taken, the baby was intubated with smallest size microcuffedendotracheal tube in left lateral position being very careful not to rupture the cyst. After the procedure was over, the baby was extubated uneventfully and shifted to ICU for observation.

Conclusion:-

Anaesthesia for rigid diagnostic bronchoscopy is a significant challenge.Anaesthetic plan and goals must be discussed before-hand with the team members.Careful vigilance should be maintained throughout the procedure, especially during recovery.So, in order to face this challenge it is important to be prepared beforehand and act promptly in case of an untoward event.

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